

Synchronous Thyroid Carcinoma and Thyroglossal Duct Carcinoma Arising from an Endolaryngeal Thyroglossal Cyst: A Case Report

Senkron Tiroid Karsinomu ve Endolaringeal Uzanımlı Tiroglossal Duktus Kisti Karsinomu Olgusu

¹ Mehmet Ekrem ZORLU^a, ² Abdulhalim AYSEL^a, ³ Onur ÇORAKÇI^a, ⁴ Uğurtan ERGÜN^a,
⁵ Çiğdem ÖZKAN^b, ⁶ Özden ÖZ^c, ⁷ Togay MÜDERRİS^d

^aDepartment of Otorhinolaryngology, Head and Neck Surgery,
University of Health Sciences İzmir Bozyaka Training and Research Hospital, İzmir, Türkiye

^bDivision of Endocrinology and Metabolic Diseases,
University of Health Sciences İzmir Bozyaka Training and Research Hospital, İzmir, Türkiye

^cDepartment of Pathology, University of Health Sciences İzmir Bozyaka Training and Research Hospital, İzmir, Türkiye

^dDepartment of Otorhinolaryngology, Head and Neck Surgery, İzmir Bakırçay University Faculty of Medicine, İzmir, Türkiye

ABSTRACT Although thyroglossal duct cysts (TGDC) are the most common congenital neck masses, their intralaryngeal extension is quite rare, and there are 24 cases reported in the literature. In intra-laryngeal extension, patients may present with symptoms such as hoarseness, dyspnea, and dysphagia, and it is challenging to differentiate endolaryngeal TGDCs from laryngeal cystic masses such as saccular cysts and laryngoceles. Also, malignancies arising from TGDCs are uncommon, and there is no clear consensus on their treatment. This study aimed to emphasize that TGDCs may also be present in the differential diagnosis of cystic laryngeal lesions by presenting our case of TGDC papillary carcinoma with intralaryngeal extension in a 37-year-old female patient.

ÖZET Tiroglossal duktus kistleri (TGDK) konjenital boyun kitlelerinden en sık görüleni olmakla birlikte intralaringeal uzanımı oldukça nadir olup, literatürde bildirilen 24 olgu bulunmaktadır. İntralaringeal uzanımlı olgularda ses kısıklığı, yutma güçlüğü ve nefes darlığı görülebilmekte olup laringesel ve sakküler kistlerle karışabilmektedir. TGDK'den kaynaklı kanserler ise yine oldukça nadir olup, tedavisi konusunda net bir görüş birliği bulunmamaktadır. Biz bu çalışmada, 37 yaşında kadın hastada intralaringeal uzanımlı TGDK papiller karsinomu olgumuzu sunarak, laringeal kistik lezyonların ayırıcı tanısında TGDK'nin de olabileceğini vurgulamayı amaçladık.

Keywords: Thyroglossal duct cyst; papillary thyroid cancer

Anahtar Kelimeler: Tiroglossal duktus kisti, papiller tiroid kanseri

Thyroglossal duct cysts (TGDCs) are the most common cause of congenital cysts in the midline of the neck, and they occur due to incomplete involution of the thyroglossal canal.¹ They usually present with painless swelling or fistula in the 2nd or 3rd decade. On

the other hand, TGDCs with intra-laryngeal extension are extremely rare, and only 24 patients have been reported in the literature.² In the presence of intra-laryngeal extension, patients may present with symptoms such as hoarseness, dyspnea, and dysphagia.³

Correspondence: Mehmet Ekrem ZORLU

Department of Otorhinolaryngology, Head and Neck Surgery,
University of Health Sciences İzmir Bozyaka Training and Research Hospital, İzmir, Türkiye
E-mail: m_ekrem_zorlu@hotmail.com



Peer review under responsibility of Journal of Ear Nose Throat and Head Neck Surgery.

Received: 10 Apr 2022

Received in revised form: 21 Jun 2022

Accepted: 11 Sep 2022

Available online: 15 Sep 2022

1307-7384 / Copyright © 2022 Turkey Association of Society of Ear Nose Throat and Head Neck Surgery. Production and hosting by Türkiye Klinikleri.
This is an open access article under the CC BY-NC-ND license (<https://creativecommons.org/licenses/by-nc-nd/4.0/>).



FIGURE 1: Cystic lesion (arrow), 29×33×33 mm in size, starting from the left pyriform sinus, pushing the epiglottis medially, extending to the posterior of the hyoid bone in the inferior section, and a well-circumscribed solid lesion measuring 10 mm with calcification in the right periphery of the cyst. Axial (a), coronal (b), sagittal (c) computed tomography scans with contrast enhancement.

Malignancies arising from a TGDC are also uncommon. TGDC carcinomas (TGDCa) make up less than 1% of all TGDCs, and papillary-type thyroid cancer is seen over 90%.⁴ In this study, we present a case of TGDC papillary carcinoma originating from a TGDC with intra-laryngeal extension, which, as far as we know, the first encountered in the literature.

CASE REPORT

A 37-year-old female patient was admitted to our clinic with nearly one year of hoarseness, dysphagia and dyspnea. The patient had no systemic disease except mild obesity. Fine needle aspiration biopsy (FNAB) had been taken from the 25 mm nodule in the left thyroid, which was followed up by another surgery clinic, once in 2014 and twice in 2018. All FNAB results were reported as benign and the patient, had no familial thyroid cancer and radiotherapy history. On flexible nasopharyngeal laryngoscopy, a submucosal mass extending into the left aryepiglottic fold was observed. Computed tomography (CT) of the neck revealed a cystic lesion, 29×33×33 mm in size, and a well-circumscribed solid lesion measuring 10 mm with calcification in the right periphery of the cyst (Figure 1).

The cyst considered as a TGDC because of its proximity with the hyoid bone was removed with the central portion of the hyoid bone by the Sistrunk procedure (Figure 2). We completed the surgery without the need for tracheotomy and without breaching mucosa of the pharynx. The patient's complaints of



FIGURE 2: The cyst started from the thyroid notch and extended from the posterior of the hyoid to the preepiglottic space and pharyngeal mucosa. Before the cyst (arrow) removal during the Sistrunk procedure.

hoarseness and dysphagia improved on the first post-operative day.

Pathological examination of the cyst revealed a TGDC, and the adjacent solid lesion was a follicular variant of papillary thyroid carcinoma (Figure 3). Thereupon, FNAB was repeated from the 2 cm nodule in the left thyroid lobe. The pathology result, which was previously considered to be benign several times, was reported as papillary thyroid carcinoma. Neck ultrasonography showed multiple hypoechoic lymph nodes with a maximum diameter of 1.5 cm in the bilateral level 2 and left supraclavicular region. Although FNAB and thyroglobulin (Tg) washout from lymph nodes were not diagnostic, malignancy could not be excluded. As the ultrasonographic images of the lymph nodes in the neck

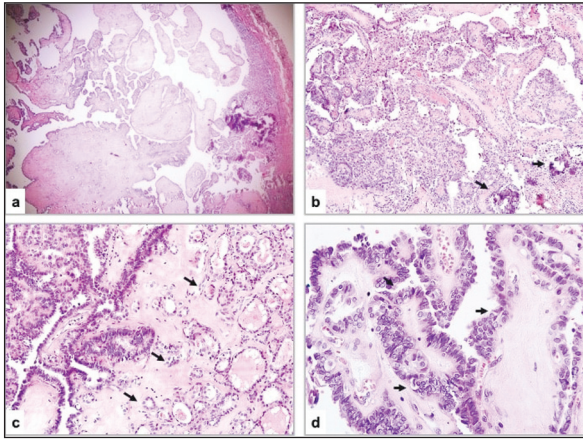


FIGURE 3: Histopathological results of classical papillary carcinoma of the thyroid originating from a thyroglossal duct cyst with hematoxylin and eosin (H&E) staining: a) Relationship with cyst wall and tumor mass, x4 b) Psammoma body-like foci of calcification at the bottom of tumor mass (black arrow) x10 c) The relationship between normal-appearing thyroid follicles (black arrow) and tumoral area, x20 d) Coffee bean-like nuclei typical of papillary carcinoma of the thyroid, x40.

suggested that it might be metastatic, performing central and lateral neck dissections was decided by the thyroid council of our hospital despite the negative Tg washout. Thus, the patient underwent total thyroidectomy, and bilateral central and lateral neck dissections. There were no complications such as hypocalcemia, recurrent nerve palsy, or hematoma after the surgery. The post thyroidectomy pathological analysis revealed a follicular variant of multifocal papillary carcinoma in the bilateral thyroid lobes and lymphocytic thyroiditis. There were no metastases in the central and bilateral neck dissections apart from a positive 0.5×0.6 cm prelaryngeal lymph node. After the surgery, the patient was treated with radioactive iodine, and her follow-up is being continued without any recurrence for 9 months.

Written informed consent was obtained from the patient to report this case for publication. This study was approved by the local ethics committee of İzmir Bozyaka Research and Training Hospital with the September 25, 2021 dated and 154 numbered decision.

DISCUSSION

During the thyroid gland development, the thyroglossal duct, which is formed as the gland moves

from the foramen cecum to the pretracheal region, involutes at the 10th week of gestation. If the canal between the thyroid isthmus and foramen cecum is not regressed, thyroglossal duct remnants, mostly cysts, can develop anywhere along the tract, including the larynx, thyroid gland, and tongue base. Although thyroglossal cysts are often found infrahyoid (61%), they can also be found suprahyoid (24%), suprasternal (13%), and intralingual (2%).⁵

It is challenging to differentiate endolaryngeal TGDCs from laryngeal cystic masses such as saccular cysts and laryngoceles by endoscopic examination. However, as suggested by Bando et al., while TGDCs are close to the hyoid bone, the close relationship of laryngoceles and saccular cysts with ventricle can be evaluated with magnetic resonance imaging or CT.³ Preoperative FNAB is very useful in predicting malignancy, especially in heterogeneously enhancing masses, and distinguishing from laryngeal cysts if it contains follicular thyroid cells.^{3,6} In the meta-analysis published by Rayess et al., it was found that only 20% of TGDC cancers were diagnosed with FNAB before the operation, but when it was done, the FNAB sensitivity was between 56-62%.⁴ Performing FNAB to all patients with TGDC is not cost-effective, especially when children are considered, and a negative FNAB result does not exclude malignancy. However, in the presence of calcification or solid nodule in the cyst on CT scan, in cysts larger than 2 cm, and when invasion to adjacent structures is suspected, FNAB may be very valuable in preventing delays in diagnosis and preventing recurrent surgeries. FNAB was not performed before the first operation in our case since thyroid FNAB had been taken from the patient twice by other departments, and the results were benign. However, due to the presence of multinodular goiter and a suspicious solid lesion near the cyst, it would be more accurate to repeat FNAB from the cyst and thyroid nodules before the first operation.

Laryngeal symptoms can be seen in cysts with endolaryngeal extension, and they may cause erosion of the thyrohyoid membrane and thyroid cartilage due to compression. There was usually a palpable mass in the neck in the cases previously reported in the literature, but neck examination was unremark-

able in our patient, probably due to the obesity.² Even though there were patients who underwent tracheotomy due to airway narrowing and difficult intubation in the literature, we did not have such a requirement.³ Disruption of airway integrity is a significant risk in these patients, but we were able to excise the cyst entirely without damaging the airway epithelium.

TGDCs are the most common congenital midline neck masses, but malignancies arising from TGDCs are uncommon, with papillary thyroid cancer consisting 92.1% and squamous cell carcinoma consisting 4.3%.⁴ In addition, in the presence of TGDCca, synchronous thyroid cancers may accompany. Rayess et al. showed that the frequency of synchronous thyroid carcinoma was 23.4% in patients who were operated for TGDCca when total thyroidectomy was included in the surgery, in their meta-analysis.⁴ On the other hand, Wood et al. detected tumors in the thyroid gland in two of four patients who underwent total thyroidectomy in their case series.⁷ The prevailing view currently is that synchronous tumors arise de novo from TGDCs.^{4,7}

There is no clear consensus on the treatment of these tumors, as their incidence is also low. Since the Sistrunk procedure is the primary treatment method for TDGCs that decreases the chance of recurrence, it should be the least procedure that is done in the presence of cancer. Some authors recommend a routine total thyroidectomy in TGDCa to facilitate follow-up.^{8,9} However, there are also some authors who do not recommend that all the patients with TGDCa undergo a routine total thyroidectomy, because it is thought that TGDCa develops de novo.^{4,10} Patients younger than 45 years with a radiologically or clinically normal gland and negative margins can be observed.^{4,10,11} In the presence of synchronous thyroid cancer, the treatment according to the established guidelines is suggested.^{7,11} Unlike the thyroid gland, TGDCa tends to metastasize to first the lateral neck.⁴ In cases of lymph node metastases, a dissection of appropriate levels in the neck is recom-

mended in addition to the Sistrunk procedure and total thyroidectomy.^{4,12} In this case, we performed a total thyroidectomy due to the synchronous thyroid cancer. Because the appearance of the lymph nodes in the bilateral lateral neck and left supraclavicular fossa was radiologically suspicious, we also performed central and lateral neck dissections.

In conclusion, TGDCs with intralaryngeal extension which may present with laryngeal symptoms such as hoarseness and dysphagia are quite rare and can be confused with laryngoceles and saccular cysts. TGDCs should be kept in mind in the presence of endolaryngeal cystic masses. In addition, although carcinomas originating from TGDCs are uncommon, we recommend that suspicious lesions should be evaluated with FNAB before surgery, since malignancies may occur in large cysts, solid nodules and calcifications in the cysts.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Mehmet Ekrem Zorlu, Togay Müderris; **Design:** Mehmet Ekrem Zorlu; **Control/Supervision:** Togay Müderris, Abdulhalim Aysel; **Data Collection and/or Processing:** Çiğdem Özkan, Onur Çorakçı, Uğurtan Ergün; **Analysis and/or Interpretation:** Mehmet Ekrem Zorlu, Özden Öz; **Literature Review:** Onur Çorakçı, Özden Öz, Uğurtan Ergün; **Writing the Article:** Mehmet Ekrem Zorlu, Togay Müderris; **Critical Review:** Togay Müderris, Abdulhalim Aysel; **References and Fundings:** Mehmet Ekrem Zorlu; **Materials:** Mehmet Ekrem Zorlu, Çiğdem Özkan, Özden Öz.

REFERENCES

1. Ng ACW, Yuen HW, Huang XY. Atypical thyroglossal duct cyst with intralaryngeal and para-glottic extension. *Am J Otolaryngol.* 2019;40(4):601-4. [[Crossref](#)] [[PubMed](#)]
2. Bosco S, Cohn JE, Everts M, Papajohn P, Lesser R. Thyroglossal duct cyst occupying posterior hyoid space with endolaryngeal extension presenting after neck trauma. *Ann Otol Rhinol Laryngol.* 2020;129(6):628-32. [[Crossref](#)] [[PubMed](#)]
3. Bando H, Uchida M, Matsumoto S, Ushijima C, Dejima K. Endolaryngeal extension of thyroglossal duct cyst. *Auris Nasus Larynx.* 2012;39(2):220-3. [[Crossref](#)] [[PubMed](#)]
4. Rayess HM, Monk I, Svider PF, Gupta A, Raza SN, Lin HS. Thyroglossal duct cyst carcinoma: a systematic review of clinical features and outcomes. *Otolaryngol Head Neck Surg.* 2017;156(5):794-802. [[Crossref](#)] [[PubMed](#)]
5. Lübben B, Alberty J, Lang-Roth R, Seifert E, Stoll W. Thyroglossal duct cyst causing intralaryngeal obstruction. *Otolaryngol Head Neck Surg.* 2001;125(4):426-7. [[Crossref](#)] [[PubMed](#)]
6. Soliman AM, Lee JM. Imaging case study of the month. Thyroglossal duct cyst with intralaryngeal extension. *Ann Otol Rhinol Laryngol.* 2006;115(7):559-62. [[Crossref](#)] [[PubMed](#)]
7. Wood CB, Bigcas JL, Alava I, Bischoff L, Langerman A, Kim Y. Papillary-type carcinoma of the thyroglossal duct cyst: the case for conservative management. *Ann Otol Rhinol Laryngol.* 2018;127(10):710-6. [[Crossref](#)] [[PubMed](#)]
8. Kennedy TL, Whitaker M, Wadhi G. Thyroglossal duct carcinoma: a rational approach to management. *Laryngoscope.* 1998;108(8 Pt 1):1154-8. [[Crossref](#)] [[PubMed](#)]
9. Miccoli P, Minuto MN, Galleri D, Puccini M, Berti P. Extent of surgery in thyroglossal duct carcinoma: reflections on a series of eighteen cases. *Thyroid.* 2004;14(2):121-3. [[Crossref](#)] [[PubMed](#)]
10. Plaza CP, López ME, Carrasco CE, Meseguer LM, Perucho Ade L. Management of well-differentiated thyroglossal remnant thyroid carcinoma: time to close the debate? Report of five new cases and proposal of a definitive algorithm for treatment. *Ann Surg Oncol.* 2006;13(5):745-52. [[Crossref](#)] [[PubMed](#)]
11. Yaman H, Durmaz A, Arslan HH, Ozcan A, Karahatay S, Gerek M. Thyroglossal duct cysts: evaluation and treatment of 49 cases. *B-ENT.* 2011;7(4):267-71. [[PubMed](#)]
12. Kandogan T, Erkan N, Vardar E. Papillary carcinoma arising in a thyroglossal duct cyst with associated microcarcinoma of the thyroid and without cervical lymph node metastasis: a case report. *J Med Case Rep.* 2008;2:42. [[Crossref](#)] [[PubMed](#)] [[PMC](#)]