Disappearance of the spleen as a rare complication of infected pancreatic pseudocyst following acute relapsing phase of chronic pancreatitis.

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ABSTRACT Splenic involvement of an infected pseudocyst is known to be a rare complication of infected pancreatic necrosis following pancreatitis. We present herein a case of chronic alcoholic pancreatitis complicated by a major infected pseudocyst formation involving the spleen, with subsequent rupture and complete disappearance of the entire spleen. A 60-year-old Japanese man with a history of chronic alcoholic pancreatitis with a pancreatic tail pseudocyst was referred to us because of severe epigastralgia. CT revealed spontaneous rupture of the pseudocyst into the stomach and free air in the pseudocyst. The body and tail of the pancreas were disrupted due to pancreatic necrosis, and the splenic parenchyma was also partially ruptured and disrupted with large tears of the splenic surface. One month later, CT showed almost complete disruption of the splenic parenchyma. Three months later, CT showed remarkable regression of the pseudocyst and complete disappearance of the entire spleen. Although splenic involvement of pancreatic pseudocysts should be considered to be a potentially lethal complication because of hemorrhage or infection, it can possibly be treated conservatively. In the present case, complete obstruction of both the splenic artery and vein due to chronic inflammation, and spontaneous drainage into the stomach were the most probable causes for the spontaneous regression without surgical treatment. This is the first report of splenic disappearance after splenic rupture demonstrated by CT.

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INTRODUCTION

Splenic involvement of an infected pseudocyst is an uncommon complication of infected pancreatic necrosis following acute or chronic pancreatitis, and may include intrasplenic pseudocyst, abscess, hemorrhage, infarction, splenic rupture, and

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vascular injury. Because these complications can be potentially life-threatening, the course of the disease should be closely monitored by helical computed tomography (CT) or ultrasonography (US) to determine when surgical intervention is necessary [1]. We present herein a case of chronic alcoholic pancreatitis complicated by a huge infected pseudocyst formation involving the spleen, with subsequent rupture and disappearance of the spleen. This is the first report of splenic disappearance after splenic rupture demonstrated by CT.

CASE REPORT

A 60-year-old Japanese man with seven year history of chronic alcoholic pancreatitis with a pancreatic tail pseudocyst was referred to us because of severe epigastralgia. Multiple pseudocysts in the tail of the pancreas were demonstrated on contrast-enhanced (CE) CT 5 years before (Fig.1). His arterial blood pressure was 188/100 mmHg, his white blood cell count was 18100/mm^3 and serum amylase was 168 IU/L (normal range, 40–118 IU/L). He was diagnosed as being in an acute relapsing phase of chronic pancreatitis. After admission, emergency CT revealed an enlarged pseudocyst in the tail of the pancreas which directly extended into the splenic parenchyma (Fig.2). It was recommended to undergo surgical treatment. However, he rejected invasive treatments such as surgery or transabdominal drainage for some

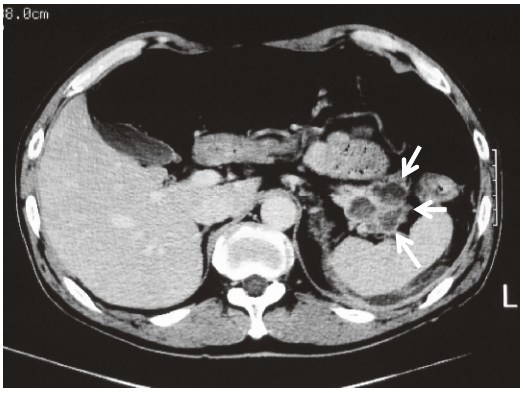

Fig. 1. Five years before, multiple pseudocysts in the tail of the pancreas were demonstrated on CE-CT (arrows).

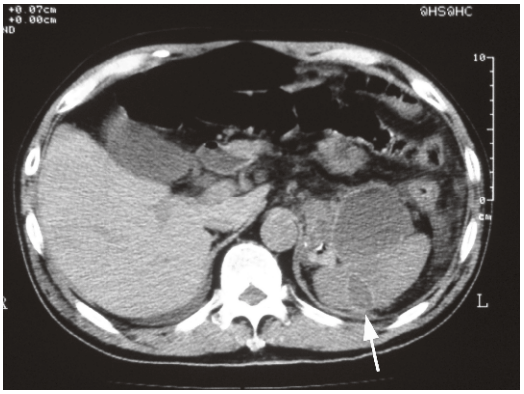

Fig. 2. On admission, emergency CT revealed an enlarged pseudocyst in the tail of the pancreas which directly extended into the splenic parenchyma (arrow).

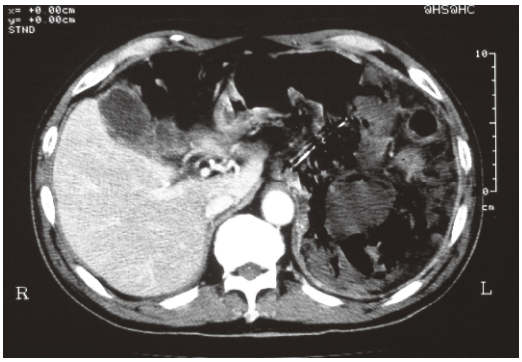

Fig. 3a. After 10 days, CE-CT showed spontaneous rupture of the pseudocyst into the stomach and free air in the pseudocyst.

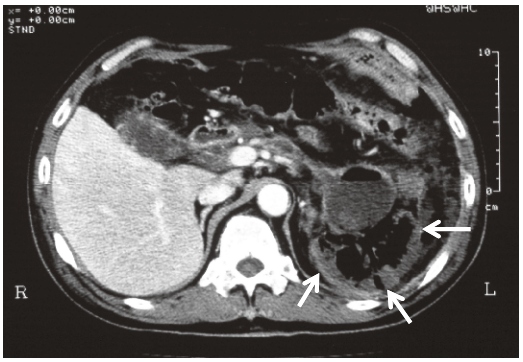

Fig. 3b. At a different level, the body and tail of the pancreas were disrupted due to pancreatic necrosis, and the splenic parenchyma was also partially ruptured and disrupted with large tears of the splenic surface (arrows).

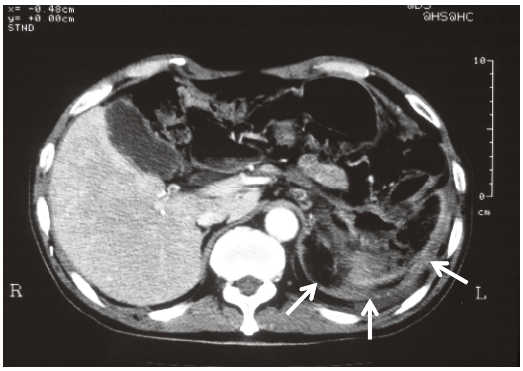

Fig. 4. One month later, CE-CT showed almost complete disruption of the splenic parenchyma (arrows).

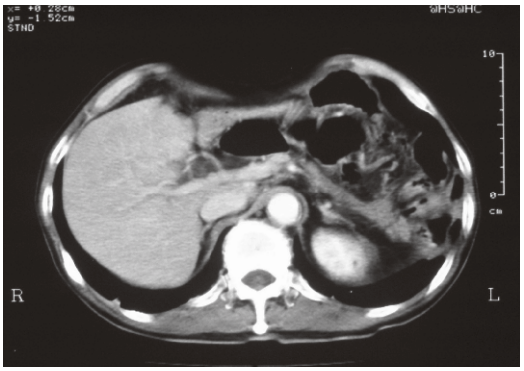

Fig. 5. Three months later, CE-CT showed remarkable regression of the pseudocyst and disappearance of the spleen.

reasons, although we informed the high mortality rate in a conservative treatment for that splenic complication. We considered endoscopic drainage or percutaneous drainage as an alternative treatment. In 2001, the endoscopic drainage was not popular and endoscopists in our institution did not have techniques to perform endoscopic drainage at that time. Also, percutaneous drainage could not be performed, because it could not avoid splenic injury. Fortunately, he was in stable condition and did not show any symptoms related to hemorrhagic complications. Medical treatment with gabexate mesilate and prophylactic antibiotics was continued and his symptoms gradually improved. Ten days later, CE-CT showed spontaneous rupture of the pseudocyst into the stomach and free air in the pseudocyst (Fig.3a). The body and tail of the pancreas were disrupted due to pancreatic necrosis, and the splenic parenchyma was also partially ruptured and disrupted with large tears of the splenic surface (Fig.3b). However, he still showed no hemorrhagic complications. One month later, CE-CT showed almost complete disruption of the splenic parenchyma (Fig.4). Three months later, CE-CT showed remarkable regression of the pseudocyst and complete disappearance of the entire spleen (Fig.5). Four months later, he returned to his daily activities. More than three years after the event, the patient is alive and free from symptom.

DISCUSSION

The incidence of splenic complications in patients with pancreatic pseudocysts has been reported to be 1 - 6%, and include intrasplenic pseudocyst, subcapsular hematoma, and splenic rupture[1-4]. A high complication rate of 79% and a high mortality rate of 8% have been reported and these high rates are an indication of the difficulties and complexities in treatment.

Splenic rupture is one of the most serious complications and usually develops as a consequence of intrasplenic pseudocysts. Intrasplenic pseudocyst is the most often complication in the spleen and a number of cases have been reported[1,5,6]. The incidence of intrasplenic pseudocysts in patients with chronic pancreatitis with distal pseudocysts has been reported to be 6 %[3]. Ueda et al reviewed 27 cases of intrasplenic pseudocyst of which 6 cases (22.2%) developed complications including 2 cases of intracystic hemorrhage, 2 cases of splenic rupture (7.4%), and 2 cases of infection[6]. Another review noted that 15 of 42 cases (35.7%) of pancreatic pseudocyst involving the spleen developed hemorrhagic complications including 7 cases of intracystic hematoma, 6 cases of subcapsular hematoma, and 2 ruptures of the spleen (4.8%)[7].

Liquefaction of splenic infarcts, proteolytic effects of pancreatic enzymes on splenic vessels or

parenchyma may present alternative mechanisms of development of intrasplenic pseudocysts[3]. A fistulous or inflammatory connection between the pancreatic tail and the spleen may also cause splenic complications. In addition, four pathogenetic mechanisms of splenic rupture associated with pancreatic pseudocysts have been suggested; (a) encasement of splenic vessels, causing infarction; (b) intrasplenic dissection of pancreatic enzymes, such as elastase and trypsin; (c) damage to small intrasplenic vessels causing intrasplenic or subcapsular hematoma; (d) If the hematoma is large enough, laceration, capsular disruption, or actual rupture of the spleen may occur[4,8]. Moreover, the presented case did not show any hemorrhagic complications including hemoperitoneum, hematemesis, or bleeding pseudocyst. This specific condition strongly suggests complete obstruction of both the splenic artery and vein due to extensive inflammation and compression around the vessels.

The clinical consequences of involvement of the spleen and its vasculature include splenic vein thrombosis, arterial pseudoaneurysms, splenic infarction, splenic rupture, intra-abdominal bleeding, and death[9]. Splenic infarction resulting from obstruction of the splenic artery is a rare complication of pancreatitis. The probable causes of arterial constriction and obstruction have been reported to be pancreatic fibrosis around the artery, tortuous aneurysm, splenic vein thrombosis, compression by edematous pancreatitis, and chemical inflammation leading to arterial thrombosis[10,11]. The splenic artery courses along the pancreas within the anterior pararenal space and it is highly susceptible to the effects of pancreatic inflammation.

Bernades et al reported the findings of splenoportal venous obstruction (SPVO) in 13.2 % of chronic pancreatitis cases[12]. Acute pancreatitis and pseudocysts were considered the probable causes of SPVO in 91.4 % of those cases and SPVO was found in 24.0 % of patients with previous acute pancreatitis and pseudocyst. The pathogenetic hypothesis of SPVO relates it to many factors: (a) venous constriction by fibrosis; (b) intimal injury and venous thrombosis; (c) extension of thrombosis[13,14].

CT and/or US are necessary in making a diagnosis of splenic complications and in looking for other risk factors including necrosis, abscess, bleeding in the cyst, hemoperitoneum, and splenic vein thrombosis. CT is the primary radiographic method of diagnosis. Earlier detection of complicated pseudocysts may contribute to a decreased number of patients presenting with acute life-threatening hemorrhage[4]. Care must be taken in dealing with high-risk pseudocysts involving the splenic parenchyma.

The application of surgical treatment for splenic complications of pancreatic pseudocysts is controversial. Usually, in the case of the absence of spontaneous regression or expansion of the pseudocysts, a distal pancreatectomy with a splenectomy is recommended. Some investigators have suggested that surgery is mandatory and the only definitive treatment[2,4,8]. Percutaneous drainage has also been reported to be for intrasplenic pseudocysts[15]. However, recently the principles of treatment for pancreatic pseudocysts have become more conservative[16,17]. Rypens et al reported on 12 of 16 patients with splenic complications who were conservatively and successfully managed[18]. They suggested that most splenic parenchymal complications of pancreatitis regress spontaneously and might be managed conservatively. In the present case, we did not perform surgery, because the patient rejected surgery and did not develop any serious complications such as hemoperitoneum or gastrointestinal bleeding. Conservatively, the huge pancreatic pseudocyst regressed after the rupture and disappearance of the spleen.

Pancreatic pseudocyst drainage by endoscopy

has been employed with increasing frequency. In a retrospective study, surgical or endoscopic management resulted in similar success rates of 50% and 52%, respectively, after an average period of 33 months follow up[19]. However, the majority of these patients had simple, uncomplicated pseudocysts. Recently, endoscopic treatment of more complicated pancreatic pseudocysts, including ductal disruption[20], pancreatic necrosis, and pancreatic pseudoaneurysms[21], has been reported. They utilized endoscopic transenteric drainage (endoscopic cysto-gastrostomy), endoscopic transpapillary drainage and stenting[22]. In the present case, we could not perform endoscopic drainage, unfortunately. However, if the endoscopic drainage for the pseudocyst had been performed earlier, disappearance of the spleen might not be happened.

In conclusion, we reported a case of complete disappearance of the entire spleen after rupture due to involvement of a pancreatic pseudocyst which was treated without surgery. Although splenic involvement of pancreatic pseudocysts should be considered to be a potentially lethal complication because of hemorrhage or infection, it can possibly be treated conservatively. In the present case, complete obstruction of both the splenic artery and vein due to chronic inflammation, and spontaneous drainage into the stomach were considered the most probable causes for spontaneous regression without surgical treatment. However, if early treatments could have been done, those complications might not be happened.

DISCLOSURE STATEMENT

None of the authors have any conflicts of interest to declare. This work was not funded by any educational or commercial organization.

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