

Splenik Arter Psödoanevrizmasının Senkop ile Kendini Gösteren Rüptür Sonrasında Rekürrensi: Transarteriyel Embolizasyon ile Tedavi

Re-Emergence of A Splenic Artery Pseudoaneurysm Following Rupture Presenting as Syncope: Management with Transarterial Embolization

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ÖZ

Splenik arter psödoanevrizması nadir ancak potansiyel olarak ölümcül bir bozukluktur. Transarteriyel embolizasyon bu bozukluğun tedavisinde tercih edilen yöntem haline gelmiştir, ancak psödokistlerin de içerisinde yer aldığı duvarlı pankreatik sıvı birikimlerinin varlığı bu tedavi yöntemini zorlaştırabilir. Bu yazıda, splenik arter psödoanevrizmasının duvarlı bir pankreatik nekrotik sıvı birikimi içerisine yırtılmasından dolayı senkop şeklinde bulgu veren 40 yaşında kadın olguyu sunuyoruz. Yırtılmayı takiben daha büyük boyutta yeniden ortaya çıkan psödoanevrizma, başarısız bir girişimin arkasından endovasküler yöntemle dolaşımdan uzaklaştırılmıştır.

Anahtar Kelimeler: splenik arter, psödoanevrizma, psödokist, embolizasyon

ABSTRACT

Splenic artery pseudoaneurysms are rare but potentially lethal disorder. Transarterial embolization has become the preferred mode of therapy for the disorder, however, accompanying walled-off pancreatic fluid collections including pseudocysts can make the management more challenging. Herein, we present the case of a 40-year-old woman presenting with syncope due to the rupture of a splenic artery pseudoaneurysm into a walled-off pancreatic necrotic collection. The pseudoaneurysm that re-emerged at a larger size following the rupture was endovascularly excluded from circulation after a failed attempt.

Keywords: splenic artery, pseudoaneurysm, pseudocyst, embolization

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INTRODUCTION

There is considerable diversity in the presentation of splenic artery pseudoaneurysms (SAPAs), with incidental findings and acute hemodynamic collapse representing the two ends of the spectrum (1). Hemorrhage is the most common finding in patients with SAPA, and the site and amount of bleeding are the main factors leading to the different clinical presentations (1, 2). Pancreatic pseudocysts, which coexist in 41% of cases, are among the sites with potential for bleeding, but not the most common (1-7). A rupture into a pseudocyst may lead to either a larger pseudoaneurysm or bleeding that can eventually exteriorize into the gastrointestinal tract or intra-/retroperitoneal space (1, 7). While transarterial embolization (TAE) has become the treatment option of choice for SAPAs, particularly in patients at high surgical risk, its suitability for the management of ruptured pseudocyst-associated SAPAs remains unclear (1-4, 6).

Herein, we present the case of a 40-year-old woman managed endovascularly for a SAPA that re-emerged in a larger size following a rupture into a pseudocyst, and discuss the suitability of TAE for such cases.

CASE REPORT

A 40-year-old Middle-Eastern woman presented with an 8-month history of epigastric pain radiating to her back. An abdominal examination revealed distention with tenderness over the epigastrium. She had no history of chronic disease, and never drank alcohol. A laboratory blood test showed slightly decreased levels of the main serum electrolytes, as well as albumin, total protein and hemoglobin. Serum amylase, lipase, ALT and AST levels were normal. The levels of pancreatic amylase and CRP were moderately elevated. Depending on the abdominal CT scan that had been obtained in another center, the patient was admitted to the hospital with the diagnosis of chronic pancreatitis-associated pseudocyst, and appropriate management was initiated.

On the 12th day of inpatient care, the patient suffered a collapse while visiting the bathroom, and was found unconscious with signs of having vomited blood. Her arterial blood pressure was 80/50 mmHg,

and pulse was 118 bpm. There was a drop in hemoglobin level to 4.6 gr/dL. An abdominal CT scan performed immediately after the event revealed clotted blood within a walled-off necrotic collection, 118×136×157 mm in size, replacing the pancreas, aside from the tail part, and an irregular focal narrowing in the middle third segment of the SA (Figure 1a-b).



Figure 1 (a, b). Contrast-enhanced abdominal CT, coronal multiplanar reconstruction (a) and curved maximum-intensity projection (b). Clotted blood within the collection (star in a) and an irregular focal narrowing of the splenic artery (arrow in b) are shown.

Re-evaluating the previous CT scan, a pseudoaneurysm measuring 4 mm in the same segment was noted (Figure 2a), and the narrowing mentioned above was attributed to a vasospasm associated with hemorrhage. An upper endoscopy performed the next day showed that the stomach had filled with blood, although the source of bleeding could not be located. On the third day of bleeding, she was admitted to the angiography suite. A selective coeliac arteriography was performed, showing a faint filling of the SA, however, the pseudoaneurysm couldn't be recognized. Superselective catheterization of the SA using different diagnostic catheters was not possible. The patient was discharged to the ward, and an endoscopic cystogastrostomy was performed the following day. On the 12th day of bleeding, the patient underwent a control CT scan and was identified with a recurrent pseudoaneurysm measuring 10×15 mm (Figure 2b).

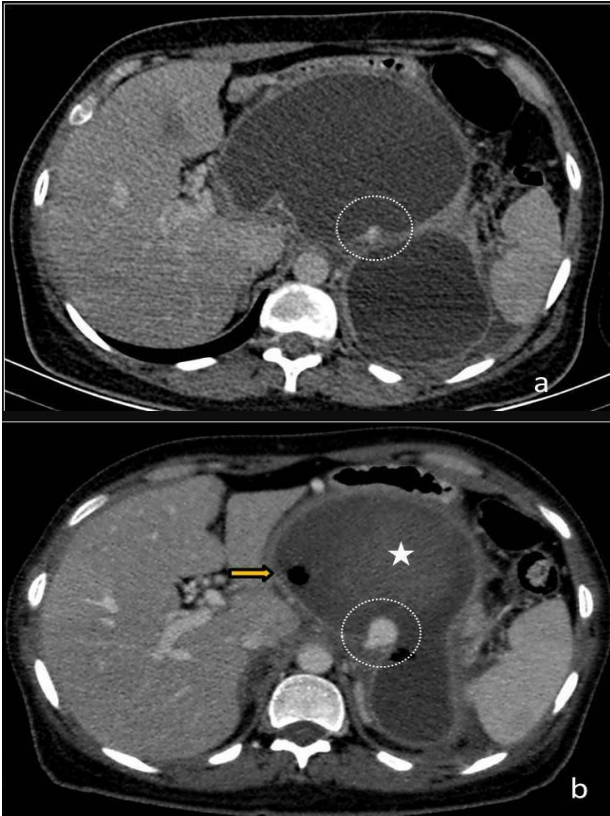


Figure 2 (a, b). Contrast-enhanced abdominal CT, axial slices. Appearance of the pseudoaneurysm (dotted circles) before (a) and after (b) the rupture. Note hematoma (star in b) inside the collection. Note also air bubbles (arrow in b) permeated into the lesion via the cystogastrostomy catheter.

She was readmitted to the angiography suite. A selective coeliac arteriography revealed an imperceptible filling of the SA. At this time, superselective catheterization of the SA was achieved using a microcatheter. At first, a filling coil was uneventfully deployed within the normal portion of the SA, distal to the pseudoaneurysm (Figure 3a), then a second coil was introduced immediately next to the first one. Unfortunately, this resulted in a rapid distal migration of the first coil (Figure 3b). The deployment of the second coil was carried out mainly within the proximal portion of the artery, while a short section of the coil was deployed within the pseudoaneurysm sac. Subsequently, the microcatheter was withdrawn proximal to the stack of coil, and a total volume of 1 mL 1:3 cyanoacrylate glue/iodized oil mixture was injected (Figure 3c). The final angiogram demonstrated total occlusion of the SA from orifice to the segment harboring the pseudoaneurysm. Despite filling of a short distal segment of the SA via the collateral circulation, there

was no flow into the pseudoaneurysm sac (Figure 3d).

With continuous supportive care, the patient showed gradual improvement in her general condition. A control CT scan revealed the complete exclusion of the pseudoaneurysm with a downsizing of the collection. Albeit diminished, splenic blood flow was maintained. Eventually, the patient was discharged from the hospital 38 days after admittance.

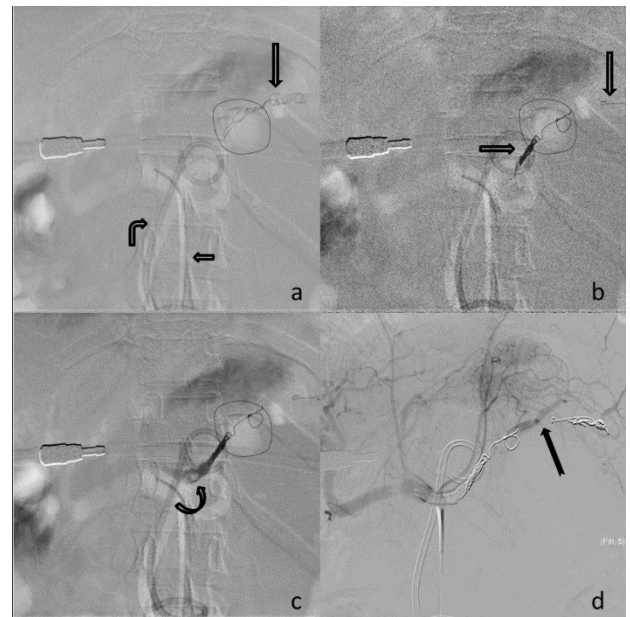


Figure 3 (a-d). Digital subtraction angiography. Primary position (a) and distal migration (b) of the first coil (vertical arrows in a and b) following the deployment of the second coil (horizontal arrow in b). Glue cast (curved arrow) within the proximal part of the splenic artery (c). Complete occlusion of the pseudoaneurysm and partial filling of the splenic artery (oblique arrow) by collateral vessels (d). Note that the pseudoaneurysm is marked with a dotted circle in a, b and c. Note also the double pigtail cystogastrostomy catheter superposed on the vertebral column (curved arrow in a) and Cobra-shaped angiocatheter (horizontal arrow in a) within the coeliac trunk.

DISCUSSION

The management of SAPAs and true splenic artery aneurysms has long been discussed under the same headings in most studies, although etiological factors, histopathologic features, prevalence as well as gender predominance differ (1, 2, 8). There is no debate on the point that a rapidly enlarging or ruptured aneurysm requires treatment, whether true or false (8, 9). That said, debates on the management of asymptomatic, silent aneurysms are ongoing. Several authors on this controversial topic have

suggested that the mechanism behind the formation of the aneurysm is not a determining factor in the decision making process (9). Albeit less common, some such aneurysms are false, and their natural history may not be well-delineated, leading a significant number of authors to recommend the repair of all SAPAs, regardless of size and symptoms (1, 3, 8, 10). We strongly advocate this view, having witnessed the devastating rupture of a SAPA that was overlooked due to its small size. Given the reported mortality rate of bleeding from a pseudoaneurysm or pseudocyst exceeding 10% (4), it is very fortunate that the patient survived. It would be that the coexistence of a pseudocyst that induces extreme fragility of the pseudoaneurysm wall attributed to proteolytic enzymatic digestion, as well as direct compression leading ischemia (4), is an expediting factor in the rupture of SAPAs, whether or not they are small or silent.

In addition to the rising mortality rate, it has been suggested that pseudocysts reduce the success rate of endovascular interventions, being approximately 85% (1-3), and for this reason, the suitability of TAE for the patients with ruptured pseudocyst-associated SAPA is another source of dispute (2, 9). In several reports and in many centers, TAE is not regarded as the first choice treatment for ruptured SAPAs (8). Moreover, TAE is not advised as the rupture may not be limited to the borders of a pseudocyst (6). Given that 58% of the patients with bleeding are hemodynamically unstable at presentation, the time to complete endovascular ablation is considered an obstacle to this modality in such cases (1, 6, 9). In contrast, although TAE carries a relatively increased risk of failure, as mentioned previously, some authors consider it to be an effective and safe approach to such disorders in its control of bleeding, enabling interventionists to improve hemodynamic status and decrease the risk of an urgent surgical intervention (1-4). In the event of pseudoaneurysm rupture into the gastrointestinal tract, peritoneal cavity or retroperitoneal space, which emerges more often secondary to bleeding into the pseudocyst lumen (7), surgery comes with an increased perioperative complication rate due to hemodynamic instability, or may be complicated, since inflammatory changes including adhesions can prevent the location of the source of bleeding (2, 3).

Logistical facilities in this respect and experience-based management preferences are decisive when choosing the method of treatment. Given that the primary goal of the therapeutic approach is to ensure hemostasis as soon as possible, especially in hemodynamically unstable patients, TAE is still considered the optimum approach due to its minimally invasive nature, the fact that it doesn't require general anesthesia and its facilitation of repeated interventions with a shorter hospital stay. The case presented here stands as a good example of this, since the patient showed gradual improvement in her general condition following the procedure, without supplementary surgical treatment.

Concerning the embolization technique, there is general consensus that TAE should exclude the neck of the pseudoaneurysm from the circulation by occluding the normal portions of the SA both distal and proximal to the pseudoaneurysm, rather than solely packing the sac (3, 4). Our management plan was to achieve this goal, however, anchoring the first coil in the desired position was not possible. Although not initially planned, we were compelled to use glue in the absence of any suitable coil left at that moment to complete the embolization. The use of glue allowed the embolization procedure to be completed a probable delayed migration was prevented (4). This experience showed us the importance of having the capacity to manage complications through other embolization techniques.

CONCLUSION

All patients with walled-off peripancreatic fluid collection (including pseudocysts) should be followed up radiologically so as not to miss the formation of a pseudoaneurysm in the peripancreatic arteries. Irrespective of the underlying cause, size or symptoms, all SAPAs should be repaired. Although the existence of pseudocyst has been blamed for reducing the success rates, TAE may be attempted prior to surgery even in hemodynamically unstable patients with ruptured pseudocyst-associated SAPA, as its morbidity is lower and allows repeated interventions.

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