Incidental Parathyroid Carcinoma Detected during Thyroidectomy for Papillary Thyroid Carcinoma: Report of a Case

Mehmet KILIÇ*, Mehmet KEŞKEK*, Tamer ERTAN*, Özge HAN, Erdal GÖÇMEN***

* V. Department of Surgery, Ankara Numune Hastanesi
** Department of Pathology, Ankara Numune Hastanesi, ANKARA

ABSTRACT

A 60-year old woman with a history of nodular goiter was admitted to hospital. Fine needle aspiration cytology (FNAC) of dominant nodule was suspicious of papillary thyroid carcinoma. Preoperative serum calcium level was normal in biochemical analysis. Cervical exploration revealed an aberrant tissue inferior to left thyroid gland which was reminiscent of parathyroid adenoma or an enlarged lymphadenopathy. Total thyroidectomy and en-bloc resection of this tissue was performed. Histopathological examination of the thyroid gland and aberrant tissue revealed papillary thyroid carcinoma and parathyroid carcinoma which was silent preoperatively. As the authors, we think that any abnormally enlarged parathyroid gland detected during thyroidectomy for malignancy should raise the suspicion of a second primary in the parathyroid gland.

Key Words: Thyroid carcinoma, Parathyroid carcinoma

ÖZET

Papiller Tiroid Kanseri için Tiroidektomi Sırasında Saptanan Paratiroid Kanseri: Olgu Sunumu

Nodüler guatr öyküsü olan 60 yaşındaki bir kadın hasta hastaneye kabul edildi. Dominant nodülden yapılan ince iğne aspirasyon biyopsisi (İİAB) papiller tiroid kanseri açısından şüpheliydi. Ameliyat öncesi serum kalsiyum düzeyi normal sınırlardaydı. Servikal eksplorasyonda sol tiroid lobunun aşağısında paratiroid adenomu veya lenfadenopatiyi düşündürecek anormal bir dokuyla karşılaşıldı. Total tiroidektomi yapıldı ve bu doku bir bütün olarak çıkartıldı. Histopatolojik incelemede papiller tiroid kanseri ve ameliyat öncesi bulgu vermeyen paratiroid kanseri saptandı. Otörler olarak, kanser nedeniyle tiroidektomi yapılırken saptanan büyümüş paratiroid bezlerinin ikinci bir primer şüphesini uyandırmasını düşünmekteyiz.

Anahtar Kelimeler: Tiroid kanseri, Paratiroid kanseri

INTRODUCTION

Parathyroid carcinoma is a rare entity and represents 0.1% to 5% of patients with hyperparathyroidism (1). Most cases are sporadic with a roughly equal male to female incidence and most patients present in the third to sixth decades. The rarity of parathyroid carcinoma has limited the accumulation of data on its natural history and etiologic factors. Parathyroid carcinomas are difficult to diagnose preoperatively; however, they may present with symptoms like nausea, vomiting, polyuria, generalized weakness, and weight loss or abnormalities in laboratory values. Presenting signs and symptoms may include sub cortical bone resorbtion, urolithiasis, parenchymal renal disease, pancreatitis, and peptic ulcer disease. Characteristically, the serum concentrations of calcium, parathyroid hormone (PTH), and alkaline phosphatase are markedly elevated compared to concentrations in patients with benign hyperparathyroidism. The presence of a palpable neck mass in a patient with hyperparathyroidism should raise the suspicion of parathyroid carcinoma. The diagnosis is most accurately made on histological examination when there is local invasion of surrounding tissues or metastases to regional lymph nodes or distant metastases. Intraoperative findings of local invasion are highly suspicious for malignancy, but frozen section evaluation has not been found to be helpful in confirming the diagnosis of malignancy (1). The best operative procedure is the en-block resection of the primary lesion.

Concomitant thyroid disease is an usual condition in patients undergoing cervical exploration for primary hyperparathyroidism. But, reports of synchronous thyroid and parathyroid carcinoma are extremely few.

CASE REPORT

A 60-year old woman was admitted to Ankara Numune Training and Research Hospital for surgical treatment of multinodular thyroid goiter. She had nothing worth in previous medical history. On admission, she was complaining of neck pain for the last 6 months. Physical examination revealed both thyroid glands to be enlarged with palpable, mobile, and hard nodules constituting almost whole glands. No cervical lymph nodes were palpable.

Laboratory investigations revealed thyroid function tests to be in normal limits, normocalcemia (a serum calcium concentration of 9.92 mg/dl; normal values, 8.10-10.40 mg/dl), and alkaline phosphatase level of 83 U/l; normal values, 38-155U/l. Thyroid ultrasonography demonstrated both thyroid glands to be enlarged with nearly 60 mm length and hight, heterogeneous, solid-cystic nodules filling both glands with no cervical enlarged lymph nodes. Subsequent technetium-pertechnetate-99m scanning revealed no activity in right thyroid gland (huge hypoactive nodule). Fine needle aspiration cytology (FNAC) performed from the nodule on the right side was suspicious of papillary thyroid carcinoma.

Surgical exploration revealed both gland to be composed of huge semi-solid nodules and total thyroidectomy was performed. During the exploration of left side there was an aberrant tissue inferior to thyroid gland but separate from it, and giving the impression of parathyroid adenoma or enlarged lymph node. This tissue was also resected and sent to pathology. Except this abnormal tissue, no cervical enlarged lymph node was detected.

Pathological examination of the right gland revealed a nodule 3 cm in diameter. Microscopically, there was a tumor with papillary configurations, and tumor cells had ground glass nuclei and nuclear groove. There was no tumoral finding in the left gland (Figure 1). Pathological examination of aberrant tissue revealed it to be a parathyroid gland with 2 x 1 x 1 cm in size. Microscopical examination revealed a tumor which perforated the capsule and infiltrated the surrounding tissues. Tumor mostly consisted of chief cells which extensively formed microfollicles. Tumor stroma was rich in vascular network. Very little mitotic activity was detected (Figure 2).

The patient had transient hypocalcemia in the postoperative period and was put on calcium preparations. Normocalcemia was achieved in the fourth postoperative month. The patient also received 100 mCi radioactive iodide as adjuvant treatment to eliminate any residual disease. At the end of 1 year she had total body scan with I^{131} and no evidence of any metastatic foci detected. Serum calcium levels and neck ultrasonography failed to show any

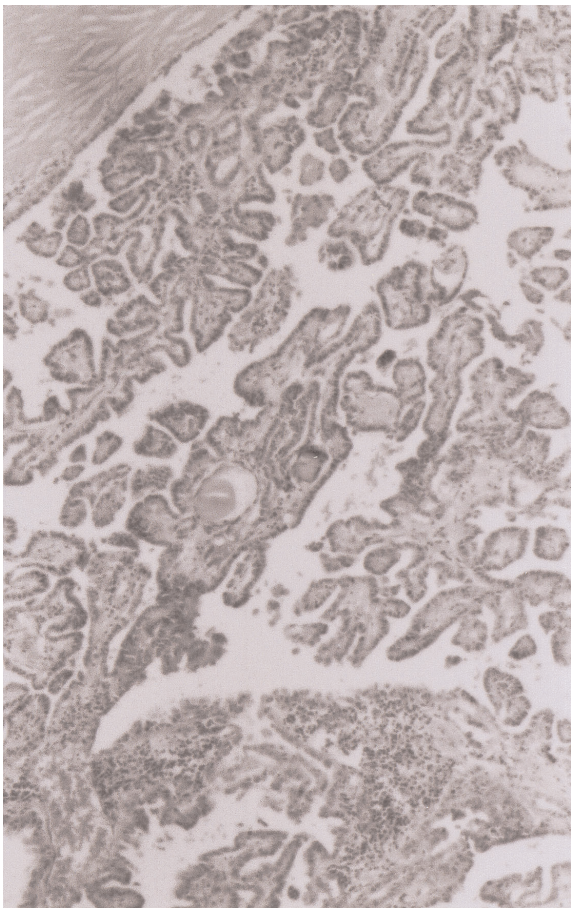

Figure 1. Papillary thyroid carcinoma surrounded by a fibrous capsule.

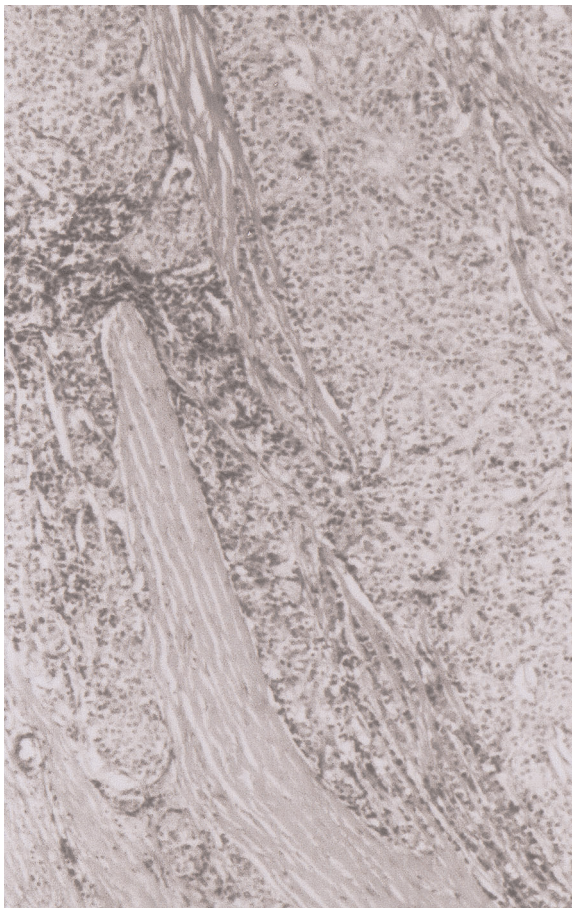

Figure 2. Parathyroid carcinoma with capsular invasion.

parathyroid cancer recurrence at regular follow-ups performed at 6, 12, and 18 months postoperatively.

DISCUSSION

Parathyroid cancer is a rare entity and natural occurrence is less than 1% of all primary hyperparathyroidism cases. The etiology is largely unknown but some associations are familial hyperparathyroidism, multiple endocrine neoplasia type 1, and previous irradiation to the head and neck (3).

It is a slow-growing, persistent, locally recurrent tumor. Therefore, most of these lesions are likely to present with a palpable cervical mass and signs of severe hypercalcemia affecting primarily the renal and skeletal system. Rarely, nonfunctioning parathyroid carcinomas, with normal serum levels of PTH, do occur but in less than 2% of all parathyroid carcinomas (3,4). Any parathyroid gland with a grey appearance, firm texture, or adherence to surrounding tissue must be treated as carcinoma. Frozen section diagnosis is very difficult, therefore, malignancy cannot be ruled out with certainty (5). Clinical suspicion of parathyroid malignancy guides the extent of surgery and performance of an adequate en-bloc removal of the primary lesion when appropriate offer the best local cure for a patient with this unusual malignancy.

Patients with parathyroid carcinoma represent a heterogeneous group. Some patients are cured for as long as a decade or more, whereas some with aggressive tumor experience recurrence early with local and distant spread. Approximately 50% of patients have recurrent disease and of these patients 50% have metastatic spread most commonly to lung, liver, and bones (1,6). Lifetime monitoring of patients with parathyroid carcinoma, with periodic measurement of serum calcium levels is, therefore, advised.

The association of papillary carcinoma and parathyroid adenoma and/or hyperplasia has been described by Prinz (7) and Burmeister (8). Most of the patients in these series had previous history of neck irradiation and none of the cases had parathyroid carcinoma.

The number of the concomitant parathyroid carcinoma and thyroid carcinoma is very few (2,3,9). In Schoretsanitis' case the patient had hypercalcemia, and preoperative ultrasonography demonstrated a mass at the left lower thyroid pole but was unable to evaluate whether the mass originated from the thyroid or the parathyroid parenchyma. Koea's patient and Savli's patient had no signs of hypercalcemia preoperatively. Both authors detected the enlarged parathyroid glands intraoperatively. In all three cases en-bloc resection of enlarged parathyroid glands were performed and histopathological examination confirmed the diagnosis of parathyroid carcinoma. All cases presented here succeeded uneventful recovery after the operation. Two of these patients had the previous history of irradiation to neck region. Our case is the second case of concomitant thyroid and parathyroid carcinoma without any previous neck irradiation in literature. As the authors, we think that any abnormally enlarged parathyroid gland detected during thyroidectomy for malignancy should raise the suspicion of a second primary in the parathyroid gland.

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Corresponding Author:
Dr. Mehmet KEŞKEK
Reşat Nuri Sok. No: 102/16
Yukarı Ayrancı
ANKARA

Tel: (0.312) 310 30 30
Faks: (0.312) 310 34 60
E-mail: keskekm@yahoo.com