Primary Hyperparathyroidism: A Rare Cause of Spinal Cord Compression: Differential Diagnosis

Primer Hiperparatiroidi: Spinal Kord Basısının Nadir Bir Sebebi Olabilir

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ABSTRACT A 66-year-old woman was admitted to our hospital with difficulty in walking. Magnetic resonance imaging revealed mass lesions at T2- T3 vertebrae causing deviation of the spinal cord. The positron emission tomography (PET) scan of the body showed a hypermetabolic state in the right maxillary sinus and multiple metastatic lesions in the skeletal system indicating a metastatic tumor of unidentified origin. The patient was operated immediately for cord compression that could lead to a neurological problem. Pathological examination of the operation specimen was consistent with a giant cell tumor. The patient's serum calcium level was elevated (11.6 mg/dL; reference limits: 8.5-10.5) and phosphorus level was below the normal ranges. Parathyroid hormone was extremely high; 2097 pg/mL (reference limits: 15-65 pg/mL). Sonographic examination of the neck revealed multinodular goiter and a lesion at the inferior region of the right thyroid lobe suggesting a parathyroid adenoma. Skeletal X-rays suggested Brown tumors of long bones. The case was diagnosed as primary hyperparathyroidism and was referred for parathyroidectomy.

Key Words: Hyperparathyroidism, primary; spinal cord compression; hypercalcemia, giant cell tumors

ÖZET 66 yaşında bayan hasta hastanemize yürümede zorlanma yakınması ile yatırıldı. Çekilen manyetik rezonans görüntülemesinde T2-3 omurlar seviyesinde kitle lezyonları izlendi. Bu lezyonlar spinal kord üzerinde deviasyona sebep olmaktaydı. Yapılan pozitron emisyon tomografi (PET) görüntülemede sağ maksiller sinüste hipermetabolik aktivite ve iskelet sisteminde ise çok odaklı metastatik lezyonlar tespit edildi. Bulgular odağı bilinmeyen bir tümör metastazını düşündürmekteydi. Sinir dokusu basısının nörolojik sekel geliştirme riski nedeniyle hasta acilen ameliyat edildi. Operasyon materyalinin patolojik incelemesi dev hücreli tümör ile uyumluydu. Hastanın serum kalsiyum düzeyleri (11.6 mg/dL; normal sınır: 8.5-10.5 mg/dL) yükselmiş, fosfor düzeyleri ise azalmış olarak bulundu. Paratiroid hormon düzeyi (2097 pg/mL; normal sınır: 15-65 pg/mL) çok yüksekti. Sonografik incelemesinde multinoduler guatr ile sağ tiroid bezi inferior kesiminde bir lezyon tespit edildi. İskelet sistemi direkt grafi incelemesi uzun kemiklerin Brown tümörü ile uyumluydu. Olgu primer hiperparatiroidi tanısı alarak paratiroidektomiye yönlendirildi.

Anahtar Kelimeler: Hiperparatiroidi, primer; spinal kord kompresyonu; hiperkalsemi, dev hücreli tümör

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Primary hyperparathyroidism (PHPT) is a calcium metabolism disorder caused by overproduction of parathyroid hormone (PTH) by the parathyroid glands.[1,2] It is characterized with elevated serum calcium and alkaline phosphatase and low phosphorus levels.[3] In symptomatic patients, dysfunction of CNS, peripheral nerves and/or muscles, gastrointestinal tract and/or joints may occur.[4]

Patients with mild hypercalcemia are usually asymptomatic, however those with moderate to severe hypercalcemia often complain of nausea and vomiting. Muscle weakness, impaired concentration and memory, difficulty with cognitive function, stupor and coma can also be observed in affected individuals. Constipation and electrocardiographic alterations may be the manifestations of this disorder, as well.[1,2]

Over-excretion of parathyroid hormone in PHPT can lead to an imbalance of osteoclastic and osteoblastic activity in the fibrous stromal matrix in multiple skeletal lesions and can be presented as Brown tumors. This is not a true tumor, being an unusual, reactive lesion in association with primary or secondary hyperparathyroidism.[3] Spinal complication of both primary and secondary hyperparathyroidism is rase and affects 15% and 13% of the patients, respectively.[5] Radiological features of multiple bone lesions may sometimes mimic multiple metastatic tumors and suggest the terminal stage of a malignancy.[3] Involvement of the spine with neural compression is very rare.[5]

In this report, we present the case of a woman with Brown tumors mimicking multiple metastases leading to spinal cord compression.

CASE REPORT

A 66 year-old-woman was admitted to our hospital with difficulty in walking. She complained of upper back pain and weakness in both lower extremities. Her pain was reported to start two months before admission and was unresponsive to standard analgesics. Her medical history was remarkable for a peptic ulcer operation and she had a weight loss of approximately 7-8 kg within the last six months. Physical examination revealed a tender and soft mass in the thoracic region without warmth and erythema. Neurological examination showed decreased pinprick sensation on both sides and brisk deep tendon reflexes in both lower extremities. Emergency magnetic resonance imaging showed mass lesions at the corpus of T2 and T3 vertebrae, involving also posterior elements and leading to expansion of the bone mass. These lesions extended into the epidural space and lead to deviation of the spinal cord. The positron emission tomography (PET) scan revealed a hypermetabolic state of the right maxillary sinus and multiple metastatic lesions at the skeletal system. These signs suggested a metastatic tumor of unknown origin. Due to risk of neurological cord compression that could lead to a neurological sequele, the patient was operated immediately. The mass was removed and laminectomy of the T2, T3 and T4 vertebrae was performed. Pathological examination of the excised mass was consistent with giant cell tumor (Figure 1). A detailed biochemical analysis was performed in order to evaluate the underlying pathological disorder. The patient's serum calcium level was found slightly elevated (11.6 mg/dL; reference limits: 8.5-10.5 mg/dL), and phosphorus was low (1.55 mg/dL, reference limits: 2.7-4.5 mg/dL). Serum parathyroid hormone level was extremely high (2097 pg/mL, reference limits: 15-65 pg/mL) and an endocrinology consultation was requested. Sonographic examination revealed a multinodular goiter and a lesion in the inferior region of the right thyroid lobe indicating a parathyroid adenoma, which was confirmed by scintigraphic evaluation. Numerous lytic defects in the femur and humerus of the patient on direct X-rays suggested focal Brown tumors. Relying on all these findings, the case was diagnosed as primary hyperparathyroidism and was referred to the surgical department for parathyroidectomy. Pathological examination of the excised specimen was compatible with a parathyroid adenoma (Figure 2).

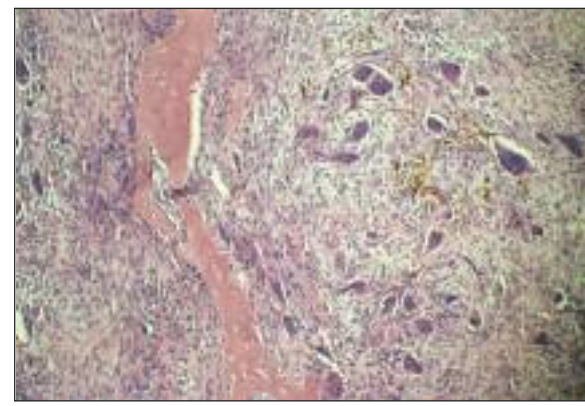

FIGURE 1: Multinucleated osteoclasts (giant cells) in the specimen obtained from T3 and T4 vertebrae (Hematoxilen-eosin x100).

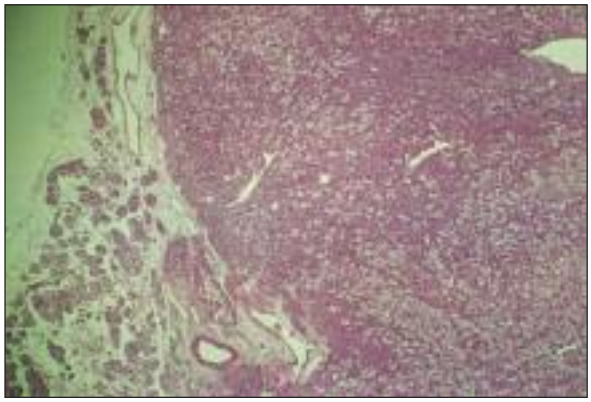

FIGURE 2: Parathyroid adenoma staining (Hematoxilen-eosin x40).

DISCUSSION

Primary hyperparathyroidism is a common endocrine disorder and is characterized by hypercalcemia and hypophosphatemia.[3,5] It has various clinical manifestations and may primarily affect kidney, skeletal system and the gastrointestinal tract.[4] Symptoms due to hypercalcemia such as drowsiness, anorexia, abdominal pain, constipation, nausea, and vomiting, as well as renal calculi are common.[6]

Brown tumors are benign focal bone lesions caused by increased osteoclastic activity and fibroblastic proliferation. They may be observed in primary or rarely in secondary hyperparathyroidism. Nowadays such extensive and destructive bone disease due to hyperparathyroidism is hardly seen because of early diagnosis and successful treatment of hyperparathyroidism. Brown tumors of hyperparathyroidism may appear in any bone, however they are frequently seen in the facial bones and jaws, sternum, pelvis, ribs and femur.[7,8] Spinal cord compression has been reported very rarely in PTHP.[4] Although involvement of the spine is rare, it can require emergent surgery for the prevention of a neurological sequela, as it happened in our patient.[7] She was operated due to the high risk of permanent neurological impairment.

The characteristic findings of hyperparathyroid bone on microscopic examination are increased osteoclastic activity, peritrabecular fibrosis and giant cells. Brown tumor and neoplastic giant cell tumor are histologically similar, however the diagnosis of Brown tumor rests on the presence of biochemical hyperparathyroidism.[8]

Depending on literature that we mentioned above, our patient was diagnosed as primary hyperparathyroidism, and a parathyroid adenoma was detected at the surgical exploration of her neck. Following surgery, her serum PTH and calcium levels returned to normal and she gradually began to walk without help.

In conclusion, primary hyperparathyroidism has a wide spectrum of manifestations and the Brown tumors leading to spinal cord compression are rare. However these tumors can cause severe neurological deficits. Hyperparathyroidism should be kept in mind in the differential diagnosis of patients presenting with symptoms such as anorexia, abdominal pain, constipation, nausea, vomiting, history of urolithiasis and difficulty in walking.

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