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# Endoscopic management of intralingual thyroglossal duct cysts: Case series and systematic review

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ABSTRACT

*Objective:* To discuss our institutional experience with endoscopic management of intralingual thyroglossal duct cyst (TGDC) and review cases in the published literature in a systematic review.
*Methods:* Pediatric patients with intralingual TGDC treated with endoscopic surgery at our institution from 2009-2019 were identified. Metrics from our case series were then compared to those in the literature in a systematic review to assess pooled outcomes of endoscopic or transoral management. Patient demographics, age of presentation, presenting symptomatology, size of cyst on imaging, type of surgery, and post-operative outcomes were assessed.
*Results:* We identified 5 institutional cases of intralingual TGDC and 48 cases of intralingual TGDC described in the literature. The average age of presentation was 20.36 months. 69.8% (N=37) of patients presented with at least one respiratory symptom, 22.6% (N=12) presented with dysphagia, 9.4% (N=5) presented with an identified mass in the oropharynx, and 15.1% (N=8) had the cyst discovered as an incidental finding. Three patients required revision surgeries due to prior incomplete TGDC excisions and one patient experienced a recurrence >6 months after primary excision requiring a second procedure. Our data pooled with published case series in systematic review confirms that endoscopic or transoral management are excellent options for definitive management of intralingual TGDC.
*Conclusions:* Intralingual TDGC is a potentially life-threatening variant of TGDC. Our results pooled with published series in a systematic review suggest that endoscopic or transoral management of intralingual TGDC are excellent minimally invasive treatments with a low risk of recurrence. Postoperative surveillance up to one year is recommended.



## 1. Introduction

Thyroglossal duct cyst (TGDC) is a common congenital pathology that arises due to persistence of the embryonic thyroglossal duct. Most commonly, TGDCs develop adjacent to the hyoid bone, presenting as a soft, nontender anterior midline neck mass that moves vertically with swallowing or protrusion of the tongue [1,2]. Intralingual TGDC, is a rare presentation of this congenital anomaly, accounting for approximately 0.5-2% of cases of TGDC [3,4]. While cysts in the anterior neck may initially be asymptomatic, intralingual thyroglossal duct cysts are more likely to be symptomatic, causing airway obstruction or dysphagia [5–8]. The risk of airway obstruction is increased in neonates due to the size of the lesion relative to that of the airway [9].

Diagnosis of intralingual TGDC is multi-faceted. In the otolaryngologist's office, cysts can be detected initially with flexible fiberoptic laryngoscopy showing a mass or fullness in

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the base of tongue. Further evaluation should include cross-sectional imaging to more accurately characterize and localize the lesion. Given that the most common site of ectopic thyroid tissue is the base of the tongue, lingual thyroid should also be included on the differential diagnosis for an intralingual TGDC, and presence of cervical thyroid tissue should be confirmed [1].

Surgical management of a TGDC generally consists of complete resection of the cyst and its tract, using generous margins to best prevent recurrence [10]. Complete excision, however, may not always be possible, and in these cases, it is important to remove as much cyst and cyst wall as possible without injuring the lingual arteries or additional lingual musculature. These procedures remove the majority of the lesion and, although microscopic residual cyst may be left behind, recurrence is rare because any microscopic cyst remnant is externalized. The Sistrunk procedure for TGDC excision includes resection of the midline portion of the hyoid bone. This procedure has been demonstrated to significantly reduce recurrence rates and remains the preferred method of excision in cases of cervical TGDC. With regard to intralingual thyroglossal duct cysts, however, the standard of care is not as well established. Several studies have demonstrated the feasibility of transoral or endoscopic excision in intralingual TGDC without significant recurrence rates [7,8,11]. Recurrence rates following the Sistrunk procedure range from 2-6%, with increased recurrence rates being associated with postoperative infection or incomplete resection [12,13, 24]. Though data on endoscopic or transoral excision is more limited, previously published case series looking exclusively at endoscopically or transorally managed intralingual TGDC found no recurrence among patients in the respective cohorts [7,12–14]. Due the rarity of intralingual TGDC, large series are not widely available in existing literature.

Although rare, intralingual TGDC represents a serious, potentially life-threatening pathology in the pediatric population that warrants a cohesive plan for clinical approach. Combining cases seen at a single pediatric tertiary care institution from 2011-2020 with cases gathered from a systematic review of the literature, this study aims to quantify outcomes of intralingual TGDC and identify factors that contribute to recurrence.

## 2. Methods

### 2.1. Search Strategy

A comprehensive search was performed to identify studies related to the evaluation and management of intralingual TGDCs.

Searches were initially run on May 1, 2020, in the following databases: PubMed; Cochrane Library; and Scopus. Search terms included all subject headings and associated keywords for “lingual thyroglossal duct cyst” and “intralingual thyroglossal duct cyst”. There were no language or article type restrictions on the search strategy.

### 2.2. Study Selection Criteria

Two independent reviewers screened (N.P., M.D.). Discrepancies were discussed and resolved by consensus. Titles and abstracts were reviewed against pre-defined inclusion/exclusion criteria, in accordance with the PICOS framework [15]. Articles considered for inclusion were all English-language studies that evaluated surgical management of pediatric intralingual TGDCs. Duplicate articles were excluded. Full text was then reviewed for 46 selected studies for a second round of eligibility screening. Subsequently excluded articles were those that: presented adult patients; did not include individual patient data; documented a TGDC with components involving the neck or hyoid bone; had death as the presenting symptom of the case; and did not include follow up data regarding recurrence.

### 2.3. Data Extraction

Data were gathered from the identified cases at the authors’ home institution as well as extracted from the included studies from the literature search. Measures of interest included age at presentation (months), presenting symptoms, maximum dimension of the cyst (cm), excision method, recurrence, and follow up period (months). Presenting symptoms were classified generally as respiratory, dysphagia, mass, incidental finding, or some combination of the four. Excision method was classified as endoscopic or transoral based on the documented use of an endoscope or microscope vs. direct visualization.

### 2.4. Statistical Analysis

Individual cases from the authors institution along with identified cases from the literature were analyzed as one collective sample of patient observations. Descriptive statistics were utilized to describe the cohort of patients using N (%) and median [IQR] for categorical and continuous factors, respectively. Association between clinical variables of interest (age at presentation, cyst size, and surgical approach) and outcomes of interest (presenting symptoms and recurrence) were analyzed by using T-test/Wilcoxon-rank sum test for continuous variables and Chi-square test/Fisher’s exact test for categorical variables.

## 3. Results

### 3.1. Study Selection

The initial search yielded a total of 368 articles from all databases. After duplicates were removed 262 articles were screened by title and abstract. Forty-six articles underwent independent full text review by both investigators. A total of 16 articles were selected for final review (Figure 1).

### 3.2. Characteristics of Included Studies

Within the 16 articles selected for inclusion in the review, 48 cases of intralingual TGDC were identified in the liter-

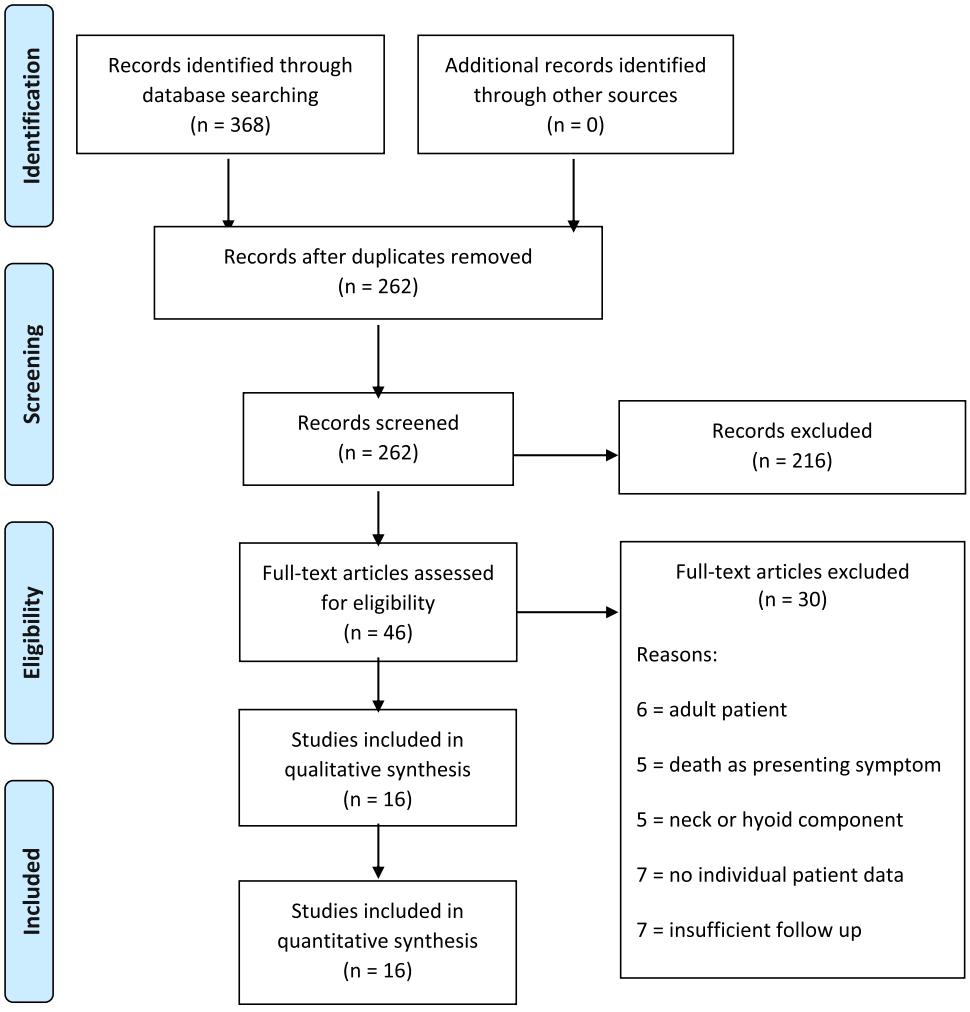


**Fig. 1.** PRISMA 2009 Flow Diagram

ature. These cases were added to 5 cases from the author's home institution yielding 53 cases for analysis (Table 1). All the included articles were case series, with numbers of included patients ranging from 1 to 16. No studies included a control group.

The average age of presentation was 20.36 months. 69.8% (N=37) of patients presented with at least one respiratory symptom, 22.6% (N=12) presented with dysphagia, 9.4% (N=5) presented with an identified mass in the oropharynx, and 15.1% (N=8) had the cyst discovered as an incidental finding (Figure 2). One institutional patient presented initially to an outside hospital in respiratory arrest due to obstruction from the TGDC. The average maximum dimension of the cysts, based on imaging or intra-operative reports, was 1.66 cm. Patients who presented with no respiratory symptoms were usually older than one year and had a significantly larger cyst size (2.84 cm) compared to patients presenting with at least 1 respiratory symptom, who tended to present at a younger age with smaller cysts (1.30 cm, $p = 0.006$). The average age at presentation was significantly younger in patients presenting with at least one respiratory symptom (11.05 months) compared to patients with no respiratory symptoms (41.9 months, $p = 0.02$).

### 3.3. Risk of Bias

Table 2 shows the risk of bias assessment for each study, assessed by two independent reviewers (N.P., M.D.), according to the MINORS instrument [16].

### 3.4. Intervention Outcomes

Within the patient cohort extracted from published series as well as our own, 52.8% (N=28) underwent transoral excision while 47.2% (N=25) underwent endoscopic excision.

Surgery for recurrence, defined as an event in which gross cyst was initially excised and the cyst then recurred after the immediate postoperative period requiring a repeat surgery, was reported in 1.9% (N=1) of cases. Average follow up period was 40.42 months. Because of limited data on TGDC recurrence, conclusions could not be drawn regarding the relationship of excision method to risk of recurrence. Given the heterogeneity, small sample size and risk of bias present in the included studies, meaningful quantitative evaluation and meta-analysis were not feasible.

Three cases (5.7%) in the series were revision surgeries, defined as an event when gross cyst was incompletely excised

**Table 1. Descriptive features of intralingual TGDC cases.** N = 53 total cases: 5 cases from the authors' home institution and 48 cases identified in the literature.

| Study | # of Patients | Age of Presentation (mos) | Presenting Symptoms | Max Dimension (cm) | Excision Type | Recurrence | Follow Up (mos) |
|---|---|---|---|---|---|---|---|
| Institutional Cases | 5 | 2 | Respiratory | 0.9 | Endoscopic | Y | 101 |
| | | 3 | Respiratory | 0.7 | Endoscopic | N | 59 |
| | | 1.75 | Respiratory | 1 | Endoscopic | N | 40 |
| | | 2 | Respiratory | 1.4 | Endoscopic | N | 10 |
| | | 3 | Respiratory | 1.3 | Endoscopic | N | 9 |
| Weldon and Krafcik (2000) [25] | 1 | 1.3 | Respiratory | 1 | Endoscopic | N | 12 |
| Sameer et al (2012) [6] | 3 | 48 | Mass, Dysphagia | 3 | Transoral | N | 30 |
| Burkart et al (2009) [17] | 16 | 48 | Incidental | NR | Endoscopic | N | 96 |
| | | 60 | Incidental | NR | Endoscopic | N | 96 |
| | | 24 | Incidental | NR | Endoscopic | N | 84 |
| | | 18 | Incidental | NR | Endoscopic | N | 84 |
| | | 3 | Respiratory | NR | Endoscopic | N | 72 |
| | | 4 | Respiratory | NR | Endoscopic | N | 60 |
| | | 2 | Respiratory | NR | Endoscopic | N | 48 |
| | | 2 | Respiratory | NR | Endoscopic | N | 48 |
| | | 1 | Respiratory | NR | Endoscopic | N | 48 |
| | | 6 | Incidental | NR | Endoscopic | N | 36 |
| | | 16 | Incidental | NR | Endoscopic | N | 24 |
| | | 7 | Respiratory | NR | Endoscopic | N | 24 |
| | | 96 | Incidental | NR | Endoscopic | N | <12 |
| | | 192 | Mass | NR | Endoscopic | N | <12 |
| | | 3 | Respiratory | NR | Endoscopic | N | <12 |
| | | 96 | Mass | NR | Endoscopic | N | <12 |
| Carroll et al (2016) [8] | 1 | 72 | Respiratory | 2.5 | Endoscopic | N | 11 |
| Kayhan et al (2013) [26] | 1 | 2 | Respiratory, Dysphagia | 1.5 | Endoscopic | N | 8 |
| Lindstrom et al (2003) [27] | 1 | 0.06 | Dysphagia | 2.5 | Transoral | N | 33 |
| Harumatsu et al (2019) [28] | 12 | 0.25 | Respiratory | 1.2 | Transoral | N | 9 |
| | | 0.6 | Respiratory | 1 | Transoral | N | 38 |
| | | 1 | Respiratory | 1 | Transoral | N | 73 |
| | | 2 | Respiratory | 0.7 | Transoral | N | 50 |
| | | 2 | Respiratory | 1.1 | Transoral | N | 37 |
| | | 2 | Respiratory | 1 | Transoral | N | 36 |
| | | 2 | Respiratory | 1.2 | Transoral | N | 55 |
| | | 2 | Dysphagia, Respiratory | 1.4 | Transoral | N | 32 |
| | | 2 | Respiratory | 1 | Transoral | N | 20 |
| | | 2 | Respiratory | 1.2 | Transoral | N | 43 |

(*continued on next page*)

**Table 1** (*continued*)

| Study | # of Patients | Age of Presentation (mos) | Presenting Symptoms | Max Dimension (cm) | Excision Type | Recurrence | Follow Up (mos) |
|---|---|---|---|---|---|---|---|
| | | 2 | Respiratory | 1 | Transoral | N | 33 |
| | | 6 | Respiratory | 1.3 | Transoral | N | 68 |
| Turhan and Bostanci (2019) [29] | 1 | 3 | Dysphagia, Respiratory | 3 | Transoral | N | 6 |
| El Korbi et al (2018) [30] | 1 | 0.16 | Mass | 3.7 | Transoral | N | 8 |
| Yagasaki et al (2014) [31] | 1 | 2 | Respiratory | NR | Endoscopic | N | 12 |
| Gupta et al (2011) [32] | 1 | 48 | Dysphagia | 3 | Transoral | N | 24 |
| Kuint et al (1997) [4] | 2 | 0.033 | Respiratory | 2 | Transoral | N | 24 |
| | | 0.033 | Respiratory | NR | Transoral | N/A | N/A |
| Leach and Jonas (2018) [33] | 1 | 1.25 | Dysphagia | 4.8 | Transoral | N | 12 |
| Jing et al (2013) [34] | 1 | 2.75 | Mass | 1.4 | Transoral | N | 2.25 |
| Kayhan et al (2017) [35] | 4 | 2 | Respiratory, Dysphagia | NR | Transoral | N | 73 |
| | | 48 | Respiratory, Dysphagia | NR | Transoral | N | 39 |
| | | 144 | Respiratory, Dysphagia | NR | Transoral | N | 41 |
| | | 75 | Respiratory, Dysphagia | NR | Transoral | N | 16 |
| Urao et al (1996) [14] | 3 | 0.75 | Respiratory, Dysphagia | 1.5 | Transoral | N | 60 |
| | | 1.25 | Respiratory | NR | Transoral | N | 48 |
| | | 14 | Incidental | 1.5 | Transoral | N | 48 |

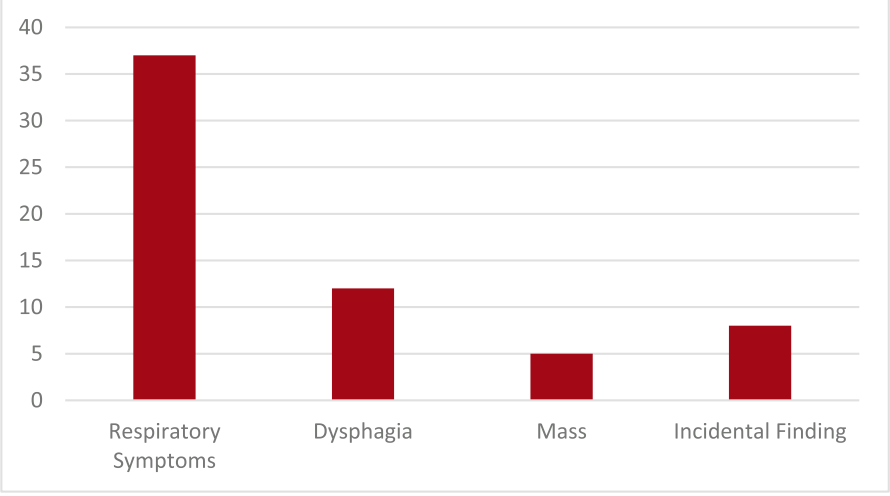


**Fig. 2.** Presenting symptoms of intralingual thyroglossal duct cyst. Each symptom was recorded individually for patients that had multiple presenting symptoms.

at the initial surgery requiring an additional surgery in the immediate postoperative period. Two were reported in Burkart et al. as revisions of procedures (marsupialization and an open procedure) at outside hospitals,[17] and 1 was from the authors' institution where TGDC could not be completely identified on the initial procedure thus necessitating a revision endoscopic procedure within one week postoperatively.

## 4. Discussion

The thyroid primordium develops during the 4$^{th}$ week of gestation, proliferating in the foramen cecum of the developing tongue before descending through the tissues of the neck via the thyroglossal duct. The cranial portion of the duct involutes starting during the 5$^{th}$ week of gestation and continues to obliterate in a cranial to caudal manner as the thyroid tissue migrates caudally to its final position by the 7$^{th}$ week. During the course of normal development, a depression at the foramen cecum is the only remnant of the thyroglossal duct, however, persistence of the duct can lead to the development of cysts, which can be secondarily infected leading to rupture, fistulae or sinuses, as well as airway complications [18].

While most TGDC present in the neck either at the level of (60%), above (24%), or below (13%) the hyoid, some 0.5-2% may present intralingually as a base of tongue lesion

**Table 2.** Risk of Bias Assessment according to Methodological Index for Non-Randomized Studies (MINORS) criteria. Each item scored from 0 to 2. Criteria for comparative studies not included.

| Individual MINORS Items | Clearly Stated Aim | Inclusion of Consecutive Patients | Prospective Collection of Data | Endpoints Appropriate to Study Aim | Unbiased Assessment of Study Endpoint | Follow-up Period Appropriate to Study Aim | Loss to Follow-Up Less than 5% | Prospective Calculation of Study Size | Total Score | Risk of Bias |
|---|---|---|---|---|---|---|---|---|---|---|
| Institutional Cases | 2 | 2 | 0 | 2 | 2 | 2 | 1 | 0 | 11/16 | Moderate |
| Harumatsu et al (2019) | 1 | 2 | 0 | 1 | 2 | 2 | 2 | 0 | 10/16 | Moderate |
| Turhan and Bostanci (2019) | 0 | 0 | 0 | 0 | 1 | 1 | 2 | 0 | 4/16 | High |
| Leach and Jonas (2018) | 0 | 0 | 0 | 0 | 1 | 1 | 2 | 0 | 4/16 | High |
| El Korbi et al (2018) | 0 | 0 | 0 | 0 | 1 | 1 | 2 | 0 | 4/16 | High |
| Kayhan et al (2017) | 2 | 0 | 0 | 2 | 2 | 2 | 2 | 0 | 10/16 | Moderate |
| Carroll et al (2016) | 0 | 0 | 0 | 0 | 1 | 1 | 2 | 0 | 4/16 | High |
| Yagasaki et al (2014) | 0 | 0 | 0 | 0 | 1 | 1 | 2 | 0 | 4/16 | High |
| Jing et al (2013) | 0 | 0 | 0 | 1 | 1 | 0 | 0 | 0 | 2/16 | High |
| Kayhan et al (2013) | 0 | 0 | 0 | 1 | 2 | 1 | 2 | 0 | 6/16 | High |
| Sameer et al (2012) | 1 | 1 | 0 | 1 | 2 | 1 | 1 | 0 | 6/16 | High |
| Gupta et al (2011) | 0 | 0 | 0 | 2 | 2 | 2 | 2 | 0 | 8/16 | Moderate |
| Burkart et al (2009) | 2 | 2 | 0 | 2 | 2 | 2 | 2 | 0 | 12/16 | Moderate |
| Lindstrom et al (2003) | 2 | 0 | 0 | 2 | 1 | 2 | 2 | 0 | 9/16 | High |
| Weldon and Krafcik (2000) | 1 | 0 | 0 | 1 | 1 | 2 | 2 | 0 | 7/16 | High |
| Kuint et al (1997) | 1 | 0 | 0 | 0 | 1 | 2 | 2 | 0 | 6/16 | High |
| Urao et al (1996) | 2 | 0 | 0 | 2 | 2 | 2 | 2 | 0 | 10/16 | Moderate |

[3,4,19,20]. In our systematic review, patients with intralingual TGDC appear to present most often with upper aerodigestive symptoms including respiratory distress (70%) and/or dysphagia (23%). Intralingual TGDC may also be found incidentally (15%). In cases of incidental discovery, it is imperative to follow the patient closely and strongly consider surgery prior to development of symptoms. Once an intralingual TGDC becomes infected or undergoes rapid growth due to an upper respiratory infection or internal hemorrhage, the patient can quickly develop respiratory obstruction and experience respiratory arrest and possible death [21,22].

One infant in our series experienced respiratory demise. The patient had been transferred to our institution intubated from an outside institution after a respiratory arrest in the field with subsequent cardiac arrest and prolonged time until return of spontaneous circulation due to difficulty securing the airway. In this case, intralingual TGDC was discovered on imaging performed for neurologic imaging after the arrest. The intralingual TGDC was successfully removed with an endoscopic procedure and there were no neurological deficits detected after extubation. This life-threatening presentation illustrates the potential emergent nature of presentation with intralingual TGDC and the imperative of early removal to prevent life-threatening complications.

As illustrated by this case, airway management for elective removal of intralingual TGDC can be complex, and these types of procedures therefore should be managed at tertiary care institutions, with pediatric anesthesiologists. For endoscopic or transoral management, nasotracheal intubation is preferred to allow for adequate access to and visualization of the base of tongue [11].

Management of cervical TGDC is usually accomplished with the Sistrunk procedure, first described in the 1920s, which includes removal of the cyst, remnant thyroglossal duct tract, as well as the central portion of the hyoid bone and adjacent base of tongue tissue [23]. This procedure reduced recurrence rates from up to 50% for simple cyst excision to 2-6% [12,13,24]. To accomplish the same principle for intralingual TGDC via an open approach would require a cervical incision, hyoid resection, coring of the base of tongue to the foramen cecum, and subsequent drain placement.

Endoscopic and transoral techniques present appealing minimally invasive options for management of intralingual TGDC, however, there are few head to head studies comparing the two. For children <1-year-old, endoscopic or transoral management is less invasive and less morbid than an open approach. Due to superior visualization, the endoscopic approach is the preferred method at our institution as previously described [11]. Our systematic review found an overall recurrence rate of 1.9% using endoscopic or transoral techniques, which is less than the range documented in the literature for the Sistrunk procedure in cervical TGDC.

In young infants, endoscopic or transoral excision can be challenging, especially if the cyst is not superficial and is deep in the tongue base musculature. At the risk of damaging the lingual arteries, it is safer to stay conservative during endoscopic or transoral excision. This is reflected in one institutional case in our series where remnants were left behind requiring a revision procedure in the perioperative period. This can also lead to recurrence, which occurred in one of our institutional patients 5 months after the initial procedure. Both patients were successfully managed with a subsequent endoscopic procedure without recurrence and without an open procedure required. That the aforementioned TGDC recurrence occurred within 6 months of the initial procedure highlights the importance of following patients who undergo surgical

excision of an intralingual TGDC with serial flexible fiberoptic laryngoscopy, every 3 months, for up to 1 year to monitor for recurrence. The systematic review also demonstrated a 5.7% rate of revision surgery for endoscopic management of an intralingual TGDC underscoring the importance for close surveillance after surgery.

## 5. Conclusions

Intralingual TDGCs represent a rare but potentially life-threatening variant of TGDC, especially in infants. Delay in treatment can lead to catastrophic outcomes including death. Intralingual TGDCs can be managed successfully via transoral or endoscopic approaches with a low recurrence rate. Postoperative surveillance up to one year is recommended.

## Declaration of Competing Interest

The authors have no financial support or conflicts of interest to declare.

## Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.anl.2022.05.014.

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