Journal of Surgery
Militerno G, et al. J Surg 7: 1564
www.doi.org/10.29011/2575-9760.001564
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Case Report

Incidental Finding of Pancreatic Cystadenoma in Splenic Cavernous Hemangioma

Giuseppe Militerno[1*], Giuseppe Surfaro[1], Claudio Cimmino[1], Marco Maione[1], Gianluca Benassai[1], Francesco Arienzo[2], Miriam Piccirillo[3], Ciro Fittipaldi[3]

[1]Department of Emergency and General Surgery, Ospedale del Mare, Napoli 80147, Napoli, Italy

[2]Department of Radiology, Ospedale del Mare, Napoli 80147, Napoli, Italy

[3]Department of Anesthesia, Ospedale del Mare, Napoli 80147, Napoli, Italy

[*]**Corresponding author:** Giuseppe Militerno, Department of Emergency and General Surgery, Ospedale del Mare, Napoli 80147, Napoli, Italy

Citation: Militerno G, Surfaro G, Cimmino C, Maione M, Benassai G, et al. (2022) Incidental Finding of Pancreatic Cystadenoma in Splenic Cavernous Hemangioma. J Surg 7: 1564. DOI: 10.29011/2575-9760.001564

Received Date: 01 September, 2022; **Accepted Date:** 12 September, 2022; **Published Date:** 15 September, 2022

Abstract

Background: Cavernous hemangioma, also called cavernous angioma, venous malformation, or cavernoma, is a type of venous malformation due to endothelial dysmorphogenesis of a lesion present at birth. Splenic cavernous hemangioma is a very rare lesions, with fewer than 100 cases reported in the literature. They are often asymptomatic, and their diagnosis is occasional, or after a symptom such as compression if it is large, bleeding, or other complications. Pancreatic Cystic Neoplasms (PCNs) are liquid-containing tumors that can grow to a considerable size and most commonly affect women. PCNs are estimated to occur in 2-45% of the general population and comprise a clinically challenging entity as their biological behavior varies from benign to malignant disease. They may be asymptomatic or show symptoms of compression or pancreatitis. Depending on the case, follow-up or resection may be useful. Follow-up is always recommended due to their potential for malignancy.

Case Summary: We report the case of a 46-year-old Chinese woman who was referred to our emergency department for diffuse abdominal pain. Radiological investigations reported the presence of a suspected ruptured splenic aneurysm. Due to the worsening of the patient's clinical conditions, an exploratory laparotomy was performed. At the opening of the peritoneum, an aneurysm of the splenic artery of approximately 10 cm was found with active bleeding. The aneurysm was in close continuity with the pancreatic tail and the splenic hilum and was very difficult to separate from the pancreas. We performed the resection en bloc of the aneurysmatic neoformation.

Conclusion: The management of splenic cavernous angioma is determined by hemodynamic. The simultaneous presence of a pancreatic cystic neoplasm makes this case very rare. Our aim is to report on this particular individual case and help to understand therapeutic management.

Keyword: Cystadenoma; Distal Pancreato-splenectomy; Pancreatic cystic neoplasm; Splenic hemangioma; Splenectomy

Introduction

Cavernous hemangioma, also called cavernous angioma, venous malformation, or cavernoma, is a type of venous malformation due to endothelial dysmorphogenesis of a lesion present at birth. Despite its designation as a hemangioma, a cavernous hemangioma is not a tumor, as it does not show endothelial hyperplasia. The abnormal tissue causes a slowing of blood flow through the cavities, or "caverns". Blood vessels do not form the necessary junctions with surrounding cells, and the structural support from the smooth muscle is hindered, causing leakage into the surrounding tissue. It is blood leakage, known as hemorrhage, that causes a variety of symptoms known to be associated with the condition. Splenic hemangiomas are the most common benign splenic lesions and are usually relatively small (< 4 cm). Hemangiomas are often cavernous, but may be capillary. Most splenic hemangiomas are discovered incidentally, but have been associated with spontaneous rupture. Splenic cavernous hemangioma is rare, with less than 100 cases reported in the literature [1]. They are often asymptomatic and their diagnosis is mostly occasional, but in some cases they can be found during investigations of generic abdominal pain, compression, or bleeding [2]. Spontaneous rupture has been reported to occur in up to 25% of these patients, especially when the tumor diameter is greater than 4 cm, requiring splenectomy as the only modality of treatment [3].

Pancreatic Cystic Neoplasms (PCNs) are liquid-containing tumors that can grow to considerable size and most commonly affect women. PCNs are estimated to occur in 2-45% of the general population. PCNs represent a clinically challenging entity, as their biological behavior varies from benign lesions to malignant disease [4,5]. The most common subtypes of PCN are serous, mucinous, Intraductal Papillary-Mucinous Neoplasms (IPMN) and cystadenocarcinomas, but their classification is wider and more complicated [5]. They can be asymptomatic or show symptoms such as compression or pancreatitis. Depending on the case and, moreover, on the histology of the PCN, follow-up or resection may be useful. Mucinous cystic neoplasms should undergo surgery if they are > 40 mm or if they are symptomatic or have risk factors regardless of their size. Otherwise, if they are smaller, asymptomatic, or do not have risk factors, follow-up should be performed due to their potential for malignancy. Serous cystic neoplasms are always a benign entity and in the literature cases of malignant transformation are not reported, so they should undergo follow-up and be resected only in case of symptoms of compression of other abdominal organs. IPMNs should be investigated because their treatment depends on their dimensions and radiological subtype [5].

Case Presentation

A 46-year-old woman came to our emergency department with diffuse abdominal pain. A total abdomen ultrasound was performed, which reported the presence of a suspected fissured aneurysm of the splenic vessels. We followed up with an abdominal CT scan without contrast that confirmed what the ultrasound showed (Figure 1).

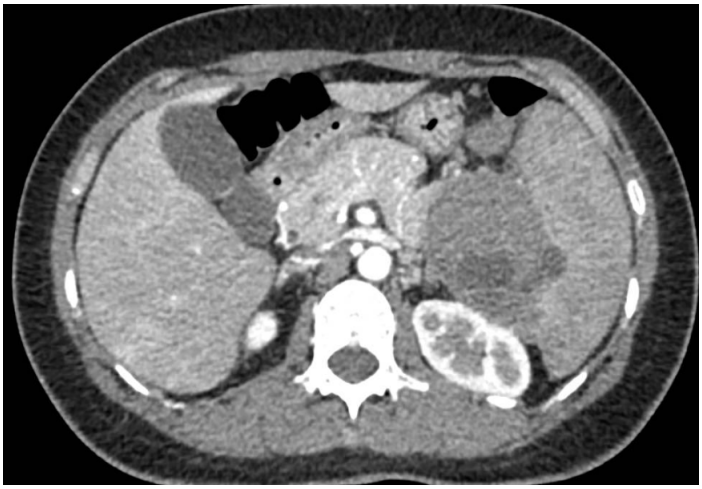

Figure 1: Abdominal CT.

Due to the patient's unstable clinical condition, we decided to perform an emergency laparotomy. We performed a left subcostal laparotomy. At the opening of the peritoneum, an aneurysm of the splenic artery was found of around 10 cm with active bleeding. The aneurysm was closely in continuity with the pancreatic tail and the splenic hilum and very difficult to separate from the pancreas. After the ligation of the splenic vessel, we performed a resection in bloc of the aneurysmatic neoformation and the pancreatic tail with ultracision. A drainage was left in the splenic lodge due to the inflammatory status found. The clinical condition of the patient was stable on the following days, but on Postoperative Day 7 (POD), due to purulent liquid present in the drainage, an increase in White Blood Cells (WBC) and C-Reactive Protein (CRP), we performed a new abdominal CT scan that reported the presence in the splenic lodge of a voluminous fluid collection with gaseous nuclei (Figure 2), compatible with a pancreatic fistula, resolved spontaneously 8 days later.

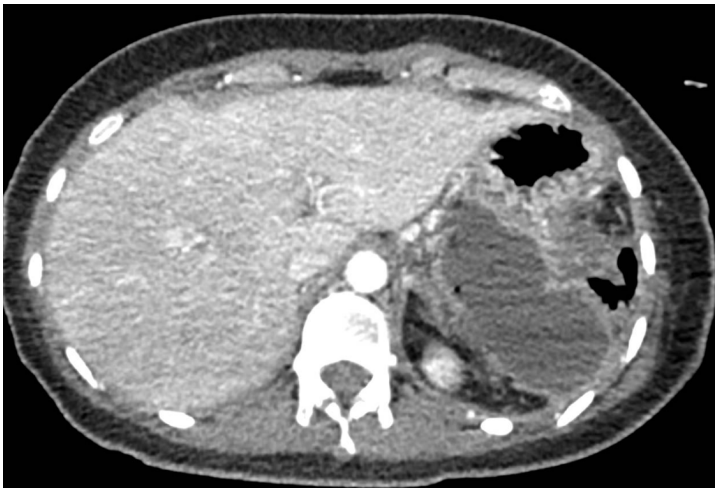

Figure 2: CT abdomen showing collection in pancreatic lodge.

Discussion

Fourteen days after the operation we received a report of histological examination which diagnosed cavernous angioma of the splenic vessels and, in addition, a borderline cystic mucinous neoplasm of the pancreas. We performed a contrast-exposed abdominal CT scan that showed no other lesions or secondarisms. The patient was discharged 24 days after surgery. An abdominal CT scan and tumor markers were repeated after discharge from the patient and were negative. The diagnosis of a PCN should always be followed by a thorough evaluation of the patient to discover indirect signs of malignancy, except in the cases of a serous cystic neoplasm. This subtype of tumor has never shown signs of malignant transformation in the literature, and its diagnosis requires only the patient's follow-up. However, we must remember that the presence of tumor-related symptoms provides a direct indication of surgery, independently of the subtype of PCN. In the other subtypes of PCN, surgery is indicated not only in the presence of tumor-related symptoms, but also in the case of dimensions > 40 mm or in the presence of risk factors (such as a mural nodule or a worrying annual increase rate). However, in this case, the unstable condition of the patient did not allow an accurate assessment of the mass and a discussion of the case during a multidisciplinary team (MDT) session. Even if not planned before, the surgeon's decision not to spare the pancreatic tail, due to its tenacious adhesion to the cavernous hemangioma of the splenic tail, has been the most appropriate. In this way, was faster and safer for the patient. However, the bleeding of the mass made the histological demarcation between the splenic cavernous hemangioma and the pancreatic mucinous neoplasm very difficult; therefore, we do not know the exact dimension of the mucinous mass (which deprives us of the dimensional resection criteria). However, bleeding itself of the mass (even if the origin of bleeding has not been defined) even more justifies aggressive surgical management. Furthermore, a posteriori, the histological classification of the mucinous neoplasm as 'borderline' would have led to another surgery if the first intervention had been incomplete.

Conclusions

The rupture of cavernous angiomas is a dangerous event that, if not properly treated can lead to death. The management of cavernous angioma rupture, even if it is a rare pathological condition, consists of an emergency laparotomy, especially if the patient's conditions are unstable. In some pediatric cases, an arterial angioembolization may be used, only if clinical conditions are stable, to avoid surgical stress and reduce hospitalization [6]. Sometimes bleeding can come from tumors, which in an ordinary situation would require diagnostic investigations and, if possible, complete surgical resection. Our aim is to report on this particular individual case and help understand therapeutic management.

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