# Thyroglossal Duct Cyst - An Observational Study from Visakhapatnam, Andhra Pradesh

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#### **ABSTRACT**

#### **BACKGROUND**

Thyroglossal duct cyst is a developmental cyst that occurs in 7 % of the population. These cysts are most commonly seen in paediatric patients. They occur due to failure of thyroglossal duct to involute and atrophy. Majority of them are found in infrahyoid region. The purpose of this research was to summarise our three years of clinical experience in different features of thyroglossal cysts and their surgical results, with an emphasis on the naked eye extent of a patent thyroglossal duct if present.

#### **METHODS**

This observational study was carried out in the Department of ENT, GVP IHC & MT – Visakhapatnam district, Andhra Pradesh for a period of three years from January 2016 to 2019. In our study, twenty patients were enrolled. Patients with cysts were initially diagnosed based on medical history, clinical examination, and ultrasound sonography (USG) reports.

## **RESULTS**

Patients' clinical and surgical data, including cyst size and position, presence or absence of the thyroglossal duct, and so on, were analysed. The average age was 11 years. The majority (73.5 percent) were under the age of 15. Males accounted for 75 percent of the population, while females accounted for 25 %. Midline neck swelling was found in most of the patients (95 %). Majority (84.5 %) of cysts were located in the sub-hyoid region. Erythema over swelling was seen in 14.5 % of patients. Thyroglossal ducts were found to be patent at various lengths and areas. Majority of patients (75 %) had tract that began from cyst and ended at superior border of hyoid body while two patients (10 %) had patent thyroglossal duct from the cyst to the vallecular mucosa. Majority (70 %) cysts had size between 1.6 cm and 3 cm. Intraoperatively 15 % of cyst got ruptured. Most of them were present with visible midline neck swelling. None of the cysts had malignant characteristics in our study.

### **CONCLUSIONS**

In most cases, a patent duct just disappeared at the superior border of body of hyoid. Complete patent thyroglossal duct from cyst to tongue musculature was rare. None of the cysts had malignant characteristics in our study.

#### **KEYWORDS**

Thyroglossal Cyst, Neck Swelling, Thyroglossal Duct

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## BACKGROUND

Thyroglossal duct cyst is the most common and well recognized midline cervical developmental abnormality which constitutes about 70 % of all congenital neck swellings. It occurs most commonly in paediatric population. The foramen caecum connects the anterior two-thirds and posterior thirds of the tongue, and the thyroglossal duct extends to the thyroid's anatomical resting place from the foramen caecum. After the fifth embryonal week, it normally involutes. The thyroglossal duct remnant or cyst, which is normally located at the level of the hyoid bone, persists due to failure to involute and atrophy. A thyroglossal cyst may form at any point along the duct. The common sites in the order of frequency of occurrence being; between hyoid bone and thyroid cartilage (60 %), suprahyoid (24 %), suprasternal (13 %), intralingual (2 %).

Thyroglossal cyst usually presents as a midline, non-tender, palpable mass that moves with swallowing and elevates on protrusion of the tongue. It is usually 2-4 cm in diameter and gradually increases in size.<sup>3</sup> Recurrence and potential for malignancy are the two main clinical problems. Reported incidence of malignancy in thyroglossal duct cyst is less than  $1\,\%.^4$ 

The thyroglossal duct cysts is lined by pseudostratified ciliated columnar epithelium. Occasionally, the cysts may become devoid of epithelium.<sup>5</sup>

In most of the cases correct diagnosis can be achieved by clinical history and physical examination. Preoperative evaluation must include examination of midline cystic swelling moving up with protrusion of tongue and deglutition. The base of tongue has to be examined for lingual thyroid followed by examination of neck for lymphadenopathy and thyroid gland pathology. Screening of thyroid stimulating hormone (TSH) to evaluate for elevated TSH levels suggestive of hypothyroidism. To confirm the diagnosis imaging plays an important role. Fine needle aspiration cytology (FNAC) findings in thyroglossal duct cyst are variable, but when correlated with clinic-radiological findings, accurate diagnosis can be made in most cases. It's helpful to identify the presence of functioning thyroid tissue in the neck and to detect any possibility of malignant change within the cyst.<sup>6</sup> A preoperative sonographic examination is necessary to determine if the cyst has any unanticipated complications, such as a solid component or a fistula. Computed tomography (CT) and magnetic resonance imaging (MRI) are only used to help delineate the structure of the lesion more precisely.7 A benign thyroglossal cyst shows up on a CT scan as a midline, fluid-attenuated mass near the hyoid bone with a thin, smooth wall.8 But imaging techniques or FNAC alone may not be adequate for pretreatment assessment in all cases of thyroglossal duct cysts. Apart from imaging studies, all patients must be subjected to image-guided FNAC for early and definitive diagnosis of Preoperative evaluation also radionuclide isotope scanning with Technetium - 99 m (Tc-99m)/radioiodine (I - 131) in cases of solid mass: to rule out a median ectopic thyroid, in cases of elevated TSH levels and in cases of absence of normal thyroid gland on ultrasonography. When median ectopic thyroid is present, all of the patient's functional thyroid tissue can be located within the cyst, and its removal would render the patient lifelong dependent on exogenous thyroid hormone. Preoperative informed valid consent should be taken for lifelong thyroid hormone replacement.<sup>9</sup>

Differential diagnosis includes tumours of the pyramidal lobe of thyroid gland, ectopic thyroid tissue, inflammatory or malignant lymphadenopathy, epidermoid cyst and submental dermoid cyst. Thyroglossal cyst mimics plunging ranula.

Surgical treatment for problematic cysts such as recurrent infection or inflammation has long been established and was first described by Sistrunk in 1920. Sistrunk's procedure includes removal of cyst along with a tract, body of hyoid bone and core of tissue along the tract up to the foramen caecum. 10 Recurrence rates with Sistrunk's procedure, when properly performed, are less than 3%, and usually present within one year following excision. 11 Modification of Sistrunk's procedure is commonly practised that involves classic removal of the central hyoid bone avoiding the coring out of the suprahyoid tissues as described by Sistrunk. 12 The perioperative complications include haemorrhage, infection, damage to adjacent structures – superior laryngeal nerve, hypoglossal nerve and lingual nerve; tongue oedema which would require intubation or tracheotomy, Oro-cutaneous fistula when oropharynx is entered through vallecula. Late complications include recurrence 4 % for first operations with Sistrunk, higher if portion of hyoid is not removed. Revision surgery is performed after magnetic resonance imaging to know the extent of tissue to be removed after the first surgery. Oro cutaneous fistula due to post-operative infection. Hypothyroidism due to the median ectopic thyroid removal along with cyst. Follow up regularly with thyroid function tests (TFT) and exogenous thyroid hormone replacement.

### Objective

We wanted to report 3 years clinical experience of thyroglossal cysts and thyroglossal ducts at a semi urban setup in terms of clinical features and surgical findings.

## **METHODS**

This is an observational study which was done in the Department of ENT, GVP Institute of Health Care and Medical Technology, Visakhapatnam district, Andhra Pradesh for a period of three years from January 2016 to January 2019. The research included twenty patients with thyroglossal cysts. Patients were diagnosed with cysts based on their clinical history, diagnosis, FNAC findings, and USG findings. Fresh cases, normal thyroid on USG, and FNAC indicative of cyst were the inclusion criteria. History of previous surgery in neck, any other congenital anomaly were the exclusion criteria.

Modified Sistrunk's procedure was carried out in all patients, in which the cyst, as well as the body of the hyoid and the tract (of whatever length it was) were removed. An incision was made in a skin crease over the cyst or elliptical

incision around draining sinus, without puncturing the cyst. In the subplatysmal plane, superior and inferior flaps are elevated. The superior flap should be raised to around 2 cm above the hyoid bone's body. The infrahyoid strap muscles, which are spread over the cyst's surface, are described. The cyst is exposed after the infrahyoid strap muscles are separated. The body of the hyoid bone is then bared of all soft tissue using a diathermy. The suprahyoid strap muscles (mylohyoid, geniohyoid) are divided, using a diathermy, just above the body of the hyoid and remaining between the lesser cornua of the hyoid to avoid injury to the hypoglossal nerve or lingual arteries. The thyroglossal duct cyst's inferior attachments are separated, and the cyst's deeper part is mobilised with sharp dissection from the thyrohyoid membrane to the hyoid bone. The body of the hyoid bone is divided about 1 cm to each side of the midline with heavy scissors (children) or bone cutter. The suprahyoid thyroglossal duct is dissected with a 2 cm wide core of a tongue tissue (hyoglossus) towards the foramen caecum in continuity with the remainder of the operative specimen, including the hyoid bone. The platysma muscle, as well as the supra and infrahyoid muscles, are approximated in a transverse plane, and the skin is closed over a drain. Careful haemostasis and drainage are particularly important since oozing of blood and post-operative swelling can lead to acute laryngeal obstruction and suffocation. Histopathological examination of the specimen is mandatory in all cases to rule out malignancy. The size and position of the cyst, the presence or absence of the thyroglossal tract, and other surgical findings were reported. Cysts were sent for histopathological analysis.

## **Statistical Analysis**

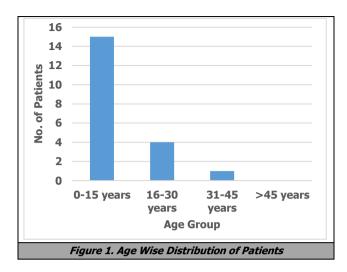
This is an observational study that included histopathology positive data for thyroglossal cyst that was analysed and formulated in tables.

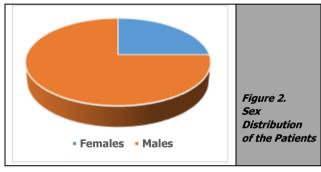
#### **RESULTS**

The average age was 11 years. The vast majority (75 %) were under the age of 15. (Figure 1). Twenty percent of the patients were between the ages of 16 and 30. Males accounted for 75 % of the population, while females accounted for 25 %. (Figure 2). Ninety-five percent of patients had swelling in the midline. In 15 % of patients, there was erythema/redness over the swelling, and others had tenderness. Movement with protrusion of tongue and while swallowing was seen in 100% of patients (Table 1). Cysts were noticed in a variety of locations. The majority of cysts (90 %) were subhyoid in location (Figure 3), with suprahyoid cysts accounting for 10 %. Most of the cysts (95 %) were midline.

For various lengths and locations, thyroglossal ducts were found to be patent (Table 2). The majority of patients (75 %) had a tract that began in the cyst and ended at the superior border of the hyoid body, while 10 % had a patent thyroglossal duct from the cyst to the vallecular mucosa.

One patient (5 %) had a fully patent thyroglossal duct from cyst to base of tongue. In two patients (10 %), a complete absence of tract was observed.





| Sign/Symptom                           | No. of Patients | Percentage |  |
|--|-----------------|------------|--|
| Non tender neck swelling               | 18              | 90         |  |
| Tenderness                             | 2               | 10         |  |
| Erythema/redness over swelling         | 3               | 15         |  |
| Movement with protrusion of tongue     | 20              | 100        |  |
| Movement with swallowing               | 20              | 100        |  |
| Midline swelling                       | 19              | 95         |  |
| Table 1. Clinical Features of Patients |                 |            |  |



| Extend   | No. of Patients | Percentage |  |  |
|--|-----------------|------------|--|--|
| Extension from cyst to base of tongue                            | 1               | 5          |  |  |
| Extension from cyst to vallecular mucosa                         | 2               | 10         |  |  |
| Absence of tract   | 2               | 10         |  |  |
| Extension from cyst and disappearing on superior border of hyoid | 15              | 75         |  |  |
| Table 2. Persistent Thyroglossal Duct                            |                 |            |  |  |
| and Its Course on Surgical Exploration                           |                 |            |  |  |

Cyst size varied in our study. Majority (70 %) of cysts were having size between 1.6 and 3 cm. Only one cyst was seen with size greater than 4.6 cm. Intraoperatively 15 % of cyst got ruptured. We did not encounter any patient having concomitant malignancy.

## **DISCUSSION**

A thyroglossal duct cyst is a congenital anomaly that arises from an epithelial remnant of the thyroglossal duct and is the most common midline neck mass in children. <sup>13</sup> During the eighth and tenth weeks of gestation, the thyroglossal duct typically shrinks and disappears. Failure to involute and atrophy causes a thyroglossal duct to persist as a fibrous cord or a minute epithelial tube, or if the duct persists due to repeated local infection or inflammation, secretion from the epithelial lining can accumulate, resulting in the formation of a thyroglossal cyst. <sup>12</sup>

Majority (75 %) of our patients were under 15 years of age while 20 % patients were in the 16 – 30 age group. This is in accordance with Soni et al. study<sup>13</sup> which also showed increased occurrence of thyroglossal duct cyst in paediatric patients. In our sample, the average age of clinical presentation was 11 years. This was in accordance with a research by Kepertis et al.14 who discovered a mean age of 6.125 years unlike Asmat et al. 15 and Ubayasiri et al. 16 who reported the mean age as 16.7 and 21 years respectively. This demonstrates that thyroglossal duct cysts are most prevalent in children, followed by patients in their second decade. The thinning of adipose tissue during this time of childhood is the most likely cause. It is uncommon in elderly patients, with an estimated incidence of 0.6 percent in the sixth decade. In our study, the thyroglossal duct cyst was noticed commonly in males (75 %). This is in contrast to Allard's study<sup>17</sup> which reported no sex predilection.

Painless neck swelling was the common presentation in majority (90 %) of our patients which was in accordance with Telander and Deane. <sup>18</sup> In contrast to our finding, Moorthy et al. <sup>19</sup> in their study found typical painless swelling only in about 41.6 % of the patients. Only 10 % of our patients presented with painful swelling. In a study by Abuabara, <sup>20</sup> one fourth of adult patients presented with draining sinus that resulted from spontaneous drainage or surgical drainage of abscess. Erythema/redness over swelling was seen in 15 % of our patients.

Thyroglossal duct cysts normally present as a painless, mobile fluctuant cystic swelling near the hyoid bone that moves with deglutition and tongue protrusion. The cyst normally moves cranially with swallowing and tongue protrusion because of its connection to both the hyoid bone and the foramen caecum. In small children, these manoeuvres can be difficult to elicit. In 100 % of the

patients, we observed movement with tongue protrusion and swallowing. In our study 95 % of the cases had palpable midline swelling. This was in contrast to the study by Kepertis et al.<sup>14</sup> who reported only 63.6 % of cases with palpable midline cystic mass.

Thyroglossal duct cyst can be found anywhere in midline from submental region to suprasternal notch. The majority of cysts we found were sub-hyoid (83.3 %), followed by suprahyoid (10 %), suprasternal and over hyoid (3.35 %). The majority of the cysts were found in the midline, with one on the left side. This may be explained by the fact that the levator glandulae thyroideae muscle is ordinarily found on the left. This predominance of sub-hyoid location has been reported in many studies. Kutuya and Kurosaki<sup>7</sup> also reported more cases in infra hyoid region unlike a study by Ali et al.<sup>21</sup> who found suprahyoid region as the most common location. Wadsworth and Siegel<sup>22</sup> found that thyroglossal duct cysts were more common at the level of hyoid. Although we did not find any unusual cyst locations, rare cyst locations such as suprasternal, lingual (foramen caecum), mediastinum, intrahyoid, and intrathyroidal have been recorded in the literature. We performed a naked eye inspection of the remnants and patent thyroglossal ducts during surgical exploration and discovered some interesting findings. In various patients, the thyroglossal duct was patent for varying lengths and regions. A duct was seen coming out of the thyroglossal cyst in the majority of patients (75 %) and vanishing at the superior boundary of the body of the hyoid in two patients (10 %).

A complete patent thyroglossal duct was seen in one patient (5 %) from cyst and entering base of tongue. Complete absent tract was seen in two patients (10 %), this was in accordance with the study by Patigaroo et al. 12 The reason for not seeing whole patent duct in majority of cases is the fact that the thyroglossal duct frequently branches and forms duplications around the hyoid bone which increases the difficulty for the surgeon to recognize all of them. Lingual thyroglossal duct cysts are uncommon variants that appear in the central tongue base. These patients can develop a life-threatening airway obstruction if they are not treated.

Ubayasiri et al. 16 in their case series found that a narrow approach (dissecting close to the thyroglossal cyst, hyoid bone and suprahyoid tissues) rather than a wide approach, as described in Sistrunk's original description, leads to increased recurrence. Removing hyoid body is necessary to significantly reduce recurrence. Nonetheless, current published data showed a recurrence rate of over 4 %, owing to the fact that an updated Sistrunk's procedure is now performed, which does not involve coring out suprahyoid tissue up to the base of the tongue. Instead, simply follow the tract above the hyoid before it breaks off or disappears. As a consequence, the surgeon may be able to leave tissue that contains remnant ducts or branches. Marshall and Becker2 described a recurrence rate of just 1.3 % in their series of 310 cases using classical Sistrunk's technique. To reduce the risk of recurrence, different modifications of the classic surgical technique have been identified. Perkins et al. described a procedure for recurrent thyroglossal duct cyst called "suture guided trans hyoid pharyngotomy," which

involves removing tissue block from the foramen caecum under clear visualisation.  $^{23}$ 

Median cervical fistulas may be caused by the perforation of the thyroglossal duct through the skin, by spontaneous perforation of median thyroglossal cyst due to infection or by surgery. A probe can be introduced into the fistula as far as the body of the hyoid bone. The lesion can be delineated with ultrasonography. The size of thyroglossal duct cysts was reported to range from 0.5 to 6 cm in diameter, but most are between 1.5 and 3 cm. The majority of the cysts in our sample were between the sizes of 1.6 and 3 cm, with 15 % being between 0 and 1.5 cm. In certain rare cases as reported by Mortaja et al.<sup>24</sup> thyroglossal duct cyst was reported as a large neck mass measuring about 6.0 X 6.0 cm.

Recurrence and potential for malignancy are the two main clinical problems. In case of recurrence, revision surgery is performed after magnetic resonance imaging to know the extent of tissue to be removed after the first surgery.<sup>25</sup> Reported incidence of malignancy in thyroglossal duct cyst is less than 1 %. We did not encounter any malignant changes in our patients. Thyroglossal duct cyst carcinomas are usually of two types: thyrogenic carcinoma and squamous carcinoma. Squamous carcinoma forms usually from metaplastic columnar epithelial cells, and thyrogenic carcinomas arise from thyrogenic rests in the duct or cyst wall. Papillary carcinomas are common and usually seen in younger women. These carcinomas are usually asymptomatic and not suspected preoperatively in most instances. **Imaging** techniques such ultrasonography and CT scan alone are usually unable to diagnose malignant disease in thyroglossal duct cyst preoperatively. Although the diagnostic criteria of papillary thyroid carcinoma (PTC) on FNAC have been defined, diagnostic pitfalls in giving definitive diagnosis of papillary thyroid carcinoma arising in thyroglossal duct cyst are very common. FNAC yielded correct results in only 50 - 60% of cases. Definitive diagnosis of PTC could not be made in this case for the following reasons: smaller size of the lesion, suboptimal sample, and dilution by cystic fluid leading to hypocellular smears. So, we feel all thyroglossal duct cyst cases shall be subjected to USG-guided FNAC for early and accurate diagnosis of PTC.<sup>26</sup>

### CONCLUSIONS

Thyroglossal duct cyst is the most common midline congenital neck swelling. It is most commonly seen in paediatric males. In few cases cyst can rupture after repeated infections leading to sinus formation. Majority of them are found in the sub-hyoid region. These cysts are usually 1.5-3 cms in diameter and are asymptomatic. A patent duct simply disappears at the superior boundary of the hyoid body in the majority of cases. On surgical exploration, only a few cysts can rupture.

Data sharing statement provided by the authors is available with the full text of this article at jebmh.com.

Financial or other competing interests: None.

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