



Hormones and Signaling

Elixir Hor. & Sig 148 (2020) 54951-54953

Elixir
ISSN: 2229-712X

Torticollis Revealing Brown Tumor of Cervical Spine: Case Report

El Ghorfi S., Edderai M., Chaoui, B., Zamani O., Moumna S., I. En-Nafae, Lahkim M., Fenni J. and Saouab R

Imaging department of Military Hospital, Mohamed V.-Rabat.

Faculty of Medicine and Pharmacy, Mohammed V University in Rabat-Morocco.

ARTICLE INFO

Article history:

Received: 12 August 2020;

Received in revised form:
15 October 2020;

Accepted: 25 October 2020;

Keywords

Brown Tumor,
Osteitis Fibrosa Cystica.

ABSTRACT

Brown tumor also known as **osteitis fibrosa cystica** is a focal bony lesion due to bone remodeling from either primary or secondary hyperparathyroidism. Torticollis is an unusual clinical presentation of the tumor. We report a 65-year-old female patient who had been on hemodialysis for chronic renal failure, admitted with severe cervical pain and torticollis due to a minor cervical trauma. The imaging revealed multiple bony lesions that turned out to be brown tumors.

© 2020 Elixir All rights reserved.

Introduction

Brown tumor (BT) is a focal osteolytic lesion secondary to hyperparathyroidism. These lesions are termed 'brown tumors' because of their brownish coloring due in part to the presence of hemosiderin deposits. These tumors are commonly seen in patients with chronic kidney disease on dialysis who have repressed ability to convert 25 hydroxycholecalciferol to 1,25 dihydroxy-cholecalciferol, resulting in prolonged hyperparathyroidism[1]. BT is found in 3 and 1.5% of patients with primary and secondary hyperparathyroidism, respectively [2]. BT can be multiple or solitary. Radiographically, when multiple, brown tumors present similar findings as bone metastasis [3]. Multiple BT rarely locates in vertebra [4]. In this report, a case of multiple BT located in maxilla and cervical vertebra is presented.

Case report

A 65-year-old female patient admitted to the emergency for severe neck pain and torticollis following a fall while walking. The patient was on hemodialysis for chronic renal failure. His hemodynamic status was stable and the physical examination showed no neurological abnormality.

Computed tomography of the cervical spine showed a displaced fracture of the odontoid process, a fracture through left pedicle of C2 secondary to an expansile lytic lesion of C2 vertebral body with blown-out, thinned cortex. Other incidental findings were lytic bone lesion involving the hard palate and the scapulae. Because of the clinical manifestations and the radiological aspects a biological assessment is required. Laboratory investigations revealed elevated serum parathyroid hormone (PTH) level of 304 pg/mL (normal range 15–70 pg/mL). We concluded that the lytic lesions were in fact brown tumors.

The patient was referred to the neurosurgery department and was transferred to nephrology clinic for medical treatment.

Discussion

Brown tumors represent one of the three histological forms of renal osteodystrophy[5]. They don't represent neoplastic process, but focal bony lesions due to bone remodeling from either primary or secondary hyperparathyroidism[6]. Elevated PTH levels leads to bone microfractures and hemorrhages which cause the influx of osteoclasts and the emergence of reactive medullary fibrosis, thus forming a tumor mass.

The lesions are most often found in the mandibles, ribs, clavicles, pelvis, skull and facial area[7]. They are rarely located in the vertebrae, especially the cervical spine. In the literature, 22 cases of vertebral BT caused by primary hyperparathyroidism [8] and 27 cases caused by secondary hyperparathyroidism [9] are reported.

Clinically, brown tumors are often asymptomatic, but they can be revealed by palpable bony swelling, bone pain, pathological fractures or can also lead to a noisy neurological sign according to their locations. In our case, torticollis was additional to the pain.

Radiologically, BT can be solitary or multiple with eccentric or cortical location. They appear as Well-defined, purely lytic lesions. The cortex may be thinned and expanded, but will not be penetrated [10]. On CT, attenuation values will be in the range of blood and fibrous tissue. On MRI, The lesions may be solid, cystic, or mixed. Solid components are intermediate to low intensity on T1- and T2-weighted images, while the cystic components are hyperintense on T2-weighted images and may have fluid-fluid levels

A solitary brown tumor might be confused with solitary bone cyst, aneurismal bone cyst, giant cell tumor or giant cell reparative granuloma. With multiple brown tumors, differential diagnosis includes osteolytic metastasis and multiple myeloma. The presence of sclerotic margin excludes metastasis and additional finding of chondrocalcinosis suggested the possibility of hyperparathyroidism.

Histologically, BT don't have a specific appearance. They appear as multinucleated giant cells in a spindle cell matrix containing hemosiderin deposits [11]. There are no histological criteria to differentiate giant cell tumors [12]. In our case a biopsy wasn't performed. The coexistence of multiple osteolytic lesions and hyperparathyroidism lead us to the diagnostic.

Conclusion

BTs represent a rare manifestation of prolonged hyperparathyroidism. They can be solitary or multiple, rarely located in spine. The diagnosis should be considered specially in the context of chronic renal failure to avoid invasive and unnecessary investigations.



Figure 1. sagittal (a,b) and axial (c) CT demonstrating an expansile lytic lesion of C2 vertebral body causing a displaced fracture of the odontoid process and a fracture through left pedicle of C2

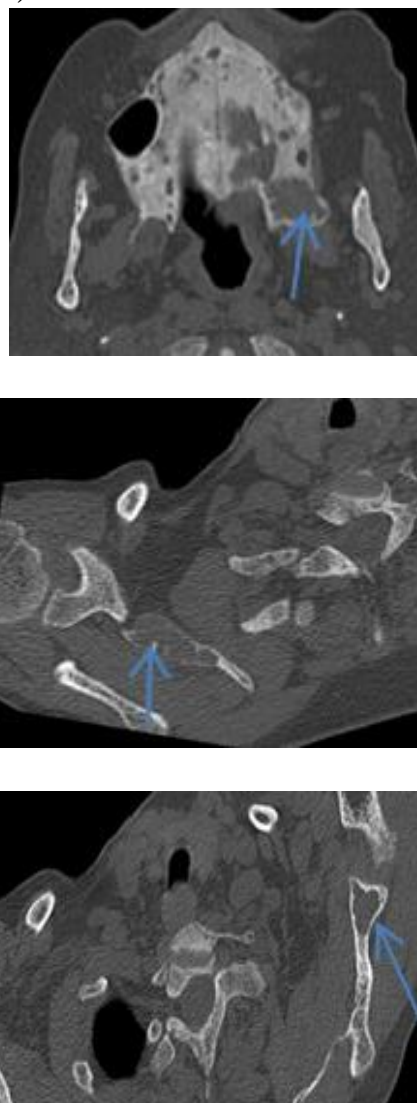


Figure 2. Axial CT scan showing lytic bone lesion of the hard palate and the scapulae

References

1. Metastasizing Carcinoma of the Parathyroid Gland with Osteitis Fibrosa Cystica and Extensive Calcinosis. Ellis JT, Barr DP Am J Pathol. 1951 Jun; 27(3):383-405. [PubMed] [Ref list]
2. Contribution of multimodality imaging for positive and aetiological diagnosis of multiple brown tumours. Grégoire C, Soussan M, Dumuis ML, Naggara N, Martin A, Dhote R, Audard V, Neuman A, Weinmann P Ann Endocrinol (Paris). 2012 Feb; 73(1):43-50.
3. A.W. Su, C.F. Chen, C.K. Huang, P.C. Chen, W.M. Chen, T.H. Chen Primary hyperparathyroidism with brown tumor mimicking metastatic bone malignancy J Chin Med Assoc, 73 (2010), pp. 177-180, 10.1016/S1726-4901(10)70035-6 [PMID: 20231005]
4. Fine-needle aspiration of brown tumor of bone: cytologic features with radiologic and histologic correlation. Pavlovic S, Valyi-Nagy T, Profirovic J, David O Diagn Cytopathol. 2009 Feb; 37(2):136-9.

5. Moe S, Drüeke T, Cunningham J, Goodman W, Martin K, Olgaard K, Ott S, Sprague S, Lameire N, Eknoyan G, Kidney Disease: Improving Global Outcomes (KDIGO) Definition, evaluation, and classification of renal osteodystrophy: a position statement from kidney disease: improving global outcomes (KDIGO) Kidney Int. 2006;69(11):1945–1953. doi:10.1038/sj.ki.5000414. [Abstract] [CrossRef] [Google Scholar]
- 6.[Multiple costal lesions in a hemodialysis patient].Meriglier E, Roblot P, Fritz O, Le Mao G, Bachelet-Rousseau C, Leroy F Rev Med Interne. 2014 Jun; 35(6):405-6.[PubMed] [Ref list]
- 7.El Harraqui R, Karimi I, Chemlal A, Ismaili FA, Haddiya I. Mode de révélation particulier d'une tumeur brune chez une patiente hémodialysée chronique [Particular mode of revelation of a brown tumor in a chronic hemodialysis patient]. Pan Afr Med J. 2014;18:223. Published 2014 Jul 17. doi:10.11604/pamj.2014.18.223.4051
8. Hu, Jinbo MD; He, Shaohui MD; Yang, Jian MD; Ye, Chen MD; Yang, Xinghai MD*; Xiao, Jianru MD* Management of brown tumor of spine with primary hyperparathyroidism, Medicine: April 2019 - Volume 98 - Issue 14 - p e15007 doi: 10.1097/MD.00000000000015007
- 9.Asian J Neurosurg. 2014 Jan-Mar; 9(1): 40–44. Brown tumor as an unusual but preventable cause of spinal cord compression: Case report and review of the literatureHakan Tayfun, Orakdöğen Metin, Somay Hakan, Berkman Zafer, and Aker Fügen Vardar1
- 10.Alfawareh MD, Halawani MM, Attia WI, Almusrea KN. Brown tumor of the cervical spines: a case report with literature review. Asian Spine J. 2015;9(1):110-120. doi:10.4184/asj.2015.9.1.110
- 11.Vaishya R, Agarwal AK, Singh H, Vijay V. Multiple 'Brown Tumors' Masquerading as Metastatic Bone Disease. Cureus. 2015;7(12):e431. Published 2015 Dec 23. doi:10.7759/cureus.431
- 12.Vandenbussche E, Schmider L, Mutschler C, Man M, Jacquot C, Augereau B. Brown tumor of the spine and progressive paraplegia in a hemodialysis patient. Spine (Phila Pa 1976) 2004;29:E251–E255. [PubMed] [Google Scholar]