

# A rare location of papillary carcinoma: Thyroglossal duct cyst

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#### **ABSTRACT**

The inadequate closure of the thyroglossal tract paves the way for a thyroglossal cyst. Thyroglossal duct cyst (TDC) malignancy is quite rare. A thirty-nine-year-old female patient was admitted to the polyclinic with a complaint of palpable mass in the neck. Findings compatible with TDC were determined in the patient's neck screening and it was considered to be malignant due to irregular margins, apparent vascularization and punctual calcifications. Fine needle aspiration biopsy was unremarkable. TDC was excised by Sistrunk procedure and frozen examination was performed. Total thyroidectomy was performed additionally since the result of the frozen examination was found to be compatible with the primary papillary carcinoma of TDC. If preoperative biopsy does not provide a diagnosis, frozen section study will be beneficial in terms of both providing the early diagnosis and directing the operation strategy during the surgery in clinically or radiologically suspected patients.

Keywords: Frozen section, papillary carcinoma, sistrunk procedure, thyroglossal duct, thyroidectomy

#### INTRODUCTION

While the thyroid gland progresses from foramen caecum to the region in front of the thyroid cartilage during the embryologic period, it leaves an epithelial trace called the thyroglossal tract in the region it has passed. The inadequate closure of this thyroglossal tract paves the way for a thyroglossal cyst (1). Thyroglossal duct cyst (TDC) is the most common congenital abnormality in the neck region (2). It usually presents with midline neck mass during the early childhood, while it is rarely observed during adulthood. It is often benign, but malignant tumor may develop by 1% (3).

Here, we aimed to present a patient who was diagnosed with primary papillary carcinoma of the TDC, with the intraoperative frozen section.

### **CASE REPORT**

A thirty-nine-year-old female patient was admitted to the general surgery polyclinic with a complaint of palpable mass in the neck. The patient had no history of smoking, alcohol consumption, radiotherapy and familial history of thyroid cancer. During the physical examination, we palpated a mass approximately 1.5 cm in diameter between the submental region and thyroid cartilage, which was mobile by swallowing. There was no cervical lymphadenopathy. The thyroid function tests of the patient were normal. Neck ultrasonography (USG) revealed multinodular goiter containing nodules the largest of which was 6 mm in size, and TDC that was 12 mm in size in the midline between the hyoid bone and thyroid cartilage. TDC was considered to be malignant due to irregular margins, apparent vascularization and punctual calcification. Findings compatible with TDC were determined in the patient's neck magnetic resonance imaging (MRI) (Figure 1). The result of the fine-needle aspiration biopsy (FNAB) was atypia of undetermined significance. The Sistrunk procedure (SP) was planned in the preoperative period. In operation, TDC was resected and frozen examination was performed. Since the result of the frozen examination was found to be compatible with the papillary carcinoma of TDC, total thyroidectomy (TT) was performed additionally. Postoperative period was un-

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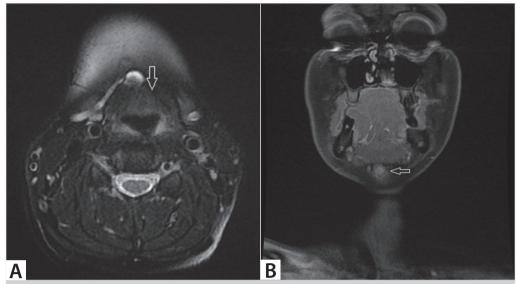
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**Figure 1. A.** Thyroglossal duct cyst (arrow) in fat-supressed T2-weighted axial sections. **B.** Thyroglossal duct cyst (arrow) in contrast enhanced fat-supressed T1-weighted coronal sections in Magnetic Resonance Imaging.

eventful and the patient was discharged on the postoperative day two without any complications.

In the histopathological examination of the tumor, it was reported that the size of the localized papillary tumor in TDC was 8 mm and that the surgical margins were intact. No malignancy was observed in the thyroidectomy material. The metastasis scans of the patient were negative, and thyroid suppression treatment was initiated. The patient had remained well for two years with no evidence of recurrence or distant metastasis.

Written informed consent was obtained from the patient for this study.

## **DISCUSSION**

TDC malignancy is quite rare. Just like in thyroid cancers, papillary cancer is observed in more than 80% of patients. Mixed papillary-follicular (8%) and squamous cell carcinoma (6%) are the most common types after papillary cancer (4). TDC malignancy may be seen in an isolated embryological thyroglossal duct remnant or develop secondary to metastasis of thyroid carcinoma (1). According to a review study published in 2016 (5), the mean age is 39 at the time of diagnosis, and it emerges more frequently in females (68.3%) compared to males. The most common cause of admission is the asymptomatic neck mass (95.1%), while less than 5% of patients are symptomatic. The patient reported here was a female at the age of 39 and was admitted to the hospital due to an asymptomatic neck mass. No malignancy was detected in the thyroidectomy material, and there was a primary tumor that originated from TDC.

USG, computed tomography (CT) and MRI can be used for the diagnosis. The presence and size of the cyst, the presence of

a solid or cystic component, accompanying thyroid or lymph node pathologies can be detected by means of USG. The presence of a solid component, wall thickening and especially calcification means suspected malignancy (6). CT can demonstrate the cyst in which the fluid level is observed, increasing intensity in the cyst wall, and calcification foci. MRI can demonstrate the cyst wall with the increased signal intensity compared to surrounding tissues both in T1 and T2 sections (7). FNAB is recommended if there is doubt in the imaging. In a review study, Rayess et al. (5) reported that FNAB was performed in 78.6% of patients and malignancy could be demonstrated in only onefourth of these patients as a result of biopsy. In the same study, it was stated that frozen examination provided a diagnosis in only 6% of patients. In our patient, FNAB was performed due to the suspicious malignant findings detected in imaging, but it did not provide diagnosis. On the other hand, intraoperative frozen section provided a final diagnosis and changed surgical strategy.

SP is applied in the treatment of TDC. However, a consensus has not been achieved regarding the need for thyroidectomy, central or lateral lymph node dissection and radioactive iodine treatment in the presence of TDC malignancy. Many authors recommend performing TT in addition to SP by considering the applicability of radioactive iodine treatment after TT and the presence of accompanying thyroid malignancy in one-third of patients (5, 8). In the patient reported, early diagnosis was obtained with the intraoperative frozen section study, and TT was performed in the same session since it was not possible to make a discrimination between primary or secondary TDC malignancy by considering multiple nodules located in the thyroid tissue.

#### CONCLUSION

TDC malignancy is a pathology that is rarely observed and is difficult to diagnose before surgery. If FNAB does not provide a diagnosis, we believe that a frozen section study will be beneficial in terms of both providing the early diagnosis and directing the operation strategy during the surgery in clinically or radiologically suspected patients.

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#### **REFERENCES**

Peretz A, Leiberman E, Kapelushnik J, Hershkovitz E. Thyroglossal duct carcinoma in children: case presentation and review of the literature. Thyroid 2004; 14: 777-85.

- Vera Sempere F, Tur J, Jaén J, Perolada JM, Morera C. Papillary thyroid carcinoma arising in the wall of a thyroglossal duct cyst. Acta Otorhinolaryngol Belg 1998; 52: 49-54.
- Yang SI, Park KK, Kim JH. Papillary carcinoma arising from thyroglossal duct cyst with thyroid and lateral neck metastasis. Int J Surg Case Rep 2013; 4: 704-7.
- Proia G, Bianciardi Valassina MF, Palmieri G, Zama M. Papillary carcinoma on a thyroglossal duct cyst: diagnostic problems and therapeutic dilemma. Acta Otorhinolaryngol Ital 2014; 34: 215-7.
- 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.
- Taori K, Rohatqi S, Mahore DM, Dubey J, Saini T. Papillary carcinoma in a thyroglossal duct cyst - a case report and review of literature. Indian J Radiol Imaging 2005; 15(4): 531-3.
- Glastonbury CM, Davidson HC, Haller JR, Harnsberger HR. The CT and MR imaging features of carcinoma arising in thyroglossal duct remnants. AJNR Am J Neuroradiol 2000: 21: 770-4.
- 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: 121-3.



# **OLGU SUNUMU-ÖZET**

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# Papiller karsinomun nadir yerleşimi: tiroglossal kanal kisti

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## ÖZET

Tiroqlossal kanal trasesinin yetersiz kapanması, tiroqlossal kist oluşumuna yol açar. Tiroqlossal kanal kisti (TDC) malignitesi oldukça nadirdir. Otuz dokuz yaşında kadın hasta boyunda palpabl kütle şikayeti ile polikliniğe başvurdu. Hastanın radyolojik taramasında TDC ile uyumlu bulgular belirlendi. Düzensiz kenarlar, belirgin vaskülarizasyon ve noktasal kalsifikasyonlar nedeniyle lezyon malign kabul edildi. İnce iğne aspirasyon biyopsisi tanısal değildi. TDC Sistrunk prosedürü ile eksize edildi ve frozen inceleme yapıldı. Frozen incelemenin sonucu TDC'nin primer papiller karsinomu ile uyumlu bulunması üzerine total tiroidektomi ameliyata eklendi. Ameliyat öncesi biyopsi tanısal olmazsa, hem klinik hem de radyolojik olarak şüpheli hastalarda ameliyat sırasında erken tanı ve operasyon stratejisinin yönlendirilmesi açısından frozen kesit çalışmanın yararlı olacağı kanaatindeyiz.

Anahtar Kelimeler: Frozen inceleme, papiller kanser, Sistrunk prosedürü, tiroglossal kanal, tiroidektomi

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