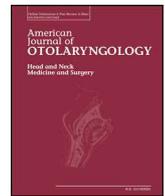




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## TORS excision of lingual thyroid carcinoma: Technique and systematic review

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### ABSTRACT

**Objective:** Clinically significant lingual thyroid tissue has a prevalence of 1/3000–10,000, and in 70% of these individuals, the lingual thyroid is their only thyroid tissue. Malignant transformation is exceedingly rare. Herein, we present a case of lingual thyroid carcinoma with a systematic literature review and description of our treatment technique.

**Data sources:** PubMed, Ovid.

**Review method:** The primary author performed a search of the literature for reports of lingual thyroid carcinoma or ectopic thyroid carcinoma associated with the tongue. Articles that did not present novel data, presented cases of ectopic thyroid carcinoma outside the tongue, non-malignant cases, non-thyroid carcinomas, or were non-English articles were excluded. Studies were limited to those published in the last 60 years.

**Results:** There are 39 cases reported in the literature. 23 cases occurred in females. Age at diagnosis ranged from 12 to 86; cases were more commonly diagnosed in the second decade of life, then in the 5th and 6th decades of life. Dysphagia, globus sensation, episodes of bleeding, voice changes, and presence of a neck mass were common symptoms at initial presentation. Nearly all patients underwent some form of pre-operative imaging, but practices varied as to the type of imaging. Treatment included surgical excision of the tumor in all but one case that was successfully treated with radioactive iodine therapy alone.

**Conclusions:** Surgeons should be aware of lingual thyroid, its presentation, workup, and carcinoma treatment. Tumors are amenable to surgical excision, possibly followed by radioactive iodine therapy. Advances in robotic and endoscopic surgery over the past decade now allow for less morbid excisions of lingual thyroid tumors.

### 1. Introduction

The prevalence of lingual thyroid tissue is variably reported in the literature. Some post-mortem studies have found lingual thyroid tissue in up to 10% of individuals [1,2]. Clinically significant lingual thyroid tissue, however, is rare; it occurs at an estimated prevalence of 1/3000 to 1/10,000 [3], and in 70% of individuals with clinically significant lingual thyroid tissue, the only thyroid tissue they have is at the base of tongue [3,4]. Malignant transformation among lingual thyroid tissue is also rare, occurring an estimated 1% of the time [5,6], making lingual thyroid carcinoma exceedingly rare. There are fewer than 60 total cases of lingual thyroid carcinoma reported in the literature. Historically, they have been treated with surgical excision via morbid open approaches, often requiring a midline split of the mandible or tongue. Herein, we report a rare case of lingual thyroid carcinoma successfully treated less morbidly with trans-oral robotic surgery (TORS). We describe our technique and present a systematic literature review.

### 2. Methods

#### 2.1. TORS technique

Patients are placed under general endotracheal anesthesia via orotracheal intubation; we have found that nasotracheal intubation is unnecessary and can lead to epistaxis with bleeding into the operative field. Patients are then prepped and draped in the usual sterile fashion, a standard mouth gag is placed, and the robot brought into the field. The tongue base is visualized, and a midline excision is performed with electrocautery, taking care to avoid lateral dissection that may injure the lingual arteries. In our experience, the lingual thyroid tissue can be excised without dissection into the native base of tongue. Following, hemostasis is achieved and the tongue is examined for edema. Not uncommonly, there is significant oropharyngeal swelling that can acutely compromise the airway, so we have a low threshold for placing a temporary tracheostomy for airway control.

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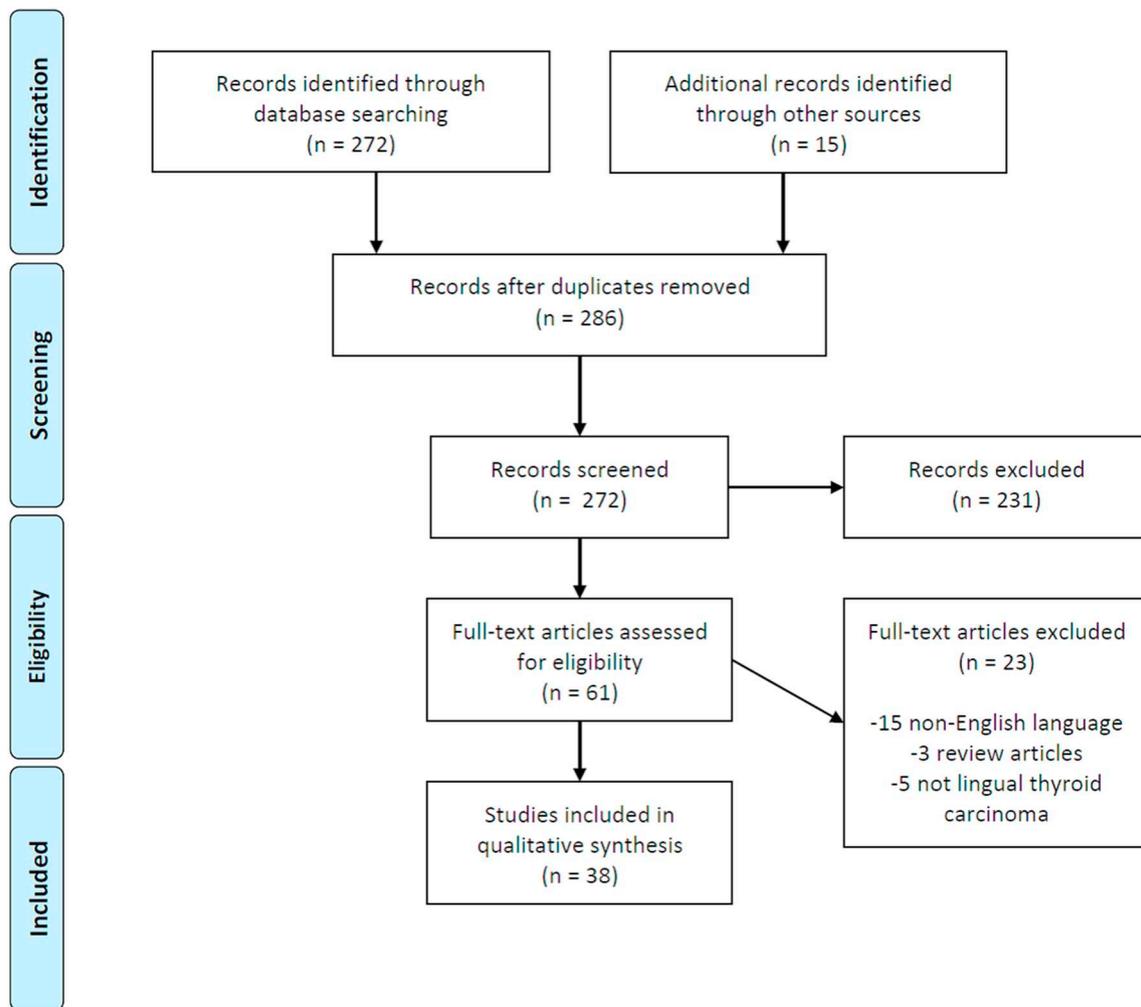


Fig. 1. PRISMA flow-diagram of our systematic literature review.

## 2.2. Literature review

We received institutional review board approval to review the case presented below. We performed a search of the literature in PubMed and Ovid for reports of lingual thyroid carcinoma or ectopic thyroid carcinoma associated with the tongue (Fig. 1) using the search terms “lingual thyroid” and “ectopic thyroid.” Our initial search identified 272 peer-reviewed articles. An additional 15 articles were identified from references of reviewed papers that could not be found through electronic searches of PubMed and Ovid. We excluded review articles that did not present novel data, cases of ectopic thyroid carcinoma in other locations than at the tongue base, cases of lingual thyroid tissue without malignant transformation, cases of non-thyroid carcinomas, articles published more than 60 years ago, and articles in languages other than English.

## 3. Results

There are less than 60 total reports of lingual thyroid carcinoma published in any language. We identified 38 reports describing 38 patients with lingual thyroid carcinoma published in the English literature (Table 1); 23 of these have been published in the past 60 years [7–27]. All publications were case reports with the exception of one that included two patients [9]; one of these patients had previously been reported by Potdar in 1971, however [8]. We have one additional case to report, making the total number of cases 39.

## 3.1. Case

Our patient is a 41 year old otherwise healthy female who presented initially with complaints of dysphagia. She also complained of several weeks of globus sensation and throat clearing. Flexible fiberoptic laryngoscopy demonstrated a posterior tongue mass (Fig. 2A). The patient was taken to the operating room for direct laryngoscopy and biopsy of the mass which was consistent with normal thyroid tissue (Fig. 1). An ultrasound of the neck did not find any other thyroid tissue present, and serum thyroid hormone levels were within normal limits. No other imaging or lab investigations were performed for workup. The diagnosis of lingual thyroid was discussed with the patient who elected to proceed with surgical excision. She was taken back to the operating room for successful TORS excision of lingual thyroid tissue and tracheostomy (Fig. 2B). Histologic examination of the excised lingual thyroid tissue demonstrated two separate 3 mm foci of papillary thyroid carcinoma (Fig. 2C–D). The patient successfully decannulated shortly after surgery and her pre-operative dysphagia improved. Post-operative I<sup>131</sup> scan did not demonstrate any orthotopic thyroid tissue or other remaining ectopic thyroid tissue. The patient underwent 125 mc of radioactive iodine therapy. After completion of therapy, she was followed with in-clinic exams, ultrasounds of her neck, periodic I<sup>131</sup> scans, and serum thyroid labs, and she remains without evidence of disease six years after completion of treatment.

**Table 1**

Previously-reported cases of lingual thyroid carcinoma in the English literature. The best level of evidence is 5, as all reports are case reports or small case series.

First author/year published	Age/sex	Level of evidence
Gunn 1910	18 F	Case report
Rutgers 1910	21 F	Case report
Brentano 1911	42 F	Case report
Seeyle 1916	18 F	Case report
Ray 1918	74 M	Case report
Hofer 1920	50 M	Case report
Tyler 1923	54 M	Case report
Ashurst 1925	56 M	Case report
Peracchia 1927	56 M	Case report
Marchal 1935	59 M	Case report
Levi 1935	21 F	Case report
Watson 1948	86 F	Case report
Cromartie 1941	52 F	Case report
Wapshaw 1942	20 F	Case report
Willis 1953	26 M	Case report
Mill 1959	24 F	Case report
Potdar 1971	12 M	Case report
Kamat 1979	40 F	Case series (2)
Singh 1979	28 F	Case report
Goeder 1980	40 F	Case report
Diaz-arias 1992	23 F	Case report
Winslow 1997	23 M	Case report
Casella 1999	66 M	Case report
Massine 2001	57 M	Case report
Seoane 2002	58 F	Case report
Kao 2002	70 M	Case report
Goldstein 2002	48 F	Case report
Perez 2003	28 M	Case report
Falvo 2005	30 F	Case report
Kennedy 2007	25 F	Case report
Bigotti 1997	30 M	Case report
Hari 2009	49 F	Case report
Mogi 2018	40 M	Case report
Stokes 2018	84 F	Case report
Yoo 2016	41 F	Case report
Chen 2013	42 M	Case report
Raju 2009	45 F	Case report
Yaday 2008	45 F	Case report

### 3.2. Clinical presentation

Of all reports in our systematic review including our own, 23 cases occurred in females and 16 in males. Age at diagnosis ranged from 12 to 86 with a bimodal distribution; cases were more commonly diagnosed in the second decade of life, then in the 5th and 6th decades of life (Fig. 3). 24 reports from the past 60 years provided more information on patient presentation and were analyzed further. Dysphagia was the most common symptom at the time of diagnosis, occurring in 27% of patients (Fig. 4). 16% of patients complained of globus sensation, some alone and some in addition to dysphagia. 14% of patients reported episodes of bleeding from the throat or mouth, and 11% of patients reported a history of voice changes or presence of a neck mass. Two (5%) patients complained a chronic cough or dyspnea at presentation. Other, less common complaints at the time of presentation included pain at the angle of mandible, snoring, or a visible mass in the mouth without other accompanying symptoms. 21 reports described a physical exam, and in all but 1 (5%) case an abnormal mass at the base of tongue was identified either with direct visualization, palpation, or with a flexible laryngoscope.

### 3.3. Imaging and diagnosis

Nineteen (79%) of 24 reports over the past 60 years, including our case herein, obtained one or more forms of pre-operative imaging. Of those who performed pre-therapy imaging, 8 (42%) included a radio-tracer uptake scan, 7 (37%) obtained a CT scan, 4 (21%) obtained an MRI, and 4 (21%) obtained plain film radiography. The majority of

patients underwent fine needle aspiration, excisional biopsy, or incisional biopsy before proceeding to definitive therapy, and the majority underwent radiotracer uptake scanning post-operatively to evaluate for residual disease. Of note, presence or absence of orthotopic thyroid tissue was reported in 21 reports; it was present in 7 (33%) cases. Eight (38%) patients had metastases at the time of diagnosis or developed regional or distant recurrence of disease after initial treatment of 21 reports that mentioned presence or absence of metastases.

### 3.4. Treatment

One (4%) patient in the past 60 years underwent radioactive iodine therapy alone (with 161 mc) with curative intent; the other 23 (96%) underwent surgical excision with or without adjuvant radioactive iodine therapy. Surgical approaches varied; earlier approaches included a midline split of the tongue or midline split of the mandible to gain access. *Trans-hyoid* and lateral pharyngotomy approaches were also employed. More recent cases have been excised with trans-oral robotic surgery or with endoscopic visualization. 14 (61%) of the 23 cases that underwent surgical excision also underwent adjuvant radioactive iodine therapy, with doses ranging from 80 to 151 mc.

### 3.5. Pathology

Five (21%) cases from the past 60 years were found to be follicular carcinoma. 16 (66%) cases, including our report herein, were papillary thyroid carcinoma, one (4%) was Huerthle cell carcinoma, one (4%) was medullary cell carcinoma, and one (4%) was malignant thyroid carcinoma not further specified.

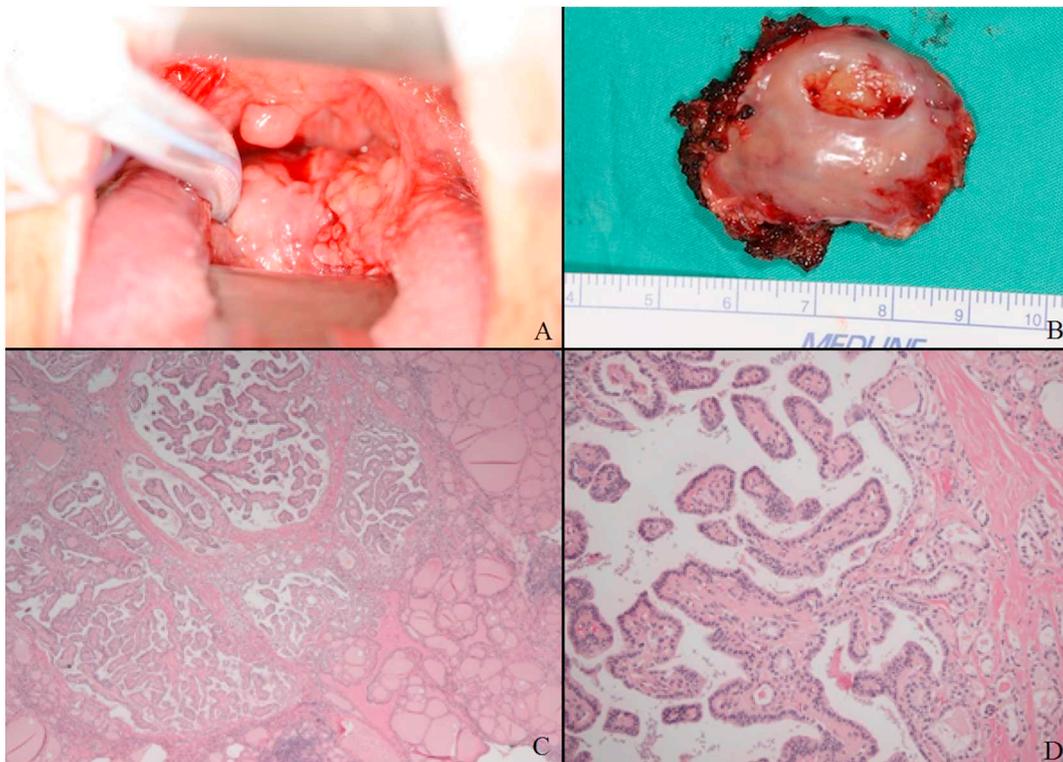
## 4. Discussion

The incidence of lingual thyroid is clinically rare, and the transformation to lingual thyroid carcinoma is even rarer, with only 39 reports in the English literature dating back to 1910. Patients with lingual thyroid carcinoma will most commonly present with dysphagia, though globus sensation, episodes of bleeding, voice changes, and having a neck mass are also common. Further, over a third of patients will develop regional or distant metastases. As with orthotopic thyroid carcinoma, papillary was the most common among lingual thyroid carcinoma, followed by follicular. Medullary carcinoma of a lingual thyroid is only reported in one case, and there are no reported cases of anaplastic carcinoma in a lingual thyroid.

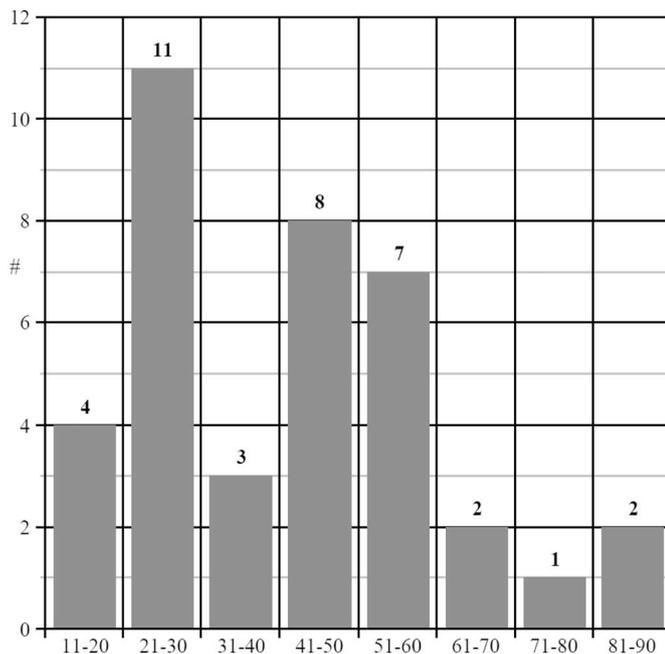
Some form of pre-operative imaging of the neck is useful before proceeding to surgery to fully evaluate for the presence of abnormal adenopathy which would also require surgical excision. In our series, multiple different modalities were employed, including plain film, CT, MRI, and ultrasound. An ultrasound of the neck in experienced hands can accurately identify nodal disease, can be quickly performed in clinic, and spares the expense of an MRI and the radiation dose of a CT. When ultrasound is indeterminate, a CT, Neck with contrast or MRI can reveal greater detail.

Similar to thyroid carcinoma presenting in an orthotopic gland, lingual thyroid carcinoma can be successfully treated with surgical excision possibly followed by adjuvant radioactive iodine therapy. Similar guidelines to those employed in managing orthotopic disease should be applied to carcinoma of a lingual thyroid. Fortunately, the development of TORS and advances in endoscopic techniques over the past decade now allow for less morbid surgical excision of lingual thyroid carcinomas than historically, when large open surgeries were required to access tumors.

TORS was developed as a minimally-invasive surgical technique and was first successfully used in humans in 2005 [28–29]. The most commonly used platform today is the daVinci System (Intuitive Surgical), approved in 2009 by the FDA for head and neck surgery [29]. Initially, TORS was employed in the head-and-neck for resection of



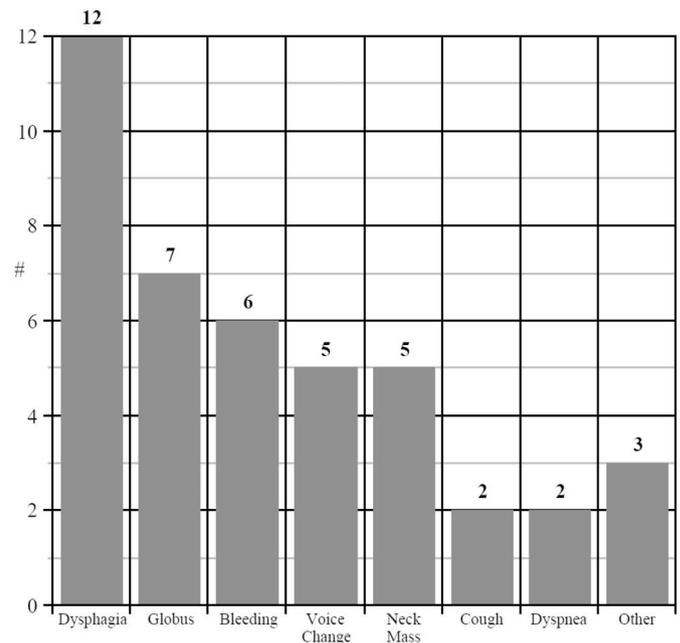
**Fig. 2.** 41 year old female with lingual thyroid tissue apparent on examination (A). Surgical excision removed 4 × 3 cm of lingual thyroid tissue. Two 3 mm foci papillary thyroid carcinoma were found, shown at low power (C) and high power (D) on hematoxylin-eosin stain.



**Fig. 3.** Histogram of counts for age at diagnosis of cases of carcinoma in a lingual thyroid with a bimodal distribution.

oropharyngeal and thyroid masses, but its indications have been continuously growing over the past few years [30].

A main advantage of TORS is that it improves access and visualization of the upper airway and allows surgeons to resect tumors that cannot be directly visualized, such as those at the base-of-tongue, supraglottis, and hypopharynx, without splitting the mandible or employing other open and morbid approaches [30–31]. By obviating the need for an open surgical resection, it also has less morbidity and



**Fig. 4.** Histogram of counts demonstrating common symptoms at initial presentation. Other symptoms include pain at the angle of mandible, a visible mass in the mouth, and snoring as the primary complaint.

improved cosmesis post-operatively [30]. Also, it can facilitate the successful initial staging of primary disease [31]. It is commonly used today for the resection of early stage (T1 and T2) oropharyngeal carcinomas, but it can also successfully be used for resection of other masses. A disadvantage of TORS is the cost of the robot itself, the need for additional surgeon training on the operator platform, and decreased tactile feedback compared to open approaches.

There are only two previously reported cases of TORS excision of lingual thyroid carcinoma [22,24]. One case required adjuvant radioactive iodine therapy, while the second was successfully treated with surgery alone. In both cases, there were no reported complications, and both patients recovered uneventfully after surgery. While mortality from lingual thyroid carcinoma and subsequent surgical treatment at the turn of the 20th century was high, mortality in modern days is negligible, and morbidity has significantly improved with minimally-invasive techniques such as TORS.

## 5. Conclusions

Lingual thyroid carcinoma is a rare entity commonly presenting with dysphagia. It can be successfully treated with surgical excision and possible adjuvant radioactive iodine therapy. Improvements in robotic and endoscopic surgical techniques over the past decade have decreased the morbidity associated with surgical removal of these tumors often obviating the need for a large, open surgical approach.

## Disclosures

None of the authors have any relevant conflicts of interest to disclose.

## Conflicts of interest

The authors have no conflicts of interest to disclose.

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