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Successful treatment of oncogenic osteomalacia with bone cement: An effective treatment option

Short title: Successful treatment of oncogenic osteomalacia with bone cement

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A 28-year-old male patient was admitted to orthopaedic department with a 4-year history of severe systemic pain, especially in the chest, bilateral hips and back. There were no other constitutional symptoms, and no history of trauma or surgery during the past years. On physical examination, the patient showed a slightly decreased muscle strength in bilateral limbs. The result of general physical examination was also remarkable in terms of sternal tenderness, tapping pain, and chest crushing pain. Routine laboratory examination and endocrine tests were comprehensively ordered and demonstrated hypophosphatemia, elevated \( \beta \)-C-terminal telopeptide of type I collagen, and elevated serum alkaline phosphatase. X-ray of knee joint showed significant quasi-circular high density shadow in the right proximal tibia (FIGURE 1A). Computed tomography and magnetic resonance imaging revealed the tumor with irregular hyperintense signals on T1-weighted image and T2-weighted image involving right tibial plateau (FIGURE 1B - 1D). \(^{68}\)Ga-DOTA-TATE PET/CT and \(^{99}\)m Tc-octreotide scan identified a high-intake nodule in the right tibial plateau, with high suspicion of oncogenic osteomalacia (FIGURE 1E). After detailed analysis, we performed osteoplasty with bone cement and postoperative posteroanterior radiograph of the hip showed cement augmentation was satisfactory (FIGURE 1F). The pathology results confirmed as oncogenic osteomalacia with evidence of phosphaturic mesenchymal tumor (FIGURE 1G). His serum phosphate returned to normal following surgery, and the symptoms improved significantly compared to the preoperative status. Moreover, visual analogue scale score of his bone pain improved to 1-2 points compared to the preoperative status, 6 points. Postoperatively, the patient declined adjuvant treatment and was monitored as an outpatient. This is an extremely unique surgical management of tumor-induced osteomalacia with cement augmentation.

Oncogenic osteomalacia (OO) was first reported by McCance in 1947 and only 500 cases have been reported worldwide [1,2]. The pathogenesis of OO is that the disease-causing
tumor can produce phosphatoninins including fibroblast growth factor-23, which affects phosphorus reabsorption and vitamin D metabolism by regulating proximal renal tubular sodium, phosphorus co-transporter and influencing the function of l-α hydroxylase, thus lowering blood phosphorus, increasing urine phosphorus and impairing bone mineralization [2,3]. Surgery is proved to be the main treatment of first choice for patients with OO, which requires complete removal of tumors and sufficient resection scope to cure radically [4]. Blood phosphorus would gradually increase after resection of the pathogenic tumors, generally returning to normal level in 2-7 days after surgery, and the clinical symptoms of the patients can be significantly relieved [5]. However, surgery may also seriously affect the local skeletal stability of patients, and bring about limitations of local motor function at the same time. Bone cement has the potential to inhibit the growth of tumors and is expected to become an effective treatment option for patients with oncogenic osteomalacia who can not undergo appropriate surgical procedure or decline completely open surgery. Cementoplasty has its own advantages over other surgical treatment options in terms of surgical injury and post-operative rehabilitation [4,5]. The present case highlights the importance of accurate diagnosis and proper treatment for patients with oncogenic osteomalacia.

ARTICLE INFORMATION

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CONFLICT OF INTEREST None declared.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

REFERENCES


FIGURE 1: A - Preoperative X-ray showing significant quasi-circular high density shadow in the right proximal tibia. B-D - Computed tomography and magnetic resonance imaging revealing the soft tissue tumor involving right tibial plateau. E - $^{68}$Ga-DOTA-TATE Positron emission tomography-computed tomography identified a high-intake nodule in the right tibial plateau, with high suspicion of oncogenic osteomalacia. F - Postoperative X-ray showing the position of bone cement was satisfactory. G - Histopathology confirmed tumor-induced osteomalacia with evidence of phosphaturic mesenchymal tumor.