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Brown Tumor of Lumbar Spine in a Patient with Primary Hyperparathyroidism: Case Report

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ABSTRACT

Background Data: Brown tumor is a lytic bone tumor and has variable symptoms according to the affected site. It is called brown because of its characteristic brown color believed to have occurred after accumulation of blood, fibrous tissue, and hemosiderin. Hyperparathyroidism can result in a brown tumor whether it is primary or secondary, but this case is much rarer in primary hyperparathyroidism. It rarely affects the spine, while it is extremely rare to compromise the spinal canal.

Purpose: To report a rare case of lumbar spinal brown tumor with primary hyperparathyroidism.

Study Design: A case report of brown tumor of the lumbar spine (5th lumbar vertebrae).

Patients and Methods: Our case was a forty-year-old female patient presenting with persistent dull aching low back pain. There was no history of any chronic illness. She had a 5-month history of slowly progressive left lower limb radicular pain and not associated with sphincter disturbance. Magnetic resonance imaging of the spine demonstrated lesion affecting the 5th lumbar vertebrae invading the left pedicle and compressing the spinal canal.

Results: Team did neural decompression, biopsy harvesting, and partial corpectomy with reconstruction of the affected lumbar vertebra (L5) followed by fixation by rods and screws. The patient showed relief of the radicular pain and back pain on VAS. Histopathological analysis of the biopsy showed evidence of giant-cell tumor (brown tumor). Parathyroid survey was done and showed normal calcium levels. By revising the literature, we claim that this present study is reporting the 10th case of brown tumor of lumbar spine in patient with primary hyperparathyroidism.

Conclusion: The diagnosis of brown tumor of spine could be considered in patients with lytic spine lesions. (2019ESJ192)

Keywords: Giant cell tumor, Lumbar spine, Brown tumor, Primary hyperparathyroidism

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INTRODUCTION

Brown tumor is a rare bone tumor that occurred more commonly in secondary hyperparathyroidism (HPT) but much less in primary hyperparathyroidism.[1] There were only 24 patients of brown tumor of spine in primary HPT were reported in the English literature: of them, nine patients had lumbar spine affected by the tumor. We claim that our case report is the 25th reported case among all spine levels and the 10th case in the lumbar spine only reports.[4]

Brown tumor is rare nonneoplastic focal lytic lesions. It is also termed "osteoclastoma", more widely and famously known as 'brown tumor" due to the high level of hemosiderin deposition and vascularity that is concentrated within the osteoclasts within the lesion.[2,9] It can occur as solitary focal or multifocal discrete lesions and involve any bony structure such as pelvis, ribs, clavicle, mandible, upper and lower limbs; however, its involvement of the spine is extremely rare.[6] The abnormal level of parathyroid hormone induces reactive process because of bone resorption and remodeling.[6,7]

Here, we present one case of brown tumor that underwent operation in our department and review the literature of other similar reports

CASE REPORT

A forty-year-old female patient had no chronic illness, with average body weight presenting with persistent slowly progressive dull aching low back pain that was relieved temporarily with analgesics and increased by exercise for about 8 months. Left lower limb sciatica in the form of numbness and tingling extended to the big toe started to develop followed by bilateral neurogenic claudication. There was no report of gait disturbance, sphincter abnormality, fever, or marked loss of weight.

On examination, the patient showed normal intact skin of the back, average lordotic curvature of the lumbosacral spine, no muscle spasm, full motor power, normal reflexes, and preserved sensation bilaterally of both upper and lower limbs.

Radiological investigations were required. Computed tomography (CT) of the lumbosacral spine showed lytic changes within the 5th lumbar vertebral body extending to the left pedicle with preserved height and both upper and lower endplates. Magnetic Resonance Imaging (MRI) of the lumbosacral spine showed hyperintense lesion that was hyperintense in TW2 and isointense in TW1 that was invading the 5th lumbar vertebral body and the left pedicle with marked neural compression on the theca and lateral recess on the left side (Figure 1).

The team prepared the patient for surgery taking in mind facing bony tumor that mostly would be aneurysmal bone cyst tumor. The goal of the surgery was fixation, decompression, fusion, and in situ reduction of the tumoral tissue with harvesting particles for histopathological analysis. Under general anesthesia, in prone position, low back midline skin incision and dissection and insertion of paired polyaxial screws at the level above L4 and level below S1, the affected vertebra (L5), were performed (Figure 3). The team started by anchoring the rod in between the screws in the right side and then opened the left transpedicular partial corpectomy of L5 with excising most of the lesion which was dark red suckable mild vascular tissue. Formal L5 laminectomy was done followed by insertion of transpedicular polyaxial screw on the right side only of L5 and insertion of bone cement when it was semisolid in consistency to augment preservation of the vertebral height and fusion. Neurolysis of the spinal roots and completing the fixation system (rods and screws) were employed (Figure 1).

The excised tumoral tissue biopsy was sent to the histopathological analysis that revealed mass formed of lobules of plump fibroblasts, extravasated red blood cells, hemosiderin-laden macrophages, and osteoclast-type giant cells that cluster in areas of hemorrhage. The lobules are separated by septa composed of reactive fibrous

tissue and trabeculae of woven bone. The adjacent bone showed osteoclasts resorbing centers of bony trabeculae and no evidence of malignancy (Figure 1).

The team revised the literature and the patient was sent to complete the required laboratory and radiological investigations to search for the evidence of HPT which in this case would be primary as the patient had no history of any chronic illness. All the results (parathyroid hormone, serum phosphorus, calcium, and ionized calcium) were within normal range except the neck ultrasonography that showed an oval-shaped left parathyroid mass 7x3x3 mm with no detectable cystic or calcified areas (Figure 1).

Postoperatively, the patient showed marked immediate relief from the back pain and sciatica. After the final result of the pathology and excellent recovery and ambulation of the patient, the team decided to transfer the patient to the endocrinology department to complete the medical treatment and follow-up periodically in our outpatient clinic.

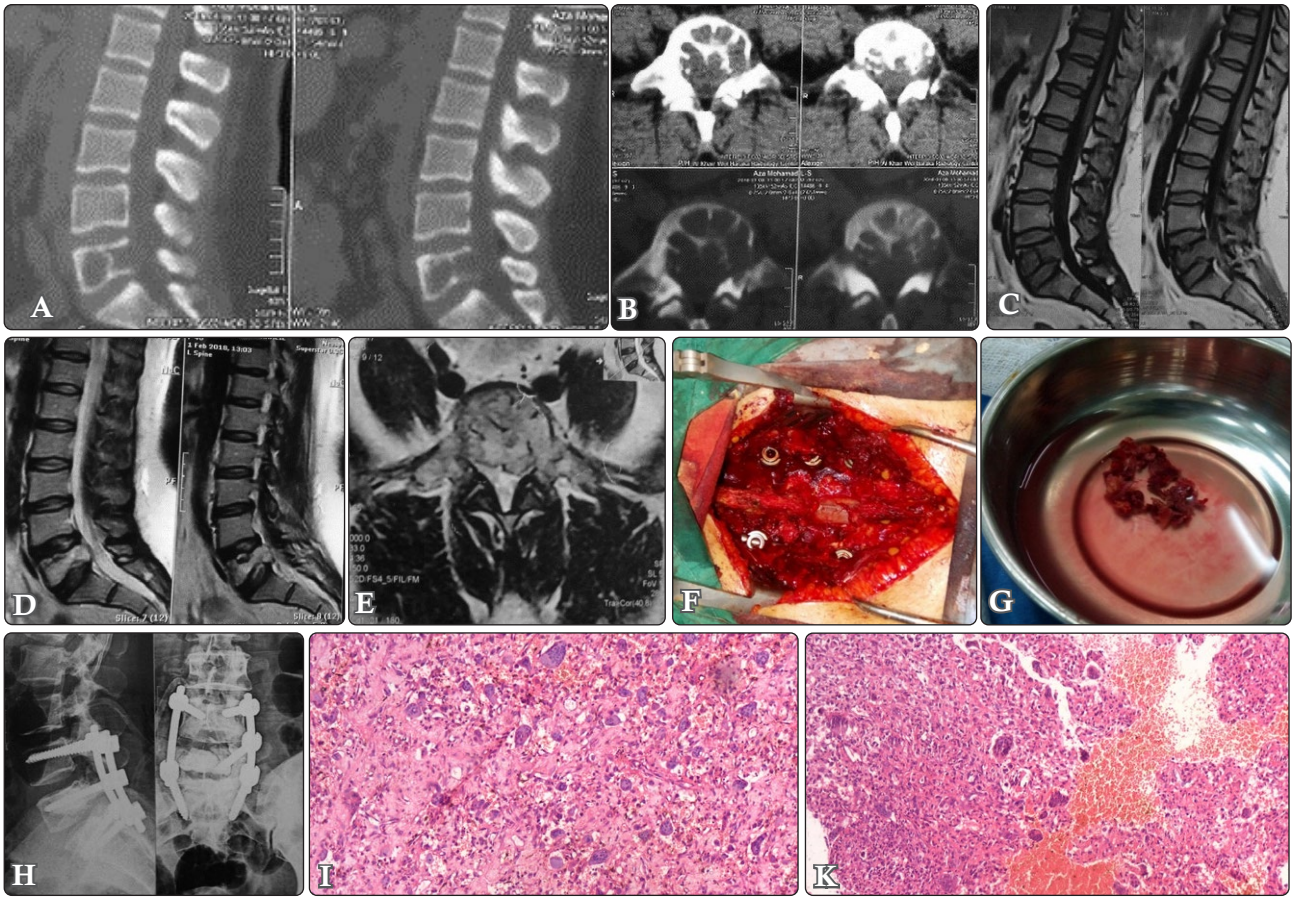

Figure 1. The images of our patients showing the following: (A) lumbosacral spine CT sagittal cuts and (B) axial cut views show hypodense areas of the body of 5th lumbar vertebra with preservation of the height of the body and compromise on the left pedicle. (C, D) Lumbosacral spine MRI T1 and T2 sagittal images and axial images (E) showing the lesion affecting the body of the 5th lumbar vertebra isointense in T1 and hyperintense in T2 and invading the body and the left pedicle with compromise of the left lateral recess compressing the spinal canal. (F) Intraoperative images show the exposure with pedicle screws inserted at L3, L4, and L5 on the right side and at L3 and L5 on the left side with formal laminectomy. (G) pink to dark red particles of the tumor tissue that were harvested for biopsy histopathological analysis. (H) Postoperative X-ray lumbosacral spine lateral and anteroposterior views show the transpedicular screw fixation system with jet hyperdense area inside the body of L5 vertebra of the bone cement that was used in the vertebroplasty. (I, K) Microscopic view of the harvested biopsy showing reactive-appearing hypervascular fibrous tissue, multinucleated giant cells that resemble osteoclasts but do not border bone trabeculae, extravasated red cells, and hemosiderin pigment which accounts for the brown color of the lesion.

DISCUSSION

Brown tumors are uncommon and those that involve the spine are very rare. Patients mostly have secondary HPT because of the increased osteoclastic activity and fibroblastic proliferation in the affected bone and most of them have chronic renal failure. Here, in our paper, we report a case of brown tumor in a patient without any chronic illness. In 2013, Witt et al.[10] published a case report of one case who had brown tumor affecting lumbar spine with primary HPT. In 2014, Khalatbari et al.[6] reported only 4 cases with primary HPT between 2000 and 2013, as follows: lesion was located in cervical (1 case), thoracic (1 case), and lumbar (2 cases) spine. So, and according to the literature, Khalatbari et al.[6] analyzed the reported cases of brown tumor with primary HPT and found 9 cases involving the thoracic spine, 6 cases the lumbar spine, 2 cases the sacral spine, and only one case the cervical spine. In 2015, Sonmez et al.[8] reported one case of brown tumor involving the thoracic spine with primary HPT.

Brown tumor is a space-occupying erosive lesion that appears to be brown grossly.[4] It is most common in the hands, facial bones, pelvis, ribs, and femur and rarely in vertebrae. Moreover, it appears in patients with untreated HPT. These tumors appear as reactive-appearing hypervascular fibrous tissue, multinucleated giant cells that resemble osteoclasts but do not border bone trabeculae, extravasated red cells, and hemosiderin pigment resulting in the brown color of the lesion. Brown tumor is thought to represent aberrant repair reactions in bone altered by HPT and is identical to what Jaffe named giant cell reparative granuloma.[6]

Diagnosing the brown tumor in patients with normocalcemia as mentioned here in our case report is very challenging and this can be the initial manifestation of the HPT.[4] Adenoma hyperplasia or carcinoma can cause primary HPT.[6] It is first reported in spine in 1978 by Ericsson et el.[4] Like any bony spinal tumor, the patient presented with variety of symptoms according to the affected site, but persistent mild to moderate back pain is very common. However, in primary HPT, about one-third of the patients are asymptomatic.[4,8] Radiologically, it is lytic lesion occurring because of bone resorption. Differential diagnosis according to the radiological images mostly includes metastatic tumor, multiple myeloma, and giant-cell tumour.[3,5] Management consists of surgical aspect and medical aspect.[1,6] From our point of view, the neurosurgical rule is the neural decompression with maintaining integrity of the neural elements and the stability of spine, then reaching the final diagnosis, and consulting the endocrine and general surgery departments for long-term follow-up.

CONCLUSION

We report a rare case of brown tumor affecting lumbar spinal vertebra in patient with normal calcium level. We also advise the spine surgeons to keep in mind the possibility of occurrence of the brown tumor even in the neurologically intact patients whose radiological investigations show expansive erosive spine lesions as brown tumor can be the first manifestation of the primary HPT disease.

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الملخص العربي

ورم بنى بالفقرات القطنية لمريض به زيادة أولية في هرمون الجاردرقية: تقرير حالة

البيانات الخلفية: الورم البنى هو واحد من الأورام المنهارة التي تصيب العظام الهيكلية بالجسم ويصيب أيضا فقرات العمود الفقري وهو وطيد العلاقة بحدوث خلل في التوازن بين الكالسيوم والفوسفور الحادث عن زيادة إفراز الغدة الجاردرقية سواء أسباب أولية أو ثانوية. ولكن الحدث الأكثر ندرة هو حدوث ها الورم في مرضى بهم معدلات طبيعية من الكالسيوم.

تصميم الدراسة: عرض حالة وتقريرها.

الغرض: هدفت الدراسة إلى إقرار حالة فريدة ونادرة عن حدوث الورم البنى بالفقرات القطنية (الفقرة القطنية الخامسة).

المرضى والطرق: حالة لسيدة تبلغ من العمر 44 سنة تعانى من ألم مزمن بأسفل العمود الفقري لمدة حوالى ثمانية أشهر تبعه حدوث آلام بالطرف السفلى الأيسر, وتم توقيع الكشف الطبي عليها وإجراء الفحوصات الطبية والتي أظهرت وجود ورم ممتد من خارج الفقرة القطنية الخامسة ضاغط على القناة الشوكية ومخارج جذور الأعصاب الشوكية.

النتائج: تم إجراء التدخل الجراحي وتثبيت الفقرات القطنية الثالثة والرابعة والخامسة واستئصال جزء كبير من الورم بالفقرة الخامسة مع رفع الضغط من على القناة الشوكية وتسليك مخارج جذور الأعصاب الشوكية وعمل حقن إسمنتي مفتوح للفقرة القطنية الخامسة حتى يتم التئام ولحام الفقرات والحفاظ على ارتفاع الفقرة الخامسة المصابة بعد استئصال جزء كبير منها متضمن جزء شبه كامل من الورم .

الاستنتاج: يعد الورم البنى بفقرات العمود الفقري حدث نادر ولا سيما في وجود زيادة أولية في هرمون الغدة الجاردرقية، وننصح بإدخاله في قوائم الأمراض المحتمل حدوثها عند وجود دليل إصابة أو ورم منهار بأجسام فقرات العمود الفقري عامة والقطنية خاصة كما تم ذكره في الدراسة وذلك يستلزم تحويل المريض بعد تشخيصه النهائي من قبل تحليل الأنسجة إلى قسم أمراض الباطنة والغدد الصماء لاستكمال العلاج الدوائي.