Case Report

Journal of Orthopedic Education
Volume 8 Number 3, September-December 2022
DOI: http://dx.doi.org/10.21088/joe.2454.7956.8322.6

Bilateral Intertrochanteric Fractures Secondary to Tumours Induced Osteomalacia: Rare Case Report

R B Uppin[1], S K Saidapur[2], Raunak Pareek[3]

How to cite this article:
R B Uppin, S K Saidapur, Raunak Pareek/Bilateral Intertrochanteric Fractures Secondary to Tumours Induced Osteomalacia: Rare Case Report/J Orth. Edu. 2022;8(3):105–107.

Abstract

Tumor induced osteomalacia also known as oncogenic osteomalacia is a rare paraneoplastic syndrome of abnormal phosphate and vitamin metabolism caused by typically small endocrine tumors that secrete the phosphaturic hormone, fibroblast, GF-23. We present a case report of bilateral intertrochanteric fracture treated with DHS which was secondary to tumor induced osteomalacia.

Methods: 36 yrs old male, complained of difficulty in walking, weakness in both lower limbs and developed sudden onset of severe LBA radiating to buttocks and legs. Patient presented with h/o pain after he fell down from stairs and was managed conservatively elsewhere and presented to KLEH for the same complaints after 1 month. Routine investigation was done and the CT scan s/o lesion in the femur head. DOTA SCAN s/o DOTA and sclerotic lesion in the right femur head. He was started on Joule's scan. B/L DHS application and biopsy was done followed by RFA. Post-Op phosphorus levels were 2.1 mg%. Repeat DOTA SCAN showed no uptake.

Result: Patient had normal RFT, biochemical evaluation showed persistent hypophosphatemia, raised ALP levels and was started on Joule's solution. Post-surgery along with RFA, repeat DOTA SCAN showed no uptake and after removal of DHS, patient had no difficulty in walking and patient's weakness and pain improved.

Conclusion: Oncogenic osteomalacia is uncommon yet curable cause of osteomalacia. The key to cure the individual cases remains reserved to the physician and his intelligence. Definitive treatment is the surgical removal of tumor with dramatic and satisfying results.

Keywords: Tumor induced osteomalacia; Paraneoplastic syndromes; Osteoporosis; Oncogenic osteomalacia.

Author Affiliation: [1]Professor, [2]Associate Professor, [3]Post Graduate, Department of Orthopedics, KLE'S Academy of Higher Education and Reaserch, J.N. Medical College, KLE'S Dr Prabhakar Kore Hospital and Medical Research Center, Belagavi 590010, Karnataka, India.

Corresponding Author: R B Uppin, Professor, Department of Orthopaedics, KLE'S Academy of Higher Education and Reaserch, J.N. Medical College, KLE'S Dr Prabhakar Kore Hospital and Medical Research Center, Belagavi 590010, Karnataka, India.

E-mail: uppinrajendra@rediffmail.com

Received on: 06.07.2022 **Accepted on:** 10.08.2022

INTRODUCTION

Tumor induced osteomalacia also known as oncogenic osteomalacia is a rare paraneoplastic syndrome of abnormal phosphate and vitamin metabolism caused by typically small endocrine tumors that secrete the phosphaturic hormone, fibroblast, GF-23. Biochemical hallmarks of the disorder are hypophosphatemia due to renal phosphate wasting, inappropriately normal or low 1,25-dihydroxy vitamin D, and elevated or inappropriately normal plasma FGF23. TIO is counted among the ranks of endocrine neoplasms

© *Red Flower Publication Pvt. Ltd.*

that have a striking presentation and, when resected, a dramatic and satisfying resolution, Tumor induced osteomalacia is a rare disease as so far less than 400 cases have been reported in the world. We present a case report of bilateral intertrochanteric fracture treated with DHS which was secondary to tumor induced osteomalacia. TIO is characterized clinically by bone pain and fracture, renal phosphate wasting, hypophosphatemia, low or normal serum 1,25(OH)2D concentrations, and elevated serum alkaline phosphatase levels. Fibroblast growth-23 (FGF-23), a phosphatonin, has been identified as a major pathophysiological factor responsible for phosphate wasting in TIO.

CASE REPORT

A 36 yrs-old male complained of difficulty in walking, weakness in both lower limbs and developed sudden onset of severe low back pain radiating to the buttocks and legs, which started approximately 1 year before his presentation. The patient used to walk in small strides and had difficulty in climbing and getting down from stairs. Patient presented in a local hospital with history of fall while walking and had pain in the left lower limb for which he was conservatively managed with analgesics and calcium supplements for 1 month. The patient presented to our hospital and was diagnosed to have Bilateral intertrochanteric stress fracture. MRI lumbosacral spine was suggestive of generalized posterior bulge of L3-L4 and L4-L5 disc. He denied fever, joint pain, oral ulcers, alopecia, sicca symptoms, Raynaud's phenomenon, skin rash, red eye, chest pain, shortness of breath, abdominal pain, diarrhea or hematochezia. He was managed with bilateral DHS plating. DEXA Scan showed osteoporosis at that time and so he was prescribed with calcium and vitamin D3 supplements. Bone scan revealed high uptake in all her joints, i.e., bilateral shoulders, knees, ankles and feet, as increased uptake in the skull, sacroiliac joints, and anterior ribs. The pattern was characteristic of a metabolic bone disease. Based on the imaging studies, the differential diagnosis was limited to hyperthyroidism, renal osteodystrophy, or osteomalacia. Given that the patient had normal renal function, a workup for osteomalacia and hyperparathyroidism was sent. Biochemical evaluation on follow-up showed persistent hypophosphatemia, raised ALP levels with slightly elevated PTH levels for which he was started on Joule's solution. MIBI scan showed no evidence of functioning parathyroid lesion. DOTANOC scan showed DDOTA and sclerotic lytic lesion was noted in head of right femur. CT scan prior to DHS insertion was reviewed and that lesion was found to be present. MRI confirmed the lesion in right femur head. Patient underwent biopsy followed by RFA of the lesion and post op phosphorus levels were 2.1 mg% without any oral phosphorus supplements. Repeat DOTA scan post procedure showed no evidence of uptake in the left femoral neck. After removal of bilateral DHS patient has no difficulty in walking and patient's weakness and pain has improved.

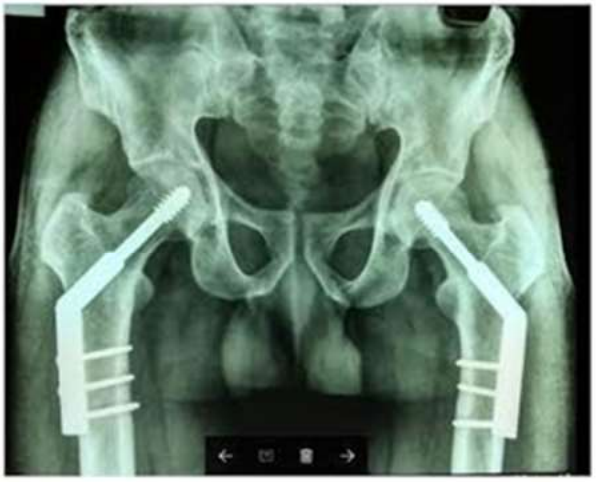

Fig. 1: B/L DHS for both hips

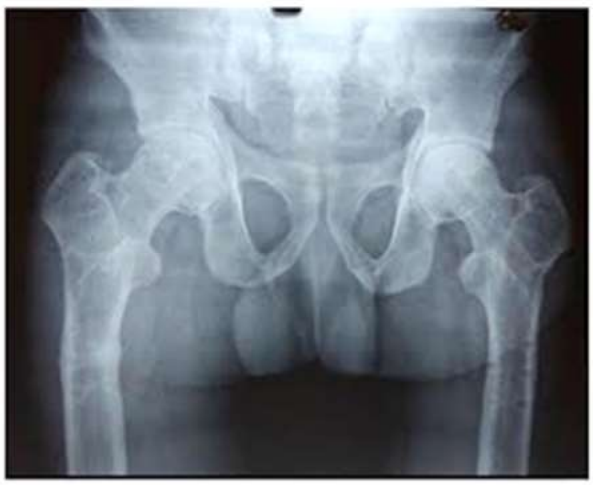

Fig. 2: Post DHS removal

DISCUSSION

We report a patient who presented with disabling low back pain that eventually. With the help of our radiological and pathological studies, was diagnosed with tumor induced osteomalacia and, with surgical intervention, was cured. Tumor-induced osteomalacia is characterized by the

triad of phosphaturia, hypophosphatemia, and low serum levels of 1,25 dihydroxy vitamin D. It clinically mimics X-linked or autosomal dominant hereditary hypophosphatemic rickets. The tumors that cause oncogenic osteomalacia are mostly of mesenchymal origin and usually benign. Peculiarly, the offending neoplasm is often inconsequential in the patient's clinical presentation, and both diagnosis and treatment are usually delayed because of the difficulty in recognizing the neoplasm and its relation to the patient's complaints. In our case, the pathology of the tumor was consistent with phosphaturic mesenchymal tumor, the most common tumor histology associated with oncogenic osteomalacia. Diagnosis of the disease id often challenging, and careful evaluation of both laboratory and radiologic testing are essential as severe disability an even death can be avoided with the surgical removal of the causative tumor. Bone and indium-111octreotide imaging have become key diagnostic radiologic tools when tumors are small and not detectable on routine physical examination or radiography. Although not useful in tumor localization, technetium-99 methylene diphosphonate scintigraphy has also been useful in revealing areas of active osteomalacia or pseudo-fractures. Recent studies have isolated fibroblast growth factor-23 as a possible phosphaturic substance may be produced by tumors that induced osteomalacia. FGF-23 genes are expressed at higher levels in tumors causing tumor-inducing osteomalacia.

In summary, oncogenic osteomalacia is uncommon yet curable cause of osteomalacia. The key to cure the individual cases remains reserved to the physician and his intelligence, who must pick up on the subtle clues leading to the diagnosis and often occult tumors. The definitive treatment is the surgical removal of tumor with dramatic and satisfying results. Recent studies and research into the pathogenesis and the role of FGF-23 is very exciting and will hopefully be a starting point in unravelling the mystery of tumor induced osteomalacia.

CONCLUSION

Tumor induced osteomalacia is yet curable unless detected. While the tumors can be difficult to locate, a stepwise approach that involves functional imaging, followed by anatomical imaging, and, if necessary, selective venous sampling or aspiration for confirmation is usually successful.

REFERENCES

1. Yu WJ, He JW, Fu WZ, Wang C, Zhang ZL.J Bone Miner Metab. 2017 May;35(3):298-307. doi: 10.1007/s00774-016-0756-9. Epub 2016 Apr 16.PMID: 27085966.
2. Hu FK, Yuan F, Jiang CY, Lv DW, Mao BB, Zhang Q, Yuan ZQ, Wang Y.Chin J Cancer. 2011 Nov;30(11):794-804. doi: 10.5732/cjc.011.10013. PMID: 22035861.