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Case Series

Octreotate avid intrapancreatic lesions-intrapancreatic splenunculi masquerading as pancreatic neuroendocrine tumours

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ABSTRACT

Splenunculi (heterotopic spleens or accessory splenic tissue), are relatively common (occurring in ~10-30% of the population) and can occur in a variety of locations, including within solid organs such as the pancreas, kidney and the liver. The majority of intrapancreatic splenunculi (IPS) are located within or adjacent to the tail of the pancreas. Differentiating IPS from other pancreatic lesions, specifically those with malignant potential such as pancreatic neuroendocrine tumours (pNETs) can be challenging based solely on radiological appearance. Both IPS and pNETs have similar characteristics on contrast enhanced CT and octreotate-PET imaging. IPS are benign and largely asymptomatic. They require no surgical intervention or follow-up. pNET's are relatively rare neoplasms which require active surveillance or surgical resection depending on their size and other patient factors. Current international guidelines do not routinely recommend definitive tissue diagnosis prior to proceeding with surgical resection of presumed pNET lesions. Here, we present a case series of four patients with tail of pancreas (ToP) lesions, initially suspected to be pNETs based on CT and octreotate PET imaging, who subsequently underwent further, more sensitive investigations (denatured red cell scan or endoscopic ultrasound (EUS) guided fine needle aspirate (FNA) and were confirmed to have IPS. Accurate diagnosis with further investigations such as EUS guided biopsy, is valuable in guiding surgical management of octreotate avid ToP lesions and avoiding potentially harmful and unnecessary surgical intervention.

Keywords: Case series, Intrapancreatic splenunculi, Pancreatic neuroendocrine tumour, Octreotate PET

INTRODUCTION

Splenunculi (heterotopic spleens or accessory splenic tissue), are relatively common (occurring in ~10-30% of the population) and can occur in a variety of locations, including within solid organs such as the pancreas, kidney and the liver.[1] The majority of IPS are located within or adjacent to the tail of the pancreas. Differentiating IPS from other pancreatic lesions, specifically those with malignant potential such as pNETs can be challenging based solely on radiological appearance.[1,2] Both IPS and pNETs have similar characteristics on contrast enhanced CT and octreotate-PET imaging.[2]

IPS are benign and largely asymptomatic. They require no surgical intervention or follow-up. pNET's are relatively rare neoplasms which require active surveillance or surgical resection depending on their size and other patient factors.[3]

Current international guidelines do not routinely recommend definitive tissue diagnosis prior to proceeding with surgical resection of presumed pNET lesions.[4]

Here, we present a case series of four patients with ToP lesions, initially suspected to be pNETs based on CT and octreotate PET imaging, who subsequently underwent further, more sensitive investigations EUS guided FNA and were confirmed to have IPS.[3,5]

Accurate diagnosis with further investigations such as EUS guided biopsy, is valuable in guiding surgical management of octreotate avid ToP lesions and avoiding potentially harmful and unnecessary surgical intervention.

CASE SERIES

Case 1

A 68-year-old female presenting with flushing and deranged liver function tests (LFTs) was found to have a dilated pancreatic duct and 13 mm intensely enhancing mass in the ToP on CT. Her chromogranin A (CgA) level was 87 and CA 19-9 was 4. A denatured red cell scan was negative for reticuloendothelial activity, and octreotate PET demonstrated intense octreotate avidity in the ToP lesion, consistent with a pNET (Figure 1). The patient underwent EUS plus FNA to further characterise the lesion, which demonstrated a 12mm hypoechoic lesion adjacent to the ToP and spleen. FNA cytopathology confirmed benign splenic parenchyma. The patient remains well and has been discharged from the hepatobiliary team.

Case 2

A 44-year-old female was investigated by her GP for generalised abdominal pain and distension. CT and MRI pancreas identified a 15mm ToP lesion, reported radiological differentials were an IPS or pNET. A denatured red cell scan showed no significant activity in the ToP lesion, which is atypical for IPS. Octreotate PET demonstrated an octreotate avid lesion in the ToP, suggestive of a pNET (Figure 1). EUS demonstrated a well-defined 14.5×11.7 mm hypoechoic ToP lesion, in an otherwise unremarkable pancreas. FNA cytology confirmed the lesion to be benign splenic tissue. No further hepato-biliary follow-up was required.

Case 3

A 65-year-old-male was found with an incidental finding of a 24mm ToP mass lesion on CTPA, conducted in the context of new diagnosis of multiple pulmonary emboli. Subsequent CT and MRI pancreas confirmed a 24mm spiculated lesion with central cystic component suspected to be either a pNET or pancreatic adenocarcinoma. The lesion showed no significant FDG activity but demonstrated moderate octreotate avidity on PET imaging (Figure 1). The patient subsequently underwent EUS with FNA biopsies to obtain a cytopathological diagnosis, however the results were indeterminate with evidence of gastric mucosal contamination and strips of glandular epithelium raising concerns for a possible mucinous cystic lesion. Molecular testing of the FNA sample was negative for KRAS mutation. Following discussion at the upper gastrointestinal multidisciplinary team meeting the patient was recommended to have distal pancreatectomy. A successful spleen preserving distal pancreatectomy was performed without complication and the resected specimen demonstrated a benign squamous epithelial cyst within heterotopic splenic tissue.[6,7] The patient recovered well post-operatively and has no ongoing surgical concerns.

Case 4

A 61-year-old male undergoing routine radiological surveillance following treatment for oesophageal squamous cell carcinoma was incidentally found to have a 24 mm solid ToP lesion and adjacent 11 mm lymph node on CT, raising concern for metachronous pancreatic malignancy. His tumour markers were normal; CA 19-9 of 12 and CEA of 4. The patient completed a denatured red cell scan which confirmed focal intense uptake in both the mass lesion and adjacent ‘lymph node’ reporting that both lesions were consistent with splenunculi. No further follow-up was required for his IPS, however he remains under surveillance for his oesophageal cancer.

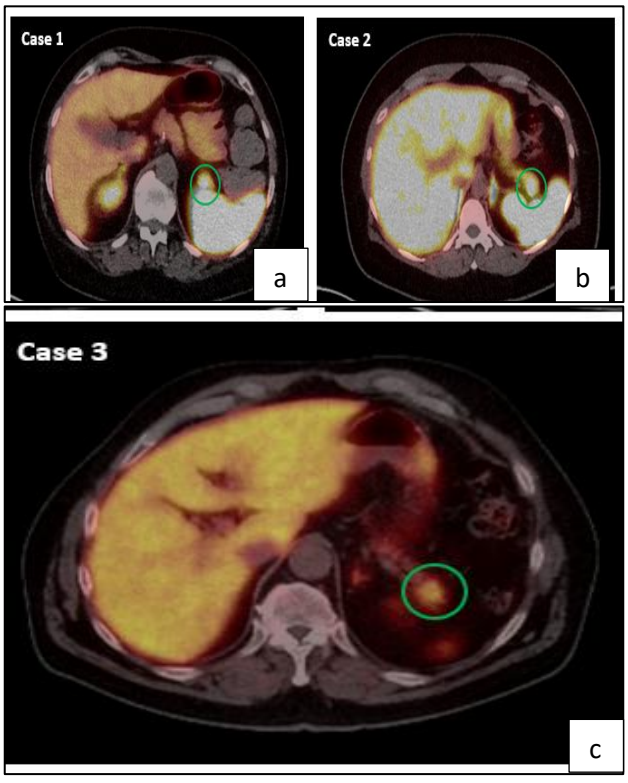

Figure 1 (A-C): Octreotate PET images showing ToP lesions with positive uptake.

DISCUSSION

In utero the spleen develops from multiple mesenchymal buds, failed fusion of these buds results in small nodules

of benign splenic tissue outside of the spleen.[8] These congenital abnormalities are called splenunculi (heterotopic spleen or accessory splenic tissue) and occur in 10-30% of the population in various locations, most frequently the splenic hilum, adjacent to the ToP or the lower pole of the left kidney.[9] Although less common, splenunculi can also occur within the parenchyma of nearby solid organs such as the pancreas, IPS almost exclusively occur in the tail of the pancreas.[7] Splenunculi consist of benign splenic tissue, are largely asymptomatic and commonly discovered as incidental findings on abdominal imaging.

pNETs are relatively rare neoplasms, broadly categorised into functional and non-functional tumours based primarily on their clinical presentation. The tumour grade (WHO classification) provides the best prognostic indicator and is determined by the Ki67 proliferation index and mitotic count.[10] While conservative management with active surgical surveillance may be an option for non-functioning and low grade pNETs <2 cm in size, the recommended management, and only curative treatment available, is surgical resection.[10,11] Overall pNETs have a good prognosis following surgical resection, with a 5-year survival rate of 65-86%.[7]

Contrast enhanced CT imaging of splenunculi demonstrates characteristic features of small (on average <20mm in size) hypervascular round lesions with clear margins and homogenous enhancement, like CT findings of normal splenic tissue.[12,13] CT findings of IPS can however mimic other hypervascular lesions such as pNETs or metastatic deposits. Furthermore, both pNETs and IPS demonstrate avidity on ^{68}Ga-Octreotate PET imaging, as splenic tissue displays intense physiological uptake of the tracer.[14] Further assessment with functional imaging such as SPECT-CT after injection with ^{99m}Tc-labelled red blood cells is able to detect splenic tissue due to its physiological uptake of RBC's.[15] However, its sensitivity is affected by proximity of the lesion to the spleen and lesion size, leading to false negatives as seen in cases 1 and 2, making it difficult to determine definitive diagnosis on radiological findings alone.

The most sensitive investigation of ToP lesions is EUS plus FNA to obtain cytopathological diagnosis.[10] Accurate diagnosis of ToP lesions is paramount in guiding appropriate management, in three of the four cases discussed the definitive diagnosis of IPS prevented unnecessary surgical resection. Two of these diagnoses were obtained via EUS biopsies and pathology demonstrating benign splenic tissue. We recommend EUS with FNA prior to resection of all ToP lesions.

CONCLUSION

Over-reliance on imaging without tissue confirmation can lead to unnecessary pancreatic surgery with inherent risks. Accurate preoperative tissue diagnosis is crucial to differentiate IPS from pNETs. When available EUS-FNA biopsy should be considered when evaluating octreotate avid ToP lesions and can avoid unnecessary patient morbidity.

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