

CASE REPORT

Simultaneous existence of papillary carcinoma in the thyroid gland and thyroglossal duct cyst in two patients

İki olguda tiroit bezinde ve tiroglossal duktus kistinde eşzamanlı papiller karsinom

Deniz DEMİR, M.D.,¹ Yusufhan SÜOĞLU, M.D.,¹ Mehmet GÜVEN, M.D.,² Yersu KAPRAN, M.D.³

Papillary carcinoma of the thyroglossal duct cyst is a rare occurrence. Two patients presenting with medial neck masses were diagnosed as having thyroglossal duct cysts by ultrasonography. The Sistrunk operation was performed. Histopathologic evaluation demonstrated papillary carcinoma in the surgical specimens of both patients. The thyroid glands were examined by ultrasonography, scintigraphy, and fine-needle aspiration biopsy. Biopsy showed papillary carcinoma, and total thyroidectomies were performed. Micropapillary carcinoma was detected in the resected thyroid glands. The patients were asymptomatic without complications after 24 and 32 months of follow-up, respectively.

Key Words: Carcinoma, papillary/pathology/surgery; head and neck neoplasms; thyroglossal duct/surgery; thyroid neoplasms/pathology/surgery.

Tiroglossal duktus kistinin papiller karsinomu nadir görülür. Boynun orta kısmında kitle ile başvuran iki hastaya yapılan boyun ultrasonografisinde tiroglossal duktus kisti ile uyumlu lezyon saptandı. Olgulara Sistrunk ameliyatı uygulandı. Histopatolojik incelemede, tiroglossal duktus zemininde gelişmiş papiller karsinom saptanması üzerine, hastaların tiroid bezleri ultrasonografi, sintigrafi ve ince iğne aspirasyon biyopsisi ile incelendi. Biyopsi sonucunun papiller karsinom ile uyumlu olması üzerine hastalara total tiroidektomi yapıldı. Tiroidektomi materyallerinde mikropapiller karsinom görüldü. İki hastanın sırasıyla 24 ve 36 aylık izlemleri sırasında asemptomatik olduğu görüldü ve komplikasyona rastlanmadı.

Anahtar Sözcükler: Karsinom, papiller/patoloji/cerrahi; baş-boyun neoplazileri; tiroglossal kist/cerrahi; tiroit neoplazileri/patoloji/cerrahi.

Primitive thyroid gland develops as median diverticle from the ventral pharyngeal wall in the fourth intrauterine week and descent adjacent to the hyoid bone. The developed thyroglossal duct takes its place on the midline of the neck. The median age for thyroglossal duct cyst carcinoma is around 40

years of age, few cases presenting before the age of 14 years.^[1] Thyroglossal duct abnormalities are the result of deficient obliteration of the duct. The duct is lined with squamous or columnar epithelium. In spite of this data thyroid tissue is detected on the walls of the duct in 62% of the cases.^[2] Malignity is

◆ Departments of ¹Otolaryngology and ²Pathology, Medicine Faculty of İstanbul University (İstanbul Üniversitesi İstanbul Tıp Fakültesi 'KBB Hastalıkları Anabilim Dalı, ³Patoloji Anabilim Dalı), İstanbul; ⁴Department of Otolaryngology, Medicine Faculty of Gaziosmanpaşa University (Gaziosmanpaşa Üniversitesi Tıp Fakültesi KBB Hastalıkları Anabilim Dalı), Tokat, all in Turkey.

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◆ Correspondence (İletişim adresi): Dr. Deniz Demir. İstanbul Üniversitesi İstanbul Tıp Fakültesi KBB Hastalıkları Anabilim Dalı, 34270 Çapa, İstanbul, Turkey. Tel: +90 212 - 414 20 00 Fax (Faks): +90 212 - 533 57 64 e-mail (e-posta): drdenizdemir@yahoo.com

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reported in over 150 cases of thyroglossal duct cyst (TDC) in the literature.^[1] Carcinoma in TDC is reported to be 1% first by Bretana in 1911 and by Uchermann in 1915.^[3] The distribution is as follows : 85.5% papillary carcinoma, 6.6% squamous cell carcinoma, 4.4% mixt papillary-follicular carcinoma, 2.2% adenocarcinoma, 1.1% follicular adenocarcinoma.^[2] In this paper we present two cases of papillary thyroid carcinoma, occurring simultaneously both in TDC and thyroid gland.

CASE REPORT

Case 1- A 64- year-old male patient applied to our clinic with the complaint of midline mass of the neck. The mass which was painless and which moved with swallowing was present for 10 years. The patient did not have complaints of dysphagia, odynophagia, hoarseness or difficulty breathing. There were no histories of neck trauma, local infection or radiation therapy. Physical examination revealed a round, mobile, medium hard, painless midline neck mass of 2x2 cm dimensions, which moved with swallowing. There was no palpable nodule in the thyroid gland. Examination of the tongue base and ear, nose and throat examination was normal.

Laboratory investigations revealed normal T3, T4 and TSH levels. By neck ultrasonography (USG), a

midline lesion that contained calcified foci, which resembled TDC, was seen. Thyroid gland had normal dimensions, but two nodules of 1 cm and 6 mm in dimensions were visible in the right and left lobe parenchymes. No cervical lymphadenopathy was detected.

After the diagnosis of TDC Sistrunk procedure (removal of cyst together with the central 1/3 of the hyoid bone together with the soft tissue around the cyst) was performed. Macroscopic examination revealed a 2.5x2x2 cm cyst with exophytic solid lesions. Histopathologic examination demonstrated a cystic lesion without an epithelial layer and towards the center of the lesion there was a papillary tumour with ground - glass - nucleated cells (Fig. 1). After this finding TC 99 nucleotide scanning of the thyroid gland was done and a hypoactive lesion in the mid-zone of the right lobe and hyperactive uptake in the medial part of the inferior pole of the left lobe was seen (Fig. 2). Fine needle aspiration biopsy (FNAB) of the hypoactive lesion revealed cytological findings consistent with papillary carcinoma. Total thyroidectomy was done 2 weeks after the first operation.

Thyroidectomy specimen contained two nodules, one of them was in 1.0x0.8 cm and the other one was a sclerotic focus of 6 mm in diameter. Microscopic evaluation revealed colloidal nodular

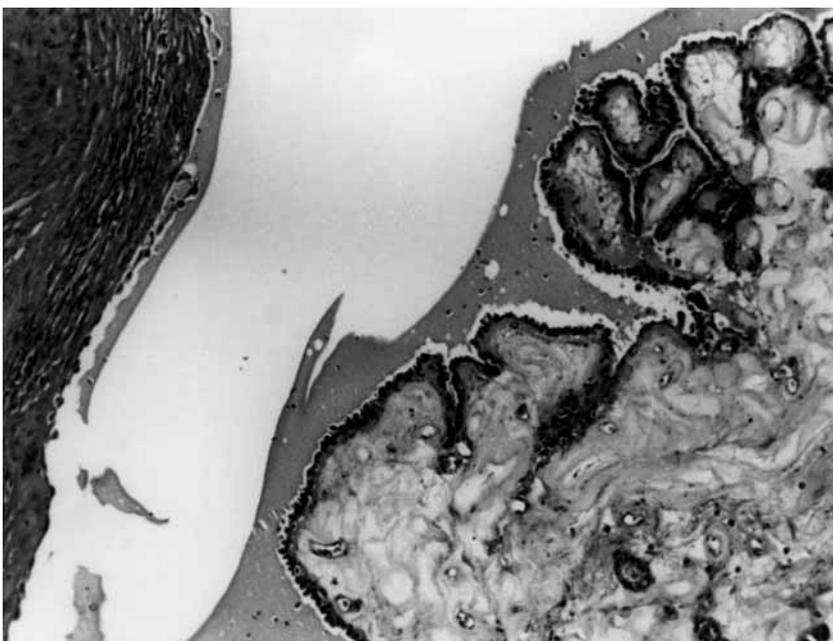


Fig. 1 - Thyroglossal cyst wall with papillary carcinoma on the left hand side (H-E; original magnification x 125).

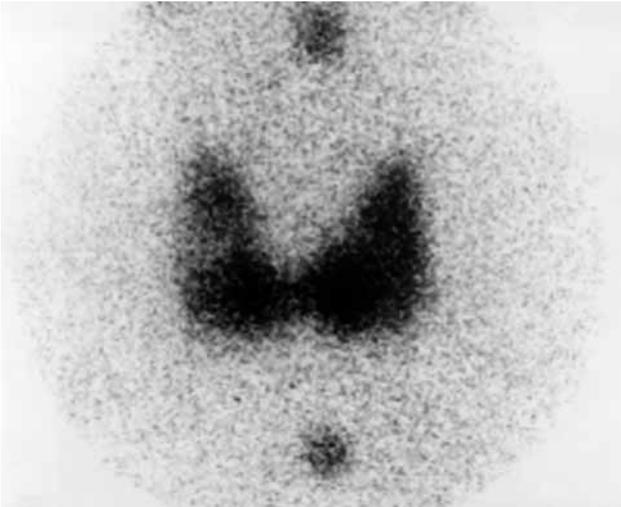


Fig. 2 - Tc 99 nucleotide scanning, hypoactive lesion in the mid-zone of the right lobe and hyperactive uptake in the medial part of the inferior pole of the left lobe was seen.

goitre and a papillary micro-carcinoma (6 mm in diameter) that did not invade the thyroid capsule. There were no postoperative complications. Thyroxine sodium 0.1 mg 1x1 treatment was administered and after four weeks 0.2 mg was continued. Radioactive iodine therapy was planned 8 weeks after the operation.

Case 2 – A 26-year-old male patient applied with midline neck mass of 13 years duration. There was

no history of dysphagia, odynophagia, hoarseness, or respiratory difficulty. There was no history of irradiation, or recent history of local infection. Head and neck examination revealed a painless midline neck mass of 5x4 cm dimensions. Neck and thyroid palpation was normal. The preoperative workups include: anterior-posterior chest radiographs, thyroid function tests, antimicrosomal antibody titers, and antithyroglobulin antibody titers. All of these were normal. USG discovered a mass consistent with TDC and numerous nodules in the thyroid gland. FNAB was applied to the dominant nodule, which revealed cytological findings consistent with papillary carcinoma. Sistrunk procedure and total thyroidectomy were performed in the same session. Histopathologic examination of the thyroid specimen revealed a 7 mm diameter papillary micro-carcinoma and cystic multinodular goitre (Fig. 3). The patient was administered to thyroxine sodium 0.1 mg therapy and after three weeks 0.2 mg was continued until now. Radioactive iodine therapy was planned 2 months after the treatment.

DISCUSSION

TDC is a painless congenital neck mass that is usually located in the midline and moves with swallowing. Differential diagnosis includes ectopic thyroid, dermoid cyst, lymphadenomegaly, sebaceous cysts, lipoma, hemangioma, and cystic hygroma.^[4] A



Fig. 3 - Papillary micro-carcinoma focus in the thyroid gland. (H-E x 32).

detailed head and neck examination, thorax x-ray, and if the thyroid is in normal position thyroid nucleotide scanning should be done for pre-operative evaluation.^[4] FNAB is necessary if the lesion is solid. Histopathologic evaluation of TDC usually reveals a mucoid purulent fluid that contains cholesterol. The walls of the cyst are made up of stratified squamous, ciliated respiratory, and pseudostriated columnar epithelium.^[2] Additionally thyroid tissue was detected in 62% of the cyst walls.^[2]

TDC is usually benign, but the incidence of malignancy is 1%.^[2] Reported cases of simultaneous papillary carcinoma of the TDC and thyroid gland are few. Total thyroidectomies were performed to 115 patients with papillary carcinoma of the TDC in a study. Only 4 of the patients had papillary carcinoma in the thyroid glands.^[1] McNicoll et al.^[4] evaluated 133 patients with papillary carcinoma of the TDC and discovered no papillary carcinoma in their thyroids.

The thyrogenic origin of thyroglossal duct carcinomas is explained by two theories. Some authors propose that the thyrogenic origin is the occult primary carcinoma in the thyroid gland and there is a metastasis from the thyroid. Another theory is de novo carcinoma of the thyroid tissue in the cyst. The tumour being multifocal explains simultaneous papillary carcinoma of the thyroid and TDC. Brown and Judd^[5] concentrates on the metastasis theory and believe that thyroglossal duct serves as a passage for the carcinoma.

According to Weiss and Orlich^[6] thyroid gland papillary carcinoma may metastasize even though no lesion is detectable in the thyroid gland. Weiss states that even though TDC papillary carcinoma is of thyroid gland origin, primary TDC carcinoma may occur and the following two criteria has to be fulfilled for this diagnosis: 1- carcinoma focus in the TDC or the tract, 2- histopathologic examination of the thyroid gland to be normal. Differential diagnosis of the thyroid gland carcinoma and TDC carcinoma is important keeping in mind the possibility of cystic metastasis of the thyroid papillary carcinoma to the anterior part of the neck.

The diagnosis of carcinoma in a thyroglossal cyst usually is not made until the post-operative histopathologic examination of the TDC specimen. We used preoperative ultrasonogram and scintigra-

phy and T3, T4 and TSH levels, FNAB, and postoperative histopathological examination; only FNAB and the histopathological examination were useful for the diagnosis of papillary carcinoma. The most common operation for benign TDC is the Sistrunk procedure.^[7] In the event of neck metastasis the treatment protocol is the same as thyroid papillary carcinoma. Resection of effected nodal groups is sufficient. In case of regional nodal involvement modified neck dissection is necessary. Radioactive iodine therapy following the total thyroidectomy is recommended because of the possibility of metastasis, thyroid involvement or carcinoma in the thyroid gland.^[3] In our cases, we did not plan neck dissection because of the fact that there was no other mass in the neck and adjuvant treatment with radioactive iodine and suppression with exogenous thyroid supplementation were applied because excision of the TDC has been combined with total thyroidectomy. Follow up of the patients with thyroidectomy should include thyroglobuline levels for detection of metastasis or recurrence.^[8] An increase in thyroglobuline levels indicates recurrence of the thyroid papillary carcinoma.^[8] A few months postoperatively, their thyroglobulin level dropped while on thyroxine, and remained low to this time. In these cases, laboratory investigations revealed normal thyroid function tests and there has been no physical evidence of recurrent carcinoma during this three years period of observation.

Carcinoma metastasis from the thyroid gland may occur as well as de nova carcinoma of the TDC or duct. When a carcinoma is detected in the TDC, USG, FNAB and nucleotide scanning for the presence of papillary micro-carcinoma should evaluate the thyroid gland. If thyroid nodule is present, FNAB is mandatory. The contemporary view is that total thyroidectomy should be done because of the possibility of the tumour being multifocal. In other words when a papillary carcinoma is detected in the TDC, the cyst should be considered as a thyroid tissue and complementary total thyroidectomy should be done.

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