RESEARCH ARTICLE

An Uncommon Case of Pediatric Lingual Thyroglossal duct Cyst: Diagnostic Challenges and Management

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ABSTRACT

Background: The most prevalent congenital neck masses in children are thyroglossal duct cysts (TGDCs), which are usually found in the midline. Rare congenital neck abnormalities known as lingual thyroglossal duct cysts (TGDCs) start along the thyroglossal tract but manifest strangely at the root of the tongue. Because of their unusual placement and proximity relationship to airway structures, pediatric manifestations provide diagnostic and surgical challenges. **Case Presentation:** While a pediatrician was examining an 11-year-old boy for fever and sore throat, an unexpected diagnosis was made. He was then sent to our hospital to receive more care. Imaging and clinical examination showed a cystic lesion that was compatible with a cyst located along the lingual thyroglossal duct. Thyroid function tests were within normal ranges, and there was no visible neck swelling. Magnetic resonance imaging (MRI) verified the diagnosis. Histopathology verified the diagnosis after transoral surgical excision was carried out. **Conclusion:** Lingual TGDCs are rare and can mostly manifest as intraoral symptoms instead of obvious neck masses. Diagnosis depends on early detection and suitable imaging. The ultimate treatment is still complete surgical excision, albeit in some circumstances a transoral approach may be more efficient and aesthetically pleasing.

Keywords: Lingual thyroglossal duct cyst; clinical examination; magnetic resonance imaging (MRI); transoral; surgical excision.

INTRODUCTION

About 40% of all primary neck tumors and 70% of congenital neck abnormalities are thyroglossal duct cysts (TGDCs), which start during the thyroid gland's development. [(1)] The neck, beneath the hyoid bone, is where 65% of TDCs occur overall. [(2)] They typically appear as soft, cystic neck lumps in the midline that rise and fall during swallowing. Sometimes, the clinicopathological characteristics of TDCs deviate from this typical appearance, particularly in patients with tongue-based TDCs, known as lingual thyroglossal duct cysts (LTDCs), which make up 1% to 2% of all TDCs. [(3)] Because LTDCs typically result in respiratory distress or potentially deadly upper airway blockage, their clinical presentation is different from that of TDCs found in other places. [(4,5)] Often, lingual TGDCs are discovered by accident or show up as nebulous symptoms like a muffled voice, trouble swallowing, or a feeling of a mass in the throat. On rare occasions, they may be confused with other cystic tumors of the oropharynx, such as dermoid cysts, lymphangiomas, or vallecular types. Surgical excision is the last resort after diagnostic imaging techniques such as ultrasonography, CT, or MRI are routinely employed. TGDCs are traditionally treated with the Sistrunk procedure, which removes the cyst and the middle portion of the hyoid bone. Transoral excision, on the other hand, is a safe and effective alternative to lingual TGDCs, particularly in pediatric patients where reduced morbidity and cosmesis are crucial components. We report an uncommon case of an 11-year-old child whose lingual thyroglossal duct cyst was effectively eliminated with transoral surgical excision.

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CASE REPORT

An 11-year-old male boy was referred by his paediatrician due to swelling at the base of his tongue. He had a history of throat soreness and fever, and he consulted a paediatrician. During a regular check, the paediatrician noticed a bulge in the midline of the base of the tongue. There was no history of odynophagia, dysphagia, dyspnoea or aspiration. Following the remission of symptoms, patients were referred to our hospital for further treatment. On throat inspection, a 1x1 cm cystic, rounded, elevated lesion was found at the midline base of the tongue. (figure 1)There was no evidence of inflammation or infection surrounding it. On palpation, there was cystic swelling. It was not tender or compressible. The patient was advised to undergo laryngoscopy to determine the extent. On laryngoscopy, it was well localised at the base of the tongue and epiglottis, with the vallecula free. (figure 2) Magnetic resonance imaging (MRI) revealed a well-defined rounded cystic lesion at the base of the tongue in the midline, causing an indentation of the epiglottis measuring 16.8 x 17.2 x 18.8 mm, with homogenous T1W hypointense and T2W hyperintense contents, no restricted diffusion, and thin peripheral regular enhancement on post-contrast sequences. (figure 3-a,b,c) These findings suggest a benign cystic lesion, most likely a lingual thyroglossal duct cyst (TGDC). The patient and his parents received counselling regarding the lesion and its effects. A transoral surgical excision of the cyst under general anaesthesia was planned when all the information was provided and valid consent was obtained. The cyst was visible in the midline during surgery. (figure 4 and 5) The cyst was completely surgically removed from the base of tongue and sent for histology. (figure 6 and 7) Hemostasis was attained. No sutures were used to close the surgical site; it remained open. Following a successful extubation, the patient was moved to the recovery area. In addition to gargles and a soft, cold, bland diet, he was kept on intravenous antibiotics for two days. Histopathological evaluation demonstrated cystic spaces lined by squamous epithelium. (figure 8 a,b,c,d) During his regular follow up for one year, child was doing well without any recurrence.

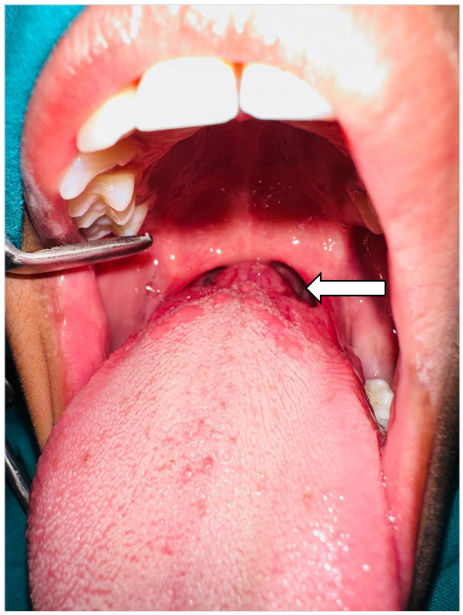

Figure 1: Clinical picture of a cystic swelling at base of tongue in the midline(white arrow).

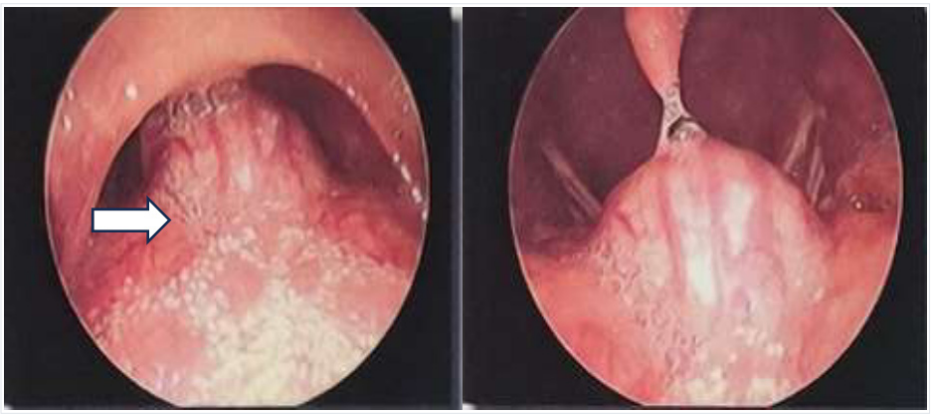

Figure 2: Laryngoscopic view of the lingual thyroglossal cyst.

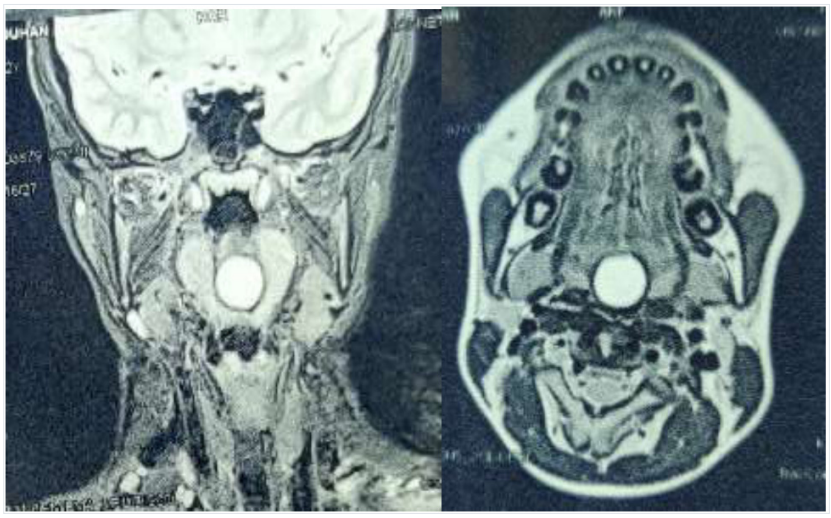

Figure 3a Figure 3b

Figure 3c

Figure 3 (a,b, c): Magnetic resonance imaging (MRI) revealed a well-defined rounded cystic lesion at the base of the tongue in the midline, causing an indentation of the epiglottis measuring 16.8 x 17.2 x 18.8 mm, with homogenous T1W hypointense and T2W hyperintense contents, no restricted diffusion, and thin peripheral regular enhancement on post-contrast sequences. These findings suggest a benign cystic lesion, most likely a lingual thyroglossal duct cyst (TGDC).

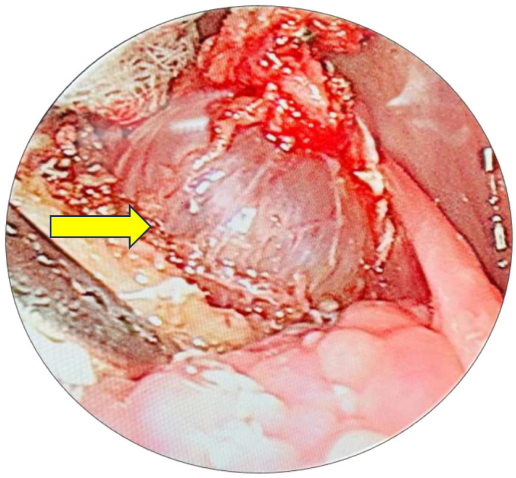

Figure 4: Endoscopic picture of midline cyst at the base of tongue

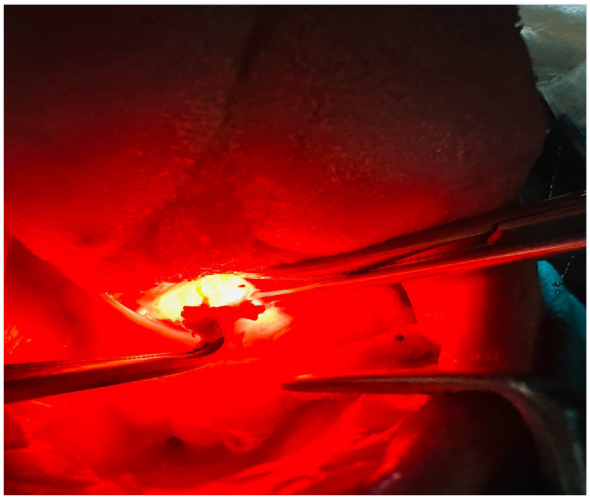

Figure 5: Intra operative transillumination test to see the content of an intact cyst.

Figure 6: Intraoperative picture showing the cyst after detached it from surrounding tissue (tip of forcep towards cyst).

Figure 7: Clinical picture after complete removal of cyst.

Figure 8: Main surgical specimen.

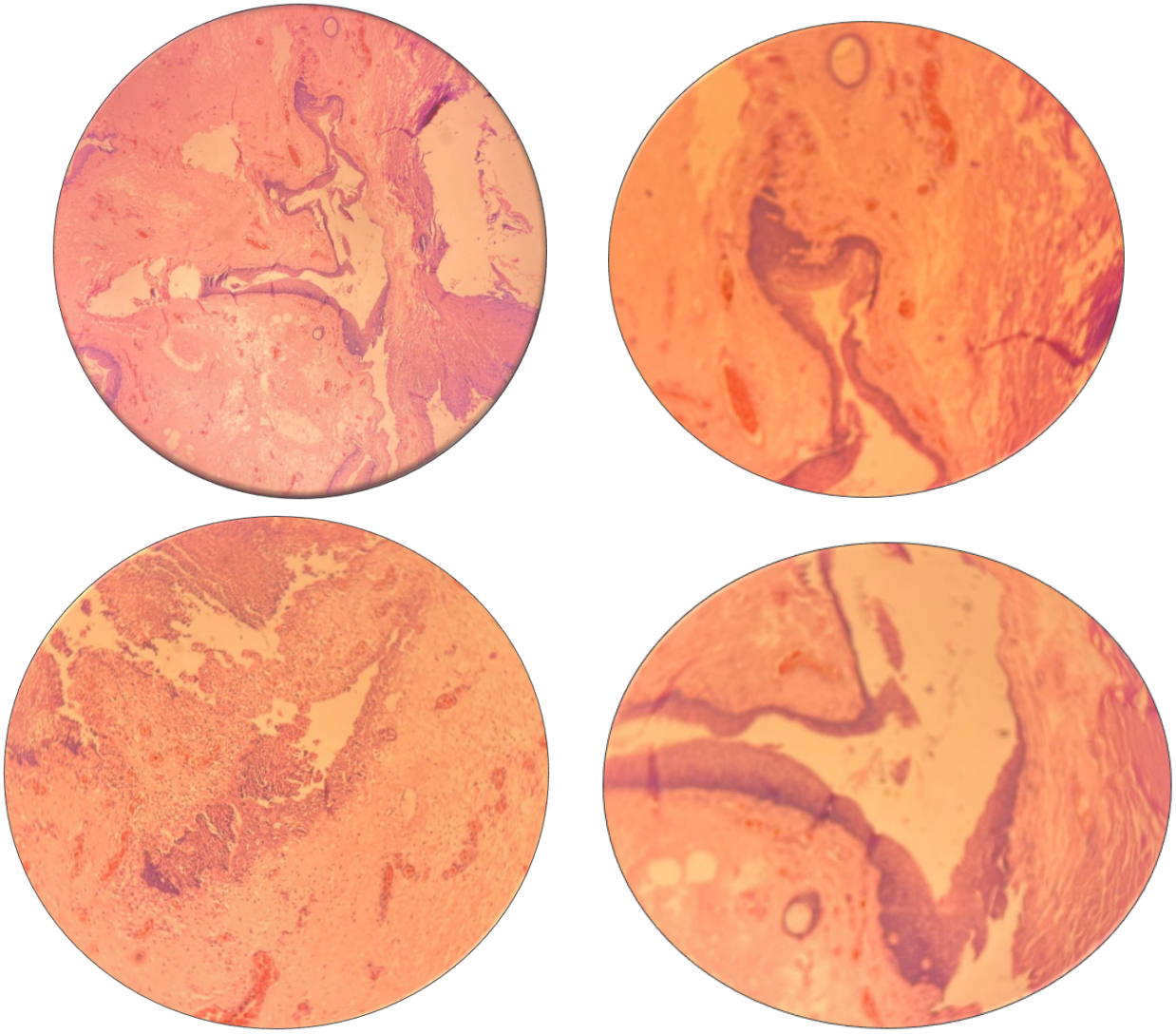

Figure 9 (a,b,c, and d): Histopathological slide pictures.

DISCUSSION

The most prevalent congenital neck abnormality is thyroglossal duct remnants [6]. The thyroglossal duct develops all over growing as the median thyroid anlage moves caudally in the direction of its ultimate pre-tracheal position. Between weeks five and eight of pregnancy, it often obliterates. [6] Remaining thyroglossal ducts can be seen anywhere throughout the tract

and are caused by the incomplete obliteration of this tract during pregnancy. Although the prevalence of thyroglossal duct remnants in the general population is estimated to be up to 7%, intra-lingual TGDC accounts for just 0.6-3% of all thyroglossal duct remnants. [6,7] TGDC is most frequent in the first two decades of life, with just 0.6% diagnosed in people over the age of 60. [7,8] Lingual TGDC can induce respiratory distress, feeding difficulties, obstructive sleep apnoea, or dysphonia, unlike other types of TGDC that manifest as a neck mass or abscess. [7] The thyroid gland develops from a foregut diverticulum in the foramen cecum of the midline tongue base, situated between the tuberculum impar and copula. The gland descends from the midline of the developing neck, anterior to the hyoid bone, and eventually reaches the cervical trachea. The thyroid gland migrates, leaving behind the thyroglossal duct. The thyroglossal duct typically involutes, but may remain for unknown causes, resulting in the development of a central neck cyst called TGDC. [9] Thyroglossal duct anomalies are the most frequent deformity in the neck, with a slight male preponderance. They account for 70% of all congenital cervical masses. Approximately 60% of cases are recognized in childhood, with the majority occurring within the first five years of life. Approximately 5% of patients have atypical presentations. [10] Although midline neck masses are prevalent, 10-24% are situated laterally, primarily to the left. The left-directed thyroidal descending embryological pathway is similar to this. [11–14] Sixty percent of patients have TGDC next to the hyoid bone, twenty-four percent have them between the hyoid and base of the tongue, and thirteen percent have them between the hyoid and pyramidal lobe; only two to three percent have them intralingually. [13-16] The precise numbers may fluctuate amongst series; in some, up to 5% have been observed in a suprasternal site [17] while in others, an unexpected 8.5% have been intralingual. [18] The thyroglossal duct's involution begins at the cranial end, which accounts for the lowest frequency of lingual position in the majority of series.

Although they can be isolated at the base of the tongue (lingual TGDC), TGDC are most frequently located (61%) in the midline of the neck, close to the hyoid bone. There have been reports of a 0.5% to 2% LTGDC rate. [9, 13, 21] Information on these less common forms of TGDC is scarce. The duct initially atrophies from the oral side, where thyroid descent first starts, which may be connected to the low incidence of lingual TGDC. Overall experience with these lesions is rare due to their low occurrence. Even though they could show up with few or no symptoms, LTGDC can have disastrous consequences. [9, 19, 20, 22, and 23] In the current study, the majority of neonates with LTGDC showed no symptoms. Many of our patients were diagnosed as a result of incidental radiography abnormalities. However, it is clear that younger patients are more likely than older ones to experience respiratory compromise. Asking particular questions about sleep issues, exercise intolerance, or dysphagia may yield information from the history, and the evaluation should be guided by these concerns. Questions on weight gain, eating patterns, breathing difficulties, apnea, or cyanotic spells are crucial when dealing with younger patients. Every time, a formal head and neck examination ought to be conducted. In many cases, LTGDC may not be visible physically but can be felt during an oral examination. Direct laryngoscopy viewing of the cyst is the most crucial technique for preoperative evaluation of LTDCs. [24] A mass coming from the oropharyngeal area may be seen by direct laryngoscopy. [25] Using direct laryngoscopy, Li et al. [26] found that LTDCs were most frequently found at the base of the tongue (53.3%), followed by the vallecula and epiglottis (33.3%). However, they were also found on the laryngeal surface of the epiglottis (13.3%); however, this was only seen in adults. It was necessary to distinguish LTDCs from masses originating from the epiglottis because laryngoscopy revealed that the majority of LTDCs in the pediatric group (83.3%) were found at the base of the tongue, whereas 66.7% of the LTDCs in the adult group were found at the posterior part of the base of the tongue.

A tongue base cyst, like an LTGDC, would be appreciated in the majority of cases. It is crucial to ascertain whether the patient has a normal thyroid at the anticipated site prior to surgery when assessing TGDC. Attempting to distinguish the origin of a tongue base mass is crucial during the diagnostic assessment. This anatomical region may have other masses in addition to an LTGDC. A vallecular cyst, dermoid, teratoma, lingual thyroid, lymphangioma, or hemangioma are a few illustrations of these potential conditions.[27] Rarely, benign tumors such as fibromas, papillomas, lipomas and adenomas may appear here. [17]

The papillomas, lipomas, and hemorrhages are all soft. The papilloma is typically pedunculated and pale, the lipoma is lobulated and non-compressible, and the haemangioma is reddish blue, blanching, and compressible. Fibromas are pedunculated, whereas adenomas are embedded, despite the fact that both are hard. In the subjacent floor of the mouth and inferior half of the tongue, lingual TGDC might be mistaken for lymphatic cysts and salivary retention cysts/mucus cysts. Similar to this, dermoid cysts and foregut duplication cysts [18] can sporadically develop on, inside, or beneath the tongue near the thyroglossal descending path.

There will be radiographic evidence of the etiology of many of these lesions. The main diagnostic method to identify the kind of mass present here is direct visualization through laryngoscopy. [27] A thicker submucosal capsule at the midline base of the tongue, immediately posterior to the foramen cecum, gives a lingual TGDC its distinctive look and makes it easy to recognize. Numerous instances of lingual TGDC excision from the base of the tongue without the need for a transcervical tract excision have been documented in the literature. A survey of the literature demonstrates a range of methods, both with and without endoscopic visualization, such as harmonic scalpel, laser, electrocautery, and microdebrider-assisted

marsupialization. A recent report on the structure of TGDCs [28] was published by Hirosawa et al. The duct above the hyoid bone is characterized in their study as a single structure that, as it approaches the foramen cecum, expands out into several ductuli. We hypothesize that atrophy of the distal section of this duct, above the hyoid bone, is the cause of the lack of lingual TGDC extension into the neck. Recurrence would be avoided if any extra ductuli were allowed to freely drain into the oral cavity following marsupialization. The lesion may be localized to the vallecula or the base of the tongue using radiographic imaging; however, lingual TGDCs and vallecular cysts would show comparable enhancement on CT or MRI. It has been demonstrated that CT is a very good diagnostic tool for examining a neck mass. Lingual TGDCs usually manifest as well-circumscribed masses with an enlarged capsule at the base of the tongue that contains 20 to 35 Hounsfield units. [28] When TDCs are examined by MRI, cysts often show low signal intensity on Tl-weighted images and high signal intensity on T2-weighted images. Cysts may also show high signal intensity on TI-weighted images, indicating TGDCs with a high protein content. [29,30] In the end, pathological analysis would differentiate between a lingual TGDC and a vallecular cyst. Practically speaking, people with LTDCs or vallecular cysts must have surgery; however, the kind of procedure differs depending on the type of cyst. While total removal is advised for lingual TGDC, marsupialization is the standard treatment for vallecular cysts.[8,25,31,32] Bai W et al. treated six of the nine lingual TGDC cases with needle puncture and cyst aspiration; two of these cases (33%), experienced recurrence. None of the three that were removed returned. [33] While ethanol ablation of thyroidal cysts has been effective, TGDC has not shown the same level of success. [34] In two cases, Gue´ Rin et al. employed a pharyngotomy and an infra hyoid approach. [16] Burkart et al. recently used transoral full excision without a further Sistrunk surgery to address all patients (original extirpation and recurrences from earlier imperfect excisions) and have not seen any recurrences. [18] We discovered a number of crucial elements in the lingual TGDC approach. The first is a precise diagnosis made using imaging analysis and laryngoscopy. Although ultrasound was useful, an MRI study was the most conclusive imaging method used in this case. The second is that during marsupialization, the entire cyst is well exposed. Third, a thorough preoperative and postoperative examination is necessary to ensure that the ductal system is extended into the neck.

CONCLUSION

Lingual TGDC are expansile lesions that can cause potentially fatal airway blockage, notwithstanding their rarity. It would be advantageous to have a clear consensus on operative management because of the possibility of significant respiratory impairment. This case report covers the typical presentation of this cyst, its clinical and radiographic evaluation, and surgical management at a tertiary care facility. Effective transoral or endoscopic complete excision reduces the risk of complications or recurrence.

COMPLIANCE WITH ETHICAL STANDARDS

The procedure performed in this case report was in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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This study is not funded by any resources.

CONFLICT OF INTEREST

The author (s) declares no potential conflicts of interest with respect to the research, authorship, and/or publication of this paper.

ETHICAL APPROVAL

For the purpose of publishing this case report, the patient's written informed consent was obtained.

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