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^{18}F -NaF and ^{18}F -FDG PET/CT in Gorham-Stout disease

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Abstract

Gorham-Stout disease (GSD) is an extremely rare skeletal disorder of unknown etiology characterized by benign proliferation of vascular or lymphatic channels, leading to progressive bone resorption. We report on a patient diagnosed with GSD affecting the right scapula and the right ribs, who underwent PET/CT scans using ^{18}F -FDG and ^{18}F -NaF. The remnant upper portion of the affected scapula did not show ^{18}F -FDG uptake, but demonstrated markedly increased ^{18}F -NaF activity. Furthermore, intense ^{18}F -NaF activity was seen on the right posterior ribs which were actively being resorbed, suggesting the potential application of ^{18}F -NaF-PET/CT imaging in GSD diagnosis and follow-up.

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FIGURE 1.

A 44 year-old-man with worsening pain and movement limitation in the right shoulder joint underwent chest X-ray, which revealed absence of the largest portion of the right scapula (Fig. 1A). Of note is that chest X-ray and MRI scans which were performed three years earlier had not shown any bone abnormality. Subsequently, chest CT scan confirmed absence of the right scapular body with preservation of only the scapular spine (Fig. 1B: coronal chest CT-image bone window; yellow arrows, Fig. 1C: 3D-reconstructed image produced by automated segmentation and volume rendering of the bones). Chest-MRI showed absence of the inferior portion of the right scapula and severe atrophy of the right subscapularis and infraspinatus muscles associated with fatty replacement (Fig. 1D: axial T1-weighted image without contrast; yellow arrows) when compared to the left side (Fig. 1D; red arrows). Given the risk of chylothorax, biopsy was avoided; clinical and imaging findings supported the diagnosis of GSD. In order to evaluate disease activity the patient underwent whole-body PET/CT scans using ^{18}F -FDG and ^{18}F -NaF. The remnant upper portion of the right scapula did not show increased uptake by ^{18}F -FDG (Fig. 1E; axial fused ^{18}F -FDG PET/CT images, white arrows), whereas it demonstrated elevated ^{18}F -NaF activity (SUVmax: 27,3) (Fig 1F; axial fused ^{18}F -NaF PET/CT images, white arrows).

FIGURE 2.

Multiple foci of intensely increased ^{18}F -NaF activity located in the right posterior ribs (Fig. 2A: ^{18}F -NaF MIP image red arrows; Fig. 2B: axial fused ^{18}F -NaF-PET/CT image white arrow, SUVmax: 84,5) were associated with areas of incipient bone resorption (Fig. 2C: axial chest CT-image bone window, white arrow) supporting the application of ^{18}F -NaF as the optimal bone-seeking PET-radiopharmaceutical to assess GSD activity.

Gorham-Stout disease (GSD) is a rare skeletal disorder of unknown etiology characterized by proliferation of endothelial-lined vessels in bone, and by progressive replacement of bone with fibrous tissue [1]. GSD can present at any age, with no clear sex predilection or inheritance pattern, and can affect any bone in the body with variable symptoms depending on the sites of involvement [2]. Diagnosis is based on combination of clinical with radiographic and histopathological features [3]. Although in some GSD cases blood vessels were reported to abnormally proliferate [4, 5] there is growing evidence that lymphatic vessels are mostly affected [6, 7]. ^{18}F -FDG is a radiolabeled glucose analog suitable for PET-imaging which enters cells via glucose membrane transporters, reflecting metabolic activity [8]. The presented findings do not indicate any increase in metabolism by the remnant bone. In contrast, ^{18}F -NaF is a bone-specific agent which is deposited on bone via ^{18}F -ions exchange with hydroxyl (OH^-)-ions on the surface of the hydroxyapatite matrix leading to formation of fluoroapatite [9]. ^{18}F -NaF bone uptake and retention reflects blood flow and processes that increase the exposed bone surface providing more binding sites [10]. Vessel proliferation present in GSD and the underlying osteolytic process which increases the exposed bone surface could explain the elevated ^{18}F -NaF activity by the remnant upper part of the right scapula in the presented case. These data suggests potential application of ^{18}F -NaF-PET/CT imaging in GSD diagnosis as well as assessment of disease activity and treatment response.