

LINGUAL THYROGLOSSAL DUCT CYST- A CASE REPORT ON ITS SURGICAL MANAGEMENT AND CHANCES OF RECURRENCE

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Abstract: The authors report a rare case of Lingual thyroglossal duct cyst (TGDC) which has been treated by the technique of intraoral wide-base excision and the authors also review the embryology, diagnosis and treatment options of Lingual TGDC. The patient was operated upon by intraoral route and the cyst was excised with a wide margin. There has been recurrence in the past 8 month follow-up even after meticulous removal. The authors urge a review of the practice of excision of Lingual TGDC.

Keywords: Lingual thyroglossal cyst, wide base excision, recurrence, sistrunk's operation.

1. Introduction

The thyroid gland arises as an out-pouching of the primitive foregut around the third week of gestation. It originates at the base of the tongue at the foramen cecum.

Endoderm cells in the floor of the pharyngeal anlage thicken to form the medial thyroid anlage that descends in the neck anterior to structures that form the hyoid bone and larynx. During its descent, the anlage remains connected to the foramen cecum via an epithelial-lined tube known as the thyroglossal duct[1]. The tract usually atrophies and disappears, although the caudal end often remains as a pyramidal lobe. Failure of the tract to involute may leave epithelial remnants or an open area of duct, which may expand into a cyst because of an accumulation of secretions.

The hyoid bone, developing later, may entrap a portion of the thyroglossal duct or draw it caudally, leaving the duct dorsal to the bone. Thyroglossal duct cysts can occur anywhere along the migratory path of the thyroid, although 80% are found in juxtaposition to the hyoid bone[2].

Lingual thyroglossal duct cysts (TGDC) are rare and account for barely 0.6–3% of thyroglossal duct remnant diseases. In these, the cranial part of the embryologic thyroglossal duct/tract persists as a sequestered cyst with/without a variable caudal extent to the pyramidal lobe of the thyroid gland. In comparison to TGDC at other locations, lingual TGDC are more often symptomatic, especially in early infancy[3].

Currently, the primary treatment for TGDC is the removal of the mass by the Sistrunk surgery. However, if

there some of the abnormal tissue remains, even a tiny amount of epithelial tissue, the cyst will recur soon after the operation.

Therefore, it is very important to correctly diagnose these cases to determine the precise locations and ranges of the masses before surgery. It is not difficult to

diagnose a typical TGDC and to determine its location and range; however, if the mass is atypical, such as a lingual cyst, it is likely to be misdiagnosed[4]. Also, the classical Sistrunk procedure may not be applicable for all lingual TGDC[3].

2. **CASE REPORT:** An 11 year old female residing in Silchar, Assam, presented with difficulty in swallowing from the last six months which increased with time and also difficulty in speech and word pronunciation from last six months.



Fig 1: Preoperative picture of the patient

On examination of the oropharynx we find a small translucent globular swelling at the base of the tongue of 1X0.5X0.5 cms dimension.

USG of the neck was done to rule out absence of thyroid or the cyst being a lingual thyroid as in some very rare cases the only thyroid tissue in the body. CT Scan of the neck and tongue base to see the extensions could not be done due to financial constraints of the patient.

3. **OPERATIVE DETAILS:** The patient was put up for surgery and was administered general anesthesia and keeping the patient in the Rose position the cyst was dissected and excised using electrocautery cold surgical

dissection. A wide base of 1mm was also excised. The whole procedure was done under endoscopic guidance.

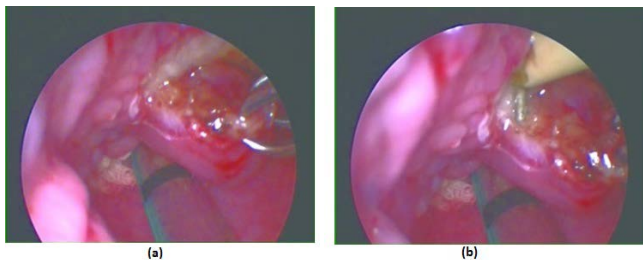


Fig 2: Intraoperative pictures showing wide base excision of the LGTDC.

There was no immediate postoperative complication like bleeding or dysphagia or respiratory distress and the patient started taking food intra-orally in the evening. The patient was discharged the very next day morning and was advised follow-up after one month.



Fig 3: Excised specimen of the LGTDC.

The excised tissue was then sent for histopathological examination the result of which confirmed the diagnosis of Thyroglossal Duct Cyst.

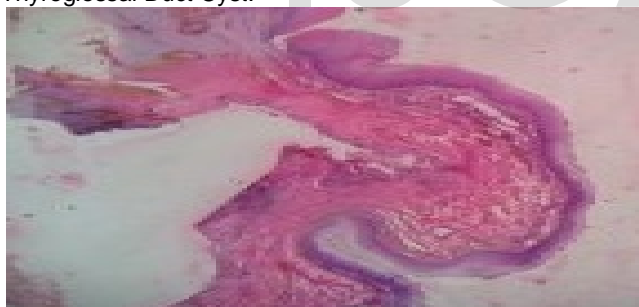


Fig 4: HPE confirms Thyroglossal duct cyst

4. **FOLLOW-UP:** On follow-up there were no complications and the wound was healing well after granulation tissue formation in the excised area.

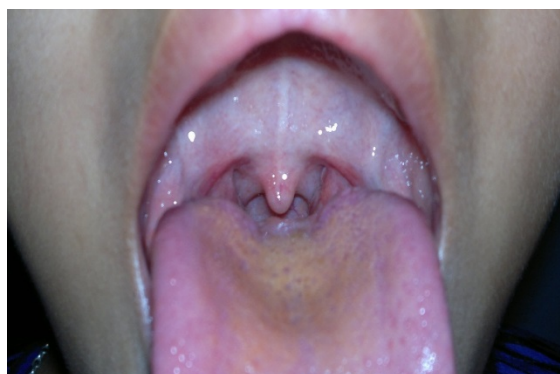


Fig 5: Post operative picture showing healing with normal granulation.

But, 8 months later the cyst recurred exactly in the same area and of same size. The patient was advised for modified Sistrunk's operation which is a combined external and internal approach in a view to remove the TGD track from base of the tongue upto the hyoid bone including part of the hyoid bone. On counseling with the patient regarding the nature of the surgery and cosmetic outcome of the surgery, the patient declined any further surgery and lost follow-up.

5. **DISCUSSION:** The thyroglossal duct is the path of the gradual decline of the thyroid primordium during embryonic development. This structure usually originates from the foramen caecum and terminates at the thyroid isthmus. The research done by Allard et al⁵ demonstrated that TDC is most frequently detected between the hyoid bone and thyroid cartilage (60.9%), above the hyoid bone (24.1%), at the suprasternal fossa (12.9%) or in the tongue (2.1%)[5]. A notable feature in the course of the descent is its proximity to the developing centrum of the hyoid. Ordinarily, the only remnant of the thyroglossal duct is a depression at the foramen caecum; the rest gets obliterated. Persistence of varying parts of the duct from the tongue to the neck leads to cysts; when these cysts get secondarily infected and rupture onto the skin surface, they form fistulas/sinuses[6]. The typical symptoms of TDCs include the absence of anterior midline cystic masses, the sudden occurrence of pharyngeal foreign body sensation, and symptoms of upper respiratory obstruction such as stridor, dyspnea, and dysphagia; these respiratory symptoms are typically worse after colds[5]. The diagnosis of a lingual thyroglossal cyst may be difficult as compared to TGDC in usual locations. The typical case is usually a clinical diagnosis but requires careful scrutiny and judicious investigations to confirm the presence of normally situated and functioning thyroid glands prior to surgery[7]. Intra oral examination and indirect laryngoscopy are useful adjuncts in the outpatient evaluation.

The diagnosis is confirmed by laryngoscopy and CECT of the region. The laryngoscopy shows a pink, cystic, round, smooth-walled, midline, non-pedunculated mass over foramen caecum. The CECT confirms the above finding and shows the mass to be non-enhancing on contrast with low intensity and constricting the air passage. Ultrasound neck or sometimes scintigraphy may be asked for, to confirm the presence of normal thyroid. This avoids excision of the only thyroid tissue present in cases of lingual thyroid. Ultrasound neck was done in our case to confirm the same[8].

Methylene blue is injected into the cyst to trace the distribution of the thyroglossal duct before surgery, which is very useful to show the range of the cyst. In 1920, Sistrunk used methylene blue in the assessment of those patients whose sinuses communicated with the outside a technique that was proven to be useful for tracking the range of a cyst[9]. It is commonly recognized that the Sistrunk surgery (involves a cervical approach with enbloc cystectomy, central hyoidectomy and excision of the entire epithelial tract with the surrounding cone of suprahyoid muscle up to the foramen caecum) is the most effective treatment for TDC. Modified Sistrunk

procedure is a combined intraoral and cervical approach has been reported to be very successful, but is associated with high morbidity and scar. Similarly, cyst excision by midline tongue-splitting incision, by lateral pharyngotomy, transhyoid approach or by mandibulotomy leads to high morbidity which is unwarranted for this benign lesion and therefore, physicians have tried many other surgical techniques, such as cystectomy and marsupialization under endoscopic view. However from our experience even after complete and meticulous removal of the cyst under endoscopic guidance with wide base excision the cyst again recurred after 8 months. And even though various studies as by K.S. Muhammed Sameer et al³ and Collin M. Burkart et al[8] mention very low rates of recurrence of LTGDC after excision, we would like to recommend the Modified Sistrunk approach for complete removal of the thyroglossal tract along with part of hyoid so as to completely prevent recurrence.

6. **CONCLUSION:** Lingual TGDC is an very uncommon variant of TGDC and its proper treatment protocol is rarely mentioned in surgical literature. It is frequently managed by cyst excision. Modified Sistrunk's procedure is suggested to prevent recurrences.

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