CLINICAL IMAGE

Positive somatostatin receptor imaging does not predict somatostatin analogue efficacy in tumor-induced osteomalacia

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A 41-year-old man, a professional truck driver, was referred to the Department of Endocrinology with a history of hypophosphatemic osteomalacia lasting several years, resulting from fibroblast growth factor 23 (FGF23) hypersecretion of an unknown origin. Positron emission tomography (PET) with ^{18}F-fluorodeoxyglucose failed to detect any lesions; however, the patient was scanned at a level of above one-third of the thighs. Thus, we decided to perform somatostatin receptor scintigraphy (^{99m}Tc-EDDA/HYNIC-Tyr3-octreotide, 730 mBq). An increased radiotracer uptake in the projection of medial condyle of the right femoral epiphysis was shown (FIGURE 1A). The possible phosphaturic mesenchymal tumor (PMT) was next visualized by computed tomography and magnetic resonance imaging (FIGURE 1B and 1C). The diagnosis was eventually confirmed by biopsy.

The first attempt to remove the tumor was ineffective, and the patient refused a more extensive surgery. Therefore, a pharmacological approach was considered. Phosphorus supplementation, although efficient in maintaining normal phosphorus levels for the last several years, now resulted in secondary hyperparathyroidism. The addition of calcium supplements or alfacalcidol was also ineffective in reducing elevated parathormone levels. As histopathology revealed positive staining for somatostatin receptor type 2 (SSTR2) in most cells and focal SSTR5 immunohistochemical reaction, we decided to introduce somatostatin analogue therapy. Octreotide (100 μg 3 times daily) for 3 days did not increase serum phosphate levels or reduce 24-hour urine phosphorus excretion. Similarly, pasireotide (600 μg twice daily) resulted only in insignificant improvement of blood phosphatemia.

Whole-body somatostatin receptor scintigraphy has been proved to be a useful tool for detecting mesenchymal tumors.[1] However, the role of somatostatin signaling in PMTs is unclear, and literature data regarding somatostatin analogue efficacy in the treatment of patients with tumor-induced osteomalacia (TIO) are inconsistent. Octreotide efficacy was shown by Seufert et al[2] in a 50-year-old man with PMT in the left thigh, although there is a considerable controversy around its wider use in this patient population. Some authors described a decrease in FGF23 levels, although with no clinically important effect.[1,3] Recently, Ovejero et al[4] reported no significant changes in blood phosphate, FGF23, 1,25-dihydroxycholecalciferol, or tubular reabsorption of phosphates (TRP) during octreotide treatment of 5 TIO patients for 3 days, concluding that these drugs are not efficient in the short term. However, from our previous experience, octreotide, both as a long- or short-acting agent, can help improve the level of blood phosphates in the preoperative period.[5]

Our study was the first attempt to use pasireotide in the treatment of a patient with TIO. Pasireotide is a potent somatostatin analogue with a 40-fold increased affinity to SSTR5 as compared with octreotide, as well as a comparable binding affinity to SSTR2. It seems that positive somatostatin receptor imaging or positive somatostatin receptor staining in histopathological specimens could serve as indicators of the possible effectiveness of somatostatin analogues. However, in our case, neither octreotide nor pasireotide was in fact effective. Concerning conflicting observations and significant costs of therapy, we conclude that before deciding on a long-term use of somatostatin analogues in TIO patients, the clinical utility of these agents should first be proved in individual cases.

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Received: August 1, 2018.
Revision accepted: August 22, 2018.
Published online: August 24, 2018.
Conflict of interest: none declared.
Pol Arch Intern Med. 2018; 128 (9): 554-555
doi:10.20452/pamw.4318

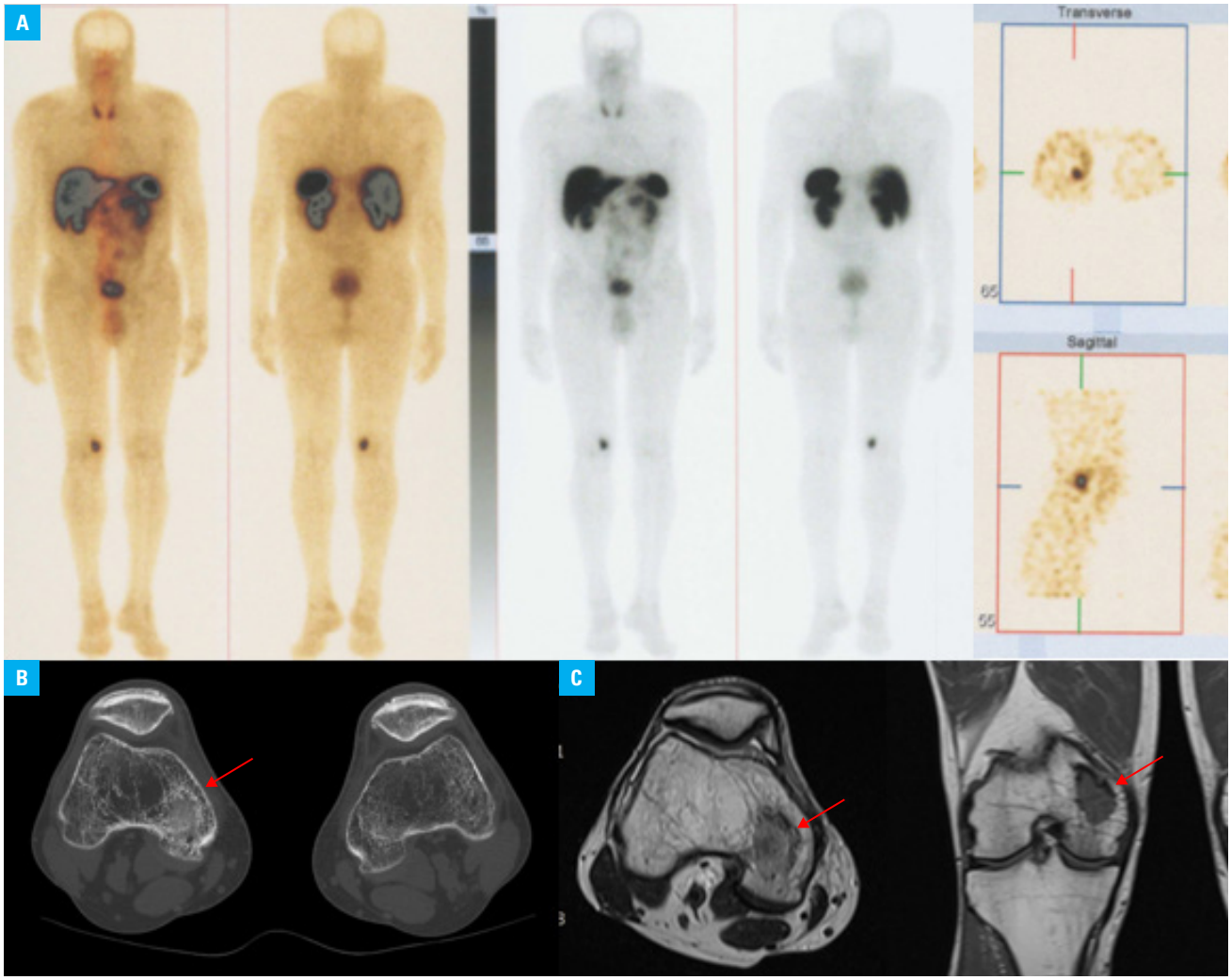

FIGURE 1 **A** – somatostatin receptor scintigraphy (^{99m}Tc-EDDA/HYNIC-Tyr3-octreotide): intense pathological radiotracer uptake in the right femoral epiphysis; **B** – computed tomography scan (transverse) of the right and left femoral epiphysis, with a visible phosphaturic mesenchymal tumor of blurred margins (25 × 18 × 14 mm) between the left epicondyle and condyle of the right femur (red arrow); **C** – magnetic resonance imaging of the right knee joint (coronal and axial sections), with a visible phosphaturic mesenchymal tumor (red arrows)

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