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Fibrous dysplasia mimicking malignancy on ^{68}Ga-DOTATATE PET/CT

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Abstract

Fibrous Dysplasia (FD) of bone is a developmental benign skeletal disorder characterized by replacement of normal bone and normal bone marrow with abnormal fibro-osseous tissue. We report on a case of a biopsy proven FD lesion in the left temporal bone, with intensely increased activity (SUVmax: 56,7) on ^{68}Ga-DOTATATE PET/CT. The presented data indicates cell surface over-expression of somatostatin receptors (SSTRs) by fibrous dysplastic cells and highlights the need of cautious management of ^{68}Ga-DOTATATE avid bone lesions, which could mimic malignancy especially in patients with history of neuroendocrine tumors (NETs).

A 72 year-old man with history of right renal cell carcinoma and gastro-intestinal stromal tumor (GIST) of the stomach which were both successfully resected with no evidence of recurrence, presented with an enlarging retroperitoneal lymph-node on consecutive follow-up imaging tests. Surgical resection of the node was performed and pathology revealed well-differentiated neuroendocrine tumor (NET) of unknown primary origin. The patient was referred to our institute for follow-up and was evaluated with whole-body PET/CT scan using ^{68}Ga-DOTATATE for the detection of additional neuroendocrine lesions, which showed intensely increased activity (SUVmax: 56,7) in the left temporal bone (Fig 1A: MIP and Fig 1B: axial fused ^{68}Ga-DOTATATE PET/CT images of the head, arrows). Subsequent CT scan showed bone expansion on the superior aspect of the left temporal bone with “ground-glass” appearance (Fig 1C: arrows; axial non-contrast CT image of the head), suggesting fibrous dysplasia (FD), although an osteosclerotic metastatic lesion could not be

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excluded. Furthermore, MRI scan showed reactive thickening of the dura and enhancement of the pathological segment of the eft temporal bone consistent with active FD (Fig 1D: arrows; axial contrast-enhanced T1-weighted MR-image). Biopsy of the left temporal bone lesion was performed revealing benign fibrotic tissue with no evidence of malignancy, establishing FD diagnosis.

FD of bone is developmental benign skeletal disorder in which normal bone and bone marrow are replaced by abnormal fibro-osseous tissue resulting in fracture, deformity, functional impairment, and pain [1]. FD may affect one bone (monostotic FD), multiple bones (polyostotic FD) or the entire skeleton (panostotic FD) and can be encountered either sporadically or within the McCune-Albright syndrome, which includes FD in association with extra-skeletal features such as café-au-lait macules and hyperfunctioning endocrinopathies [2]. Even more rarely FD may be associated with intramuscular myxomas within the Mazabraud's syndrome [3]. The radiological features of FD vary, depending on the age, the location of the affected bone and the underlying histopathology of a given lesion [4]. The introduction of ^{68}Ga-DOTA-conjugated-peptides somatostatin (SST)-analogues into clinical practice allowed SST-receptors (SSTRs) imaging with PET, and is evolving as the new imaging standard of reference for the detection and characterization of NETs and other SSTR-positive lesions [5–10]. The presented case of a biopsy-proven FD lesion with intensely increased activity on ^{68}Ga-DOTATATE PET/CT suggests cell-surface overexpression of SSTRs by the fibrous dysplastic cells and particularly SSTR-2 for which ^{68}Ga-DOTATATE has a predominant affinity. This observation is consistent with previous report referring to the findings of conventional SSTR-scintigraphy showing abnormally increased pentetreotide (SST-analogue) uptake in all sites of FD lesions in a McCune-Albright patient [11]. The presented data implies the application of PET/CT imaging with ^{68}Ga-DOTA-conjugated-peptides in assessing and monitoring FD activity and a potential role for somatostatin in treatment of this disease. Furthermore, there may be prognostic value of ^{68}Ga-DOTATATE uptake in FD patients that needs to be further explored. Finally, bone sites of intensely increased ^{68}Ga-DOTATATE uptake should be cautiously managed, since they may correspond to benign fibrotic lesions which could mimic malignancy on ^{68}Ga-DOTATATE PET/CT.

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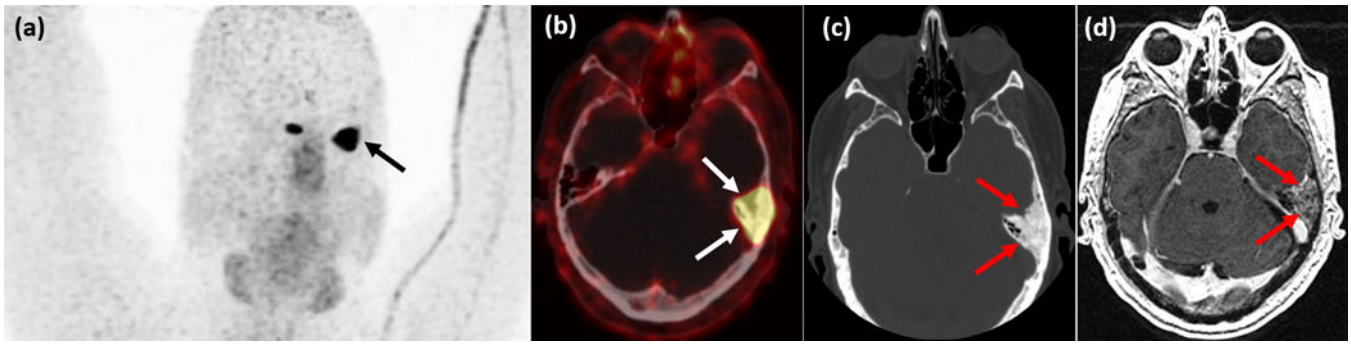

Fig 1.