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Papillary carcinoma arising from ectopic thyroid gland in the wall of a thyroglossal cyst: Report of a case and review of literature

Tiroglossal kist duvarındaki ektopik tiroid dokusundan gelişen papiller karsinom: Olgu sunumu ve literatürün gözden geçirilmesi

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A 27 year-old healthy woman was seen with a midline swelling in the neck. On physical examination this was consistent with a thyroglossal duct cyst. Histological examination following surgical excision revealed a papillary carcinoma arising in a thyroglossal duct cyst.

Primary carcinomas of thyroglossal duct tissue are rare, and usually diagnosed after surgery. In this paper we report the case of papillary carcinoma arising in a thyroglossal cyst which was diagnosed as benign thyroglossal cyst before the operation and the literature was reviewed.

Key Words: Thyroglossal cyst, papillary carcinoma.

27 yaşında sağlıklı bayan hasta boyun orta hatta şişlik şikayeti ile başvurdu. Fizik muayene bulguları tiroglossal kist ile uyumlu idi. Cerrahi eksizyon sonrası yapılan histopatolojik incelemede, tiroglossal kist dokusundan gelişen papiller karsinom varlığı tesbit edildi.

Tiroglossal kanal dokusunun pirimer karsinomu çok nadirdir ve tanısı, genellikle tiroglossal kistin cerrahi olarak çıkarılmasından sonra konur. Tiroglossal kist klinik tanısı olan bizim olgumuzda da papiller karsinom varlığını, kist eksizyonu sonrası histopatolojik incelemede saptadık ve olguyla birlikte tiroglossal kist karsinomlarını literatür ışığında inceleyip tartıştık.

Anahtar Kelimeler: Tiroglossal kist, papiller karsinom.

The thyroid gland arises from the floor of the pharynx as an ectodermal outgrowth in the third week of embryonal development. It migrates inferiorly while remaining connected to the tongue base by the thyroglossal duct, usually passing anterior, but in close proximity, to the hyoid bone. This duct is normally reabsorbed between the seventh and tenth weeks in utero [1].

Thyroglossal duct may persist from the base of the tongue to the isthmus of the thyroid gland in 7% of the cases. A persisting duct may give rise to midline cysts. It is the commonest congenital cervical abnormality, arise from the remnants of the duct and has been described as occurring anywhere from the base of the tongue to the manubrium. Ectopic thyroid tissue is found histologically in 5-45% of patients with thyroglossal cysts [2].

Primary carcinomas of thyroglossal duct tissue are rare entity. In the literature the prevalence of them is generally estimated about 1% [3]. About 155 cases of malignant neoplasm arising in the thyroglossal cyst have been reported in the literature [4,5]. Different types of neoplasia were described including thyroid papillary carcinoma in 82-87%, squamous cell carcinoma in 5-6.6%, mixed papillary-follicular carcinoma in 4.4%, adenocarcinoma in 1.7-2.2%, follicular carcinoma in 1.7%, and malignant struma, epidermoid carcinoma and anaplastic carcinoma in 0.9% of each. Medullary carcinoma has never been found in a thyroglossal cyst 6,7.

CASE REPORT

A 27-year-old healthy woman (D.D) came to the surgeon in June of 2000 with a midline swelling in the neck, which moved on swallowing. She did not complain of sore throat, dysphagia or hoarseness and had no history of neck surgery or radiation to the head and neck. It was thought clinically to be a thyroglossal cyst. A computed tomogram of the neck showed a midline cystic nodule measuring 2.8 cm in diameter. The thyroid gland was of normal size and consistency. Further examination revealed no abnormalities. There was no palpable cervical lymphadenopathy. Neck exploration was performed, and at surgery a cystic structure was removed with the central portion of the hyoid bone.

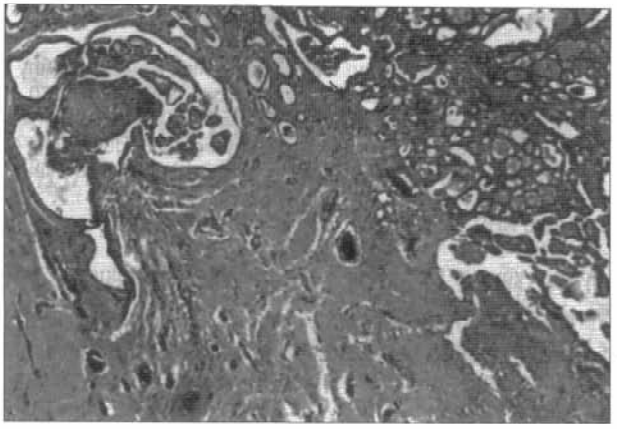

Figure 1: Remnant of thyroglossal cyst lined with squamous epithelium(arrow). On the upper, papillary carcinoma is seen within cyst wall (HE, x25).

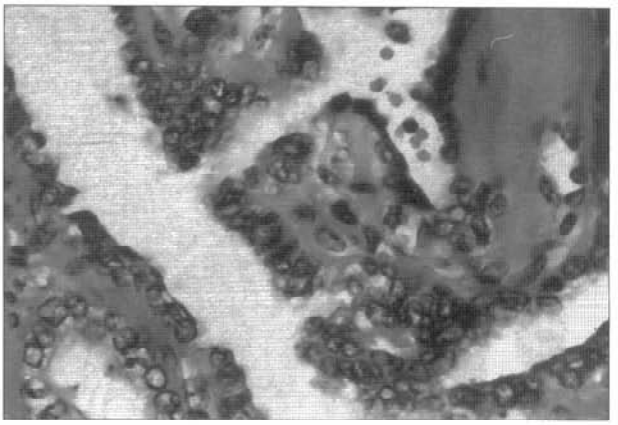

Figure 2: Papillary structures seen with optically clear nuclei and nuclear grooves in several neoplastic cells (HE, x400).

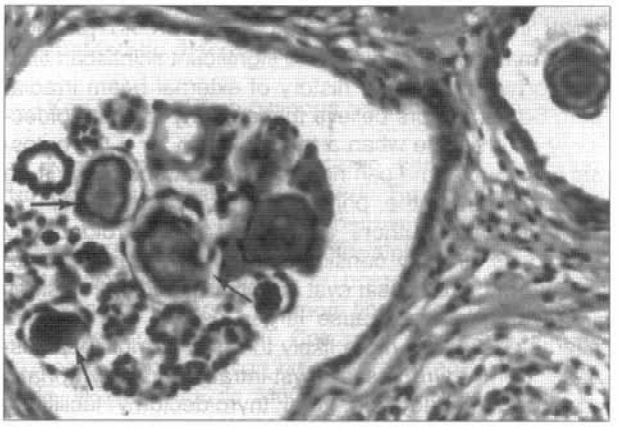

Figure 3: Photomicrograph showing psammoma bodies surrounded by neoplastic cells (arrows, HE, x200).

PATHOLOGICAL FINDINGS

The specimen (Biopsy no: 299/2000) consisted of cystic mass with a small portion of bone attached, measuring 3x2.8x2.7 cm in dimension. The cyst was well circumscribed with predominantly smooth linings. A gritty hard papillary 0.4x0.3 cm tan mass projected into the cyst.

Histological examination of the specimen showed a thyroglossal duct cyst with fibrous wall, lined with a squamous epithelium (figure 1). Sections of the papillary mass demonstrated papillary carcinoma. Optically clear nuclei and psammoma bodies were present, consistent with an origin of thyroid tissue (figures 2 and 3). The tumor invaded the cyst wall but appeared to be excised. Adjacent to the tumor there was non-tumoral thyroid tissue (figure 4).

DISCUSSION

Thyroglossal cysts usually present as a painless, mobile lump beneath the chine. Carcinoma should be suspected in any thyroglossal cyst which is hard, fixed, irregular or has undergone recent change. The presence of enlarged palpable lymph nodes in the neck, pain, weight loss should remind malignancy. But patients are usually asymptomatic in most cases. Most of the cases, as well as in our own, the diagnosis of malignancy is not suspected before the operation. Carcinoma is identified only at pathological examination[4,8,9]. Fine needle aspiration, as advocated by some authors, is the method of choice for early diagnosis. Particularly, papillary carcinoma has very characteristic cytomorphologic features that permit an accurate diagnosis on fine needle aspiration material [10,11].

Some occult papillary carcinomas of the thyroid metastasize to the medial anterior neck, become cystic and present as thyroglossal cyst carcinoma. The site of origin in the thyroid gland may be quite small. This indeed can be a problem in differential diagnosis, for often the normal lymph node architecture is completely effaced by the tumor [12]. The criteria for the unequivocal diagnosis of thyroglossal cyst carcinoma were proposed by some authors: (1) columnar or squamous epithelial lining on the cyst wall; (2) histologically normal ectopic thyroid follicles are found in the cyst wall; (3) the thyroid gland is normal [6,13]. Epithelial lining and normal ectopic thyroid tissue on the cyst wall in our own case were clearly observed. Investigation of the thyroid gland showed no abnormality.

The initial treatment of choice for such tumours is complete surgical excision, following Sistrunk's operation. This technique, consisting of dissection of the mass and thyroglossal tract, is performed superiorly to the level of the hyoid, where the midportion of the hyoid bone is resected in continuity [13,14]. If neck metastasis which is small and isolated is present, resection of the involved group of nodes is usually sufficient; but a modified neck dissection must be done if regional involvement is more extensive [15,16]. Thyroidectomy is not rou-

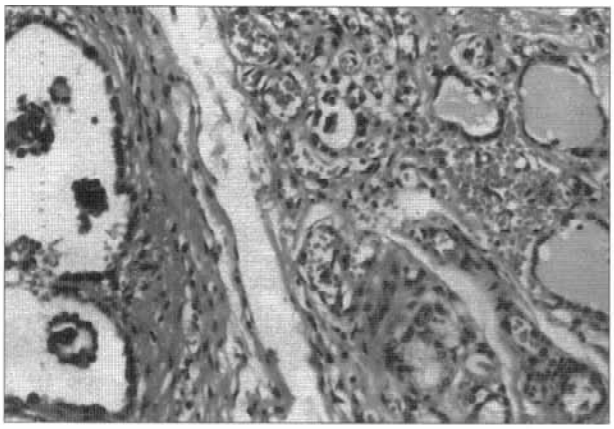

Figure 4: Benign normal-appearing thyroid thyroid follicles are observed on the right. On the left, papillary carcinoma is seen (HE, x100).

tinely indicated provided that there are no palpable abnormalities in the gland, no significant scintiscan findings or without a prior history of external beam irradiation. Some authors believe that an early total thyroidectomy is advisable when a thyroid origin for a thyroglossal cyst carcinoma can not be ruled out and there is no certainty about the primary origin of the tumor [7,8]. However, other authors advise that total thyroidectomy should be strongly considered when papillary carcinoma of the thyroglossal cyst is encountered with lesions 1 cm or greater because larger lesions with invasive characteristics are more likely to result in multifocal disease from thyroglossal cyst-intrathyroidal lymphatic spread [17]. In addition, total thyroidectomy facilitates subsequent ablation of the distant metastatic lesions with radioactive iodine and also allows a complete follow-up by measurement of serum thyroglobulin concentrations [18].

The prognosis of papillary carcinoma arising in thyroglossal cyst appears to be very good. It usually grows slowly and regional lymph node metastases are present in 7.7-14.6% of cases. However, this rate may rise up to 89% in primary papillary carcinomas of the thyroid gland. In literature, the number of patients who died of papillary carcinoma of the thyroglossal cyst is only two [3,4,19,20].

We would like to emphasize that Sistrunk's operation is the treatment of choice both in benign and malignant thyroglossal cysts. In the treatment of a carcinoma, it proves to be a curative operation in most cases and a more extensive second operation can thus be avoided.

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