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Original Research Article

EVALUATION OF RESULTS OF MANAGEMENT OF LINGUAL THYROID: A MULTI-CENTER EXPERIENCE OVER 25 YEARS

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Abstract

Background: The most common developmental abnormality of thyroid gland is a lingual thyroid. Actual prevalence of lingual thyroid is not known as most of the cases are asymptomatic. No standardized management protocol is available. We aimed at describing our experience in management of lingual thyroid. **Materials and Methods:** We conducted our study across India from 1999 to 2024 at two tertiary care centres, Kolkata and Delhi. Demographics, clinical features, thyroid profile, imaging result, treatment received are analysed. **Result:** We have got 15 cases of lingual thyroid during this time. All our cases were female. Most of our cases were in between first and second decade of life. Dysphagia was the most common presenting symptom. Two patients had severe obstructive sleep apnoea while another patient had breathing difficulty. Two ladies were referred to us from the endocrinology department who were otherwise asymptomatic. Hypothyroidism was present in 67% of our cases. Thyroid hormone replacement therapy was given in all hypothyroid patients. 4 patients have undergone surgery due to obstructive symptoms. They were also given thyroid replacement therapy post-operatively. Euthyroid patients were kept for follow-up without any intervention. **Conclusion:** Lingual thyroid is a rare entity and its possibility should be kept in mind while evaluating any patient of dysphagia or foreign body sensation in throat. Management of lingual thyroid patient should be individualized according to symptom of patient. Thyroid hormone supplementation is needed in patients with hypothyroidism whereas a small subset of patients with obstructive symptoms may need surgical excision.

Patients with mild obstructive symptoms should be first tried with suppressive thyroid hormone replacement therapy with an aim to shrink the tissue. If the obstruction is severe or medical therapy fails, surgery is the choice.

INTRODUCTION

Rarely the migration of the thyroid may become arrested anywhere along the descent pathway from the foramen caecum to low in the neck.[1] The most common abnor-mality is a lingual thyroid. Ectopic thyroid tissue may also be found in the sublingual, high cervical and mediastinal regions or within the cardiac endothelium.[2] Among the ectopic positions of thyroid, lingual thyroid is the most common. It has a prevalence of 1:100000-3,00,000 healthy individuals.[3] Most of the patients who presented with lingual thyroid are asymptomatic.[4] Among the symptoms most common are dysphagia, odynophagia, airway obstruction and difficulty while talking and sometimes bleeding. Some patients present with hypothyroidism. For patients with obstructive symptoms, thyroxine replacement should be introduced as initial therapy, to induce glandular shrinkage.[5]

Treatment of lingual thyroid vary according to the symptom of the patient. If hypothyroidism is the main symptom, thyroid hormone replacement might be enough. But if it is causing obstructive symptoms then surgery is the main solution.

MATERIALS AND METHODS

We have conducted our study in 2 centers in India from 1999 to 2024.Our study included cases from Kolkata and Delhi. We have got 15 cases during this time. All our cases were female. After thorough clinical examination thyroid profile were done in all the cases. USG of neck done was in all the cases. In

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2 cases contrast enhanced CT SCAN were done as the gland was hypoplastic and not visualised in ultrasound. Fine needle aspiration was done in all cases. Whole body thyroid scan with 99 m pertechnetate was done to localize ectopic thyroid tissue in all the patients. Management depends on the symptom of the patient. Hypothyroid patients were treated with thyroid replacement therapy. Surgical management was done in patients with obstructive symptoms.

RESULTS

Our youngest patient was 7 years old whereas oldest patient was 30 years old. Most of our patients belonged to the first and second decade of life. All of our patients were females.

Dysphagia was the most common presenting symptom, being present in 6 of our cases, 3 of whom also complained of a swelling at the base of their tongue. One patient had swelling at base of her tongue but no dysphagia. 2 patients had foreign body sensation (one of whom also had dysphagia), 2 patients had impairment of speech. Two patients had severe obstructive sleep apnoea while another patient had breathing difficulty as the chief presenting symptom. Two ladies were referred to us from the endocrinology department who was otherwise asymptomatic.

Out of the 15 cases, 10 cases had history of easy fatigue and depression, mental retardation, symptoms suggestive of hypothyroidism which was subsequently proved by serum thyroid profile.

All these patients when presented to OPD have undergone clinical examination. We found smooth round swelling pink in colour firm in consistency in the midline at the base of the tongue with intact mucosa. The thyroid gland could not be appreciated on examination of neck in 10 patients. On palpation there was no tendency to bleed. No lymph nodes were palpable on examination of the neck in any of our patients. Thyroid function test showed hypothyroidism in 10 patients. All other patients were euthyroid.

USG neck was done for all our patients. In 5 patients both lobes of thyroid glands were present in normal position without any obvious pathology. All these patients had a globular mass with distinct margin present at base of the tongue posterior to circumvallate papillae.

Fine needle aspiration done which shows follicular like cells in sheets with thick colloid material and abundant squamous cells. A thyroid scan with technetium -99m sodium was performed showing marked uptake of isotope in the area of tongue in all the patients. In one patient along with normal thyroid and lingual thyroid, ectopic thyroid tissue was present in right submandibular region.

Patients presenting with hypothyroidism were treated with thyroid hormone supplement. Surgical resection was done in 4 patients, because in these patients the swelling was causing obstructive symptom. Euthyroid and non-obstructive patients were kept for follow-up.

In one patient after surgical resection the thyroid was implanted in abdomen under rectus abdominis. When she had a pregnancy she developed a goitre which presented as a mass in abdomen.

Table 1

	Gender	Age	Main presenting symptom	Hypothyroidism	Surgical excision
1.	F	30	Asymptomatic (refer from endocrinology dept)	yes	No
2.	F	15	Dysphagia, Swelling at base of tongue	no	No
3.	F	7	Impairment of speech	yes	No
4.	F	20	Breathing difficulty	yes	No
5.	F	16	Hemorrhage from tongue	yes	Yes
6.	F	9	Impairment of speech	no	No
7.	F	11	Dysphagia	no	No
8.	F	18	Swelling at base of tongue	yes	No
9	F	7	Dysphagia,Foreign body sensation	yes	Yes
10	F	29	Obstructive sleep disorder	yes	Yes
11	F	12	Dysphagia, Swelling at base of tongue	yes	No
12	F	8	Foreign body sensation	no	No
13	F	13	Obstructive sleep disorder	yes	Yes
14	F	25	Dysphagia, Swelling at base of tongue	yes	No
15	F	10	Dysphagia	no	No

DISCUSSION

The first case of lingual thyroid was reported by Hickmann in 1869.[6] The exact prevalence of lingual thyroid is not known as most of the cases are asymptomatic. Midline ectopic thyroid tissue has been found from tongue to diaphragm.[7] Among ectopic positions of thyroid, lingual thyroid is the most common.[8] Differential diagnosis of lingual thyroid includes hypertrophied lingual tonsil, cysts of thyroglossal duct, fibroma, angioma and papilloma,salivary gland tumor.

Our series has 15 cases over a period of 25 years whereas Barbara G. Carranza Leon et al. in their Mayo clinic series identified 29 patients over a period of 35 years. Neinas FW et al reported 15 patients with lingual thyroid at Mayo clinic Rochester, Minnesota from 1907 to1971. They identified these patients at a single tertiary care referral centre whereas we have pooled our data across two different centres.

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According to literature, female to male ratio ranges from 3:1 to 7:1.[9] In our study all the cases were female. Barbara G. Carranza Leon et al. found 83% of their patients to be females.

Few researchers believe that genetic factors, specifically the genes of the transcription factors TITF-1 (Nkx2-1), Foxe1 (TITF-2), and PAX-8, play a role in thyroid gland morphogenesis and differentiation.[10]

The size of lingual thyroid tissue may be enlarged by the stimulation of elevated TSH during puberty, pregnancy, or menstruation, when the demand for thyroid hormones increases that leads hypothyroidism to develop. This explains the reason why the reported lingual thyroid is more common in females.[11-14]

During a 15-year-period, in a thyroid investigation centre in Sri Lanka comprising 16,593 cases, 8 lingual thyroids were seen. All their patients were female.[15]

We believe that similar demographic profiles of our country and Sri Lanka have led to all our patients in both series to be females, as opposed to the series from Mayo Clinic.

In our study 5 patients out of 15 have thyroid in normal position. It tallies with the literature available on lingual thyroid.

30% of reported cases of lingual thyroid have thyroid present in the normal position according to Montogomery and Buckman study done in 1935.[16] More than 70% of the patients with lingual thyroid have no thyroid tissue in the normal location.[17]

In our study 10 patients out of 15 patients have hypothyroidism. We did not get any case of hyperthyroidism. Literature search showed that 70% of lingual thyroid patient have been reported with subclinical hypothyroidism.[17]

Hypo and hyperthyroidism both can present in lingual thyroid patients according to Kaplan (1984). Barbara G. Carranza Leon et al. found hypothyroidism in 72% patients.

Most cases of lingual thyroid were asymptomatic. They present only when they have severe hypothyroidism and obstructive symptoms like dysphagia, foreign body sensation, pain over throat, dysphonia, dyspnoea and upper airway obstruction. In our study dysphagia was the common symptom. Two patients of our study had severe obstructive sleep apnoea. 5 young children were brought to us by their parents with history of failure to thrive and mental retardation.

Barbara G. Carranza Leon et al. found symptomatic patients comprised one-third of their series with the most common symptoms being cough and hoarseness. The diagnosis of lingual thyroid was incidental in 9 of their patients (31% in their series).

Treatment of lingual thyroid depends on severity of the symptoms. Medical treatment is effective in patients with mild symptoms. Subclinical hypothyroidism can be treated with hormonal therapy. Exogenous hormones can be given, which suppresses the action of gland thereby reducing the size of ectopic thyroid. But success rates are not encouraging.[18]

Barbara G. Carranza Leon et al reported Nineteen (65%) of the patients received thyroid hormone replacement, one of whom was euthyroid before receiving thyroid hormone for gland suppression. In concordance with previous report we found that we have given thyroid hormone replacement in 10 of our patients.

Surgical treatment is indicated in patients with dysphagia, dyspnoea, dysphonia, haemorrhage, uncontrolled hyperthyroidism and occasionally suspicion of malignancy. Very rarely malignant transformation of lingual thyroid has been reported.[19] Malignancy in ectopic thyroid is follicular carcinoma.[20] According to Giuseppo Santangelo et al surgery is a prudent choice due to the potential of malignant evolution of ETT.[21]

We have operated on 4 of our patients, 2 for obstructive sleep disorder, one for haemorrhage from tongue and another for dysphagia along with foreign body sensation. So in our short series, 26.7% patients have been treated by surgery. In Mayo clinic lingual thyroid series they operated on 6 patients which amounts to 17.14%. We believe the higher percentage of operated patients in our series can be attributed to more patients with obstructive symptoms like dysphagia and obstructive sleep disorders, which in turn probably stems from the fact that patients in our country often seek treatment late, by which time, the mass increases in size.

In their study RAI was administered to 2 patients. Post-operative period was uneventful in all of them. We did not use RAI in any of our patients. We have done auto-implantation in one patient which was successful.

CONCLUSION

Lingual thyroid is a rare entity and infant and young children with history of failure to thrive or mental retardation require routine screening for lingual thyroid. The possibility of lingual thyroid should be kept in mind while evaluating any patient of dysphagia or foreign body sensation in throat. Thorough examination of head, neck, base of the tongue is needed. Management of lingual thyroid patient should be individualised according to symptom of patient. Thyroid hormone supplementation is needed in patients with hypothyroidism whereas a small subset of patients with obstructive symptoms may need surgical excision.

Patients with mild obstructive symptoms should be first tried with suppressive thyroid hormone replacement therapy with an aim to shrink the tissue. If the obstruction is severe or medical therapy fails surgery becomes the treatment of choice.

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