

Lingual Thyroid

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Abstract: Ectopic thyroid gland is characterized by the presence of thyroid tissue outside its normal position resulting from a defect of the thyroid diverticulum migration from the base of the tongue until its final pre-tracheal position. The incidence of lingual thyroid is very less which varies from 1:3000 and 1:10,000. Lingual thyroid may sometimes present with symptoms of respiratory obstruction or feeding difficulties. It may lead to serious problem ranging from asymptomatic swelling to lethal condition. The authors present a 40-years-old man with an ectopic thyroid gland in the base of the tongue, found accidentally during the laryngeal intubation.

Keywords: Ectopic gland, Lingual thyroid, Lingual thyroid nodule, Thyroglossal duct

I. Introduction

The Thyroid gland (Greek term, thyroid means “Shield”) constitutes an important regulatory endocrine gland for the overall functioning of the body. This gland was first described by Thomas Wharton [1], 1616-1673, of England as the gland that wraps around the trachea (and thus acts as a shield) and the secretions of the gland are considered to play an important role in regulating the normal body activities. It's the first endocrine organ to develop at the 24th week of gestation from the endodermal germ layer. The aberrant position of the thyroid gland due to fail to follow its normal pathway with or without incomplete obliteration of the vertical tract and thus the term Ectopic thyroid is used. Ectopic thyroid was first described by Hickman in 1869 in a newborn [2]. It is the most frequent form of thyroid dysgenesis, although the true incidence is probably underestimated accounting for 48-61% of the cases [3]. Its prevalence varies between 1:100000 and 1:300000 and its clinical incidence is reported to range from 1:4000 to 1:10000 [4].

As the presence of ectopic thyroid can be anywhere along its path of descent, there are various sites where their presence can be reported. The locations have been mentioned in (Table 1). Other sites where ectopic thyroid may develop includes pituitary gland [11], palatine tonsils [12], carotid bifurcation [13], axilla [14], iris of the eye [15] and lateral cervical regions [16]. Lingual thyroid being the most common variant of ectopic thyroid [17] occurring most commonly at the level of foramen caecum in 90% of the reported cases [18]. Here we present a case report of a 40yr old male patient with presence of lingual thyroid nodule which was an accidental finding.

Table 1: This table illustrates on the various locations of ectopic thyroid and the associated clinical features

LOCATION	CLINICAL FEATURES
lingual	Usually seen in midline in the base of the tongue as a nodular mass. The surface of the lesion is usually smooth and vascularity can be seen [5].
Submandibular	nodular, non-tender, immovable pink mass with smooth surface located in the floor of the mouth [6]
Thyroglossal duct cyst	small midline anterior neck mass [7]
Intrathoracic (mediastinal, lung, and heart)	Usually asymptomatic with the tumor reported as incidental finding on chest radiograph. Sometimes they may present with respiratory symptoms [8]
Intratracheal	Usually asymptomatic with respiratory disorders, symptoms similar to bronchial asthma [9]
Adrenal glands	Asymptomatic (usually an accidental finding)
Duodenum, pancreas and intestine	Asymptomatic (usually an accidental finding)
Dual ectopy (usually lingual and infrahyoid)	Usually the patients complain of a palpable mass, growth retardation and lump sensation in the throat. Patients may have dyspnea, stridor or cough [10]

II. Case Report

A 40 year old male patient presented with a complaint of pain and swelling with right third molar region and complaints of difficulty in breathing and swallowing for 10 years that increased progressively (Fig.1). The medical history was insignificant except for a tonsillectomy 2 years previously. Physical examination revealed a 2 x 3 x 3 cm fixed, ovoid smooth surfaced pink firm mass that was covered with normal mucosa in the base of the tongue.



III. Discussion

The Thyroid gland is the first endocrine organ to develop at the 24th week of gestation from the endodermal germ layer. Various genes are responsible for the prime regulation of endoderm formation at gastrulation and are responsible for the development and descent of the thyroid gland as well. These are the NODAL factors (members of transforming growth factor b family) and a highly structured network of components including the transcription factors FOXA2, SOX17, and GATA4–6[19]. As the development precedes this endoderm gets rapidly remodelled into the germ tube, a kind of amorphous tube with no anatomical differentiation. Subsequently several morphogenetic events take place that leads to the adult gut formation. This is again done by discrete patterns of gene expression. For example, the foregut identity is closely linked to the expression of HHEX, FOXA2, and SOX2 genes [19]. Thyroid gland precursors arise at the level of the mid–hindbrain boundary closely opposed to the anterior lateral plate of mesoderm from which the heart develops[20]. The thyroid anlage is the first part to develop during organogenesis of thyroid gland. It appears at 20-22 embryonic days and consists of endodermal cells. It is later responsible for development of Thyroid follicular cells, while, the C cells arise from the ultimobranchial bodies [3].(Table 2) shows the embryonic days around which the various stages of thyroid development occurs [21]. At around 24th day the endodermal cells of the thyroid anlage form the thyroid bud by proliferation and invades the surrounding mesenchyme. The thyroid primordium subsequently develops a narrow neck that later forms the diverticulum. There is development of foramen caecum (remnant of thyroid anlage) in the form of small hole in the pharyngeal floor which is in turn connected with the migrating thyroid primordium by a narrow thyroglossal duct. The thyroglossal duct disappears between 30-40 days leading to disconnection between the thyroid primordium and the pharyngeal floor. The thyroid primordium starts growing laterally to form the lobes and the isthmus. About 5 days after, the thyroid primordium starts migrating down to reach the trachea. This final location of the thyroid gland which is distinct from its site of development suggests that the morphogenetic process involves profound cell migration. This migration could be an active or a passive movement.

Table 2 : Stages of morphogenesis of thyroid gland

Stages of morphogenesis	Embryonic Day (Human)
Thyroid anlage appears	20 – 22
Thyroid bud migration begins	24
Thyroglossal duct disappears	30-40
Thyroid migration is complete	45-50
Fusion with ultimobranchial bodies	60
Onset of folliculogenesis	70

Some studies have shown that the thyroid follicular cells are themselves responsible for the migration of the primordium [22], thus giving the idea of active migration. While others have proposed that passive movement of the cells is secondary to some factor. These factors could belong to Cadherins family. E-Cadherin is responsible for maintaining the epithelial nature of the cells and its loss leads to the conversion of epithelial cells into mesenchymal cells. Studies have shown that E-cadherin expression remains high throughout the process of thyroid gland development, thus indicating the epithelial differentiation of the thyroid cells[23]. The cells might therefore not be actively motile, but may rather reach their destination by passive translocation along with adjacent tissues. Various genes are responsible for organogenesis at early and late stages of thyroid development. The genes working in early period includes the transcription factors TITF-1(Nkx2-1), Foxe1 (TITF-2) and PAX-8 are essential for thyroid morphogenesis and differentiation. Mutation in these genes may be involved in abnormal migration of the thyroid[24]. The homeobox transcription factor HHEX has also been known to have an important role in early thyroid development[25]. Any mutation in these genes leads to the aberrant position of the thyroid gland due to fail to follow its normal pathway with or without incomplete obliteration of the vertical tract and thus the term Ectopic thyroid is used.

Its prevalence is about 1 per 100000–300000 people without the thyroid disorders but in cases with thyroid disease it rises to 1 per 4000–8000 patients [26]. Nonetheless, in autopsy studies, its prevalence ranges from 7 to 10% [27]. More than 440 cases have been reported till date. In 70–90% of cases, it is the only functional thyroid tissue present [6]. Sex predilection has also been noted. Females being more commonly affected than males (4:1) [4]. This is especially evident in populations of Asian origin [28]. It may occur at any age ranging from 5 months to 40 years, but it is most common in younger age group[17].

Lingual thyroid being the most common variant of ectopic thyroid [17] occurring most commonly at the level of foramen caecum in 90% of the reported cases [18]. The mean age of presentation is about 40.5 years and it ranges from birth to 83 years. In 70–75% of cases, lingual thyroid is the only thyroid tissue present [29]. The lingual thyroid may remain asymptomatic but may become symptomatic during increased demand of the hormones by the body. The patient may present with symptoms like dyspnoea, dysphonia, snoring, sleep apnoea and sensation of foreign body. Sometimes the mass may get ulcerated leading to haemorrhage and haemoptysis (17). Table[3] enlist the reported cases of ectopic thyroid gland.

Table 3: Review of cases of ectopic thyroid gland reported

Age	Sex	Presenting features/complaint	Location	Authors and year
32 years	female	Change in voice, difficulty in swallowing, foreign body sensation in throat	Throat	Mattakarottu Joseph Jacob, Mudalsha Ravina 2012 [30]
5 years	female	Asymptomatic swelling	Neck region	A. sood ,RK Seam et al.2011[10]
17 years	female	Asymptomatic Swelling at base of tongue that grew in size	Base of tongue	Bipul Kumar Choudhury, Uma Kaimal Saikia et al.2011[2]
15 years	Female	Accidental finding	Lingual and infrahyoid areas	Baik SH et al. 2002[31]
<2 years	female	Speech delay	Sublingual thyroid	Natasha Heather, Kate Hall et al. 2012[32]
80 years	Female	Incidental finding	Mediastinal mass	A. D.Mace, A. Taghi, et al.2011[33]
33 years	female	Progressive dyspnea	Intratracheal mass	Yon Mi Sung 2008[34]
14 years	male	Lump in throat, dysphagia, dysphonia	throat	Grant E. Ward, James R. Cantrell, and Warde B. Allan. 1954 [35]
17 years	female	Lingual mass, slight change in voice	Tongue	Grant E. Ward, James R. Cantrell, and Warde B. Allan. 1954 [35]
29 years	female	Difficulty in swallowing	Submandibular mass	Faramarz Babazade, Hamed Mortazavi, Hamed Jalalian

				and Ehsan Shahvali 2009[6]
16 years	female	Swelling of neck	Base of tongue, suprahyoid and hyoid level	Sujit Nilegaonkar, Chetna Naik, Sameer Sonar, and Deepti Hirawe 2011 [36]
19 years	female	Frequent attacks of sore throat, occasional feeling of lump in throat	Posterior part of tongue	J. F. Jarvis 1969 [37]
24 years	female	Dysphasia of 1 yr duration, some voice changes	Lingual swelling in hyoid area	J. H. Louw. 1969 [37]
19 years	female	Asymptomatic, accidental finding	Base of tongue	J. F. Jarvis, 1969 [37]
3 years	male	ENT examination for hearing problem	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
5 years	Female	Parents noted "tongue tumour" dyspnoea with exertion and lying supine	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
8 years	female	Failure to grow	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
18 years	Female	"something in throat" thick voice	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
28 years	female	"hoarseness of voice", ache in throat, hemorrhages from tongue	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
44 years	female	"Irritation" at base of tongue	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
44 years	male	Accidental finding during general examination for carcinoma of testis	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
58 years	female	"Hoarseness" of voice since 10 months, bilateral ear pain, right > left	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]
65 years	female	Mild pain in throat since 3 months, "hollow voice" for many years, left ear ache since 3 months	Lingual thyroid	Fredrick W. Neinas et al. 1973 [29]

3.1 Complications

Ectopic thyroid at unusual sites leads to its late diagnosis and thus may be source of various complications ranging from clinically evident thyroid dysfunction. This could be either hypothyroidism [38] or hyperthyroidism [30] to injudicious excision of the mass considering it as a tumour leading to lethal conditions. Other complications include dysphagia [4], dyspnoea [4], stridor [39], dysphonia [40], hemoptysis, chronic cough and development of goitrous mass [41]. Rarely, but benign [34] or malignant [42] neoplastic conditions have been reported to develop in the lingual thyroid nodules. Cases with development of Hashimoto's thyroiditis in ectopic thyroid have also been reported. In addition to all these effects, generalised growth and retardation in children have also been reported in a case report by Heather et. al [32].

3.2 Diagnosis

Various investigations can be carried out to diagnose ectopic thyroid. These are colour Doppler ultrasonography, CT scan, MRI and nuclear medicine scintigraphy along with the various other pathological investigations like T3, T4, TSH (Thyroid stimulating hormone) etc. Colour Doppler sonography permits the assessment of blood flow in addition to the depiction of morphology in thyroid imaging. It assesses the blood flow and morphology in the detection of ectopic thyroid [43]. Ultrasonography is a readily accessible, inexpensive, and radiation-free imaging method commonly used to evaluate the thyroid gland. High-frequency (10–12 MHz) US is the least invasive. It also helps in distinguishing between benign and malignant nodules of the gland. The CT scan provides us with a cross-sectional view of the gland, adding to the information about the architecture of the gland. It is also helpful in evaluating the dissemination of the malignancy if present. The main disadvantage of CT is that it requires the administration of an iodinated contrast agent to differentiate the lymph node. In addition, imaging with iodine-based nuclear medicine agents cannot be performed until six weeks after CT, because of the uptake of the iodinated contrast agent by the thyroid gland. MRI is not commonly used in daily practice because susceptibility artifacts can occur on fat suppression sequences in the anterior aspect of the neck. However, the use of eight-channel surface coils for high-resolution MRI can allow adequate evaluation of the thyroid gland and entire neck. Scintigraphic imaging using iodine-131 (I-131) and/or technetium-99 (Tc-99) provides highly sensitive image in patients with ectopic thyroid tissue and scintigraphic imaging modalities offer the possibility of whole-body evaluations [44]. Fine needle aspiration cytology (FNAC) also provides support in confirming the diagnosis of ectopic thyroid. It may be used to differentiate between a benign and a malignant lesion.

3.3 Management

There is no consensus about the optimal therapeutic strategy this might be due to the rarity of this clinical condition. The notion of surgical treatment of the ectopic thyroid gland has gained much attention by

many authors. This is based on the location of the ectopic gland, its size, patient's age, thyroid status of the patient and the possible complications that might occur during or after the surgical intervention [45]. The cases which are asymptomatic and euthyroid, regular follow-up is advised [46]. For mild symptoms and hypothyroid states, levothyroxine replacement therapy may be effective, leading to considerable mass reduction [47].

IV. Conclusion

Lingual thyroid is a rare anomaly signifying faulty migration of normal thyroid gland. The exact pathogenesis of this ectopic is not known. It is more prevalent in females than in males. It may present with various symptoms like dysphagia, dysphonia etc. or it may be asymptomatic and could be detected accidentally. Investigation include thyroid function tests, neck ultrasound scan, Technetium scanning and C.T scan. Thus, it is important from dental surgeon's point of view to consider lingual thyroid nodules as one of the differential diagnosis of swellings at base of tongue, although its occurrence is rare.

References

- [1]. Chaudhary, Concise medical physiology(New central book agency (P) LTD.Calcutta; 2006).
- [2]. Choudhury BK, Saikia UK, Sarma D, Saikia M, Choudhury SD, Barua S, and Dewri S, Dual Ectopic Thyroid with Normally Located Thyroid: A Case Report.J Thyroid Res.2011, 2011, 159703.
- [3]. Felice MD, Lauro RD, Thyroid Development and its disorders: Genetic and molecular mechanisms, Endocrine Reviews, 25, 2004, 722-746.
- [4]. Toso A, Colombani F, Averono G, Aluffi P, Pia F, Lingual thyroid causing dysphagia and dysphonia. Acta Otorhinolaryngologica Italica, 29, 2009, 213-217.
- [5]. PS Douglas, AW Baker. Lingual thyroid, Br J Oral Maxillofac Surg, 32, 1994, 123-124.
- [6]. Babazade F, Mortazavi H, Jalalian H, Shahvali E, Thyroid tissue as a submandibular mass: a case report, Journal of Oral Science,51, 2009, 655-657.
- [7]. Shin AY, Lee SH,Jung WS, Ko SH, Ahn YB. Ectopic Thyroid Nodule in Thyroglossal Duct,Korean J Intern Med, 26, 2011, 218-219.
- [8]. Pilavaki M, Kostopoulos G, Asimaki A,Papachristodoulou A, Papaemanouil S, Palladas P, Imaging of ectopic intrathoracic multinodular goiter with pathologic correlation: a case report,Cases J.,2, 2009, 8554.
- [9]. Serraj M, Ooadnoui Y, Lakranbi M, Ghalimi J, Boubou M, Tizniti S, Smahi M,Intratracheal Ectopic Thyroid Tissue, The Annals of Thoracic Surgery, 95, 2013, e13-e14.
- [10]. Sood RK. Gupta SM,Sharma DR, Bhardwaj P. Dual Ectopic Thyroid: A Case Report with Review of Literature, Iran J Radiol, 8,2011, 20-32.
- [11]. Malone Q, Conn J, Gonzales M, Kaye A, Coleman P. Ectopic pituitary fossa thyroid tissue. J Clin Neurosci,4,1997, 360-363.
- [12]. Sarin YK, Sharma AK, Ectopic tonsillar thyroid, Indian Pediatr, 30, 1993, 1461-1462.
- [13]. Rubinfeld S, Joseph UA, Schwartz MR, Weber SC, Jhingran SG, Ectopic thyroid in the right carotid triangle,Arch Otolaryngol Head Neck Surg, 114, 1988, 913-915.
- [14]. Kuffner HA, McCook BM, Swaminatha R, Myers EN, Hunt JL, Controversial Ectopic Thyroid: A Case Report of Thyroid Tissue in the Axilla and Benign Total Thyroidectomy,Thyroid, 15,2005, 1095-1097.
- [15]. Tiberti A, Damato B, Hiscott P, Vora J. Iris Ectopic Thyroid Tissue: Report of a Case, Arch Ophthalmol, 124,2006, 1497-1500.
- [16]. Choi JY, Kim JH,Case of an Ectopic Thyroid Gland at the Lateral Neck Masquerading as a Metastatic Papillary Thyroid Carcinoma,J Korean Med Sci, 23, 2008, 548-550.
- [17]. Noussios G, Anagnostis P, Goulis DG, Lappas D, Natsis K,Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity,Eur J Endocrinol, 165, 2011, 375-382.
- [18]. Basaria S, Westra WH, Cooper DS, Ectopic lingual thyroid masquerading as thyroid cancer metastases,Journal of Clinical Endocrinology and Metabolism, 86, 2001, 392-395
- [19]. Grapin-Botton A, Constam D, Evolution of the mechanisms and molecular control of endoderm formation. Mechanisms of Development, 124,2007, 253-278.
- [20]. Fagman H, Nilsson M, Morphogenetics of early thyroid development,Journal of Molecular Endocrinology, 46, 2011,R33-R42.
- [21]. Weller G,Development of the thyroid, parathyroid and thymus glands in man,Contrib Embryol, 24,1933, 93-140.
- [22]. De Felice M, Ovit C, Biffali E, Rodriguez-Mallon A, Arra C, Anastassiadis K, Macchia PE, Mattei MG, Mariano A, Schöler H, Macchia V and Di Lauro R, Amouse model for hereditary thyroid dysgenesis and cleft palate, Nat Genet, 19, 1998, 395-398.
- [23]. Fagman H, Grande M, Edsbage J, Semb H, Nilsson M, Expression of classical cadherins in thyroid development: maintenance of an epithelial phenotype throughout organogenesis,Endocrinology, 144, 2003, 3618-3624.
- [24]. Gillam MP, Kopp P, Genetic regulation of thyroid development, Curr Opin Pediatr, 13, 2001, 358-363.
- [25]. Barbera MJP, Clements M, Thomas P, Rodriguez T, Meloy D, Kioussis D and Beddington RS, The homeobox gene Hex is required in definitive endodermal tissues for normal forebrain,liver and thyroid formation,Development, 127,2000, 2433-2445.
- [26]. Ibrahim NA, Fadeyibi IO, Ectopic thyroid: etiology, pathology and management, Hormones, 10,2011, 261-269.
- [27]. Kousta E, Konstantinidis K, Michalakis C, Vorias M, Sambalis G, Georgiou M and Theodoropoulos GE, Ectopic thyroid tissue in the lower neck with a coexisting normally located multinodular goiter and brief literature review,Hormones,4, 2005, 231-234.
- [28]. Yoon JS, Won KC, Cho IH, Lee JT, Lee HW, Clinical characteristics of ectopic thyroid in Korea, Thyroid, 17,2007, 117-21.
- [29]. Neinas FW, Gorman CA, Devine KD and Woolner LB. Lingual thyroid. Clinical characteristics of 15 cases, Annals of Internal Medicine, 79,1973,205-210.
- [30]. Jacob MJ, Ravina MA, Rare case of lingual thyroid with hyperthyroidism: A case report and review of the literature, Indian J Endocrinol Metab, 16, 2012, 441- 443.
- [31]. Baik SH, Choi JH, Lee HM. Dual ectopic thyroid, Eur Arch Otorhinolaryngol, 259,2002, 105-7.
- [32]. Heather N, Hall K, Neas K, Potter H, Wiltshire E, Growth and Development in a Child with Resistance to Thyroid Hormone and Ectopic Thyroid Gland, Pediatrics,129,2012, e817-20.
- [33]. A. D.Mace, A. Taghi, S. Khalil, A. Sandison, Ectopic Sequestered Thyroid Tissue: An Unusual Cause of a Mediastinal Mass, ISRN Surg, 2011, 2011, 313626.
- [34]. Sung YM, Lee KS, Han J, Cho EU, Intratracheal Ectopic Thyroid Tissue with Adenomatous Hyperplasia in a Pregnant Woman,American Journal of Roentgenology, 190, 2008, W161-W163.

- [35]. Ward GE, Cantrell JR, Allan WB, The surgical treatment of lingual thyroid, *Ann Surg*, 139, 1954, 536-44.
- [36]. Nilegaonkar S, Naik C, Sonar S, Hirawe D, Triple ectopic thyroid: A rare entity. *Indian J Nucl Med*, 26, 2011, 194-195.
- [37]. Jarvis JF, Lingual thyroid: a report of three cases and discussion, *S.A. Medical Journal*, 1969:8-12.
- [38]. Tojo K, lingual Thyroid Presenting as Acquired Hypothyroidism in the Adulthood, *Internal Medicine*, 37, 1998, 381-84.
- [39]. Chan FL, Low LC, Yeung HW, Saing H, Case report: lingual thyroid, a cause of neonatal stridor, *Br J Radiol*, 66, 1993, 462-4.
- [40]. Tincani AJ, Martins AS, Negro AD, Araújo PPC, Barretto G, Lingual thyroid causing dysphonia: evaluation and management. Case report, *Sao Paulo Med J*, 122, 2004, 67-9.
- [41]. Zackaria M, Chisholm EJ, Tolley NS, Rice A, Chhatwani A, Multinodular goitre in lingual thyroid: case report, *J Laryngol Otol*, 124, 2010, 349-51.
- [42]. Massine RE, Durning SJ, Koroscil TM, Lingual thyroid carcinoma: a case report and review of the literature, *Thyroid*, 11, 2001, 1191-6.
- [43]. Ohnishi H, Sato H, Noda H, Inomata H, Sasaki N, Color Doppler Ultrasonography: Diagnosis of Ectopic Thyroid Gland in Patients with Congenital Hypothyroidism Caused by Thyroid Dysgenesis, *J Clin Endocrinol Metab*, 88, 2003, 5145-5149.
- [44]. Altay C, Erdoğan N, Karasu S, Uluç E, Sarsılmaz A, Mete B, Oyar O, CT and MRI findings of developmental abnormalities and ectopia varieties of the thyroid gland, *Diagn Interv Radiol*, 18, 2012, 335-343.
- [45]. Akanmu IN, Adewale OM, Lateral cervical ectopic thyroid masses with eutopic multinodular goiter: an unusual presentation, *Hormones*, 8, 2009, 150-153.
- [46]. Galizia G, Lieto E, Ferrara A, Castellano P, Pelosio L, Imperatore V, Palladino E, Ectopic thyroid: report of a case II, *Giornale di Chirurgia*, 22, 2001, 85-88.
- [47]. Farrell, ML, Forer M, Lingual Thyroid, *Aust. N.Z. J. Surg*, 64, 1994, 135-138.