A Very Unusual Pattern of Intraperitoneal and Extraperitoneal Heterotropic Splenic Tissue-Mimicking Metastases Identified on ^{68}Ga-DOTA-NOC Positron Emission Tomography/Computed Tomography and ^{99m}Tc Heat-denatured Erythrocyte Study

Abstract

The dissemination and autotransplantation of viable splenic tissue in different anatomic compartments of the body can present a diagnostic dilemma, especially when metastatic disease is suspected. We report a case of a 30-year-old male with well-differentiated gastric neuroendocrine tumor (Grade II) treated with surgery. Follow-up ^{68}Ga-DOTA-NOC demonstrated a suspicious peritoneal soft-tissue nodule in the right paracolic gutter with increased tracer uptake. In view of splenectomy 10 years ago, the patient underwent ^{99m}Tc heat-denatured erythrocyte study which showed a very unusual pattern of multiple tracer-avid foci of splenic tissue in both intraperitoneal and extraperitoneal distributions. The integration of the patient's history and complementary nuclear imaging results led to the correct diagnosis of splenosis.

Keywords: *^{68}Ga-DOTA-NOC positron emission tomography/computed tomography, ^{99m}Tc heat-denatured erythrocyte study, single-photon emission computed tomography–computed tomography, splenosis*

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A 30-year-old male with a history of gastric surgery for well-differentiated neuroendocrine tumor (NET) (Grade II) underwent ^{68}Ga-DOTA-NOC positron emission tomography/computed tomography (PET/CT) to rule out clinical suspicion of metastasis. He had a background of hemolytic anemia for which he underwent splenectomy 10 years back. The ^{68}Ga-DOTA-NOC PET/CT [Figure 1] showed a suspicious peritoneal soft-tissue nodule with intense activity in the right paracolic gutter suspicious for metastases. ^{99m}Tc heat-denatured erythrocyte scintigraphy [Figure 2] showed intense activity in this nodule confirming this to be splenosis. Interestingly, two further small foci of heterotopic splenic tissue were identified, which included a focus in the musculature of the left lower anterior abdominal wall and a further small focus in the left anterior iliac fossa abutting the left rectus sheath, both of which were not particularly avid on ^{68}Ga-DOTA-NOC PET/CT. While abdominopelvic splenosis is well described, the presence of intramuscular heterotropic splenic tissue in the anterior abdominal wall musculature is highly unusual and has never been previously described in the literature to the best of our knowledge.

The term splenosis was first proposed by Buchbinder and Lipkoff in 1939 to describe the heterotropic transplantation of splenic tissue within the abdominal cavity.[1] It occurs following traumatic or iatrogenic rupture of the spleen and is a rare finding with most cases occurring within the abdominal cavity.[2,3] The pathogenesis is still not definitively understood, however, one proposed mechanism is the spillage of the damaged splenic pulp into the adjacent cavities,[4] followed by recruitment of blood supply from the surrounding tissues and vessels, without any association to the splenic artery.[5] A second likely mechanism is hematogenous spread of splenic pulp which was suggested to explain intrahepatic splenosis.[6] The very unusual intramuscular distribution of heterotropic splenic tissue, as reported in our case, can perhaps be best

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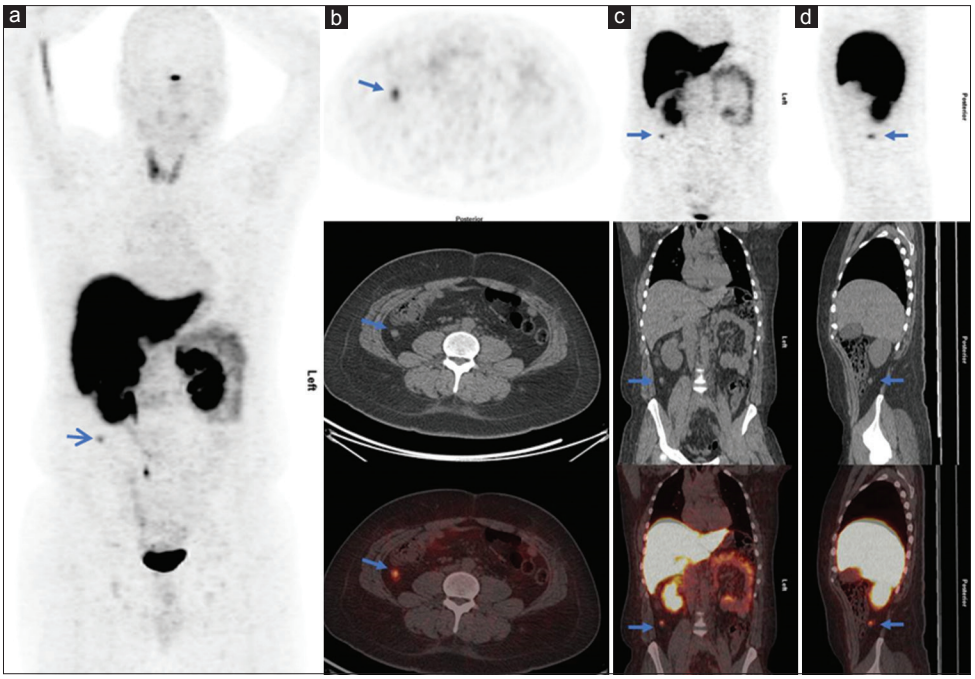

Figure 1: ^{68}Ga-DOTA-NOC photon emission tomography–computed tomography images, (a) maximum intensity projection, (b) transaxial, (c) coronal, and (d) sagittal images show a suspicious soft-tissue density nodule adjacent to the ascending colon with corresponding discrete focal tracer uptake on fused images

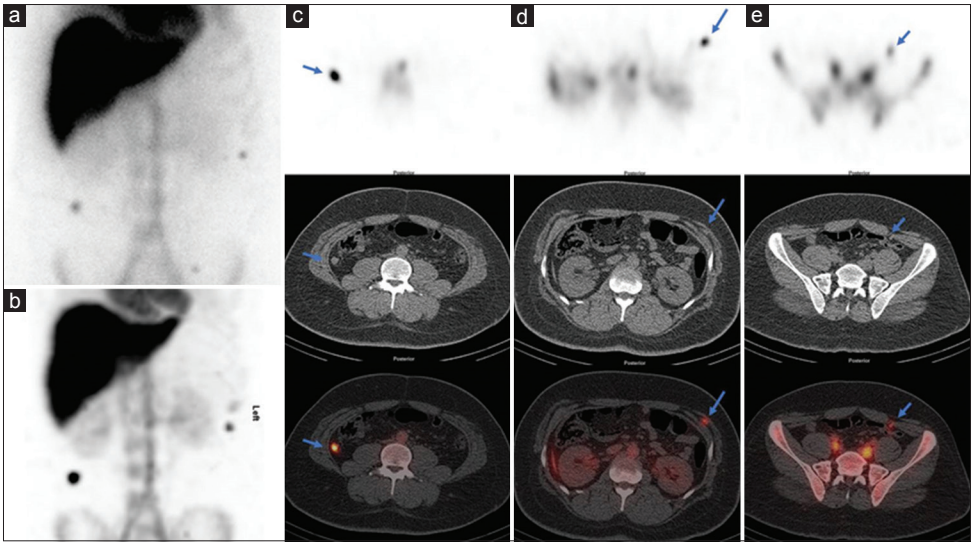

Figure 2: (a) Static view of ^{99m}Tc heat-denatured erythrocyte scintigraphy, (b) maximum intensity projection images, (c-e) transaxial single-photon emission computed tomography–computed tomography images show multiple small avid soft-tissue densities projected in the abdomen and pelvis. (c) The largest focus with most intense activity is seen in the right paracolic gutter posterior to the ascending colon at the level of L4 vertebra. (d) Further focal area of increased tracer uptake is seen in the left lower anterior abdominal wall between the internal and external oblique muscles. (e) Tiny focus is also seen in the left iliac fossa anteriorly, closely related to the left rectus sheath

explained by the second proposed mechanism, although one can postulate that splenic tissue can be seeded in the soft tissue along a standard laparotomy incision, but it is not possible to confirm this hypothesis.

The average interval reported between trauma and abdominopelvic splenosis is 10 years, with a range of 5 months to 32 years.[7] Histologically, splenosis differs from accessory spleens by the absence of elastic or smooth muscle fiber in the capsule, but heterotropic splenic tissue is still thought to perform normal splenic function similar to accessory spleen.[8] While it is usually diagnosed incidentally, splenosis can pose a diagnostic dilemma, especially when metastatic malignant disease is suspected as in the present case, when the patient was referred for restaging with ^{68}Ga-DOTA-NOC PET/CT.

^{8}Ga-DOTA-peptides bind to the somatostatin receptor (SSTR) and are a sensitive imaging tool for a

wide range of NETs.[9,10] One of its pitfalls is the expression of SSTR in normal splenic tissue. This can be a problem when ectopic splenic tissue (accessory spleens or splenosis) mimics NET or metastases. Standard cross-sectional imaging (contrast-enhanced CT and magnetic resonance imaging) can be helpful but cannot always diagnostic.[11,12] Scintigraphy with ^{99m}Tc-tin colloid is a functional test which is used to diagnose splenosis given the ability of colloid to localize in the reticuloendothelial system.[13] However, scintigraphy using ^{99m}Tc heat-denatured erythrocyte study is more sensitive and specific for splenic uptake, making it the current diagnostic test of choice.[12] Gunes *et al*.[14] demonstrated that the red blood cell (RBC) scintigraphy had a 32% greater diagnostic yield compared with the colloid study. One reason for the better accuracy with heat-denatured erythrocyte study may be that the spleen takes up only about 10% of the injected tin colloid versus a greater than 90% uptake of damaged RBCs. ^{99m}Tc heat-denatured erythrocyte study has also been shown to be more sensitive in early splenosis cases where minimal splenic tissue is present. This is probably the reason why the most unusual foci related to the anterior abdominal wall musculature [Figure 2] in the present case were not well seen on the ^{68}Ga-DOTA-NOC PET/CT. Autoradiography and immunohistochemistry studies demonstrated that SSTRs were mainly located in the red pulp of the spleen.[15] It is possible that in our case, the composition of the smaller heterotropic splenic foci influenced the nonexpression of SSTRs making these smaller foci undetectable on standard ^{68}Ga-DOTA-NOC PET/CT.

It is important to reiterate that splenosis should be included in the differential diagnosis in all oncology patients with a history of splenic trauma or splenectomy and with intra-abdominal foci which are positive on staging ^{68}Ga-DOTA-NOC PET/CT. Particular care should be taken in the interpretation of unusual intrahepatic foci as splenosis can mimic hepatic metastases on ^{68}Ga-DOTA-NOC PET/CT. A low threshold for further characterization with nuclear imaging should be exercised in these cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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