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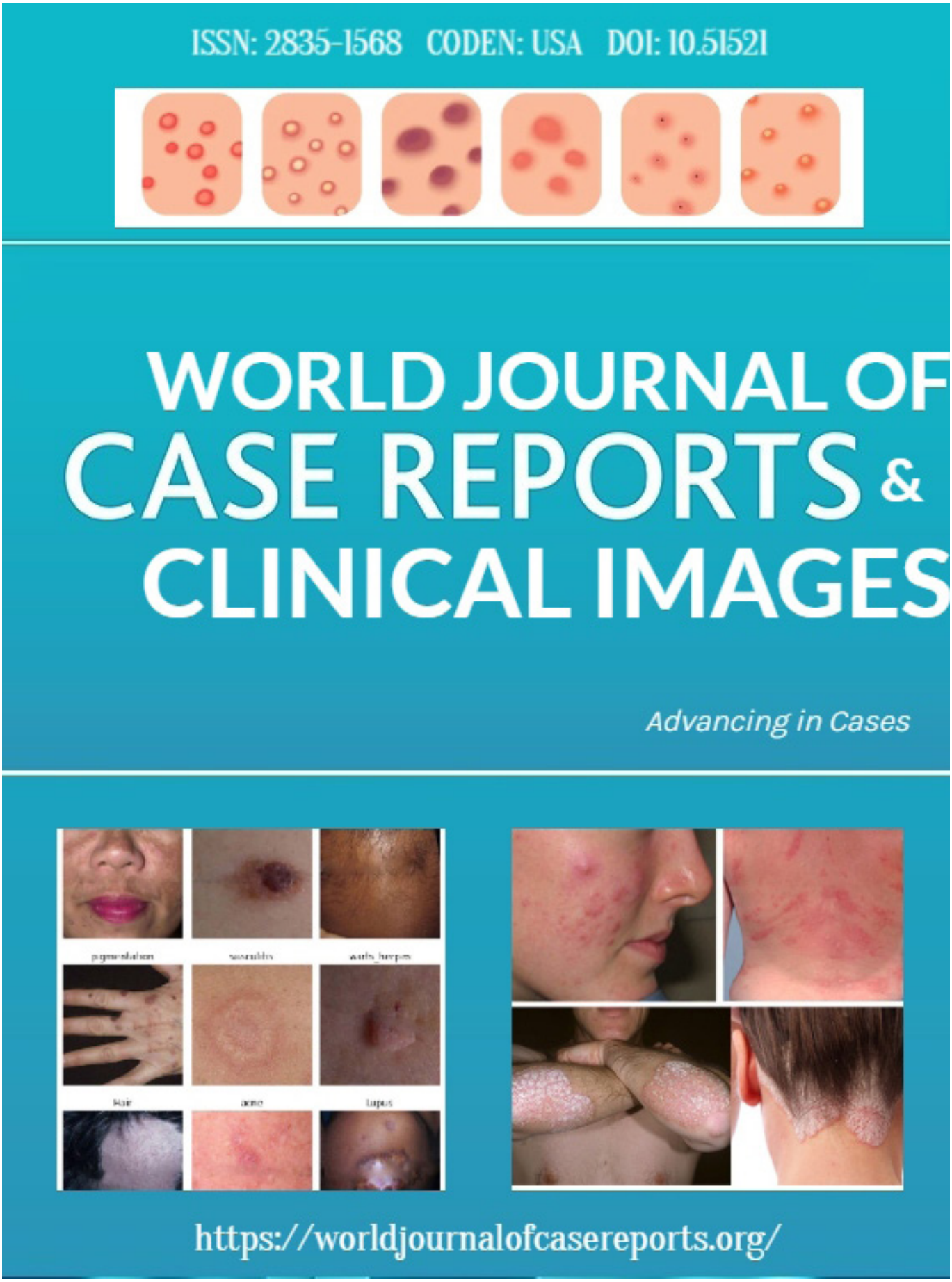

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CASE STUDY

# A Case Report of Tumor Induced Osteomalacia Due to Mesenchymal Tumor in the Temporal Bone

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## ABSTRACT

Tumor Induced Osteomalacia (TIO) is a paraneoplastic syndrome characterized by renal phosphate wasting due to excess production of fibroblast growth factor (FGF23). The average lag period between the initial clinical manifestations and the time when patient gets his correct diagnosis is 2.5-3 years and is due to non-specificity of their symptoms. Hence, it is important to diagnose the condition and localize the the tumor [1.3]. A male patient in his 40s, was referred to us as a case of osteoporosis with clinical features suggestive of proximal myopathy, Lower Motor Neuron (LMN) type facial palsy. On evaluation, the laboratory results revealed hypophosphatemia on the background of normal serum calcium levels, normal 25(OH) Vitamin D and 1,25-dihydroxy Vitamin D and elevated serum Alkaline phosphatase levels. This led to the calculation of TmP/gfr (Tubular maximum reabsorption rate of phosphate to glomerular filtration rate) which was also low indicating inappropriate renal phosphate wasting. A possibility of TIO was kept, and we were able to localise the tumor to his left temporal bone with the help of nuclear imaging. The tumor was resected and its histopathological examination showed benign spindle cell mesenchymal tumor consistent with a phsophaturic mesenchymal tumor. Postoperatively, there was significant clinical improvement and phosphate levels also returned to the normal reference range. Hypophosphatemia with normal serum calcium levels and 25(OH) Vitamin D and 1,25-dihydroxy Vitamin D levels in adults with significant musculoskeletal symptoms calls for the evaluation of TIO.

## KEYWORDS:

Tumor Induced Osteomalacia, TIO, Hypophostemic Osteomalacia, Mesenchymal Tumor, Case Report

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## INTRODUCTION

TIO is a paraneoplastic syndrome characterized by renal phosphate wasting due to excess production of FGF23. It is a rare entity with about 1000 cases that have been reported so far. Also, the average lag period between the initial clinical manifestations and the time when patient gets his correct diagnosis is 2.5-3 years due to non-specificity of their symptoms. The symptoms occur mainly because of hypophosphatemia and range from diffuse bone pains, muscle weakness to fractures. The diagnosis is usually established in the background of hypophosphatemia and renal phosphate wasting followed by searching for the phosphaturic tumor. Once the tumor is resected, the clinical as well as biochemical features resolve[2]. Hence, it is important to diagnose the condition and localize the tumor.

## CASE PRESENTATION

A male patient who was in his 40s was referred to us from orthopedics OPD for osteoporosis, based on his previously done DEXA scan. On eliciting history, his symptoms dated back to around 2 years with history of generalized bony pains followed by features of proximal myopathy including inability to stand from sitting position, followed by hearing loss and left sided facial weakness. The symptoms were gradual and progressive. He was a known case of hypertension and had been taking enalapril-losartan combination. There was no other significant past medical history, however he had undergone some surgery about 3 years back for his ear issues, the details of which were not available with him. On examination, patient had left Lower Motor Neuron (LMN) type facial weakness, proximal muscle weakness with power of Grade IV- bilateral lower limbs and Grade V in both upper limbs and a waddling gait, while rest of the CNS (Central Nervous System) functions were normal. One of his previous laboratory investigations had low serum phosphorus levels which hadn't been evaluated further. To confirm this, we repeated his calcium profile which was as under.

**Table 1:** Initial Laboratory Investigations.

| Investigation | Reference Ranges | Values |
|---|---|---|
| Serum calcium | 8.6-10.2mg/dl | 8.6mg/dl |
| Serum phosphate | 2.5-4.5 mg/dl | 1.6mg/dl |
| Alkaline phosphatase | 25-145 IU/L | 419 IU/L |
| 25-hydroxy Vitamin D | 30-50ng/ml | 42.2 ng/ml |
| 1,25-dihydroxy Vitamin D | 47.76- 190.32pmol/L | 51.1 pmol/L |
| Serum albumin | 3.5-5.2 gm/dl | 4.5gm/dl |
| Intact parathyroid hormone(iPTH) | 15-65 pg/ml | 100.4pg/ml |

**Table 2:** Investigations Done for the Evaluation of Hypophosphatemia.

| Investigation | Result |
|---|---|
| Spot Urinary Creatinine | 53.87 mg/dl |
| Spot Urinary phosphorus | 20 mg/dl |
| Serum creatinine | 0.7 mg/dl |
| Fractional excretion of phosphorus | 19.99% (<15%) |
| Tubular reabsorption of phosphorus | 0.81 |
| TMP/GFR | 1.053mg/dl (2.6-3.8) |

Both serum phosphate and TMP/GFR were low indicating inappropriate renal phosphate wasting. ABG values were pH= 7.426; pCo2= 38, Po2= 75.7, HCO3= 24.5, So2= 99.5%. Also, urine for glucose and aminoaciduria was negative. Considering inappropriately low 1,25 (OH)2 Vitamin D in the background of normal 25(OH)Vitamin D levels and renal phosphate wasting at the tubular level, FGF23 levels were measured and were found to be elevated, making a likely diagnosis of FGF23 excess. As the most common cause of acquired hypophosphatemia is tumor induced osteomalacia, we subjected the patient to 68Ga DOTATE PET scan which showed post-operative changes in the left temporal bone with common cavity formation of the external middle ear and mastoid air cells. DOTANOC avid (SUV max. 24.75) and diffuse soft tissue opacification with dysmorphic

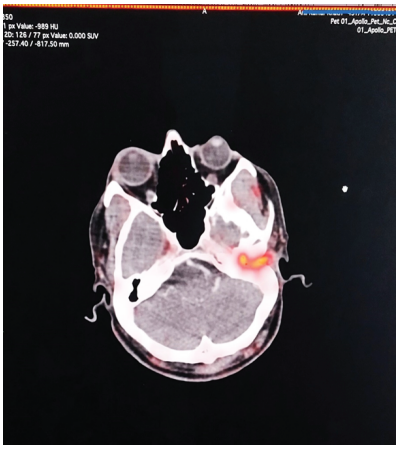

Figure 1

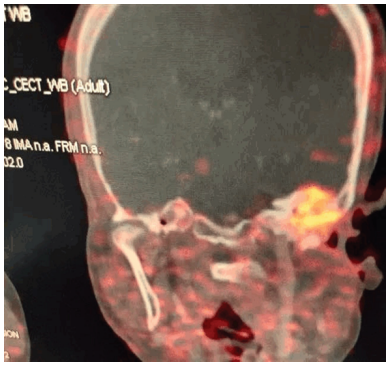

Figure 2

**Figure 1 and 2:** DOTANOC avid diffuse soft tissue opacification with dysmorphic calcification seen within the operative bed extending into the region of external ear and middle ear.

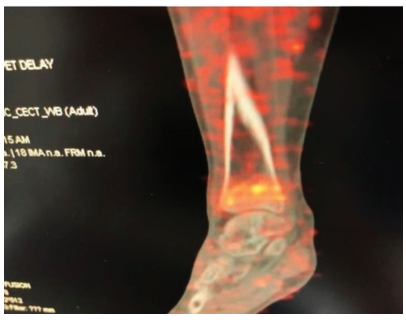

**Figure 3:** Mildly increased DOTANOC uptake in the lower end of right tibia.

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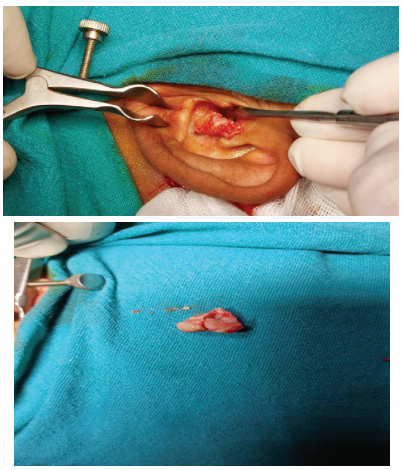

**Figure 4 and 5:** Left mastoidectomy with subtotal petrosectomy with blind sac closure with Facial nerve decompression.

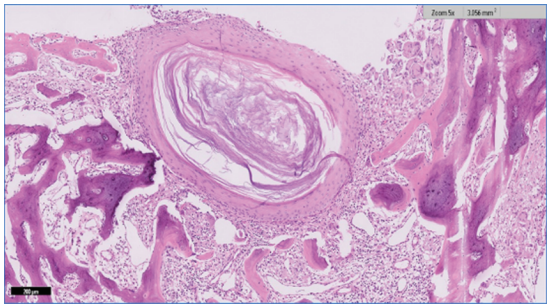

**Figure 6:** Histopathology of surgical specimen showing Chronic otitis media with cholesteatoma and very focal involvement by a benign mesenchymal neoplasm (in 5X zoom).

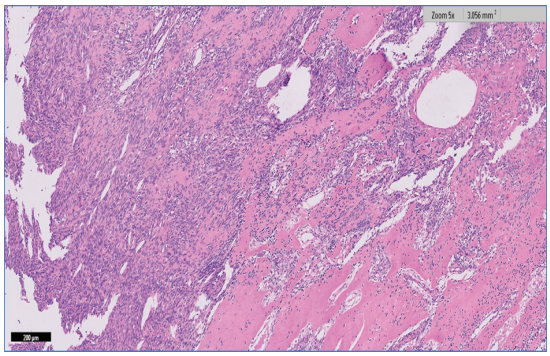

**Figure 7**

calcifications were seen within the operative bed extending into the region of external ear and middle ear (Figures 1 & 2). Non DOTANOC avid fractures were seen in right 6th-10th ribs with mildly increased DOTANOC uptake (SUV max 4.70) in the lower end of right tibia (Figure 3). As per recommendations, in cases where multiple tumors are seen on imaging, selective venous sampling is useful to identify which tumor actually produces FGF23 and hence aids in the diagnosis. The otorhinolaryngology department was consulted regarding selective venous sampling and also for

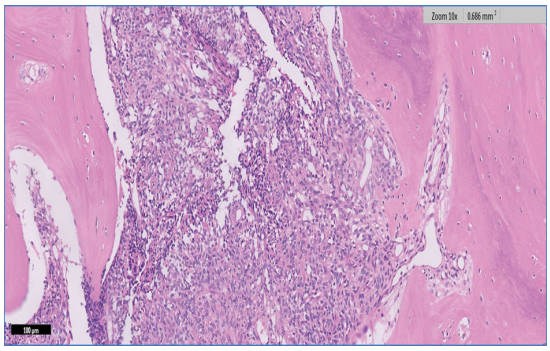

**Figure 8**

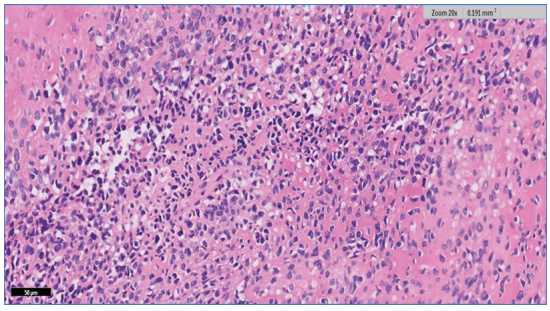

**Figure 7-9:** Benign spindle cell mesenchymal tumor consistent with a phosphaturic mesenchymal tumor in 5X, 10X, 20X.

the further management. It was performed on left Internal Jugular Vein and right popliteal vein and FGF23 levels were 143.40 pg/ml (23.4-95.4 pg/ml) and 76.40pg/ml (23.4-95.4pg/ml) respectively (Figures 4-9).

## TREATMENT AND INTERVENTION

Since FGF23 levels were significantly elevated in left internal jugular vein than right popliteal vein, the responsible tumor was identified as the one in left temporal bone. The otorhinolaryngologist did his left petrosectomy with facial nerve decompression and surgical specimen was sent for histopathological examination which showed benign spindle cell mesenchymal tumor consistent with a phsophaturic mesenchymal tumor.

## OUTCOME AND FOLLOW UP

Post operatively, patient's clinical profile improved with the Serum phosphorus 3.2mg/dl without any phosphate supplements. He initially used to walk completely with a walking stick, which gradually improved such that his power both lower limbs improved to Grade V from Grade IV-. However residual facial palsy was present. Patient has been asked to be on regular follow up with us.

## DISCUSSION

TIO is a rare paraneoplastic syndrome usually caused by phosphaturic mesenchymal tumors which are small, benign, and slow-growing neoplasms affecting bone or soft tissues, that produce FGF23 and, rarely, other phosphatonins. Increased levels of phosphatonins lead to increased renal

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phosphate wasting and hypophosphatemia and hence debilitating symptoms. The identification of tumor causing TIO might be a challenging task, as it is not always possible to identify these tumors on imaging, because of the small size and variable locations. Also, where multiple tumors are identified on imaging, to ascertain the source of TIO and that a given tumor is producing FGF23, selective venous sampling is done and FGF-23 levels are measured. Surgery is the first line management for these with resolution of symptoms, but that is possible only if tumor site has been correctly identified. In imaging negative cases, patient is managed using either radiotherapy, chemotherapy, radiolabelled somatostatin receptor-based therapies or the newer options like burosumab [3].

## WHAT IS USUALLY DONE?

- Confirm the diagnosis of hypophosphatemia
- Do TmP/GFR and TRP (will establish the distinction between low phosphate intake or increased renal wasting)
- If renal wasting is established, do FGF23 to establish its dependency
- Find the source of FGF23

## WHERE DOES THE LAG COME?

Non-specificity of symptoms and difficulty in locating tumor

## LEARNING POINTS

- Prolonged variable musculoskeletal symptoms can be due to hypophosphatemia
- Finding the cause of hypophosphatemia is important, which in adult population, can be due to tumor induced osteomalacia.

Every attempt should be made to search for the tumor location as tumors are mostly benign and can help patients in overcoming the disabling symptoms

## PATIENT PERSPECTIVE

I have been experiencing the generalized body pain initially followed by pain in both legs which progressively increased so much that I was unable to walk on my own followed by weakness of the face. I have been receiving various treatments since past 2 years, but were not of any major help. Thankfully, I have received the correct diagnosis and after surgery my symptoms are better than before and I am able to walk without support.

## AUTHOR CONTRIBUTIONS:

All authors made individual contributions to write this. Dr Varsha Kachroo and Dr Chandar Batra were involved in the management and follow-up of the patient. Dr Ameet Kishore performed the surgery of the patient. Dr Vikas Kashyap has reviewed the pathology slides and helped in clinching the ultimate diagnosis. Dr Dhruv Mishra helped in collecting the data and relevant information. Also, I would like to thank the patient for supporting this publication.

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**Informed Consent:** Signed informed consent was taken from the patient

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