

Papillary carcinoma of the thyroglossal duct cyst: Is thyroidectomy necessary?

Tiroglossal duktus kisti papiller karsinomu: Tiroidektomi gerekli mi?

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Abstract

Thyroglossal duct cyst is the most frequent midline congenital neck anomaly. Carcinoma of the thyroglossal duct cyst is a rare entity, and papillary carcinoma is the most frequent malignancy of this uncommon entity. There are a limited number of studies on this subject in the literature, and in some series, it is associated with papillary thyroid carcinoma. There is no consensus on prophylactic thyroidectomy after diagnosis. In this study, a case, operated for thyroglossal duct cyst and diagnosed with papillary carcinoma and our approach consisting of follow-up without thyroidectomy are presented in light of the current literature.

Keywords: Thyroglossal duct cyst, Papillary carcinoma, Thyroidectomy

Öz

Tiroglossal kanal kisti en sık görülen orta hat konjenital boyun anomalisidir. Tiroglossal kanal kisti karsinomu nadir görülen bir durumdur. Papiller karsinom, bu nadir görülen varlığın en sık görülen malignitesidir. Literatürde bu konuyla ilgili kısıtlı sayıda çalışma vardır ve bazı serilerde bu durum tiroid papiller karsinomu ile birliktelik göstermektedir. Tanıdan sonra profilaktik tiroidektomi konusunda fikir birliği yoktur. Bu çalışmada, papiller karsinom tanısı konan tiroglossal kanal kisti için opere edilen ve tiroidektomi olmadan takip yaklaşımımız güncel literatür bilgisi ışığında sunulmuştur.

Anahtar kelimeler: Tiroglossal kanal kisti, Papiller karsinom, Tiroidektomi

Introduction

Thyroglossal duct cyst (TDC) is a congenital anomaly developing from thyroglossal duct residues and is encountered in 7% of the young adult population [1]. Carcinoma in the TDC is extremely rare and is diagnosed in less than 1% of cases [2,3]. The most frequent primary malignancy of TDC is papillary carcinoma [4]. It is usually diagnosed by the pathological examination of the removed tissue. In this study, a case operated for TDC and diagnosed with papillary carcinoma and our approach of follow-up without thyroidectomy is presented in light of the current literature.

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Case presentation

A nineteen-year-old female patient was admitted to our clinic with the complaint of swelling in the midline of her neck for three years. There was a painless mass of 2x1 cm (Figure 1). No pathological lymph nodes were detected in the neck. Thyroid function and laboratory tests were normal. Ultrasonography (US) identified a 2 cm multilocular cystic structure. To plan a potential surgical procedure, a computed tomography (CT) imaging scan was requested, which revealed similar evidence (Figure 2). No other pathological lesions were seen in the neck and the thyroid gland. Histopathological examination of the fine-needle aspiration sample showed cells of benign cytology. The patient was offered surgery with a preliminary diagnosis of TDC. The surgical procedure was uneventful. Pathologically, microscopic examination revealed a cystic tumor tissue with a fibrous wall lined with neoplastic-looking epithelium. Normal thyroid tissue was found in the cyst wall. The papillary structures were lined with neoplastic cells with clear nuclei and intranuclear inclusion bodies, and nuclear clefts. The tumor was encapsulated and had developed within the thyroid tissue, which showed neoplastic properties. Histopathological result of the specimen was reported as papillary carcinoma originating from the cyst floor. The patient was followed up in three to six month-intervals with neck and thyroid US and recently completed the 30-month follow-up uneventfully. Informed consent was obtained from the patient before surgery.

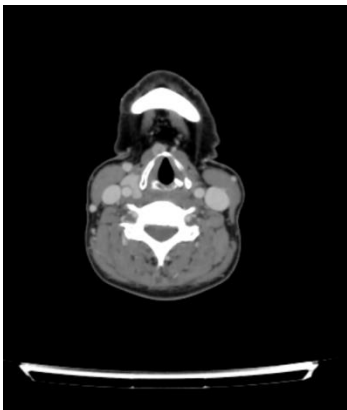


Figure 1: CT image of the patient



Figure 2: The mass in the middle of the neck

Discussion

The thyroid gland, the first endocrine gland that starts to develop in embryogenesis, completes its migration to the anatomical site of the tongue root at 7 weeks. The thyroglossal duct formed during this migration disappears by losing its channel feature over time. However, some parts of the duct may persist and TDCs develop between the hyoid and the thyroid gland because of continuing secretions of the cells that form the duct [5]. The TDC wall is covered with ciliated respiratory epithelium and / or squamous epithelium and contains more than 62% of regular thyroid tissue. Although the development of carcinoma in TDC is rare, 85% of malignant cases is constituted by papillary carcinoma, 7%, by mixed papillary follicular carcinoma, 5%, by squamous cell carcinoma, 2%, by adenocarcinoma and 1%, by anaplastic carcinoma [6]. Mechanism of thyroid malignancy in the TDC is still controversial. Some authors believe that it is caused by normal

islets of thyroid in the remains of the thyroglossal duct. This theory is strengthened by the determination of ectopic thyroid clusters in histopathological results of 62% of the TDC. TDC papillary carcinoma can synchronously accompany the papillary carcinoma of the thyroid gland and can also be detected incidentally as an isolated focus [7].

For the diagnosis of primary papillary carcinoma of TDC, squamous or respiratory epithelium and normal thyroid follicles should be found on the cyst wall and the thyroid gland should be clinically normal [8,9]. The inner surface of the cystic lesion is surrounded by cuboidal and squamous epithelium, and the existence of normal thyroid tissue in the cyst wall show that papillary carcinoma originated primarily from the cyst floor. Since the benign cyst of the thyroglossal duct and carcinoma of the thyroglossal duct can hardly be distinguished, the diagnosis of carcinoma often remains after surgery [5].

However, preoperative fine-needle aspiration biopsy can diagnose papillary carcinoma. CT is important in the differential diagnosis, and solid cyst and calcification findings may be interpreted in favor of malignancy [5,8].

The treatment approach for papillary carcinoma of the TDC is also controversial. The debate on the origin of the tumor continues. The most common approach in benign cysts of the thyroglossal duct is the Sistrunk procedure. The discussion in the treatment of papillary carcinoma arising from the cyst focuses only on whether the Sistrunk procedure will be sufficient and what to do with the thyroid gland. In his study, Kristensen recommends Sistrunk procedure and thyroid suppression treatment in the postoperative period in cases where normal ectopic thyroid follicles are present in the cyst wall, there is no tumor invasion or spreading to the cervical lymph nodes beyond the cyst wall, and the thyroid gland is normal [3]. Defenders of the metastatic propagation theory recommend total thyroidectomy for long-term follow-up when there is no tumor invasion or signs of spread in the cyst wall. Doshi presented a large study (14 cases) in the literature [10]. According to the results of this study, only Sistrunk surgery is sufficient when a negative biopsy is obtained from a suspected lymph node or thyroid gland mass. As a result, primary papillary carcinoma developing on the background of TDC is very rare and its diagnosis is generally made postoperatively by histopathological examination. If the diagnosis of primary thyroglossal cyst carcinoma is trusted, Sistrunk operation is sufficient in treatment. In our case, we decided that only Sistrunk surgery would be sufficient due to the well-limited and smooth surface of the cyst, no tumor invasion outside the cyst wall, negative clinical and radiological lymphadenopathy in the neck, and findings that showed that the tumor originated primarily from the cyst floor. However, we concluded that a regular and close follow-up is essential for these cases.

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