

Thyroglossal duct cyst various clinical presentations and modified Sistrunk procedure: A single-center study in India

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Abstract

Background: Thyroglossal duct cysts (TGDCs) are the most common congenital cervical anomalies that account for about 2-4% of all midline neck swellings. They may present as a cyst, abscess, or sinus in the anterior neck region. The cysts are managed by the standard or a modified Sistrunk operation with the least chance of recurrence when compared to the removal of the cyst alone. This study aimed to present our experience in the modified Sistrunk operation over thyroglossal duct cyst anomalies with particular emphasis on the clinical diversity in presentation, surgical technique, and management of postoperative complications.

Methods: This prospective observational study was carried out on 25 newly diagnosed cases of TGDC, which were confirmed histologically and radiologically over a period of 5 years.

Results: All patients underwent a modified Sistrunk operation under general anesthesia and followed up for 2 years. The surgery consisted of complete excision of the cyst with the removal of the central portion of hyoid bone and macroscopically evident duct tissue with intraoperative T-shaped closure of the muscular layer to restore the function of hyoid bone and reduce most postoperative complications, such as recurrence and swallowing difficulties.

Conclusion: The results indicate that the modified Sistrunk operation is one of the best surgical procedures for the treatment of TGDC anomalies limited up to hyoid without lingual extension, especially to prevent a recurrence. More attention should be paid to supra and infra hyoid muscular closure to restore the normal function of the hyoid

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Highlights:

What is current knowledge?

Ventilator-associated pneumonia (VAP) is associated with pathogenic microorganisms in the pediatric intensive care unit.

What is new here?

The most common pathogens isolated from children with VAP are *K. pneumoniae* and *A. baumannii*, which may be a risk factor for death in children with VAP.

Introduction

Thyroglossal duct cysts (TGDC) are the most common congenital cervical anomaly, with a prevalence of 7% in the general population. They are often found as midline neck swellings and account for about 2 to 4% of all neck masses (1). These cysts can be found anywhere along the route of migration of the thyroid from the base of the tongue to the inferior part of the neck.

The thyroid gland is the first endocrine organ to develop in the embryo. It develops from the endodermal epithelium of the median surface of the developing pharyngeal floor during the 4th week of gestation. At the junction of the anterior two-thirds and posterior one-third of the tongue lies the foramen caecum. During the fourth week of gestation, the thyroid anlage, or primordium, develops as a small, solid mass of endoderm. The thyroid anlage descends along the ventral portion of the neck anterior to develop hyoid bone and laryngeal cartilages while maintaining its attachment to the foramen caecum via the thyroglossal duct. However, in 30% of cases, the duct may descend posteriorly to the hyoid, which is very important while treating TGDCs. The thyroid is divided into two lobes connected by isthmus by the end of the 5th week of gestation and reaches its final position inferior to the cricoid cartilage by the 7th week of gestation. The thyroglossal duct starts involuting from the 7th to the 10th intrauterine week. In 50% of individuals, the thyroglossal duct remains as a pyramidal lobe superiorly projecting from the thyroid gland. The patency of the thyroglossal duct anywhere along the course produces TGDC. Even multiple branching from the main duct posterior to the hyoid with blind termination has been reported. The thyroglossal duct is lined by epithelium ranging from squamous epithelium to pseudostratified ciliated columnar epithelium with sometimes walls of salivary or thyroid tissue. The cysts near the foramen caecum

walls are lined by stratified squamous epithelium, whereas those nearer to the thyroid gland are lined by thyroidal acinar cells. Due to repeated local infections or inflammations, epithelial secretions accumulate and form TGDC (2, 3).

Although they are known to be the most common pediatric masses, they also present in adults with varying frequency (4). They present at the level of suprahyoid (20-25%), hyoid (15-20%), and infrahyoid (25-65%) (5, 6).

Thyroglossal duct cysts are often asymptomatic and present as painless, slightly mobile, asymptomatic soft masses in the midline close to the hyoid bone. However, they may present as an abscess or intermittently draining sinus. The thyroglossal duct cyst moves with the protrusion of the tongue and during swallowing. We have divided the TGDC based on the location in relation to hyoid bone such as suprahyoid, infrahyoid, and anterior or posterior to hyoid bone. The differential diagnosis for TGDC is a sebaceous cyst, lipoma, dermoid cysts, branchial cleft cysts, thyroid nodules, ectopic thyroid gland, and lymphadenopathy. Based on the literature, TGDC carcinomas are exceedingly rare and account for only 1% of all TGDC cases. They are usually diagnosed after surgery and have a papillary origin and rarely present as squamous cell carcinoma. Treatment for TGDC is surgical removal with a standard or modified Sistrunk operation. The Sistrunk operation is the gold standard surgery for the excision of the cyst with resection of the duct above the hyoid bone with the portion of the muscle surrounding the foramen caecum. Modified Sistrunk's operation involves the removal of the cyst and/or fistula and resection of the middle portion of the hyoid bone with dissection above it, only with macroscopic evidence of duct epithelium (7).

This study aimed to present our experience in the modified Sistrunk operation over TGDC anomalies with particular emphasis on different heterogeneity of presentation, surgical technique, and postoperative complications including wound infection, recurrence, and swallowing difficulties. In this case series, different types of TGDC/fistula were successfully managed by the modified Sistrunk operation, without recurrence, by performing a T-shaped closure of suprahyoid and infrahyoid muscles after the removal of the hyoid body to restore its function and reduce post-operative complications.

Methods

This prospective observational study was done in the postgraduate department of ENT& HNS of Narayanan Medical College (Nellore, India) between 2015 and 2020, with a follow-up period of 2 years postoperatively. The study protocol was approved by the Narayana Medical College.

New clinically and radiologically diagnosed cases that had been histopathologically confirmed as TGDC were enrolled. Intralingual extension cases and those with other congenital anomalies of the head and neck as well as recurrent cases were excluded from the study.

The patients' complaints ranged from painless midline swelling, discharging sinuses, and difficulty in swallowing. All patients were diagnosed based on their clinical history and physical examination. Ultrasound-guided fine needle aspiration cytology (FNAC), contrast-enhanced computed tomography (CECT) of the neck, and thyroid profile were done to assess the location, extent, and normal functioning of the thyroid gland. The locations of swellings or sinuses were in the midline at the suprahyoid, subhyoid, and anterior or posterior to the hyoid bone.

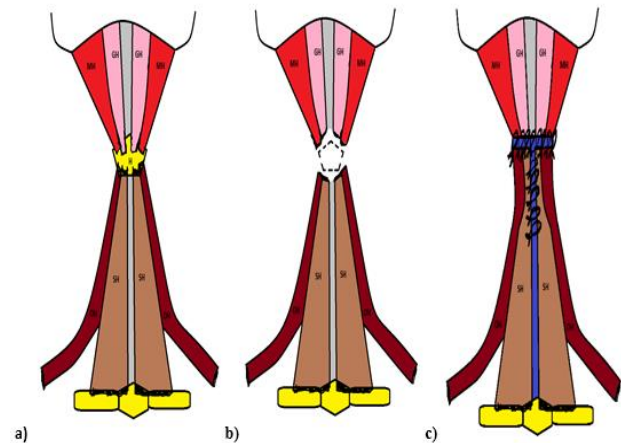


Figure 1. Diagrammatic sequences of the T-shape muscular closure after removal of central portion of hyoid bone. a) Normal hyoid bone attachment of GH– MH muscles above and SH-OH muscles below. b) Posthyoid bone removal status of suprainfra muscular tendinous insertions. c) Vertical sutures done in between right and left of SH-OH muscles and horizontal sutures done between tendinous insertions of above GH-MH muscles and below SH-OM muscles (H: hyoid bone body; MH: mylohyoid; GH: geniohyoid; SH: sternohyoid; OM: omohyoid).

All patients underwent a modified Sistrunk operation performed by a single surgeon. The postoperative care routine was done with neck drain and therapy with antibiotics, anti-inflammatory, and antacid drugs for two weeks.

In all the cases, T-shaped muscular closure was done by suturing the tendinous insertion of counteracting tendons of geniohyoid–mylohyoid above and omohyoid–sternohyoid muscles below to replace the hyoid bone function (Figure 1). This technique plays an important role in preventing post-operational long-term complications, such as difficulty in swallowing, speech difficulty, snoring, and obstructive sleep apnea, which was not mentioned in earlier studies.

Results

The study included 25 patients (4 females and 21 males) aged between 7 and 70 years (Table 1). Of these patients, 22 presented with midline neck swelling, while 3 presented with discharging sinuses. Previous incisions and drainage had not been done in any of our cases. All patients underwent a modified Sistrunk operation under general anesthesia. There was no intra-operative or immediate postoperative complication. None of the patients experienced recurrence after the surgery during the 2-year follow-up. Table 2 shows a summary of the epidemiological and clinical presentations of patients with TGDC.

Table1. Age and sex distribution of patients

Age group (years)	Number of patients	Males	Females	Percentage
7 – 20	13	12	1	52%
20 – 40	8	6	2	32%
40 – 60	3	2	1	12%
>60	1	1	0	4%

The majority of TGDCs were located in the subhyoid region (Table 3). Figure 2 shows preoperative images of a large TGDC in an elderly female. Figure 3 shows CECT images of a patient with TGDC. Figures 4 and 5 show intraoperative images of TGDC in a female and a discharging sinus.

Table 2. Summary of the epidemiological and clinical presentation of patients with TGDC

Symptoms and clinical features	Number	Percentage
Painless swelling	25	100%
Midline swelling	22	88%
Movement with protrusion of tongue	25	100%
Movement with swallowing	25	100%
Erythema	3	12%
Discharging sinus	3	12%
Difficulty swallowing	1	4%

Table 3. The location of TGDCs in relation to the hyoid bone

Location	Number	Percentage
Sub hyoid	22	88%
Anterior to hyoid	2	8%
Posterior to hyoid	2	8%

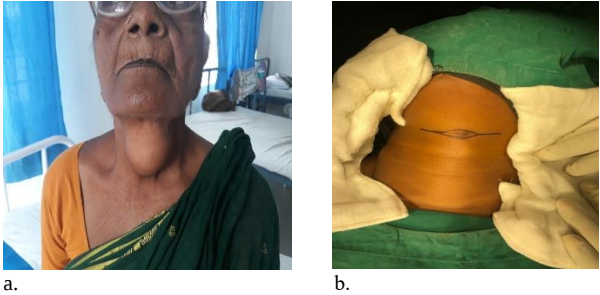


Figure 2. a) Preoperative image of a huge TGDC in an elderly female. b) Preoperative image of a TGDC discharging sinus

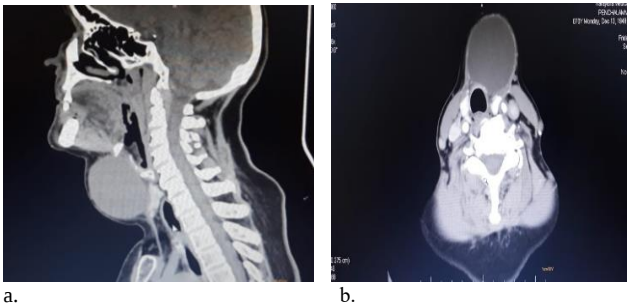


Figure 3. CECT image showing sagittal (a) and axial (b) cuts of TGDC



Figure 4. Intraoperative images of TGDC in an elderly female

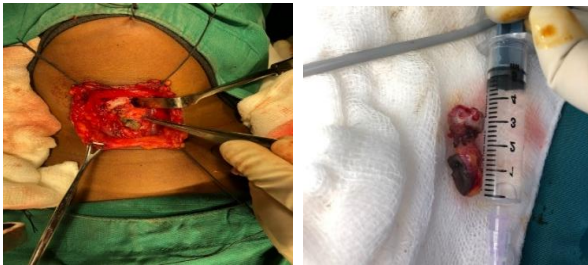


Figure 5. Intraoperative images of TGDC discharging sinus

Discussion

This was a prospective study of TGDC excision using a modified Sistrunk operation over a period of 5 years with a 2-year follow-up. The study evaluated the T-shaped closure of the muscular layer to help maintain the hyoid bone function and reduce post-operative complications. Overall, in our study, the post-operative complications were very low and there was no recurrence.

It has been demonstrated that TGDCs account for more than 75% of midline congenital swellings of the neck. Usually, TGDCs are diagnosed in childhood more than half are diagnosed in the second decade of life few in adulthood (8).

As mentioned earlier, TGDC usually presents as a painless midline swelling in the neck. In our study, all patients had a painless midline swelling, which moved on protrusion of the tongue, and 3 patients presented with erythema and a discharging sinus, while a single patient presented with difficulty swallowing. In a study by Patigaroo et al., 90% of patients presented with painless swelling (9).

In our study, the prevalence of male patients was higher than females and the majority of them were in the 7-20 years age group. In a meta-analysis by Allard et al. on 1,316 patients with TDCs or fistulae, 31.5% of the patients were under 10 years of age, 20.4% were in their 2nd decade of life, 13.5% were in their 3rd decade, and 34.6% were older than 30 years (10).

In the present study, the most common location of TGDC was in the subhyoid region (88%), but in one patient, TGDC was found to have an extension both anterior and posterior to the hyoid with a bilobed presentation (Figures 6 and 7) which is rare.

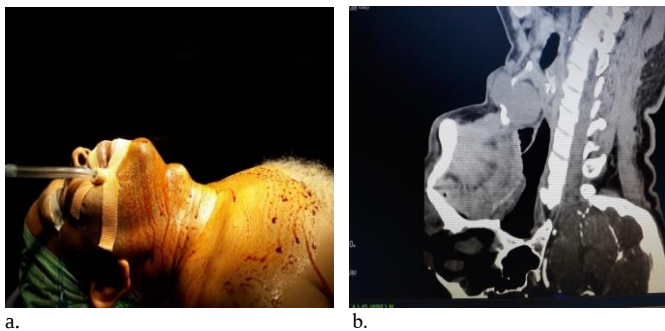


Figure 6. Preoperative image of bilobed TGDC (a) and the CECT image of the neck of bilobed TGDC (b)

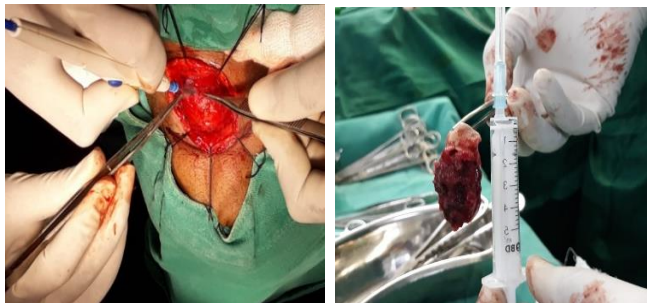


Figure 7. Intraoperative images of bilobed TGDC

Usually, TGDCs appear as midline cervical masses that lie either directly above the hyoid bone or just below it. However, their location may vary; approximately one-third of TGDCs may present as submental or low-level cervical masses. Less than 1% are located off the midline (10-13).

In our center, all patients underwent a modified Sistrunk operation with the removal of the hyoid bone and without removing the above until the foramen caecum. Initially, TGDCs were treated by simple excision or incision and drainage, which resulted in recurrence rates of greater than 50% (14). In 1893, Schlange (15) proposed the excision of the cyst and central body of the hyoid bone, thereby reducing recurrence to 20%. In 1920, Sistrunk first described the procedure that is commonly performed today for the excision of TGDCs. It was based on the principles of embryology that were known at that time and included resection of the central hyoid, including a cuff of tongue musculature toward the foramen caecum, and has proven to be the most efficacious way of eliminating recurrences. The effectiveness of the Sistrunk procedure is most evident because, when it is properly performed, recurrence rates are extremely low i.e. less than 3% (16, 17).

Arda et al. (18) found no statistical difference between the standard and modified Sistrunk operations, and no risk factor for recurrence was identified. Pucher et al. (19) concluded that the modified Sistrunk operation is the preferred surgery over central neck dissection in the pediatric population with uncomplicated TGDC.

A patent thyroglossal duct was not visualized in any of our cases. Complete excision of the duct macroscopically visible and resection of the hyoid bone was done (Figure 8).



Figure 8. Intraoperative images of the T-shaped muscular closure after removal of the central portion of the hyoid bone

This maintains the lever action between the supra- and infrahyoid muscles, which prevents the back prolapse of the tongue and decreases the long-term complications, including dysphagia, snoring, and obstructive sleep apnea. All biopsy specimens were also sent to histopathology and confirmed (Figure 9).

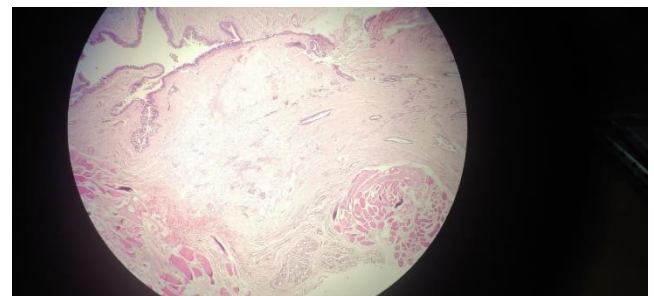


Figure 9. Microscopy and histopathology image of a TGDC. Scanty cellularity is composed of ciliated columnar epithelial cells and tiny clusters of thyroid follicular epithelial cells. Occasional cyst macrophages were noted against a thin colloid mixed hemorrhagic background

The intraoperative and postoperative period was uneventful, and there was no complication. Generally, the most commonly encountered complication is recurrence, which was reported to have a 10% incidence rate (20). According to the mentioned study, the reasons for recurrence were incomplete excision, intraoperative rupture, surgical proficiency and experience, and the presence of infection. Similar to our findings, a retrospective study by Maddalozzo et al. (21) also reported no recurrence.

A laryngotracheal injury and hypoglossal nerve injury are very rare and potentially devastating complications of the Sistrunk procedure. Patients were regularly followed for 2 years postoperatively except for a patient who was followed for only 18 months.

Ultrasound of the cyst is the first modality in suspicious cases of neck swelling. The echogenicity of the cyst ranges from hypoechoic to heterogeneous pattern with internal septa. The thick walls of the cyst represent the ongoing inflammation of the cyst (22). In our study, we found cysts with both hypoechoic, calcifications as well as hyperechoic on ultrasonography. In a study by Yang et al., FNAC showed a positive rate of 53% and a false-negative rate of 47% due to hypocellularity results from the dilution by cystic contents (23).

In our study, all patients underwent FNAC to establish the composition of the cyst, such as papillary carcinoma. None of the patients had any papillary carcinoma composition, which correlated with the histopathology of the specimen postoperatively. In addition, CECT was performed to determine the extent and size of the TGDC. In a study done by Barton et al., a mural nodule and calcification were suggestive of TGDC carcinoma (23).

Conclusion

The results of this prospective observational study indicate that the modified Sistrunk operation is one of the best surgical procedures for the treatment of TGDC anomalies limited up to hyoid without lingual extension, especially to prevent a recurrence. More attention should be paid to supra and infra hyoid muscular closure to restore the normal function of the hyoid bone.

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Ethical statement

The study protocol was approved by the Ethics Committee of Narayana Medical College, India.

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Author contributions

RB conceived and designed the experimental design, contributed to drafting and revising the article. Shamsheer and IR. D are involved in patient recruitment and follow-up. All authors read and approved the final version of the manuscript.

References

- Kurt A, Ortug C, Aydar Y, Ortug G. An incidence study on thyroglossal duct cysts in adults. *Saudi Med J*. 2007;28(4):593-7. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Chou J, Walters A, Hage R, Zurada A, Michalak M, Tubbs RS, et al. Thyroglossal duct cysts: anatomy, embryology and treatment. *Surgical and Radiologic Anatomy*. 2013;35(10):875-81. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Hillary S, Balasubramanian SP. Anatomy of the thyroid, parathyroid, pituitary and adrenal glands. *Surgery (Oxford)*. 2017;35(10):537-41. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Shah R, Gow K, Sobol SE. Outcome of thyroglossal duct cyst excision is independent of presenting age or symptomatology. *Int J Pediatr Otorhinolaryngol*. 2007;71(11):1731-5. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Ross J, Manteghi A, Rethy K, Ding J, Chennupati SK. Thyroglossal duct cyst surgery: A ten-year single institution experience. *Int J Pediatric Otorhinolaryngol*. 2017;101:132-6. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Thompson LD, Herrera HB, Lau SK. A Clinicopathologic Series of 685 Thyroglossal Duct Remnant Cysts. *Head Neck Pathol*. 2016;10(4):465-74. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Zerilli M, Scarpini M, Bisogno ML, Caterino S, Di Giorgio A, Flammia M. [The surgical treatment of thyroglossal duct cysts and fistulae]. *Ann Ital Chir*. 1993;64(6):659-63. Italian. [View at Publisher] [Google Scholar] [PMID]
- Mondin V, Ferlito A, Muzzi E, Silver CE, Fagan JJ, Devaney KO, et al. Thyroglossal duct cyst: personal experience and literature review. *Auris Nasus Larynx*. 2008;35(1):11-25. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Patigaroo SA, Dar NH, Jallu AS, Ahmad R. Thyroglossal Duct Cysts: A Clinical surgical Experience. *Indian J Otolaryngology Head Neck Surg*. 2017;69(1):102-107. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Allard RH. The thyroglossal cyst. *Head Neck Surg* 1982;5:134-46. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Guarisco JL. Congenital head and neck masses in infants and children, II. Ear Nose Throat J. 1991;70(2):75-82. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Koempel JA, Maddalozzo J. Evaluation of head and neck masses. *Indian J Pediatr*. 1997;64(6):771-6. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Pelausa ME, Furte V. Sistrunk revisited: a 10-year review of revision thyroglossal duct surgery at Toronto's Hospital for Sick Children. *J Otolaryngol*. 1989;18(7):325-33. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Schlange H. Über die fistula colli congenita. *Arch Klin Chir*. 1893;46:390-2. [View at Publisher] [Google Scholar]
- Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. *Ann Surg*. 1930;92:57-66. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Sistrunk WE. Techniques of removal of cysts and sinuses of the thyroglossal duct. *Surg Gynecol Obst*. 1928;46:109-12. [View at Publisher] [Google Scholar]
- Arda MS, Ortega G, Layman IB, Khubchandani NA, Pichardo MS, Petrosyan M, et al. Sistrunk vs modified Sistrunk procedures: Does procedure type matter?. *J Paediatric Surg*. 2021;56(12):2381-4. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Pucher B, Jonczyk-Potoczna K, Kaluzna-Mlynarczyk A, Kurzawa P, Szydlowski J. The Central Neck Dissection or the Modified Sistrunk Procedure in the Treatment of the Thyroglossal Duct Cysts in Children: Our Experience. *Biomed Res Int*. 2018. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Amos J, Shermetaro C. Thyroglossal Duct Cyst. *Stat Pearls Publishing; Treasure Island (FL):2021*. [View at Publisher] [Google Scholar] [PMID]
- Maddalozzo J, Venkatesan TK, Gupta P. Complications associated with the Sistrunk procedure. *Laryngoscope*. 2001;111(1):119-23. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Kutuya N, Kurosaki Y. Sonographic Assessment of Thyroglossal Duct Cysts in Children. *JUM*. 2008;27(8):1211-9. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Yang YJ, Haghir S, Wanamaker JR, Powers CN. Diagnosis of papillary carcinoma in a thyroglossal duct cyst by fine needle aspiration biopsy. *Arch Pathol Lab Med*. 2000;124(1):139-42. [View at Publisher] [Google Scholar] [DOI] [PMID]
- Barton F, Branstetter, Jane L Weissman, Thomas L Kennedy, Whitaker Mark. The CT appearance of thyroglossal duct carcinoma. *AJNR Am J Neuroradiol*. 2000;21(8):1547-50. [View at Publisher] [Google Scholar] [PMID]

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