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A case of parathyroid carcinoma mimicking parathyroid adenoma

Paratiroid adenomunu taklit eden paratiroid karsinomu olgusu

Semra Demirli Atıcı [1], Değercan Yeşilyurt [1], Dudu Solakoğlu Kahraman [2], Hakan Öğücü [1], Emre Dikmeer [1], Halit Batuhan Demir [1], Gökhan Akbulut [1]

[1] Department of General Surgery, University of Health Sciences, Tepecik Training and Research Hospital, Turkey
[2] Department of Pathology, University of Health Sciences, Tepecik Training and Research Hospital, Turkey

ORCID ID of the authors
SDA: 0000-0002-8287-067X
DY: 0000-0001-6938-2076
DSK: 0000-0002-4126-5326
HÖ: 0000-0003-4089-1162
ED: 0000-0001-9442-0273
HBD: 0000-0002-2746-7941
GA: 0000-0002-9054-1446

Corresponding author / Sorumlu yazar:
Semra Demirli Atıcı
Address / Adres: S.B.Ü. Tepecik Eğitim ve Araştırma Hastanesi Güney Mahallesi, 1140/1. Sk. No:1, 35180 Yenişehir, Konak, İzmir, Türkiye
e-Mail: smrdemirli@hotmail.com

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Abstract
Parathyroid carcinoma is one of the rare endocrine tumors and constitutes 1% of the cases with primary hyperparathyroidism. Because of similar imaging modalities and similar clinical findings, it is difficult to distinguish between preoperative parathyroid adenomas and parathyroid carcinoma. A 70-year-old female patient presented with fatigue and generalized bone pain. Her laboratory tests, neck and parathyroid scintigraphy were compatible with primary hyperparathyroidism with a significantly elevated level of parathormone. With the imaging methods which supported parathyroid adenoma, the patient was operated with a presumptive diagnosis of primary hyperparathyroidism due to parathyroid adenoma. The postoperative course was unremarkable and she was discharged on the postoperative fifth day. Postoperative pathology was reported as parathyroid carcinoma developed in a parathyroid adenoma. Parathyroid carcinoma and parathyroid adenoma have similar clinically and imaging methods, it is difficult to diagnose preoperatively.
Keywords: Hypercalcemia, Primary hyperparathyroidism, Parathyroid adenoma, Parathyroid carcinoma

Öz
Paratiroid karsinomu nadir görülen endokrin tümörlerden biri olup primer hiperparatiroidizmli olgularının %1'ini oluşturmaktadır. Benzer görüntüleme yöntemleri ve benzer klinik bulgular nedeniyle preoperatif paratiroid adenomları ve paratiroid karsinomu ayırt etmek zordur. 70 yaşında kadın hasta, yorgunluk ve yaygın kemik ağrısı şikayeti ile başvurdu. Laboratuvar testleri, artmış parathormon düzeyi, boyun ultrasonografisi ve paratiroid sintigrafisi, primer hiperparatiroidizm ile uyumluydu. Paratiroid adenomunu destekleyen görüntüleme yöntemleri ile hasta paratiroid adenomuna bağlı primer hiperparatiroidi ön tanısı ile opere edildi. Ameliyat sonrası problem saptanmayan hasta, postoperatif beşinci günde sorunsuz bir şekilde taburcu edildi. Postoperatif patoloji paratiroid adenomunda gelişen paratiroid karsinomu olarak rapor edildi. Paratiroid karsinomu ve paratiroid adenomu benzer klinik ve görüntüleme yöntemlerine sahiptir, preoperatif tanı koymak zordur.
Anahtar kelimeler: Hiperkalsemi, Primer hiperparatiroidizm, Paratiroid adenomu, Paratiroid karsinomu

Introduction

Approximately 1% of the cases of primer hyperparathyroidism constitute parathyroid carcinoma, which was a rare type of endocrine tumor [1]. Patients usually have symptoms of hyperparathyroidism, except that clinically parathyroid carcinoma has no significant findings. The mainly diagnosis of the disease is made with histopathological evaluation. The precise etiology of parathyroid carcinoma is unknown; radiotherapy history in neck region, some sporadic and familial tumors such as hereditary hyperparathyroidism jaw tumor (HPT-JT) and multiple neuroendocrine neoplasia type 1 (MEN1) mutations being accused in the etiology [2].

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Case presentation

A 70-year-old female presented with weakness, fatigue, and generalized bone pain. She had a history of hemigastrectomy due to peptic ulcer, cholecystectomy, and deep vein thrombosis. Her laboratory tests were reported Calcium: 11.8 mg/dL (normal range 8.8-10.6 mg/dL) Phosphor: 3.2 mg/dL (normal range 2.5-4.5 mg/dl), 25 Hydroxy Vitamin D: 15.47 ng/dL and significantly elevated Parathormone (PTH): 1459 pg/mL (normal range 18.5-88.0 pg/mL). Thyroid ultrasonography showed a 2.5x3 cm sized semisolid lesion at the posterior right thyroid lobe which was extending into the retrosternal area. A 99m Sestamibi computerized tomography (CT) scan confirmed increased uptake in the topography of the right lobe inferior, which was first evaluated in favor of parathyroid adenoma. The presumptive diagnosis of primary hyperparathyroidism due to parathyroid adenoma surgery was performed. Written informed consent which was necessary was obtained from the patient for treatment, surgery, and publication. During surgery, palpable, fixed lymph node which was enlarged was send frozen. The enlarged lymph node was reported as reactive and right parathyroidectomy was performed. On gross pathology revealed a cystic nodule which was measured as 35x28x27 mm (Figure 1). Histopathological examination showed features compatible with parathyroid carcinoma which has revealed capsular and vascular invasion (Figure 2, 3). Surgical margins of the specimen were free for tumor. Eleven lymph nodes were reactive. Postoperative first day PTH and patient's serum calcium level performed. PTH levels decreased 15.6 pg/mL and calcium levels declined to 8.4 mg/dL. The patient postoperative course was unremarkable and she was discharged on the postoperative 5th day. She was directed to the oncology department for follow-up and treatment. The patient was in follow-up at postoperative nearly one year with no local recurrence or distant metastasis.

Figure 1: Gross pathology: parathyroid carcinoma, cystic nodule (35x28x27 mm)

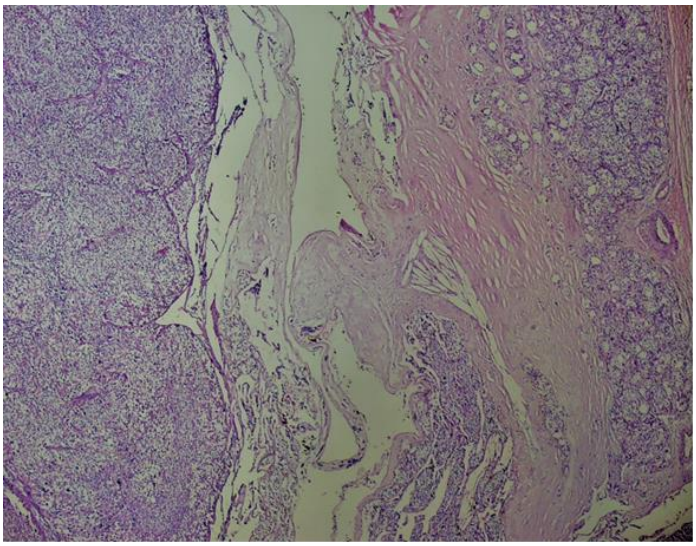

Figure 2: Right side of the figure: ordinary parathyroid tissue, Left side: encapsulated tumor. Tumor invasion into capsule and vascular space; Hematoxylin and Eosin ×40

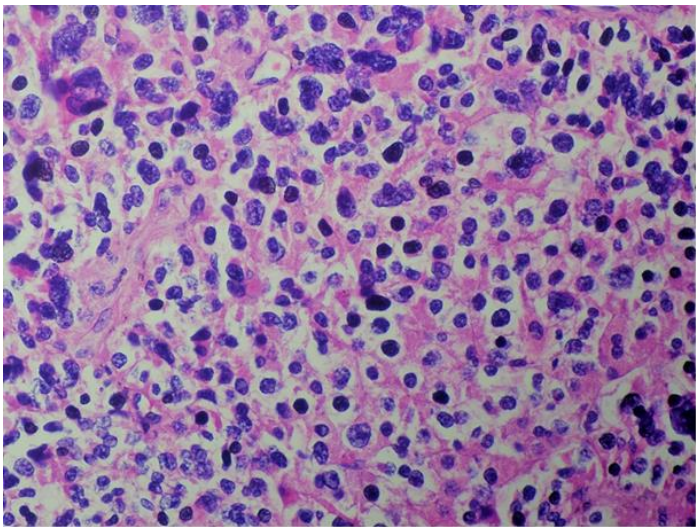

Figure 3: Tumor cells containing pleomorphic nuclei with macronucleoli. Hematoxylin and Eosin ×400

Discussion

Parathyroid carcinoma is a rare endocrine tumor with an incidence of 0.0002% [1]. The incidence of parathyroid carcinoma is equal in both men and women and especially seen fifth decade of life [2]. Parathyroid carcinoma constitutes 1% of the cases with primary hyperparathyroidism [1]. Patients usually present with complaints of weakness, fatigue, nausea, vomiting, anorexia, constipation, and common bone pain [1]. Etiopathogenesis of parathyroid carcinoma is mainly unclear. Parathyroid carcinoma incidence is increasing with the patient who has radiotherapy history in the neck region, secondary hyperparathyroidism due to renal insufficiency and some sporadic and familial tumors such as HPT-JT and MEN type 1 [2,3].

Imaging methods like ultrasound, CT, magnetic resonance imaging (MRI) is helpful in determining tumor localization but cannot help to differentiate if it is benign/malign [4]. But, MRI with gadolinium can give us detailed information, about supplement the assessment because of showing best detail on soft tissues of the neck [5].

Typically, high blood calcium levels which were >14mg/dl and high parathormone >5 times the upper limit, which were >300 pg/dl, levels should be clinically suspicious in

preoperative laboratory tests. Although a definitive diagnosis is usually made with postoperative pathology specimen reports [6]. Preoperatively with high suspicious parathyroid carcinoma diagnostic fine needle aspiration biopsy is deprecated because of the risk of seeding the tumor and also the high possibility of false negatives [7].

If we suspected about parathyroid carcinoma per-operatively frozen can be studied. However, since some pathologic features may be observed with parathyroid carcinoma in some benign adenomas, the frozen study is generally not reliable [8]. However, studies show that the best opportunity of surgical treatment for parathyroid carcinomas is en-bloc resection of the tumor with the ipsilateral thyroid lobe which provides better local disease control and improves long term-survival significantly [9]. In our case, we performed only parathyroidectomy and near one year follow up the patient has no local recurrence or distant metastasis. As our case during surgery enlarged, suspicious enlarged lymph nodes can be resected but studies shows that unnecessary prophylactic radical neck dissection can increase the risk of surgical complications [10].

For treatment of parathyroid carcinoma, except some case reports and small studies, adjuvant chemotherapy and radiotherapy didn't found effective [11]. Postoperative adjuvant radiation therapy might be useful in the treatment of patients with lymph node metastases and histologically tumor -positive surgical margin [12]. Local recurrence is very commonly seen. Most patients present with symptoms of increasing serum calcium levels and laboratory tests show accompanying high levels of PTH. Also distant metastases to bone, lung, and liver can be seen in parathyroid carcinoma. For local or distant metastasis diseases the goal of treatment is controlling hypercalcemia and symptoms of hypercalcemia. Localized diseases which were resectable, surgical resection can be performed [5,9].

In conclusion, although it is possible to obtain information about preoperative mass localization with similar clinical findings and imaging methods, it is difficult to distinguish between parathyroid adenomas and carcinomas. High parathormone levels should cause clinical suspicious in us about parathyroid carcinoma.

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