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Brown tumor in patients with secondary hyperparathyroidism due to chronic renal failure: a case series study

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(Original Research Article)

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Abstract

Brown tumor (BT) is an un-common benign bone lesion caused by hyperparathyroidism. Occurrence of BT in sitting of chronic renal failure is considered as sever form of renal-osteodystrophy. Prevalence of BT is (1.5-13%) among dialysis patient. The definite diagnosis of BT is by histopathology. However, BT can be diagnosed noninvasively by using lab and imaging-based criteria. The most frequent sites: ribs, clavicles, extremities, pelvis and facial bones. Although no malignant potential for BT, it can result in catastrophic complications if not treated. Unfortunately, no universal guideline management for BT. This necessitates recognition of clinical manifestation and imaging patterns of BT to be detected and managed properly. Aim of study: Describe the clinical features and the imaging patterns of BT among the studied population. A descriptive case series study was conducted in the central hospital of Al-Baida city /Libya from December 2021 to the end of March 2022, involved 3 hemodialysis male patients. The age ranges from 32-42 years. Parathyroid hormone level measured, Digital x-rays and Non-contrast CT-scan (chest, abdomen and pelvic) performed to all patients. All cases exhibited multifocal lesions located in spine, ribs, pelvic bone. Expansile lytic lesions found in 100% of cases. Some showed mixed solid and lytic lesions. None of studied cases received a proper pharmacological treatment, one case underwent decompressive surgery. This humble case series shed light on the pattern of BT aiming for early discovering and proper management. The findings of this study can be used as baseline for further research in future.

Keywords: Renal osteodystrophy, Brown tumor, hyper parathyroidism, lytic bone lesions, osteitis fibrosa cystica.

Introduction

Brown tumor (BT) is an un-common benign lesion of the bone caused by increase osteoclast activity and deposition of hemosidrin as consequence of uncontrolled hyperparathyroidism HPT. It can be primary or secondary to other disease (Nago et al, 2021). The secondary hyperparathyroidism usually due to end stage renal failure (ESRF) and appearance of BT in such patients is considered as severe form renal osteodystrophy (ben fatama, 2010). The prevalence of BT is (1.5-13%) among dialysis patients and the incidence increase with chronicity of renal failure (Resic, 2011). The definite diagnosis of brown tumor is by histopathology. However, BT can be diagnosed noninvasively by using the diagnostic criteria of Balon & Kalavar (1998) and Chew et al (1993) that is a lytic lesion with Hounsfield units equal to blood and fibrous tissue in end stage renal failure, patient on hemodialysis therapy with secondary hyper-parathyroidism (HPT). The most frequent sites of BT are in ribs, clavicles, upper and lower limbs, pelvis and facial bones. Clinical presentation can vary from asymptomatic swelling to diffuse bony pain and pathological fracture (Kamal et al). It could be invasive in a number of patients; however, it does not have a malignant potential (Pinto et al, 2010). The radiological features of BT can be a single or multiple and the lesions resemble the appearance of lucent bone tumors like aneurysmal bone cyst, gaint cell tumor, metastatic carcinoma and myeloma (Kamal et al, 2020). Although there is no universal guideline management for BT, according to literatures, the main treatment is control HPT effectively either by pharmacological or surgical means, and bone curettage or decompression when it necessarily (Kamal, 2020; Wiederkehr, 2020). Giving the above-mentioned knowledge, it emphasizes the significance of think about BT in patients on hemodialysis, with single or multiple lytic lesions and bone symptoms. Therefore, this case series study of three patients with secondary HPT due to renal failure in hemodialysis department in Albida Medical Center is carried out. Aiming for analyze the clinical and radiological pattern of brown tumors among the studied population. Hoping to be a nucleus for further comprehensive studies and to draw more attention for this disease as there is no screening, prevention programs or even national guideline to follow and manage this preventable disease in order to avoid the catastrophic out comes.

Methods

This descriptive case series designed according to definition of case series of Gordis epidemiology text book (Celentano & Szklo 2019) and Hennekens and their colleagues (abu-Zaidan et al. 2012). The study was conducted from 11 December 2021 to the end of February 2022. The ethical form approval to conduct the study was obtained from the radiology department\ Al-Baida Medical Centre\ Al-Baida-city. The number of studied cases is 3, all were males. Their ages range from 32-42 years. The average period for hemodialysis was 10 years (9-12years).

Data regarding demography, symptoms, reason for imaging, types of medications were collected directly from patients and medical records during the scan. Data filled manually in special paper forms, A blood samples were collected from all patients, and parathyroid hormone level analyzed at the main lab of Baida Medical Centre. The inclusion criteria followed Balon & Kaleva (1998) and Chew et al (1993), that means all patients with end stage renal failure on hemodialysis therapy, secondary hyper parathyroidism, and CT lytic lesions with Hounsfield units equal to blood and fibrous tissue, were included. The x-ray of site of interest and CT scans were performed using a FCT Speedia HD, Fujifilm, 64-MDCT scanner.

The following parameters were used; 80-120 kV, 5mm beam collimation, 1.58 pitch, 0 gantry tilt, and the FOV (347-500) depending on patient's size. The scans covered the extremities bony deformity, Chest, spine and pelvis. No intravenous contrast had been administered. Following acquisition, the images reported by two radiologists.

Results

The number of studied cases is 3, all were males. Their ages range from 32-42 years. The average period for hemodialysis was 10 years (9-12years).
The reasons for imaging were as following: for first patient, neurological deficient for which CT spine requested; the second patient, acquired bony deformities in hands, the third patient accidently discovered in Chest X ray as a part of covid-19 work up as seen in (table 1) and (figure: 1).

Table-1: The main clinical and radiological patterns among the studied population

Patient age	Duration of hemodialysis	Reason of imaging	Other symptoms	Site of lesions
32	11	Cough/accidently discovered lesion during Chest x ray examination	Bone pain	Ribs, pelvis, femur, thoracic vertebral of spine.
40	9	Quadri-paresis	Bone pain	Multiple lesion spine with Compression lesion in cervical spine,
42	10	Bony deformity hands	Bone pain	Both hands, right scaphoid, ribs, pelvis, spine, humeri

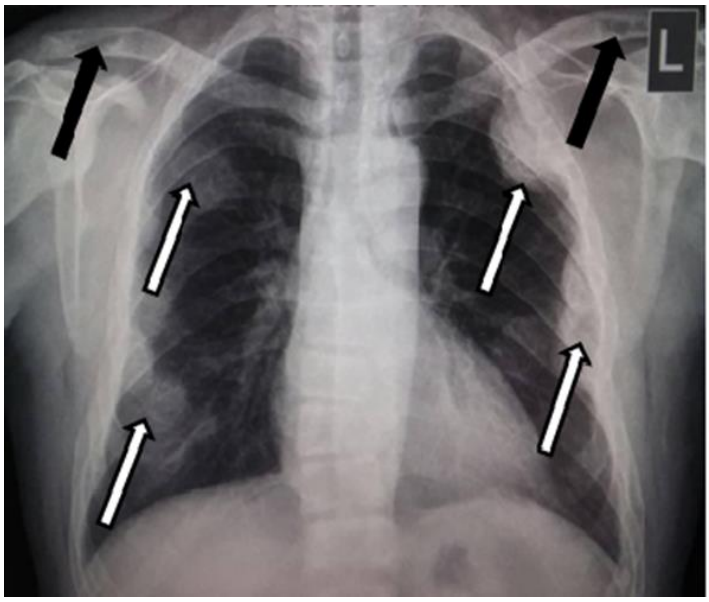

Figure (1): PA chest x ray; showing multiple expansile bony lesions along the ribs (white arrows), multiple lytic lesions clavicles (black arrows).

The most common CT patterns among the studied cases were lytic in 100% with some lesions exhibit mixed solid and lytic lesions (figure 2, 3b). The HU of lesions ranges from 25-80. Most of lesions are asymptomatic or exhibit mild bony pain. However, the spinal lesion in one patient was large enough to cause compression on cervical spinal cord leading to quadriparesis. All cases exhibited multifocal lesions located in spine, ribs, and scapulae. Pelvic bones (figure 4). One case showed brown tumors involving the carpal bones in addition to above mentioned sites (figure 3A). Although all patients were on calcium, none of them were on regular one alpha (VIT D analogue). None of them underwent parathyroidectomy. Only one patient underwent decompression surgery as a part of treatment.

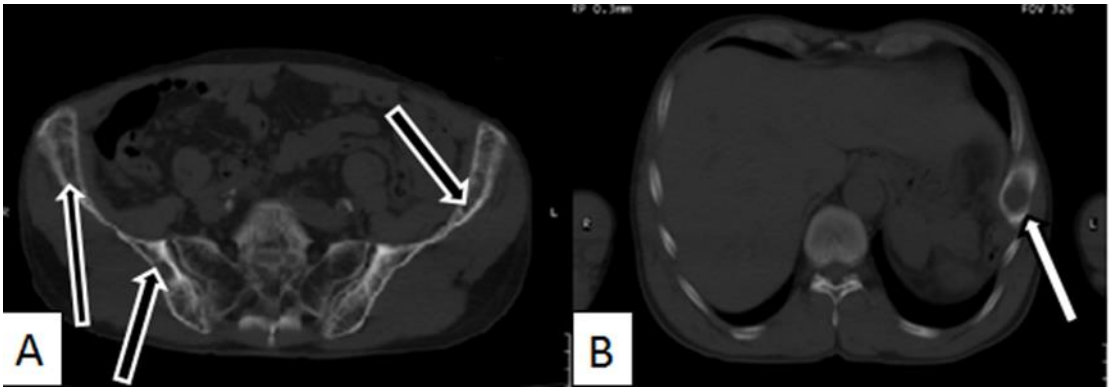

Figure (2): CT axial views; (A) multiple lytic lesions in pelvic bones (black arrows). In (B) ; expansile bony lesions along the left 9th rib (white arrow)

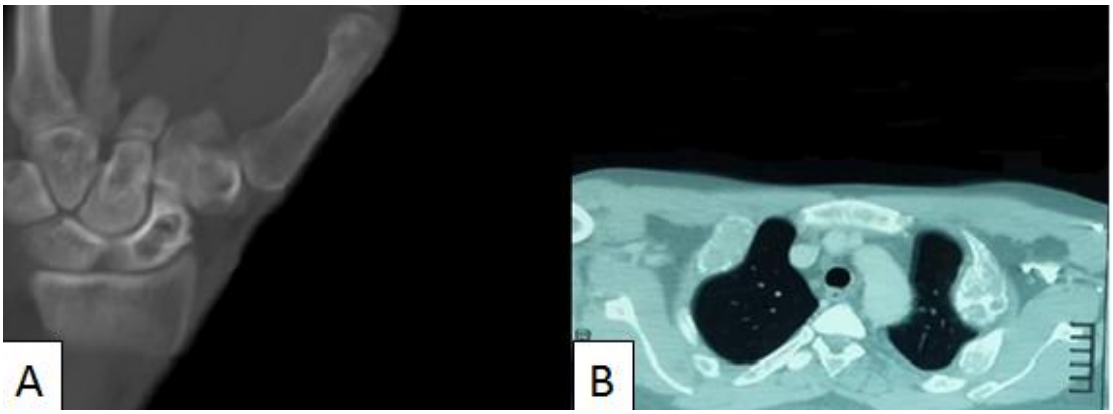

Figure (3): (A) CT for carpal bones showing lytic lesion in right scaphoid (black arrow). In (B); CT chest axial view showing expansile mixed solid\lytic bony lesions involving left 3ird ribs (white arrow.)

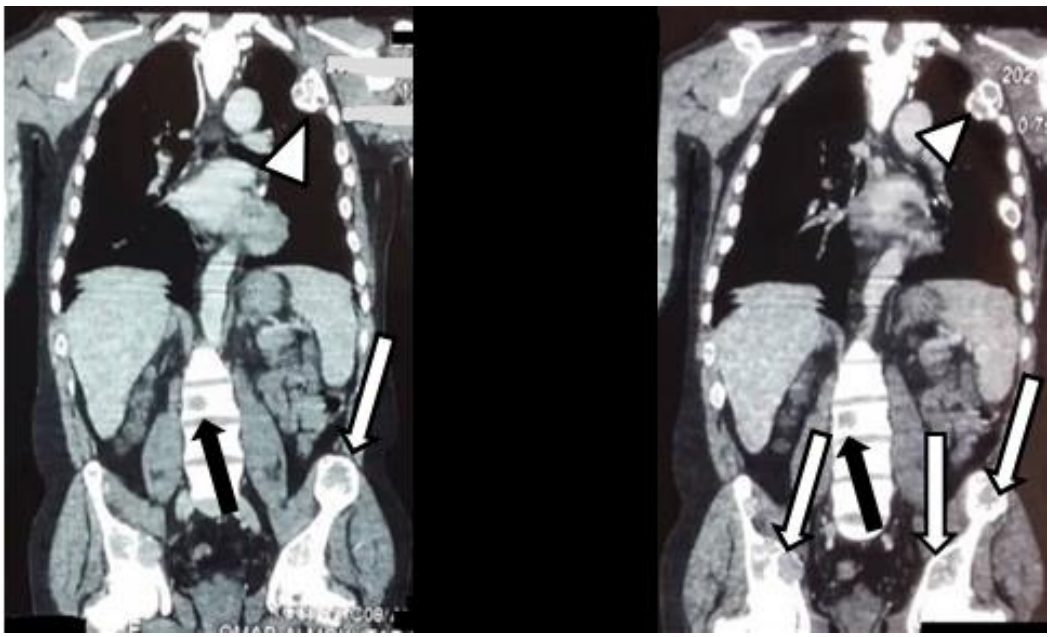

Figure (4): CT coronal reconstructions views; multiple lytic / mixed expansile bony lesions in pelvic (white arrow), lytic lesion in vertebrae (black arrow), and mixed expansile lesion in ribs (white arrow head).

Discussion

Brown tumors are a type of cystic fibrous osteitis, even though, BT are found in primary hyperparathyroidism, cases with secondary hyperparathyroidism as a result of chronic renal failure are documented with raising rate (pinto et al). Its rate is increasing due to increase survival rates in patients with chronic renal failure (pinto et al, 2010; Ben Fatma et al,2010). The incidence of BT in secondary HPT due to chronic renal disease vary from 1.5% to 13 % (Ben Fatma et al, 2010). BT in end stage renal disease occurs in patients with very elevated PTH level and over a long interval of time (WiederKehr, 2020). Increase PTH secretion changes the ratio of extra – and intra- cellular calcium, leading to increase resorption of bone thus decrease bone density and deposition of calcium in soft tissue, lesions are found in places of sever bone resorption (pinto et al, 2010). BT can be found in any bone, nevertheless, published articles are mainly in maxillofacial and dental journals because of high frequency for maxilla, mandible and hard palate, they are occurred also in ribs, femur and pelvis, and less frequently in spine (WiederKehr 2020). In our study all cases showed multifocal lesions placed in spine, ribs, scapulae and pelvic bone, one case, carpal bones are involved in addition to above mentioned places. The clinical presentation of BT is differed according to its location; they are mainly asymptomatic, although it can lead to swelling or dis-configuration, particularly in the face (WiederKehr 2020). Pain is due to the tumor itself or due to the sequel of the disease such as pain because of pathological fracture, tumor in vertebral column may cause paraesthesis, radicualar pain, more critical manifestations such as Para paresis, cauda equine syndrome and paraplegia represents a neurological emergency (WiederKehr 2020). In this study there was three patients, the lesions are mainly asymptomatic or show mild bony pain, one of the patients has spinal lesion that was large and causing compression on cervical spinal cord leading to quadriparesis. BT do not have particular laboratory or radiographic findings, most frequently cases have significant increase of PTH, phosphorus and calcium (WiederKehr, 2020). Radiographic imaging could be showed an osteolytic lesion with well—circumscribed margins with bone expansion and thinning of cortex. This may mimic bone metastasis, anurysmal bone cyst, osteosarcoma, giant cell tumor, multiple myeloma or chondroma (Kamal et al, 2020). The CT patterns amongst the studied patients were mainly lytic lesions in 100% with a few lesions display mixed lytic and solid lesions. The proper management of BT is often not easy and there is no common reference to guide the management. There is no refer to BT in published guidelines, also there is no talk on BT in standard textbooks of nephrology (WiederKehr, 2020). Management including partial or complete removal of parathyroid gland and when there is fracture or severe deformity orthopedic surgical procedures is considered (Kamal et al). Complete remission and regression of the tumor has been well reported after parathyroidectomy in both primary and secondary HPT (Pinto et al, 2010; and WiederKehr 2020). In addition, Vit D and phosphate binders are helpful preventive treatment of secondary HPT and BT (Ben Fatma et al, 2010). All of the studied cases in this study received calcium, no one of the studied patients were on regular one alpha (VIT D analogue), none of them underwent parathyroidectomy. Decompression surgery was done to one patient as a part of treatment. We should consider BT due to secondary hyperparathyroidism in patients with chronic renal disease if there is osteolytic lesions, we suppose that BT are under-reported, and its diagnosis is not simple thing to do, clinical history and examination, lab investigation and medical imaging should be correlated together to reach diagnosis.

In this study the number of cases is small, none of them underwent parathyroidectomy or received proper medical treatment, other studies with large number, receiving proper management and follow-up to monitor the effect of management is recommended.

Conclusion

Brown tumor is one of the complications of secondary Hyperparathyroidism due to chronic renal disease; however, it may become asymptomatic until cause severe destruction. Therefore, screening for secondary HPT in patients with chronic renal disease is recommended, with considering BT in differential diagnosis of any single or multiple lytic lesion(s). Multi displinary view is necessary in management cases with osteolytic lesions.

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