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A Rare Case of Tumor-Induced Osteomalacia Despite Resection of a Benign Glomangioma

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Tumor-induced osteomalacia (TIO), a rare paraneoplastic syndrome, ectopically secretes fibroblast growth factor 23 (FGF23) and prevents phosphate reabsorption by suppressing renal Na/Pi cotransporter and inhibiting 1-α-hydroxylase.

A 42-year-old male with an incompletely resected left mastoid glomangioma presented with unprovoked fractures for the past four years. He had a DEXA scan which showed osteoporosis and a bone scan which showed innumerable foci of increased activity. Previous workup for multiple myeloma was negative. A skull X-ray showed an indeterminate focal lucency in the calvarium, suggestive of Paget's disease, for which he was treated with zoledronic acid. A phosphorus level was never ordered. On exam, there was tenderness to palpation in his forearms and he ambulated with crutches. A skeletal survey revealed impending fractures of the femurs. Labs revealed calcium 8.6mg/dL (8.4-10.2mg/dL), phosphorus 1.9mg/dL (2.5-4.5mg/dL), 25(OH)D 52.2ng/mL (30-100ng/mL), $1,25(OH)_2D$ 19.1pg/mL (19.9-79.3pg/mL), PTH 80pg/mL (15-65pg/mL), alkaline phosphatase 183U/L (35-105U/L) and bone alkaline phosphatase isoenzyme 73% (12-68 %). Tubular maximum phosphate reabsorption per GFR (TmP/GFR) was low at 0.43mmol/L (0.99-1.34mmol/L) indicating high urinary phosphorus excretion. Intact FGF23 level was elevated to 93pg/mL (<22pg/mL) and genetic testing for hypophosphatemic rickets was negative. A ^{68}Ga-DOTATATE CT/PET scan revealed an intense uptake in the left temporal bone (SUV max 19.0) at the site of his glomangioma resection. Burosumab was initiated and he is planned to undergo radiation therapy to treat the residual tumor in hopes of achieving resolution of TIO.

Our patient had TIO from his glomangioma which is often difficult to localize and missed on routine imaging. Somatostatin receptor imaging such as ^{68}Ga-DOTATE CT/PET can be a useful imaging modality. Burosumab, a human anti-FGF-23 monoclonal antibody, is a therapeutic option in cases of unresectable TIO to improve fracture-healing and normalize phosphorus levels. Our case further highlights the importance of measuring phosphorus levels to evaluate metabolic bone disease.

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