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The Undescended Thyroglossal Duct Cyst: A Rare Clinical Encounter of Thyroglossal Duct Cyst in the Base of Tongue

İnmemiş Tiroglossal Kanal Kisti: Dil Tabanında Nadir Görülen Bir Tiroglossal Kanal Kisti Klinik Görünümü

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ABSTRACT

The thyroglossal duct cyst (TDC) commonly presents as an anterior neck swelling. TDCs are a congenital disorder that are usually asymptomatic. It rarely occurs the tongue or at the base of the tongue. The patient usually presents with upper airway obstruction symptoms that need immediate attention. We report a case of an infant who presented with respiratory distress symptoms, requiring immediate intubation. The computed tomography scan images showed a well-circumscribed hypodense collection located at the base of the tongue. Direct laryngoscopy and excision of the cyst were performed under general anaesthesia. The infant recovered after the procedure, and there has been no recurrence since surgery.

Keywords: Case report, thyroglossal duct cyst, laryngoscopic excision

ÖZ

Tiroglossal kanal kisti (TDK) genellikle ön boyun şişliği olarak ortaya çıkar. TDK'ler genellikle asemptomatik olan konjenital bir hastalıktır. Nadiren dilde veya dilin tabanında görülür. Hasta genellikle acil müdahale gerektiren üst solunum yolu tıkanıklığı semptomlarıyla gelir. Acil entübasyon gerektiren solunum sıkıntısı semptomlarıyla gelen bir bebek vakasını bildiriyoruz. Bilgisayarlı tomografi tarama görüntüleri dilin tabanında iyi tanımlanmış hipodens bir koleksiyon gösterdi. Genel anestezi altında doğrudan laringoskopi ve kistin eksizyonu yapıldı. Bebek işlemden sonra iyileşti ve ameliyattan bu yana tekrarlama olmadı.

Anahtar Sözcükler: Olgu sunumu, tiroglossal kanal kisti, laringoskopik eksizyon

INTRODUCTION

Incomplete obliteration of the thyroglossal duct is the root cause of thyroglossal duct cyst (TDC), a congenital anomaly. Clinically, 20% to 25% at the level of suprahyoid, 15% to 20% at the hyoid, and 25% to 60% at the infrahyoid level (1). The incidence of TDC at the base of the tongue, or the lingual TDC, is uncommon (2). It is a rare condition and may require urgent interventions. The lesion clinically manifests during infancy, and it commonly presents with obstructive symptoms. Management will require a different approach than the

other TDCs mentioned. We report a case of lingual TDC presenting with acute upper airway obstruction. The infant was subjected to airway, and surgical intervention. The patient responded well to the treatment, and no recurrence was observed.

CASE REPORT

A 1-month-old boy was brought to the casualty by his parents for recurrent choking episodes for the past 2 weeks associated with cyanosis. He has no history of foreign body ingestion, no fever, and no history of prior intubation. Antenatal and postnatal history

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were insignificant. Upon assessment, the patient was stridorous, tachypnoeic, and appeared lethargic with the presence of chest recessions. There was no noticeable neck swelling. He developed type I respiratory failure. The patient was intubated with an appropriate-sized endotracheal tube with no difficulty during the intubation process. He was diagnosed with aspiration pneumonia, and intravenous antibiotics were commenced. Attempts to wean the infant from intubation had failed as he developed stridor and respiratory distress following each extubation.

A computed tomography (CT) scan was performed. Imaging showed a non-enhancing cystic lesion at the base of the tongue, measuring $1.0 \times 0.7 \times 1.1$ cm. No septation or calcification was seen. There was no extension or involvement of adjacent structures (Figure 1a and Figure 1b). The thyroid gland was visualised and appears normal.

He was referred to the ear, nose, and throat team for a formal upper airway assessment. He was subjected to a direct laryngoscopy, examination under anaesthesia, and excision of the lesion. Intraoperatively, a cystic mass was present at the base of the tongue, displacing the epiglottis posteriorly (Figure 2). The cyst was aspirated, yielding one mL of mucoid content. The cyst wall was excised using cold instruments, and the residual cyst wall was cauterised using diathermy (Figure 3). Tracheoscopy showed no abnormality from the glottis until the carina. Post-operatively, the patient was given intravenous dexamethasone 0.1 mg/kg three times daily (TDS) for a total of 3 days. He was successfully extubated three days later without any upper airway obstruction symptoms or respiratory distress.

The cyst wall's reported tissue consisting of stratified squamous epithelium and sporadic mucous glands with scattered lymphoplasmacytic cells. This finding is consistent with TDC. He was discharged one week later without any complications. Upon follow-up, he has no recurrence of his symptoms and is thriving well. A flexible nasopharyngolaryngoscopy examination was performed during the visit, and no residual mass or cystic lesion was seen.

DISCUSSION

TDC is a congenital cervical anomaly that accounts for around 7% of the population worldwide. Embryologically, the thyroid primordium originates from the foramen caecum at the posterior third of the tongue. It descends towards the anterior neck to reach its final position at the pre-tracheal level in the seventh week of gestation (1). During the tenth week of gestation, the thyroglossal duct will involute. Failure of involution will lead to TDC formation (3).

The common site of presentation is an anterior midline neck swelling around the hyoid region (1). The cyst rarely occurs at the tongue base, also termed a lingual TDC. The incidence of lingual TDC is very low, reported as being between 2.1%-8.3%. Burkart et al. (2) reported that the incidence of lingual TDC was around 8.5%, while other studies suggested an incidence of 2.1% (2,4). Clinically, the presentation depends on the lesion's site. TDC located in the midline of the neck is usually asymptomatic. Some may be diagnosed as epidermoid cysts, plunging ranulas, lymphangioma, or recurrent neck abscesses, such as infected TDC (5). Acute obstructive symptoms such as stridor, apnea, dysphagia, and respiratory distress are more commonly seen in lingual TDC. Burkart et al. (2) reported that 44% of the lingual TDC patients presented with upper airway obstruction. In this case, the patient presented with multiple choking episodes with cyanosis. He was intubated for airway protection, and a CT scan was performed, confirming the diagnosis of lingual TDC. In patients with less acute symptoms, nasoendoscopy can be performed to exclude other pathologies, such as laryngomalacia or choanal atresia (3).

Preoperative imaging is essential to ensure the presence of the thyroid gland and to determine the site of the lesion. Ultrasound assists in determining the presence of normal thyroid gland tissue, thereby avoid accidentally removing an ectopic thyroid, which can be mistaken for TDC (6). It is a cheaper modality and is more readily available across many centres, but it requires skillful operators, and cannot rule out other anomalies if present. A CT scan or MRI is beneficial for assessing the anatomy, location, concurrent lesion, and extension of the TDC in preparation for surgery. However, a

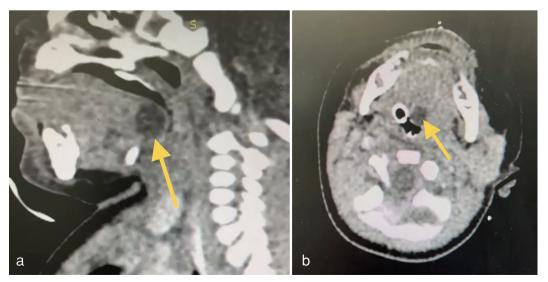


Figure 1. a) and b) a CT scan showing the cystic lesion at the base of the tongue (arrow). *CT: Computed tomography*

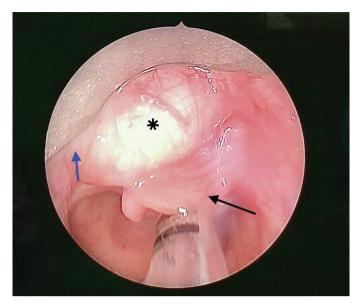


Figure 2. Direct laryngoscopy view demonstrating a cystic mass (asterisk) at the base of the tongue (blue arrow) pushing the epiglottis (black arrow) posteriorly.



Figure 3. Post excision of the base of tongue cystic mass (asterisk) via direct laryngoscopy.

CT scan is more costly, has higher radiation exposure and requires sedation for paediatric patients (7).

Urgent surgical intervention is indicated, especially in lingual TDC cases complicated by acute upper airway obstruction. Endotracheal intubation is required if the patient develops respiratory distress or has an impending airway collapse. Difficult intubation may necessitate other interventions, such as a laryngeal airway mask, needle cricothyroidotomy, or tracheostomy (8).

Sistrunk remains the gold standard surgical procedure for TDC and has the lowest recurrence rate (6). For lingual TDC, management

involves a different approach, and intervention is needed more urgently. Several surgical techniques were described in the literature. In cases of a large cyst that obstructs the airway, puncture and aspiration of the cyst's contents via direct laryngoscopy are performed to reduce the size of the cyst (9). According to Bai et al. (9) this method has effectively treated obstructive symptoms. Close monitoring is required, and if recurrence sets in, a second-look surgery and lesion excision should be performed in a controlled setting. Histopathological examination should be performed to confirm the diagnosis, and malignancy should also be excluded, although the incidence is less than one percent (10).

Another method, described by Urao et al. (11) is endoscopic marsupialisation of the cyst with direct laryngoscopy under general anesthesia. This method is complicated, as suturing the cyst wall is tedious. Endoscopic excision of the cyst using electrocautery, microscissors, or microdebriders is the most effective method (3). The cyst wall needs to be removed completely to prevent recurrence. A study applying this method reported no recurrence was encountered during the follow-up of all patients (2).

In this case, direct laryngoscopy was performed, and cyst excision was done using cold instruments. A tracheoscopy was performed to confirm the absence of other airway pathologies. IV Dexamethasone at a 0.1-0.5 mg/kg TDS dosage for 3 days can be administered to reduce laryngeal oedema post-operatively (12).

CONCLUSION

A lingual TDC is a rare condition that can present as an upper airway emergency. Urgent airway intervention is warranted should the patient develop respiratory distress secondary to upper airway obstruction. Direct laryngoscopy and examination under anaesthesia are a diagnostic and therapeutic method used to secure the airway. A complete cyst wall excision should be performed to prevent recurrence.

Ethics

Informed Consent: Consent was obtained from the parent.

Footnotes

Authorship Contributions

Surgical and Medical Practices: M.H.B.M.S., A.M.A., S.A.B., A.F.J., Concept: M.H.B.M.S., A.M.A., S.A.B., A.F.J., Design: M.H.B.M.S., A.M.A., S.A.B., A.F.J., Data Collection or Processing: M.H.B.M.S., A.M.A., S.A.B., A.F.J., Analysis or Interpretation: M.H.B.M.S., A.M.A., S.A.B., A.F.J., Literature Search: M.H.B.M.S., Writing: M.H.B.M.S., A.M.A., A.F.J.

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