Case Report / Olgu Sunumu

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A Rare Presentation of Primary Hyperparathyroidism: Generalized Brown Tumors

Primer Hiperparatiroidizmin Nadir Bir Sunumu: Yaygınlaşmış Kahverengi Tümörler

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Abstract

Primary hyperparathyroidism is a common and easily diagnosed disease at present. However, its complicated presentation as a brown tumour is rare. Here, we present the case of a patient with knee pain and common asymmetrically located brown tumours. The parathyroid adenoma was radiologically detected, followed by successful minimally invasive parathyroidectomy. However, the brown tumours regressed after surgery. Primary hyperparathyroidism can still present with common bone involvements and can be clinically and radiologically improved with appropriate treatment.

Keywords: Primary hyperparathyroidism, bone, brown tumour, parathyroidectomy

Öz

Primer hiperparatiroidizm, günümüzde yaygın ve kolay teşhis edilebilen bir hastalıktır. Neyse ki, kahverengi tümör şeklinde komplikasyonla karşılaşılması nadirdir. Aşağıda, diz ağrısı ile başvuran ve asimetrik yerleşimli kahverengi tümörleri olan primer hiperparatiroidizmli bir olgu sunduk. Paratiroid adenomu radyolojik olarak tespit edildi, ardından minimal invaziv paratiroidektomi ile başarıyla ameliyat edildi. Kahverengi tümörler ameliyattan sonra geriledi. Primer hiperparatiroidizmin hala yaygın kemik tutulumu ile ortaya çıkabildiği ve uygun tedavi ile klinik ve radyolojik olarak iyileştirilebileceği anlaşılmaktadır.

Anahtar kelimeler: Primer hiperparatiroidizm, kemik, kahverengi tümör, paratiroidektomi

Introduction

Primary hyperparathyroidism (PH) is one of the commonest endocrinological disorders in the whole world (1). It presents with hypercalcemia, hypo or normophosphatemia and high or mid-to-high parathyroid hormone level (2). Its target organs are kidney, bone and intestine, main signs and symptoms are artralgia, bone pain, urinal stones, bone fractures, cysts and/or brown tumors (3). Brown tumor, also called osteoclastoma, is a focal, benign lytic bone lesion of primary or secondary hyperparathyroidism. Today, PH can be diagnosed earlier and the incidence of this lesion has decreased (4). We present a case with brown tumors involving whole skeleton.

Case Report

A 39-year-old woman admitted to orthopedics with left knee pain. A hypoechoic lesion was detected on X-ray. Laboratory evaluation revealed mild hypercalcemia with calcium of 11.6 mg/dL. A whole-body bone scintigraphy was taken on suspicion of cancer, multiple similar lesions with increased activity were determined on right and left knees, 4th thoracal spine, left iliac crest, right symphysis pubis and between left femur's head and neck (Figure 1). An excisional biopsy was made and reported as brown tumor. After that she was directed to our endocrinology policlinic because of hypercalcemia and hyperparathyroidism. We detected that calcium: 13 mg/dL, phosphorus: 2.4 mg/dL, parathyroid hormone: 625 pg/mL, 25 hidroksi vitamin D: 8 ng/mL and we hospitalized her with the diagnosis of PH. We started intensive hydration with 0.9% NaCl and followed urine output, then gived intravenous furosemide infusion. In the one daily urine, calcium excretion was 461 mg/day. On the ultrasound, we detected a single, smooth edged, non-vascularized, hypoechoik lesion, size in 8x10x15 millimeters, under the right thyroid lobe and two thyroid nodules more than 15 millimeters. Parathyroid scintigraphy showed same lesion. Thyroid fine needle aspirations were resulted as benign thyroid nodule. We also researched multiple endocrine neoplasia (MEN) type

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1 because of young age of the patient and we did not detect any other components of this syndrome. We also excluded other reasons for hypercalcemia like malignancy. Although we used hydration and loop diuretic threapy, serum calcium did not fall enough, so we had to use calcitonin and succeeded that calcium falled under 11 mg/dL. We applied vitamin D to protect from hungry bone syndrome (HBS) and transfered the patient to general surgery for operation. Minimally invasive parathyroid adenomectomy was performed successfully and calcium and parathyroid hormone levels decreased quickly. The pathology was resulted as parathyroid adenoma. By the way, vitamin D had been replaced preoperativelly, nevertheless the patient developed HBS and she was given medication. After 6 months, the bone scintigraphy was repeated and reported that the previous lesions were regressed (Figure 2). Informed consent was obtained from the patient.

Discussion

Brown tumor or osteitis fibrosa cystic is the result of increased osteolitic activity and fibroblastic proliferation due to uncontrolled primary or secondary hyperparathyroidism (5). Its brown colour arises from bleeding and hemosiderin deposition (6). The incidence is nearly 5% of PH (7) and this has decreased because of the earlier diagnosis and therapy of the disease (8). In the case report of 100 patients from Poland, brown tumors were identified in 10 cases, the authors noted that they did not expect so high result (9). Because of the radiological view, it should be differentiated from giant cell granuloma, aneurysmal bone cyst and similar lesions. Biopsy and elevated parathyroid hormon level can help in this instance (10).

Brown tumor often occurs in pelvis, long bones and ribs. Additionally, it can be seen on face and so any part of skeleton (11). It can cause paraplegia, fracture, movement limitation, bleeding and pain according to its site (4,12). Rarely, PH can cause brown tumor with extensive parenchymal calcification in kidneys, it calls as 'putty kidney' (radiological name) (13). It can mimic other diseases like malignancy and so the diagnosis delays (14). Vaishya et al. (15) reported a brown tumor of tibial diaphyses in a young female patient. She had severe pain, tenderness and immobilization due to lesion. The radiograph showed a geographical lytic lesion like malignancy. But she had secondary hyperparathyroidism according to severe Vitamin D deficiency. After vitamin D replacement, the lesion was disappeared, so it was diagnosed as brown tumor clinically and she was prevented unnecessary surgery. In our patient, it was considered as malignancy, so whole body bone scintigraphy was performed. It showed a great number of similar lesions spreading all over the skeleton asymmetrically and move from here the pathological diagnosis was made.

Because of reporting as brown tumor, she referred our clinic and the diagnosis and treatment came step by step. During this time, we researched MEN type 1 owing to the patient's age. Because, PH is the most common component of this syndrome and MEN-1 is seen in 1-3% of PH (16). Shortly after surgery, HBS occurred despite of vitamin D replacement preoperatively so she was administered intravenous and oral therapy and she improved quickly. HBS is an uncommon complication of parathyroidectomy for severe PH. Patients with preoperative

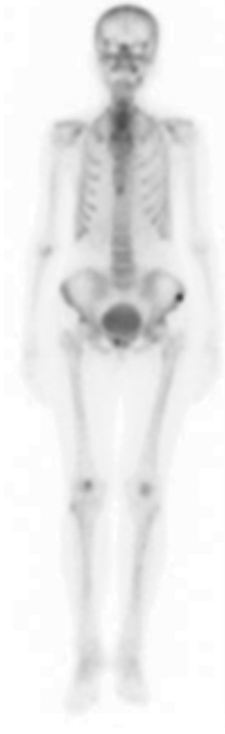

Figure 1. The wholebody bone scintigraphy

Figure 2. The whole body bone scintigraphy after 6 months from the surgery

high bone turn over-like our patient- have this risk highly. Long duration of PH is also important. The therapy and follow of this syndrome must be done quickly and carefully (17).

Conclusion

Despite of developing in diagnostic methods, formerly seen lesions like brown tumor can occur any time. It comes in front of us with a lot of symptoms. Both of clinicians and surgeons must be alert for this lesion. If it is not considered, the diagnosis delays and it can cause inappropriate medical interventions and irreversible damages to patients.

Ethics

Informed Consent: Informed consent was obtained from the patient.

Peer-review: Externally and internally peer-reviewed.

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