



# Papillary Carcinoma Developing from a Thyroglossal Duct Cyst: A Case Report

## Tiroglossal Kanal Kistinden Gelişen Papiller Karsinom: Olgu Sunumu

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### ABSTRACT

**Background:** Thyroglossal duct cyst is the most common midline congenital anomaly of the neck. Papillary carcinoma in the thyroglossal duct cyst and papillary carcinoma in the thyroid tissue can be seen together. In this article, we present a case of papillary carcinoma of the thyroglossal ductus cyst in a 43-year-old male patient. In the postoperative second year ultrasonography, our patient had multiple reactive lymph nodes in the submandibular region due to tooth infection and about 4x3 mm cystic nodule was detected in the thyroid left lobe. Total thyroidectomy was recommended to the patient when our patient wanted to exit long-term follow-up. In our case, although the focus of papillary carcinoma is less than 1 cm and the capsule is intact in the thyroglossal cyst material, the focus of 4 mm papillary carcinoma was found in the total thyroidectomy material. Even if the focus of papillary carcinoma in the thyroglossal cyst is smaller than 1 cm and the capsule is intact, it should be kept in mind that there may be a focus of papillary ca in the thyroid tissue and patients should not be excluded from follow-up.

**Keywords:** Thyroglossal cyst, papillary carcinoma, thyroidectomy

### ÖZ

Tiroglossal kanal kisti boynun en sık görülen orta hat konjenital anomalisidir. Tiroglossal kanal kistinde papiller karsinom ile tiroid dokusunda papiller karsinom bir arada görülebilir. Bu yazıda 43 yaşında erkek hastada tiroglossal duktus kistinin papiller karsinomu olgusunu sunduk. Hastamızın postoperatif ikinci yıl ultrasonografisinde diş enfeksiyonuna bağlı olarak submandibuler bölgede çok sayıda reaktif lenf nodu mevcuttu ve tiroid sol lobunda yaklaşık 4x3 mm boyutunda kistik nodül tespit edildi. Hastamızın uzun dönem takipten çıkmak istemesi üzerine hastaya total tiroidektomi önerildi. Bizim olgumuzda tiroglossal kist materyalinde papiller karsinom odağı 1 cm'den küçük ve kapsül sağlam olmasına rağmen total tiroidektomi materyalinde 4 mm'lik papiller karsinom odağı tespit edildi. Tiroglossal kistteki papiller karsinomun odağı 1 cm'den küçük ve kapsül sağlam olsa bile tiroid dokusunda papiller ca odağı olabileceği akılda tutulmalı ve hastalar takipten çıkarılmamalıdır.

**Anahtar Sözcükler:** Tiroglossal kist, papiller karsinom, tiroidektomi



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## INTRODUCTION

Thyroglossal duct cyst is seen in approximately 7% of the entire population (1). The presence of lymphatic tissues and normal thyroid follicles in the cyst wall, the cyst wall consisting of cuboidal epithelial cells, and the cyst being located in the middle region of the neck indicate that it is a thyroglossal duct cyst (2). First ultrasonography is performed for diagnosis. On ultrasonography, it may appear homogeneous hypoechoic, anechoic, heterogeneous or homogeneous hyperechoic. If calcification is seen within the cyst on ultrasonography, malignancy should be considered (3).

Fine needle aspiration biopsy is used to detect malignancy in preoperative midline neck masses (4). En-bloc excision of the cyst with hyoid bone corpus (sistrunk operation) is the treatment of thyroglossal duct cyst (5). The malignancy of the thyroglossal duct cyst is mostly diagnosed incidentally by histopathological examination after surgery (6).

Malignancy is very rare (less than 1%) in a thyroglossal cyst (1). Papillary carcinoma accounts for approximately 80% of malignancies originating from thyroglossal cysts (7). Other malignancies originating from thyroglossal cysts are squamous cell carcinoma and adenosquamous carcinomas (1). It occurs mostly in the 3<sup>rd</sup> decade and in women (8). In this article, we present a case of papillary carcinoma of the thyroglossal ductus cyst in a 43-year-old male patient.

## CASE REPORT

A 43-year-old male patient was admitted with a mass in the neck for five years. At the time of admission, the patient did not have any complaints such as dysphagia, dysphonia or pain. The patient stated that the mass had been growing slowly, more rapidly, over the last 6 months. During the physical examination of the patient, a semi-mobile and solid mass of approximately 4×4 cm was observed extending from the midline to the left. No pathology was observed in the na-



**Figure 1:** The preoperative contrast enhanced neck MR image of the patient.

sopharynx and larynx in the endoscopic examination of the patient. Thyroid function tests and routine blood tests were normal. In the desired ultrasonography results of the patient, it was reported that there was a 3.5×3×2.8 cm thick-walled cystic lesion (thyroglossal cyst?) with thick septations and calcifications in the septa. In the magnetic resonance imaging (MRI) images of the patient, a well-circumscribed, thin-walled septal cystic lesion with thyroid cartilage levels in the left paramedian, 35×30×21 mm in size, T1W hypointense, T2W hyperintense, and weak-walled contrast enhancement was reported in postcontrast examination (Figure 1).

Fine-needle biopsy of the septa was performed with ultrasound. Serous fluid was aspirated. No malignancy was detected as a result of the fine needle. An operation was planned for the patient, as benign cyst content was observed in the biopsy report. The mass hyoid bone corpus and en-bloc were excised by Sistrunk operation under general anesthesia (Figure 2). The mass pathology was reported as papillary carcinoma developing in terms of thyroglossal cyst. On microscopic examination, malignancy was observed at a distance of 3 mm from the cyst wall and approximately 9 mm in diameter. There was no tumor on the outer surface of the cyst (Figure 2).



**Figure 2:** Image of the mass intraoperatively after en-bloc removal.

No pathological lesion was observed in the thyroid and neck ultrasonography performed in the 1<sup>st</sup> month postoperatively. Considering that the papillary carcinoma was primarily from the duct cyst, thyroid and neck ultrasonography every 6 months. In the 1<sup>st</sup> postoperative year, neck MRI and thyroid scintigraphy were performed instead of thyroidectomy. In the postoperative second year ultrasonography, our patient had multiple reactive lymph nodes in the submandibular region due to tooth infection and about 4.3 mm cystic nodule was detected in the thyroid left lobe. No findings were observed in 2<sup>nd</sup> year thyroid scintigraphy. Total thyroidectomy was recommended to the patient when our patient wanted to exit long-term follow-up. The patient underwent total thyroidectomy. The patient's total thyroidectomy material was observed in a 4 mm papillary carcinoma focus. Our patient is in the low risk group and the patient's TSH was aimed to be kept between 0.5-2 mIU/l. Our patient was started on postoperative oral levothyroxine sodium 75 mcg. During follow-ups, the treatment dose was adjusted according to the TSH target value (Table 1).

## DISCUSSION

There is some thyroid tissue in 5-67% of thyroglossal cysts (9). This thyroid tissue, which is normally seen in the cyst wall, has the potential for malignancy (10). A study by Rayess et al. shows that thyroglossal duct cyst carcinomas mostly are associated with excellent prognosis and a recurrence rate of 4.3 % (10).

There are two theories explaining the development of papillary carcinoma of a thyroglossal duct cyst (11). In the first theory, it is suggested that the carcinoma spreads from the thyroid tissue via the thyroglossal duct. In the second theory, it is suggested that carcinoma develops de novo from the ectopic thyroid tissue in the cyst wall. A study by Rossi et al. suggests that the majority of the thyroglossal carcinomas most likely develop as a primary malignancy from a thyroid remnant, and not a metastasis from a primary thyroid

cancer (12). Presence of an epithelial wall of the cyst, the presence of normal thyroid tissue in the cyst, failure to show the presence of a primary tumor in the thyroid gland, and a normal thyroid gland are the necessary criteria for papillary carcinoma arising from a thyroglossal duct cyst to be considered as primary (13).

In the clinical picture, pain, sudden increase in cyst size, presence of lymphadenopathy in the neck, and hoarseness findings suggest a malignant mass (14). There was no finding suggesting malignancy in the reported case. Ultrasonography is preferred among neck kits as the initial diagnosis method. Radiologically, the presence of mural nodules within the cystic lesion, the presence of enlarged cervical lymph nodes, and microcalcification indicate malignancy. A fine needle aspiration biopsy is the most reliable method for detecting malignancy in preoperative midline neck masses (4). Findings of calcification within the cyst on preoperative computed tomography are important in the diagnosis of malignancies (15). Masses with ultrasonographic features that pose a risk of malignancy correlate with malignant cytopathology results (16). Since it was known that the patient had a cystic lesion with calcifications on ultrasonography, surgery was planned for the patient even if there was no suspicion of malignancy in the fine needle biopsy.

In the study by Miccoli et al., thyroid malignancy was also seen in 6 of 18 cases of papillary carcinoma due to a thyroglossal duct cyst (17). Therefore, the thyroid gland should be evaluated radiologically and scintigraphically in these cases. Sistrunk operation and postoperative thyroid suppression treatment are recommended in cases where there is no tumor invasion beyond the cyst wall, there is no spread to cervical lymph nodes, there is normal thyroid gland and tumor diameter is found to be less than 1 cm (18). Some authors have considered surgical removal of the entire thyroglossal duct sufficient in patients who meet these criteria and have recommended intermittent follow-up of their patients (19). In cases with lymph node involvement

**Table 1.** Patient's preoperative and postoperative laboratory results.

	Preoperative	Postoperative 1 <sup>st</sup> day	Postoperative 3 <sup>rd</sup> week	Postoperative 6 <sup>th</sup> week	Postoperative 1 <sup>st</sup> year
TSH (mIU/l) (0.27-4,2)	1	-	<b>72,48</b>	1,08	0,55
FT4 (ng/dL) (0.93 - 1.7)	1,08	-	<b>0,42</b>	1,46	1,8
Thyroglobulin (ng/ml) (3,5-77)	-	-	-	<b>0,09</b>	<b>0,08</b>
Anti-Thyroglobulin Antibody (IU/ML) (0-115)	-	-	-	15,70	12,30
ALT (IU/L) (0-40)	33	24	-	19	19
Hemoglobin (g/dl) (13,5-18)	15,23	15,9	-	14,73	15,70
Fasting blood glucose (mg/dl) (74-106)	85	95	-	94,5	95,10
GFR (ml/dk) (90-150)	131,15	103,99	-	107,88	109,01

**ALT:** Alanine transaminase, **GFR:** Glomerular filtration rate

and cyst wall invasion in the neck, it is recommended to add a modified radical neck dissection together with thyroidectomy (20). Kara et al. removed an approximately 5×2 cm thyroglossal duct in a 63-years-old patient with sistrunk surgery and followed up the patient with papillary carcinoma of the thyroglossal duct, with the capsule intact and no invasion into the surrounding tissue, without any problems (19). In our patient, at first, follow-up was deemed sufficient, since the entire thyroglossal duct was surgically removed. However, lymph nodes that were seen after dental infection and did not regress with treatment were detected. Thyroidectomy was performed upon the patient's concern and his own request. In our case, a 4 mm focus of papillary carcinoma was detected in the thyroidectomy material of the patient who had papillary carcinoma in the thyroglossal cyst material, for which sistrunk surgery alone can be considered sufficient in the literature (19).

In addition to helping predict prognosis, postoperative risk assessment is also guiding in determining the level of TSH suppression after thyroidectomy. Turkish Endocrinology and Metabolism Association adopts the American Thyroid Association (ATA) guideline in risk assessment according to histopathological subtype (21). Since our patient was in the low-risk group, we aimed to keep TSH between 0.5-2 mIU/l. Thyroid suppression treatment dose was determined according to the target.

Even if the focus of papillary carcinoma in the thyroglossal cyst is smaller than 1 cm and the capsule is intact, it should be kept in mind that there may be a focus of papillary ca in the thyroid tissue and patients should not be excluded from follow-up.

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#### Author Contributions

Conception/Design of Study: **Mehmet Ali Say**, Data Acquisition: **Mehmet Ali Say, Sami Açar**, Data Analysis/Interpretation: **Mehmet Ali Say**, Drafting Manuscript: **Mehmet Ali Say, Sami Açar**, Critical Revision of Manuscript: **Mehmet Ali Say, Sami Açar**, Final Approval and Accountability- **Mehmet Ali Say, Sami Açar**, Material or Technical Support: **Mehmet Ali Say, Sami Açar**, Supervision: **Mehmet Ali Say, Sami Açar**.

#### Conflicts of Interest

The authors have no conflict of interest to declare.

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#### Ethical Approval and Consent

Written informed consent was obtained.

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