Posterior Hyoid Space; A Brilliant Concept in Managing Thyroglossal Duct Cyst (TGDC)

**Ehab M. Oraby (MD);**
Lecturer of general surgery, faculty of medicine, Benha University, Egypt.

**Mohamed I. abdelhamid (MD);**
Lecturer of General Surgery, Faculty of Medicine, Zagazig University, Egypt.

**Taha A. Baiomy (MD);**
Lecturer of General Surgery, Faculty of Medicine, Zagazig University, Egypt.

**Hayam E. Rashed (MD);**
Assistant professor of pathology, Faculty of Medicine, Zagazig University, Egypt.

Correspondence to Ehab Mahroos Oraby, MD, department of general surgery, Faculty of Medicine, Benha University, Fareed Nada street, Benha, Egypt.

Postal code 13518
Tel: 00201003783425

e-mail: ehab.arabi@fmed.bu.edu.eg
       ehaborabyy@yahoo.com
Abstract

**Introduction:** Thyroglossal duct cyst (TGDC) and/or fistula represent the most common congenital anomaly of the neck. A wide range of procedures were described for managing thyroglossal cysts. These procedures are varying from simple cyst excision up to anterior block neck dissection.

**Objectives:** to evaluate the concept of posterior hyoid space according to Maddalozzo modification of Sistrunk operation.

**Patients and methods:** This prospective study was conducted at general surgery departments of Benha and Zagazig University Hospitals. Twenty-eight patients diagnosed to have primary TGDC were included in this study. The surgical technique as described by Maddalozzo et al.

**Results:** Twenty patients (71 %) were complaining of anterior neck cystic swelling and eight patients (29 %) were with neck fistula. After histopathological examination; the tract passed in front of the hyoid bone in all cases (100 %) while ectopic thyroid follicles were detected ventral to hyoid bone in 11 cases (39%), behind the hyoid bone in three cases (11%) and in four cases (14%), found in both ventral and dorsal position

**Conclusion:** Our results were encouraging, and this approach of PHS deserves a wider spread for more assessment of its efficacy and benefits. And also, to evaluate the possible rule of ectopic thyroid tissues in recurrence with classic Sistrunk procedure.

**Keywords:** thyroglossal cysts, posterior hyoid space, Sistrunk operation.
Introduction:

The thyroid gland originates at the foramen cecum and descends anterior to the pharynx to its final position close to the larynx [1]. During the second gestational month; this pathway is divided by the hyoid bone into upper and lower segments. A failure of thyroglossal tract obliteration, or retained epithelial cysts, are the origin of thyroglossal duct cysts (TGDCs) [2]. Knowledge of the path taken by the thyroglossal tract, particularly in relation to the hyoid bone, remains the most crucial factor in determining optimum treatment of TGDCs [3]. Although extensive descriptive data regarding the operative treatment and the pathology of surgical specimens of TGDCs are available today, only limited and contradictory information about the anatomic course of the thyroglossal tract in relation to the hyoid bone is found in the literature [4-7].

The topographic course of thyroglossal tract is a matter of debate. Sprinzl et al [8] used -for the first time ever- serially step-sectioned histologic autopsy specimens for assessment of thyroglossal tract remnants. They reported that 41.3% of specimens (24/58 specimens) revealed remnants of the thyroglossal tract or ectopic thyroid tissue. Four specimens showed complete thyroglossal tracts and ectopic thyroid tissue. The tract remained ventral to hyoid bone in all planes. On the other hand; they found ectopic thyroid follicles alone in 20 specimens. The thyroid follicles were located ventral, dorsal, and a combination of ventral and dorsal in 11, 3 and 6 specimens respectively.

Other authors believed that a persistent thyroglossal duct courses anterior to, and rarely through, the hyoid body, and often has a diverticulum that hooks below and behind the hyoid bone (Fig. 1)
This variation of understanding the embryological development is reflected in the way of how to excise the cyst.

A wide range of procedures were described for managing thyroglossal cysts. These procedures are varying from simple cyst excision up to anterior block neck dissection. Simple cystectomy was associated with a higher recurrence rate [10]. In 1893, Schlange [11] described a procedure that entailed the removal of the mid-portion of the hyoid bone in continuity with the main cyst. In 1920, Sistrunk [12] described his procedure by excision of the cyst in continuity with the central part of the body of the hyoid bone and core of tongue muscle up to the foramen cecum. Sistrunk procedure showed lower recurrence rate (about 4–14 %) and is considered a gold standard for treating thyroglossal cyst [13, 14]. Maddalozzo et al [15] modified Sistrunk approach to include skeletonization of the thyroid cartilage to identify the alae and notch of the cartilage (Fig. 2). The thyrohyoid membrane is then identified and used as a conduit to locate the posterior aspect of the hyoid bone and to evacuate a previously undescribed anatomic area and referred to it as the posterior hyoid space (PHS).

The PHS is outlined inferiorly by the inferior margin of the hyoid, superiorly by the superior margin of the hyoid and thyrohyoid membrane, anteriorly by the posterior surface of the hyoid, and dorsally by the thyrohyoid membrane (Fig. 3). This approach facilitates complete resection of the hyoid and exposing the PHS, allowing evacuation of abnormal tissue from this area. [15].

The aim of this study to evaluate the concept of posterior hyoid space according to Maddalozzo modification of Sistrunk operation.
Patients and Methods:

This prospective study was conducted at general surgery departments of Benha and Zagazig University Hospitals after obtaining approval from local ethical committee and after fully informed written consent signed by patient parents. This study was carried out since April 2013 to November 2016. This duration (about 44 months) allowed for patients’ selection and at least 12 months follow up period from the last case operated upon.

Twenty-eight patients diagnosed to have primary TGDC were included in this study after proper treatment of recent infection to be free of infection for at least 6 weeks before surgery. The diagnosis of a TGDC was made on the basis of a thorough history and physical examination (midline neck mass, which moves with deglutition and with tongue protrusion). Recurrent cases are excluded from this study. Also, cases with other pathology rather than TGDC were retrogradely excluded. Neck ultrasonography was a routine investigation for cyst assessment and to visualize the thyroid gland in its normal position. Routine preoperative laboratory tests and thyroid function tests were also done for all cases.

Surgical Technique:

The surgical technique as described by Maddalozzo et al. [15]. A horizontal incision approximately 4 cm in length was made in a skin crease inferior to the lesion and carried through subcutaneous tissue and platysma. Upper and lower flaps were created in subplatysmal plane. Superiorly the flap was elevated up to the inferior part of the submental triangle of the neck. Inferiorly the flap was
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extended to expose the thyroid cartilage (Fig 3). The strap muscles were retracted along the midline, and dissection was extended to the level of the thyroid cartilage. The alae and notch of the thyroid cartilage were exposed. The thyrohyoid membrane was exposed (Fig 4) and dissection was carried out superficial to it till identifying the posterior aspect of the hyoid bone and the space between the bone and membrane (PHS). The hyoid bone could then be clearly seen and grasped with an Allice clamp and then be transected medial to the tendon of the digastric muscle. Further suprahyoid dissection was carried out to remove a core of tissues up to lingual musculature where the specimen was transected (Fig 5). The tongue defect was repaired, and the wound was drained before closure.

All patients received prophylactic antibiotic. All specimens were examined histo-pathologically to locate thyroglossal tract with or without ectopic thyroid tissues. Also, the relation of these tissues (tract and ectopic thyroid tissues) to hyoid bone.
Results

The study group consisted of 28 patients; 16 males (57%) and 12 females (43%) with mean age 5.3 years (2.9–17.2 years) (Table 1). The history of the disease varied from 3 month to 2.7 years. Twenty patients (71%) were complaining of anterior neck cystic swelling and eight patients (29%) were with neck fistula (Table 2).

After histopathological examination; the tract passed in front of the hyoid bone in all cases (100%) while ectopic thyroid follicles were detected in 18 patients (64%) (Fig 6). Eleven patients (39%) had ectopic thyroid tissues ventral to hyoid bone, behind the hyoid bone in 3 cases (11%) and in 4 cases (14%), found in both ventral and dorsal position. In all cases, during peri-hyoid dissection, we found single tract. On the other hand, suprahyoid dissection was carried as enblock excision of 1 cm core of tissues till lingual musculature, without any trial to skeletonize the suprahyoid portion of the tract. Only two patients (7%) had superficial wound infection, and treated successfully by oral second generation cephalosporin. No other complication or recurrence was reported during the follow-up period (Table 2).
Discussion:

Thyroglossal duct cyst (TGDC) and/or fistula represent the most common congenital anomaly of the neck representing more than 70–75% of congenital midline neck masses. Post mortem examination of adult larynges suggested that TGDC remnants may be present in 7% of the population [15]. Management varies widely from simple drainage, cystectomy, tract excision, tract plus hyoid bone excision up to extended excision or even anterior block neck dissection [10, 11, 12, 17]. Simple cystectomy was associated with high recurrence rate (50%). Schlang [11] initiated hyoid bone excision with significant reduction in recurrence rate to 20%. Sitrunk [12] followed the same Schlang principles and added suprahyoid dissection and he reported more reduction in recurrence rate (4-14%) [18]. Moreover, extended dissection approach was described by Valentina and Fabio but they limited indication of this technique to recurrent cases [17]. Maddalozzo et al [15] found a brilliant pathway between all these modifications of Sistrunk operation. They introduced the concept of posterior hyoid space which was anatomically undescribed area. This concept not only ensures complete tract excision, but also eradication of abnormal ectopic thyroid tissues.

Previously, it was believed that the main cause of recurrence is unexcised residual part of the tract [19]. But even after clear tract excision, still they had a recurrence.
Sprinzl et al. [8], for the first time ever, used serially step-sectioned histologic autopsy specimens. They highlighted the peri-hyoidal distribution of abnormally ectopic thyroid tissues with a constant position of the tract anterior to the hyoid bone. The study of Sprinzl et al. can be considered as a strong support to the concept of posterior hyoid space.

This study evaluated the posterior hyoid space concept, and we found it as a brilliant modification of Sistrunk operation. This approach offers a complete eradication of a thyroglossal duct cyst, tract, hyoid bone and evacuation of the posterior hyoid space with its possible contents of ectopic thyroid tissues and it still a simple easy technique.

In this study, we found the thyroglossal tract at a constant position anterior to hyoid bone. On the other hand, Chandra et al. reported that: in approximately 30% of cases, the tract has been found posterior to the hyoid bone [20].

In this study, we have no recurrence allover 12 months follow up duration. Maddalozzo et al, used the same technique which was described here, reported 1.05% of recurrence in a series of 95 patients. Their results were still showing significant reduction in a recurrence rate if compared with classic Sistrunk operation. As reported by Mondin et al., in their extensive 2008 review, they did a meta-analysis to combine recurrence rates of TGDC after classic Sistrunk operation from 13 reported series involving 950 patients. They reported a recurrence rate of 6.6% [16].

Also, pathological studies reported the constant position of TGDC and tract ventral to hyoid bone with plane of cleavage in between. Moreover, we are also highlighting the important possible rule of ectopic thyroid tissues which were found in the peri-hyoid area. Ectopic thyroid tissues were found in 64% of cases. Its relation with hyoid bone was anterior, posterior and combined anterior and posterior to the hyoid bone in 39%, 11%, and 14% of cases respectively. These results are
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comparable with that reported by Sprinzl et al. They found that ectopic thyroid tissues are 55%, 15%, and 30% in anterior, posterior and combined anterior and posterior relation to hyoid bone [8]. This point of research is still unclear and no available data discussing it. For further evaluation in subsequent researches on a wide scale.

Our results were encouraging, as we had no recurrence over a follow up period of 12 months. The relatively small number of cases in this study is a limiting factor, and this approach of PHS deserves a wider spread for more assessment of its efficacy and benefits. And also, to evaluate the possible rule of ectopic thyroid tissues in recurrent cases with classic Sistrunk procedure.
References:


9. Johan Fagan: The Open Access Atlas of Otolaryngology, Head & Neck Operative Surgery by johannes.fagan@uct.ac.za


Table (1): Demographic data (Original):

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>2.9 – 17.2 (5.3)</th>
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<tr>
<td>Sex</td>
<td>16 males (57%)</td>
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<tr>
<td></td>
<td>12 females (43%)</td>
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Table (2): Clinical and pathological findings (Original):

<table>
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<tr>
<th>Presentation</th>
<th>20 cysts (71%)</th>
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<tbody>
<tr>
<td></td>
<td>8 fistula (29%)</td>
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<tr>
<td>Track: (single or multiple)</td>
<td>All cases had single track (100%)</td>
</tr>
<tr>
<td>Relation to hyoid bone</td>
<td>All tracks were ventral to hyoid bone (100%).</td>
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<tr>
<td>Ectopic thyroid tissues</td>
<td>Detected in 18/28 patients (64%)</td>
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<tr>
<td></td>
<td>11 cases ventral to hyoid (39%)</td>
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<tr>
<td></td>
<td>3 cases dorsal (11%)</td>
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<tr>
<td></td>
<td>4 cases combined ventral &amp; dorsal (14%)</td>
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<tr>
<td>Post-operative complications</td>
<td>2 cases wound infection (7%)</td>
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<td></td>
<td>No recurrence (0%)</td>
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**Figures legends:**

**Fig 1:** Course of Thyroglossal Tract

**Fig 2:** Autopsy & Diagrams showing PHS

**Fig 3:** Surgical steps: 1- incision, 2- subplatysmal plane, 3 raising flaps, 4-strap muscles retraction

**Fig 4:** Exposure of Thyrohyoid membrane

**Fig 5:** Surgical Steps: 1- entering PHS, 2,3- cutting of hyoid bone, 4- suprathyroid dissection

**Fig 6:** Section from ectopic thyroid tissue revealed Thyroid Follicles H&E x200.