CASE REPORT

Undiagnosed Chronic Renal Failure Presenting With Brown Tumor of Bilateral Patellae With Pathological Fracture

Mohan Kumar EG[1,2], Yathish Kumar GM[2] and Mohammed Noorudheen[2]

[1]HoD, Department Of Orthopedic Surgery, KIMS Al Shifa Hospital, Perintalmanna. E-mail: orthomohan@rediffmail.com

[2]Consultant Orthopaedic Surgeon, KIMS Al Shifa Hospital, Perintalmanna, Kerala, India

Article Info

Keywords
brown tumour
patella
bilateral

Correspondence
yathishishere@gmail.com

Source of funding
Nil

Conflict of interest
The author(s) declare that they have no conflicting interests.

Available online at
http://www.kjoonline.org/

Quick response code

Abstract

It has been reported that the common sites of brown tumors are the jaw, pelvis, ribs, femurs and clavicles. We report our experience in a case of a middle aged man with undiagnosed chronic renal failure and secondary hyperparathyroidism who presented to us with Brown tumor of bilateral patellae with pathological fracture. An initial radiograph showed an osteolytic lesion and MR images showed a mixed solid and multiloculated cystic tumor in the left patella and pathological fracture in right patella. Being a sesamoid bone, brown tumor of patella is extremely rare indicative of severe secondary hyperparathyroidism. Fracture was treated surgically by curettage, biopsy and wiring, and renal failure was treated by nephrologist. One and half months after the fixation of fracture and improved renal function, along with calcium and Vit D supplements, complete healing of fracture and rapid bone formation was observed on radiographs.

Cite this paper as: Mohan Kumar EG *et al*. Undiagnosed Chronic Renal Failure Presenting With Brown Tumor of Bilateral Patellae With Pathological Fracture. *Kerala Journal of Orthopaedics* 2016;29(1–2):39–43.

Introduction

Brown tumors are focal reactive osteolytic lesions that are encountered in patients with primary or secondary hyperparathyroidism[1–3]. It can also occur in end stage renal failure complicated by secondary hyperparathyroidism[4]. The incidence of brown tumor in renal failure patients is reported to be 1.5 to 13%[4]. Common sites affected are the mandible, ribs, pelvis and clavicle[4]. Involvement of extremities are common while patella being a sesamoid bone, involvement is extremely rare[4]. There have been a few reports on brown tumor of the patella and still fewer cases of brown tumor of patella are reported in association with secondary hyperparathyroidism in renal failure patients[5–8]. We recently treated a patient, who developed a brown tumor of the patella caused by secondary hyperparathyroidism due to an undiagnosed renal failure. The patient's clinical course is discussed in this report. An informed consent was taken.

Case Presentation

A 34 year old gentleman presented with an eight month history of chronic dull aching type of pain over the knee cap of both knees, presented acutely with inability to walk since 3 days following a trivial tripping injury. He is a known hypertensive for 5 years, but not on regular treatment. Physical examination revealed severe tenderness on right patella and mild tenderness over the superior pole of left patella (Fig. 1). Patient was not able to do straight leg raise on either sides. On general examination, patient was moderately built and nourished, his blood pressure was elevated. Plain

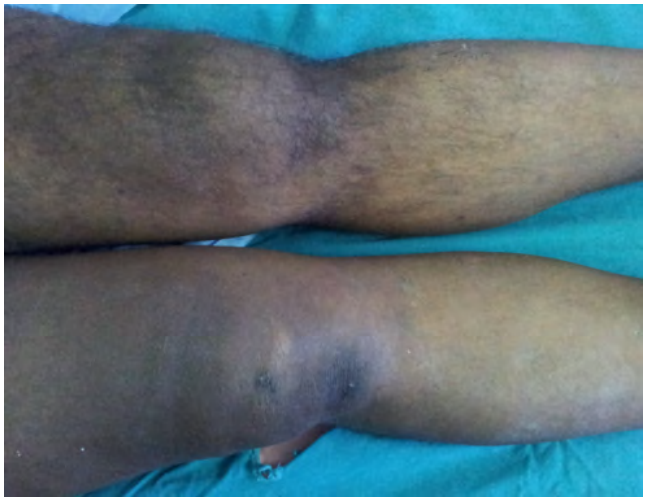

Figure 1. Clinical presentation.

radiograph of the right knee (Fig. 2) showed a lytic lesion in the patella with pathological fracture. Left knee (Fig. 3) also showed a lytic lesion in patella. MRI of the left knee was taken and axial T2-weighted image showed the mixed solid and multiloculated cystic nature of the tumor in the left patella (Fig. 4). Skeletal survey of pelvis, spine, chest, long bones and facial bones was done but no other lesions were found. Laboratory results showed corrected calcium level of 8.7 mmol/l and elevated phosphate level of 7.6 mmol/l. Serum alkaline phosphatase was also elevated (1480 IU/L), with elevated blood urea(209 mg/dl) and serum creatinine (10.84 mg/dl), and parathyroid hormone levels (924 pg/ml). An urgent nephrologist consultation was taken and he was advised haemodialysis. He had been on some unknown indigenous medications for weight reduction since 3 months. Probably this patient developed renal damage long back resulting in weight gain for which he went for indigenous treatment. The patient was provisionally suspected to have Brown tumors involving bilateral patellae with a pathological fracture on right patella, caused by hyperparathyroidism secondary to chronic interstitial nephritis based on both the imaging and laboratory findings. When patient became fit, he was taken up for surgery. Curettage of the lesion was done (Fig. 5), there was minimal articular damage (Fig. 6) and the fracture was fixed with tension band wiring (Fig. 7). The specimen consisted of brownish tissue with histopathological findings of fibrous tissue with presence of osteoclasts and multinucleated giant cells (Fig. 8), consistent with a diagnosis of Brown tumor.

Postoperatively, pain on both knees reduced. He refused arterio-venous fistula. He was advised salt restriction, low potassium and normal diet. He was put on Vit D3 60,000 IU once weekly once along

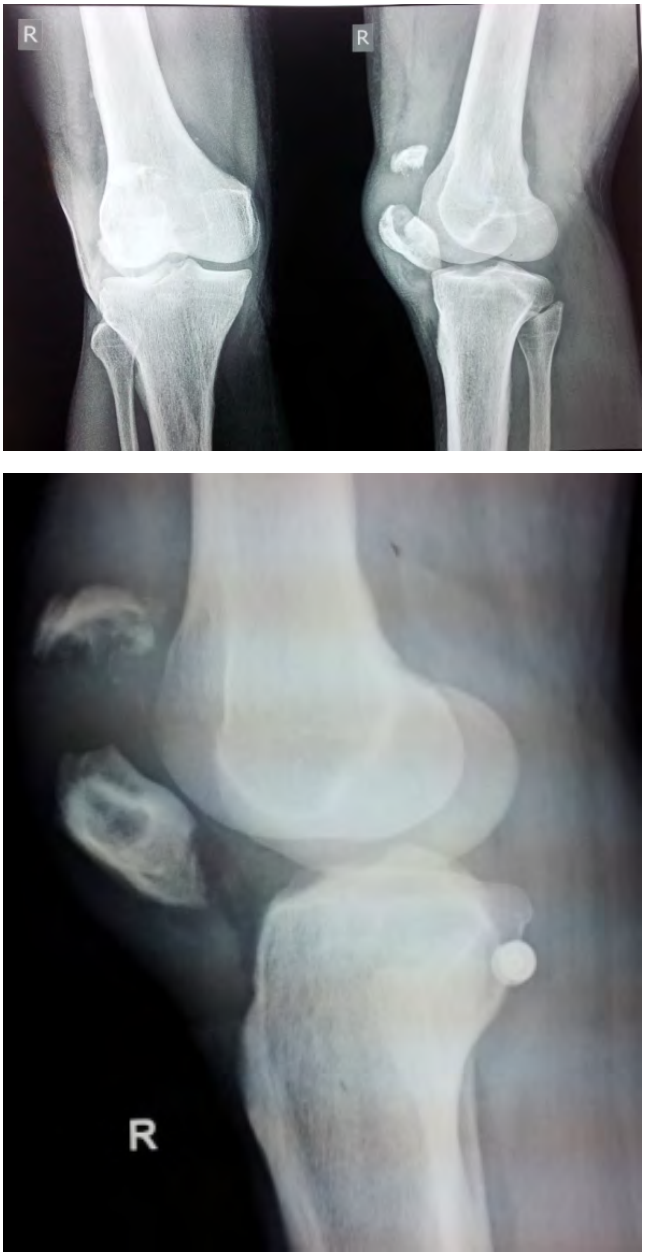

Figure 2. Plain X-ray right knee.

with calcium supplements and regular dialysis. Serum parathyroid hormone levels subsequently returned to normal. One and half months after fracture fixation and improved renal function, rapid bone formation was observed on radiographs (Fig. 9) and the fractures healed completely.

Discussion

Brown tumor is a non-neoplastic osteolytic lesion of the bone that is caused by primary or secondary hyperparathyroidism[1–3]. Brown tumors can occur as solitary or multiple lesions. They may appear in any bone, but commonly affect the pelvis, ribs, femurs,

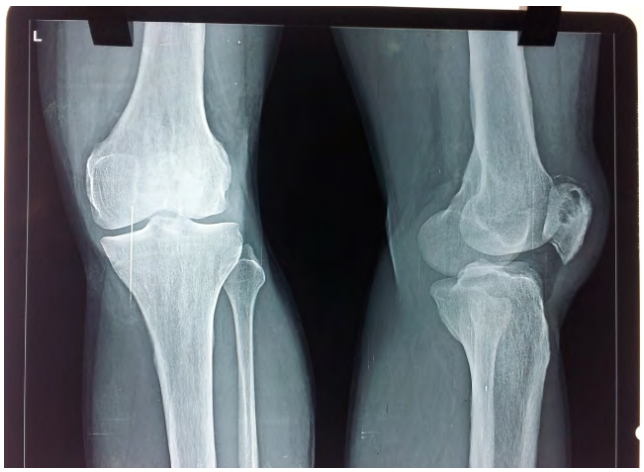

Figure 3. Plain X-ray left knee showing lytic lesion.

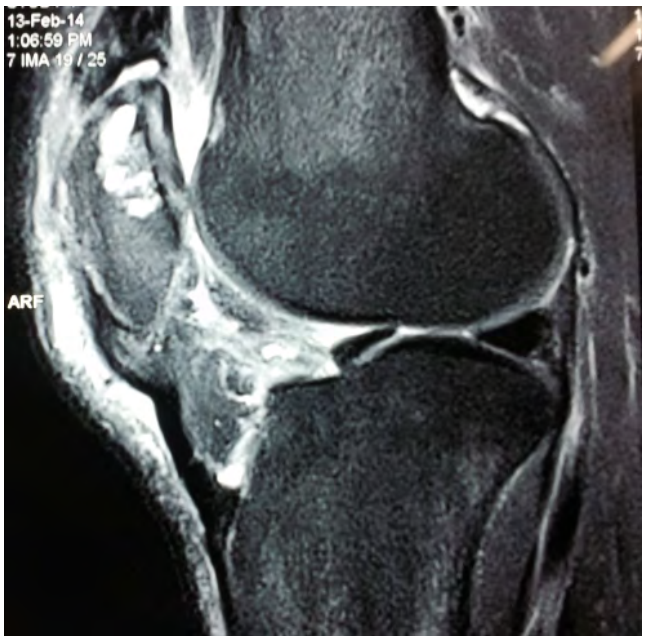

Figure 4. MRI image of left knee.

humeri and other long bones, as well as the facial bones and jaws, clavicles and spine[1–3,8–10]. Generally, brown tumors are lytic in nature with well-defined borders according to radiographs or CT scans, and similar appearances were seen in our case (Figs. 2–4). The Brown tumor lesions are heterogeneously hypo and isointense to skeletal muscle on T1-weighted MR images, although the signal characteristics depend on the level of sclerosis[6].

Singh et al. reported 59 cases of bone tumors of the patella. They described 46% of cases as non-neoplastic, 39% as benign, while 15% as malignant[4]. Giant cell tumor is the most common disease which affects the patella, and 19% of patella tumors have previously

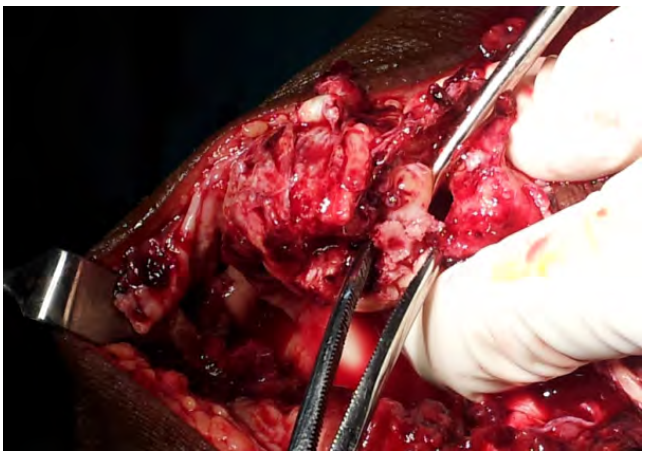

Figure 5. Brownish tissue on curettage.

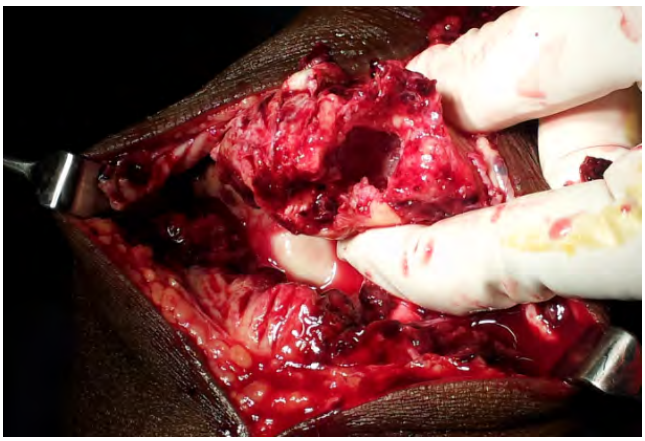

Figure 6. Showing minimal articular damage.

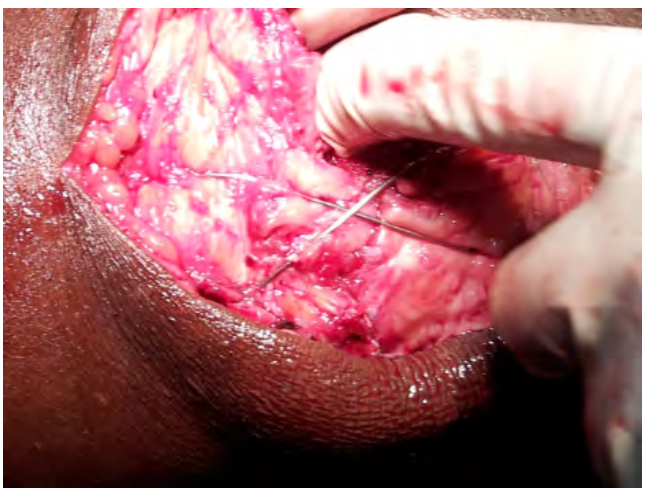

Figure 7. Tension band wiring.

been reported as giant cell tumors[4]. The second and third common diseases affecting the patella are chondroblastoma (15%) and aneurysmal bone

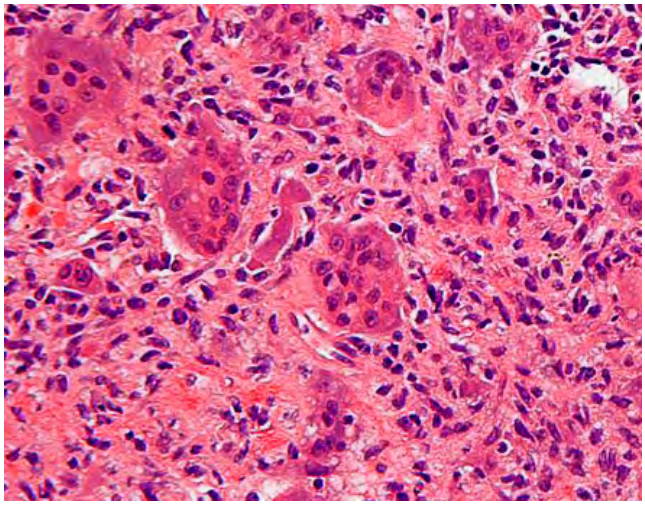

Figure 8. Histopathology showing giant cells and fibrous tissue.

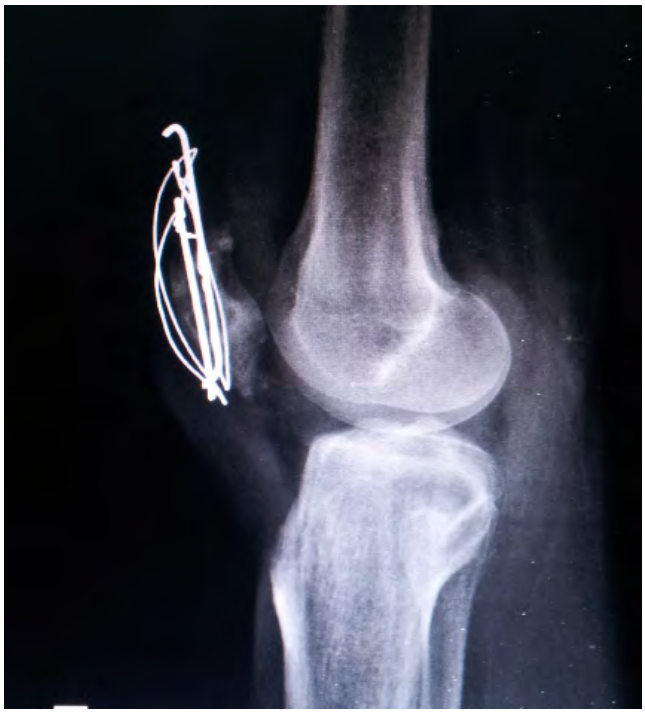

Figure 9. X-ray at 6 weeks fracture united consolidating.

cyst (10%), respectively[4]. A biopsy has the potential to lead to a misdiagnosis, since brown tumor is similar to aneurysmal bone cyst or giant cell tumor in terms of the pathological tissue[9]. Therefore, the imaging and laboratory findings are important for accurate differentiation of the tumor. A recent paper showed that fluoro-deoxyglucose positron emission tomography/CT is effective for detecting osteolytic lesions, such as Brown tumors[10]. The uptake of fluoro-deoxyglucose by osseous lesions can assist in diagnosing Brown tumors with hyperparathyroidism, and abnormal uptake of fluoro-deoxyglucose can aid in differentiation of primary neoplasms[10].

In chronic renal failure, low serum calcium and phosphate retension due to the deficiency of 1(alpha), 25-dihydroxycholecalciferol stimulates an increase in parathyroid hormone levels[4]. Elevated levels of PTH were observed in the current case, which resulted in severe osteoporosis, subperiosteal bone resorption and reactive proliferation of the fibrovascular tissue in the bone marrow caused by an increase in bone turnover[4]. Microfractures and hemorrhages commonly occur in these lesion due to their weak bony architecture. The resultant hemosiderin deposition gives the brown colour and hence the name of these tumors[4]. The diagnosis of Brown tumor requires both histological findings of fibrous tissue with multinucleated giant cells and the presence of hyperparathyroidism[4].

The definitive treatment of Brown tumors is to normalize the serum calcium and phosphate levels with calcium supplements and calcitriol, restrict dietary phosphate and phosphate binder intake along with treatment of renal failure. Success in normalizing serum calcium and phosphate then leads to remineralisation and resolution of lesion[11,12].

One and half months after the fixation of fracture and improved renal function along with calcium and Vit D3 supplements, complete healing of fracture and rapid bone formation was observed on radiographs.

As increased osteoclastic bone turnover results in fractures followed by influx of multinucleated macrophages, destruction of cortex and soft-tissue extension can be expected as imaging findings[3]. It has been reported that the risk of pathological fracture in Brown tumors correlates with the time of diagnosis[13]. Therefore, it is important to diagnose or differentiate Brown tumors from other tumors at an early stage, and as soon as possible.

Conclusion

The incidence of Brown tumor is expected to increase in line with the increased number of dialysis-dependent patients and the increase in life span of chronic renal failure patients due to improved medical treatment and the lack of available organs for transplantation. The presence of Brown tumor in sites such as the patella is rare and generally indicates a severe stage of secondary hyperparathyroidism. The active work up of a lytic bone lesion should also include investigation to rule out hyperparathyroidism. Diagnosis of hyperparathyroidism and Brown tumor in a chronic renal failure patient is easy, BUT it requires a high index of suspicion to diagnose patients presenting with bony swelling and/or skeletal pain, with lytic lesion in CKD with secondary hyperparathyroidism.

Consent

The patient has given his informed consent for the case report to be published.

Authors' Contributions

YK analyzed and interpreted the patient data, MK diagnosed the condition, performed the surgery and was a major contributor in writing the manuscript. MN assisted MK during surgery.

Acknowledgements

Sincere acknowledgement to Dr Thomas Varghis, Dr Ajay Kumar, all PG students, Umaiva, Manikantan for making substantial contributions to this study.

References

1. Takeshita T, Takeshita K, Abe S, Takami H, Imamura T and Furui S. Brown tumor with fluid-fluid levels in a patient with primary hyperparathyroidism: radiological findings. Radiat Med. 2006;24:631–634.
2. Knowles NG, Smith DL and Outwater EK. MRI diagnosis of brown tumor based on magnetic susceptibility. J Magn Reson Imaging. 2008;28:759–761.
3. Hong WS, Sung MS, Chun KA, Kim JY, Park SW, Lee KH, et al. Emphasis on the MR imaging findings of brown tumor: a report of five cases. Skeletal Radiol. 2011;40:205–213.
4. TH Low, LB Saw, MK Kwan,CYW Chan and AS Deepak. Extensive brown tumors of spine, distal femur and patella presenting with acute cord compression. A Case Report. Malaysian Orthopaedic Journal. 2011;5, doi: 10.5704/MOJ.1103.014.
5. Bhagat S, Sharma H, Bansal M and Reid R. Presentation and outcome of primary tumors of the patella. J Knee Surg. 2008;21:212–216.
6. Vernace N and Bancroft LW. Multiple brown tumors and pathologic patellar fracture in a patient with secondary hyperparathyroidism. Orthopedics. 2014;37:564–567.
7. Duran C, Ersoy C, Bolca N, Kiyici S,Yalcinkaya U, Erturk E et al. Brown tumours of the maxillary sinus and patella in a patient with primary hyperparathyroidism. Endocrinologist. 200;15(6):351-354.
8. Jouan A, Zabraniecki L, Vincent V, Poix E and Fournié B. An unusual presentation of primary hyperparathyroidism: severe hypercalcemia and multiple brown tumors. Joint Bone Spine. 2008;75:209–211.
9. Campanacci M. "Brown tumors" in primary hyperparathyroidism. In: Campanacci M, editor. Bone and soft tissue tumors. 2nd ed. New York: Springer-Verlag Wien; 1999. pp. 877–899.
10. Lacativa PG, Franco FM, Pimentel JR, Patrício Filho PJ, Goncçalves MD andFarias ML. Prevalence of radiological findings among cases of severe secondary hyperparathyroidism. Sao Paulo Med J. 2009;127:71–77.
11. Barlow IW and Archer IA. Brown tumour of the cervical spine. 1993:18:936-937.
12. Vandenbussche E, Schmider L, Mutschler C, Man M, Jacquot C and Augereau B. Brown tumor of the spine and progressive paraplegia in a hemodialysis patient. Spine 2004;29(12):251-255.
13. Vestergaard P, Mollerup CL, Frøkjaer VG, Christiansen P, Blichert-Toft M and Mosekilde L. Cohort study of risk of fracture before and after surgery for primary hyperparathyroidism. BMJ. 2000;321:598–602.