Prudence for Deformity Correction in Surgically Treated Tumour-Induced Osteomalacia Patients

Sir,

Tumour-induced osteomalacia (TIO) is a rare disease caused by phosphaturic mesenchymal tumours (PMT) originating from soft tissues. The most common location is the lower limbs, followed by the head region.[1] Although the disease is debilitating, the improvement after surgery is dramatic and motivating for the treating team. However, in a few cases of TIO, poor outcomes and mortality have been reported due to tumours that were either nonlocalized, unresectable, metastatic or unsuitable for alternative therapeutic modalities. Herein, we report a patient with a fatal outcome despite successfully removing the phosphaturic tumour.

A 54-year-old man presented to a local hospital with a history of low backache for 3 years. He was misdiagnosed with polymyositis and received glucocorticoids, which did not improve his symptoms. He developed bilateral lower limb pain and proximal muscle weakness for 2 months. Re-evaluation in our centre showed bilateral femur fractures (right femoral neck and left intertrochanteric region) along with a very low phosphate level (2.02 mg/dl; N range 2.5–5) and elevated alkaline phosphatase (466 IU/L; normal range 30–120). Other biochemical investigations were unremarkable [(corrected calcium 8.8 mg/dl; normal range 8.8–10.6), (intact parathyroid hormone 51.7 pg/ml; normal range) and (25-hydroxyvitamin D 41.28 ng/ml; normal range 20–100)]. C-terminal fibroblast growth factor-23 (FGF-23) levels were high [1334 RU/ml; normal range 0–150]. The 68-GaDOTANOC scan revealed increased somatostatin receptor (SSTR)-expressing lesions in the right thyroid lobe and right hemi-mandible [Figures 1 and 2]. He was started on phosphate and active vitamin D supplementation. He underwent a right hemithyroidectomy followed by a right segmental hemi-mandibulectomy. His serum phosphate improved after 3 days post-surgery with normalization of C-terminal FGF-23 levels. Phosphate and calcitriol were stopped. Histopathology showed PMT in the resected mandible. He was discharged from the endocrinology ward with advice to review after 3 to 6 months for subsequent orthopaedic consideration of bony deformities. However, in his anxiety to get better faster, he independently consulted an orthopaedician in the immediate postoperative phase and underwent a right-sided hemiarthroplasty and a left-sided open reduction with proximal femoral nailing. Intraoperatively, there was massive blood loss, and he was admitted to the critical care unit for stabilization. The orthopaedician reported that intraoperatively, his bones were brittle like chalk. There was a prolonged course of ventilation due to difficulty in weaning. The patient had ventilator-acquired pneumonia and pulmonary thromboembolism pre-terminally during the intensive care unit (ICU) stay. He succumbed to the complications after 3 weeks of ICU stay.

Bone remineralization starts immediately after normophosphataemia is established by the curative surgery. Although there is a rapid improvement in bone mineral density, the clinical improvement may lag and require extensive rehabilitation.[2] Death is infrequent in patients of TIO with a range of 1–10% in different case series.[3] It is more often a consequence of metastases in patients with malignant tumours. However, our patient had a fatal outcome due to the unfortunate decision to undergo early corrective orthopaedic intervention. The majority of the fractures heal without any surgical intervention after normal phosphate homeostasis.[4] Unless as a part of resecting the PMT,[5] orthopaedic interventions in patients with TIO do not have a favourable risk-benefit

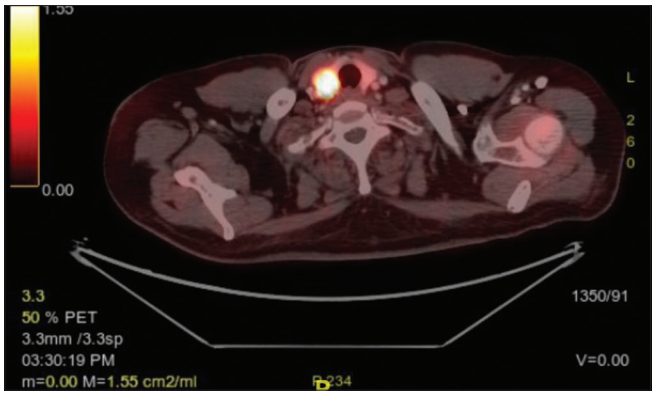

Figure 1: 68-GaDOTANOC scan showing uptake in the right thyroid lobe

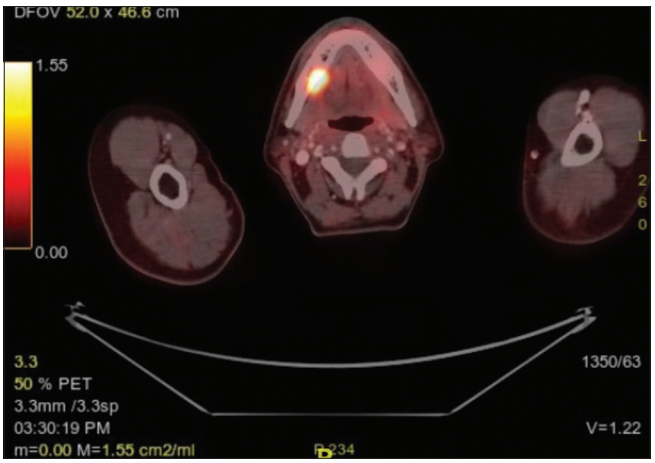

Figure 2: 68-GaDOTANOC scan showing uptake in the right hemi-mandible

ratio. We propose a time interval of 1–2 remodelling cycles, that is, 3–6 months at least before planning any orthopaedic intervention.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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Submitted: 25-May-2023 **Revised:** 17-Feb-2024
Accepted: 18-Mar-2024 **Published:** 26-Jun-2024

Access this article online

Quick Response Code:

Website: https://journals.lww.com/indjem/

DOI: 10.4103/ijem.ijem_216_23

How to cite this article: Suryadevara V, Prasad A, Penumadu P, Kamalanathan S. Prudence for deformity correction in surgically treated tumour-induced osteomalacia patients. Indian J Endocr Metab 2024;28:329-30.