



# Transoral Robotic Lingual Thyroidectomy

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Base of tongue is the most common location of ectopic thyroid. Conservative management is the mainstay of treatment, but surgical treatment is required when complications occur due to the airway obstruction, dysphagia, or hemorrhage. An 18-year-old male with dyspnea and voice change was diagnosed with lingual thyroid, and the symptoms were refractory after several months of thyroid hormone replacement. The lingual thyroid gland was surgically excised using transoral robotic surgery (TORS). Although there are many surgical approaches for the resection of the lingual thyroid, TORS has advantages of using the natural orifice, and leaving no scar on the cervical area. Herein, we report a case of TORS-assisted lingual thyroidectomy with a literature review.

**Key Words:** Ectopic thyroid, Lingual thyroid, Transoral robotic surgery, Lingual thyroidectomy

## Introduction

Lingual thyroid gland is an ectopic thyroid that accounts for almost 90% of the ectopic thyroid, and is formed when primitive thyroid gland fails to fully descend to the neck level.<sup>1)</sup> The prevalence of ectopic thyroid is reported to be 1 in 100,000 to 300,000.<sup>2)</sup> It is about 4 times more frequent in female than in male patients, especially in Asian ethnicity.<sup>3)</sup> The lingual thyroid gland is usually asymptomatic. However, as it is positioned in the upper aerodigestive tract, symptoms may develop when it becomes enlarged. Aggravations are related to puberty, pregnancy, trauma, or menopause. The lingual thyroid gland may cause symptoms such as cough, snoring, globus, sleep apnea, dysphagia, dysphonia, and dysarthria. In severe cases, bleeding and dyspnea may occur due to airway obstruction.<sup>2)</sup>

For asymptomatic patients with euthyroid hormonal status, conservative management is the mainstay of treatment plan. Levothyroxine replacement therapy

can be effective in decreasing the size of the lingual thyroid gland or elevating the thyroid hormone level, in patients with symptoms or hypothyroidism. Surgery is indicated when medical treatment is ineffective or complications occur, such as airway obstruction, ectopic thyroid ulceration, bleeding, cystic degeneration, or malignant change. Surgical methods for lingual thyroidectomy include transhyoid approach, suprahyoid approach, lateral pharyngotomy, or transoral approach.<sup>4)</sup> The development of transoral robotic surgery (TORS) has enabled to safely and effectively access the base of tongue, and perform dissection in the oropharynx. Unlike TORS, transoral endoscopic laser excision has limitations of straight vision and straight surgical instruments. However, the robotic arms and instruments can be flexible in the oropharynx, which enables transoral access to the vallecular part of lingual thyroid gland.<sup>5)</sup> TORS provides a 10-fold magnification image to view blood vessels, thyroid gland, and oropharyngeal mucosa in detail. Also, the robotic system may provide precision and agility of the instruments to enable minimally invasive surgery.<sup>6)</sup>

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Therefore, we report our experience of a case of TORS-assisted lingual thyroidectomy for a male patient who presented with dyspnea, and a literature review.

## Case Report

An 18-year-old male patient presented in the outpatient clinic with breathing difficulties that persisted for several years, and voice change that developed one month ago. The patient had a history of receiving thyroid hormone replacement for hypothyroidism few years ago. No specific family history associated with thyroid gland was identified. Laryngoscopy showed a large mass in the base of tongue was partially obstructing the airway (Fig. 1).

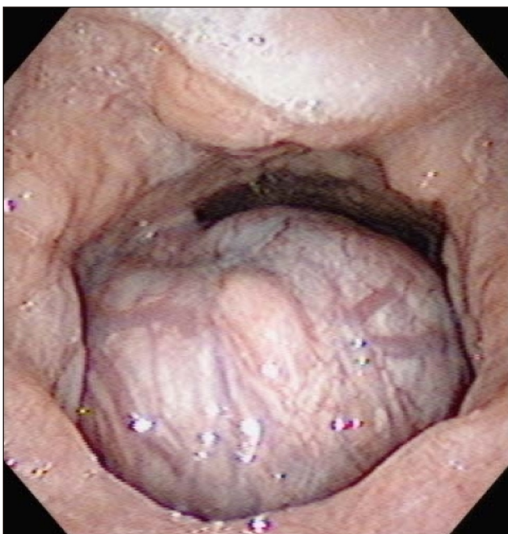
Under suspicion of ectopic thyroid, imaging studies including computed tomography (CT), ultrasonography, and magnetic resonance imaging were performed. A 2.8×2.7×2.7 cm sized heterogeneous, contrast-enhancing mass was noted in the base of tongue in CT images (Fig. 2). Tc-99m thyroid scan was performed, and heterogeneous uptake of the base of tongue was noted. In all imaging studies, including CT, magnetic resonance imaging (MRI), ultrasonography, and Tc-99m thyroid scan, normal thyroid tissue or thyroid scan uptake was not detected in the normal

thyroid location.

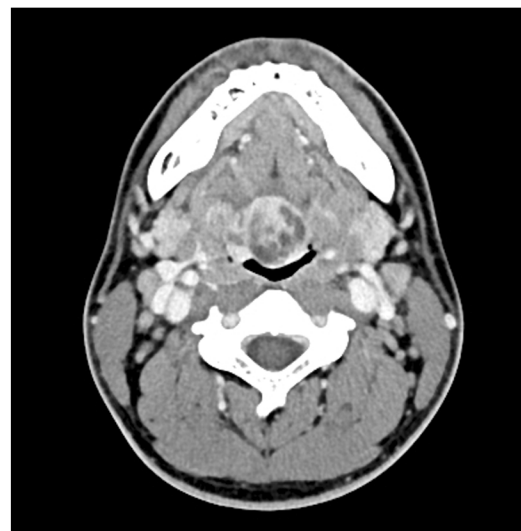
Thyroid-stimulating hormone (TSH) level and thyroglobulin antigen levels were elevated which were 28.75  $\mu$ IU/mL and 92.17 ng/mL, respectively. Free T4 level and triiodothyronine (T3) level were within the normal range at 1.03 ng/dL and 128 ng/dL, respectively. The patient was finally diagnosed as ectopic lingual thyroid gland, and was prescribed levothyroxine 50 mcg, once daily.

After 4 months of thyroid hormone replacement therapy, symptoms of dyspnea gradually worsened. Afterwards, the patient did not appear for the next clinic visit. After 20 months, he visited the outpatient clinic for aggravated symptoms of dyspnea during sleep. The lingual thyroid gland was not significantly enlarged on the laryngoscope and CT scan, but the oropharyngeal airway was significantly obstructed due to the lingual thyroid. The patient complained that dyspnea had progressed, and appealed that physical activity, and climbing the stairs were difficult due to airway obstruction. The patient strongly wanted surgical treatment, and the surgical treatment was decided.

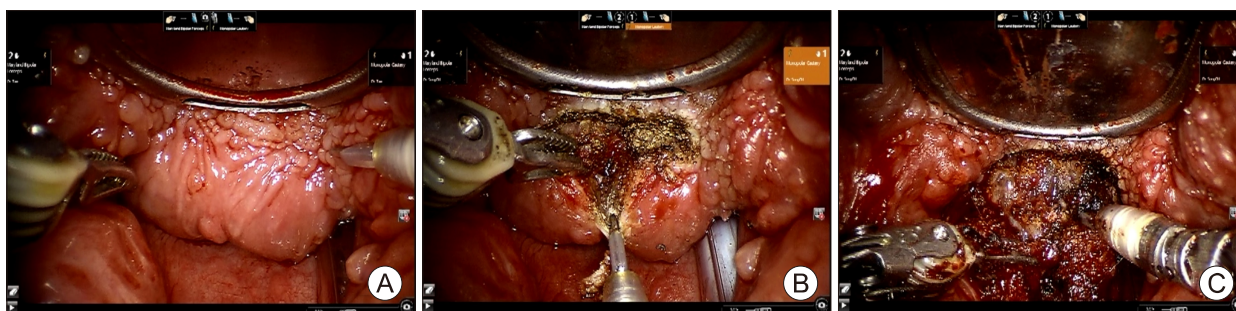
For the minimally invasive resection of the lingual thyroid, which extends to the lingual surface of epiglottis, a TORS assisted lingual thyroidectomy was planned. The possibility of tracheostomy and trans-



**Fig. 1.** Initial laryngoscopy of a lingual thyroid with engorged vessels.



**Fig. 2.** Round mass in the base of tongue with heterogeneous, contrast-enhance in axial image of CT (2.8×2.7×2.7 cm sized).



**Fig. 3.** Transoral robotic surgery assisted lingual thyroidectomy. (A) Surgical view after setting of Maryland bipolar forceps and monopolar spatula. (B) Midline incision with monopolar spatula to evaluate the depth of lingual thyroid gland (C) Surgical view after excision of lingual thyroid gland.

cervical approach were also explained to the patient. The patient underwent arterial embolization of the right lingual artery branching to the lingual thyroid under angiography 3 days before surgery.

Under supine position, nasotracheal intubation was performed. The surgical view was secured using Davis retractor. The tip of the retractor was passed along the base of the tongue and lingual thyroid was noted. The Davinci robot was docked and Maryland bipolar forceps and monopolar spatula was set, with a 30-degree endoscope facing upwards. The midline of the lingual thyroid was first dissected with monopolar cautery to control the depth, and complete resection was done without tracheostomy or transcervical approach (Fig. 3). The size of the resected mass was 3.5×3×2 cm sized, and final pathology was reported as an ectopic thyroid gland. Extubation was successfully performed in the operating room after the surgery. A 150 mcg of levothyroxine was prescribed postoperatively. At 2 months after the surgery, the upper airway was patent with no recurrence of the lingual thyroid on laryngoscopy, and the patient reported that the symptoms of dyspnea had improved.

## Discussion

Thyroid gland is of endodermal origin and originates from the primitive pharynx during the end of third embryonic week.<sup>7)</sup> It is located around the oropharynx at the level of tongue base of the embryo, and the thyroglossal duct is formed and descends to the hyoid bone at the 7th week. When the thyroid reaches its final

position, the thyroglossal duct disappears. The thyroid hormone levels begin to increase from around the fifth month, reaching full levels at the ninth month.<sup>8)</sup> The lingual thyroid is the ectopic thyroid that accounts for most of the ectopic position of thyroid. Other sites can be mediastinum, lungs, liver, esophagus, heart, breast, and trachea, and very rarely below the diaphragm such as ovaries, vaginal wall, duodenum, small intestine mesentery, and pancreas.<sup>9)</sup> The mean age is 40 years at which ectopic thyroid is detected, and in three-fourth of the cases, the only thyroid tissue is the lingual thyroid gland.<sup>10)</sup> Patients usually present with throat mass or hypothyroidism, but some present as euthyroid.<sup>11)</sup>

Scintigraphy is important for detecting ectopic thyroid and is also used to confirm the presence of thyroid in its normal position.<sup>11)</sup> Thyroid scan may also be used to detect ectopic thyroid. Both scintigraphy and thyroid scan have high sensitivity and specificity in the diagnosis of ectopic thyroid.<sup>12)</sup> However, these scans can be false-positive due to uptake in many other parts of the body. Therefore, physiological uptake, which is seen in salivary glands, breast, stomach, intestine, bladder should be confused with ectopic thyroid.<sup>13)</sup>

Ultrasonography, CT, and MRI are useful in determining the location of the ectopic thyroid. In CT images, the thyroid tissue is observed as high density, and is enhanced with contrast.<sup>14)</sup> In MRI, the ectopic thyroid appears as a round mass with a high signal on both T1 and T2 weighted MRI.<sup>15)</sup> In patients with ectopic thyroid, thyroid function tests are usually per-

formed, and TSH level tends to increase and T3 and T4 level tend to decrease.<sup>3)</sup>

There are several surgical approaches for lingual thyroid including transoral, transhyoid, suprahyoid approach, and lateral pharyngotomy. For the transoral approach, surgery using monopolar coagulation or various lasers, can achieve successful outcomes, but has limitation in approaching and visualizing the deep portion of tongue base because vision and surgical instruments have limitations in flexibility. When the ectopic thyroid is present with no cervical thyroid gland, some authors recommend transplantation of the resected thyroid tissue to preserve a euthyroid state.<sup>16)</sup> Invasive surgical approaches including lateral transcervical approach or mandibulotomy have been used conventionally. However, the emergence of TORS has enabled us to safely and effectively access and make dissection in the deep and laryngeal part of the lingual thyroid.<sup>5)</sup> In the lingual thyroid resection, as was done in our case, TORS assisted lingual thyroidectomy was possible to successfully remove the lingual thyroid through transoral approach.

In this case, after the lingual thyroid in the base of tongue was diagnosed, thyroid hormone was prescribed to suppress the proliferation of thyroid gland. However, as the patient's symptoms such as dyspnea persisted and worsened, surgical treatment was planned. Through TORS, it was possible to secure the surgical field and facilitate access to surgical instruments. This enabled safe and minimally invasive complete resection. Pre-operative arterial embolization is not a routine procedure before cervical thyroidectomy. However, lingual thyroidectomy can lead to severe airway obstruction since lingual thyroid is located in the upper airway. There is no standard procedure for lingual thyroidectomy due to its rarity. We recommend preoperative arterial embolization in cases when bleeding is anticipated after and during lingual thyroidectomy. The patient was satisfied with the surgical result which avoided transcervical incision. In conclusion, our case shows that TORS can be an effective and safe surgical option for lingual thyroid resection.

## Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

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## Robotic Lingual Thyroidectomy

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