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Brachiocephalic to left brachial vein thrombotic vasculitis accompanying mediastinal pancreatic fistula: A case report

brachiocephalic vein thrombotic vasculitis due to pancreatic fistula

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Abstract**BACKGROUND**

Pancreatitis is a severe inflammatory pancreatic disease commonly due to bile duct stones or excessive alcohol usage, with clinical manifestations of abdominal pain, nausea, fever, and fluid collections. Healthy persons with less symptomatic pancreatitis are quite rare. Herein, we report a case of a patient with an undetermined onset of pancreatitis mimicking left arm cellulitis due to thrombotic vasculitis of the brachiocephalic vein.

CASE SUMMARY

A 50-year-old woman visited our hospital for tenderness in the left arm over several recent days. She was diagnosed with cellulitis on the left arm due to left elbow tenderness. Intravenous antibiotics administration did not improve symptoms and laboratory data worsened; thus, chest and abdominal computed tomography (CT) was performed. CT demonstrated pancreatitis with pseudocyst around the pancreas extending to the mediastinum. Thrombotic vasculitis of the brachiocephalic to left brachial vein was observed, which could be the cause of left elbow pain. A pancreatic

fistula was found in the head of the pancreas by endoscopic retrograde cholangiopancreatography, so a pancreatic cyst drainage tube *via* the duodenum was placed in the pseudocyst. Cyst content culture was positive for *Escherichia coli* infection. Clinical symptoms, imaging findings, and inflammatory reactions resolved gradually after starting therapeutic intervention. The mediastinal pancreatic pseudocysts shrunk, and the venous thrombi remained but shrunk.

CONCLUSION

The case of a patient with pancreatitis with an undetermined onset that mimics left arm cellulitis is reported. Deep vein thrombosis should be kept in mind when treating patients with severe inflammatory disease.

Key Words: case report; pancreatic fistula; thrombotic vasculitis

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Core Tip: Pancreatitis is a severe inflammatory disease commonly caused by bile duct stones or alcohol usage, with clinical manifestations of abdominal pain, nausea, fever, fluid collections, *etc.* Because of its severity, it is rare to experience patients with less symptomatic pancreatitis. Herein, a 50-year-old Japanese woman was referred to our hospital due to a history of cough and fatigue for 2 mo and recent left elbow tenderness. CT showed pancreatic pseudocysts in adjacent tissues and mediastinum, with thrombotic vasculitis in the brachiocephalic to left brachial vein. A history of pancreatitis might cause systemic coagulopathy, which may result in brachiocephalic vein thrombosis.

INTRODUCTION

Pancreatitis is a severe inflammatory pancreatic disease commonly due to bile duct stones or alcohol usage, with clinical manifestations of abdominal pain, nausea, fever, fluid collections, *etc.*^[1]. Acute or chronic pancreatitis is at risk of pancreatic fistula to adjacent tissues, and it might extend to the mediastinum through the esophageal hiatus or aortic hiatus, which is called mediastinal pancreatic pseudocyst (MPP)^[2,3]. The notifying signs of MPP include chest pain, dyspnea, and systemic inflammatory changes, and diagnostic imaging modalities like computed tomography (CT) or magnetic resonance imaging (MRI) have an enormous role for its definitive diagnosis^[4,5]. Rare types of signs of MPP-like pancreatic panniculitis^[6], multiple ascending aortic mural thrombi^[7], or symmetrical peripheral gangrene^[8] were also reported; however, no case of MPP accompanying brachiocephalic thrombotic vasculitis has been reported. Herein, we present of a case of a patient with an undetermined onset of pancreatitis that resulted in MPP with brachiocephalic vein thrombotic vasculitis mimicking left arm cellulitis.

CASE PRESENTATION

Chief complaints

A 50-year-old Japanese woman with left elbow tenderness for several days was referred to the department of dermatology of our hospital.

History of present illness

She suffered from cough at night and exertional dyspnea for more than six months, bilateral lower extremity edema for two months, and fatigue, which worsened in recent months. Left elbow tenderness was also noted, so she was referred to the department of dermatology of our hospital.

History of past illness

The patient's medical history included epilepsy.

1

Personal and family history

The patient had no family history of similar illnesses.

Physical examination

2

Her body temperature was 38.9 °C, with a heart rate of 150/min, blood pressure of 120/85 mmHg, respiratory rate of 20/min, and SpO₂ of 98% at room air. Tender, erythematous, and edematous left arm was observed (Figure 1).

Laboratory examinations

The following results were observed at the initial examination: white blood cell count, 14,600/uL (segmented, 84.9%); red blood cell count, 322×10^4 /uL; hemoglobin level, 10.6 g/dL; platelet count, 53.1×10^4 /uL; total protein level, 4.7 g/dL; albumin level, 1.8 g/dL; total bilirubin level, 0.35 mg/dL; aspartate aminotransferase, 16U/L; alanine aminotransferase, 10 U/L; γ -glutamyl transpeptidase, 52 IU/L; lactate dehydrogenase, 269 IU/L; alkaline phosphatase, 352 IU/L; urea nitrogen, 8.8 mg/dL; creatinine, 0.49 mg/dL; creatine kinase, 26 U/L; uremic acid, 4.2 mg/dL; Na, 133 mmol/L; K, 3.6 mmol/L; Cl, 102 mmol/L; c-reaction protein, 22.0 mg/dL; and D-dimer, 12.7 μ g/mL.

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Imaging examinations

The CT demonstrated pancreatitis with pseudocysts around the pancreas (Figure 2) extending to the mediastinum (Figure 3). Thrombotic vasculitis of the brachiocephalic to left brachial vein was also observed (Figure 4), which was considered to be the cause of elbow pain. MPP was not seen around the brachiocephalic vein; however, it was compressed between the sternal bone and brachial artery (Figure 5). A pancreatic fistula was found in the head of the pancreas by endoscopic retrograde cholangiopancreatography (ERCP), so a pancreatic cyst drainage tube *via* the duodenum was placed in the pseudocyst (Figure 6).

FINAL DIAGNOSIS

During the next CT scan, the amylase concentration was 1108 U/L; therefore, a final diagnosis of brachiocephalic to left brachial vein thrombotic vasculitis with pancreatic pseudocysts in adjacent tissues of the pancreas and mediastinum was made.

TREATMENT

The pseudocyst around the head of the pancreas was drained with the tube inserted by ERCP. The culture of the cyst contents was positive for *E. coli* infection. MRI of MPP did not show any infection; thus, the patient was followed up with conservative treatment with antibiotics. MPP was followed up with conservative treatment with antibiotics for 32 days along with intravenous administration of heparin with 12000 U/day for 25 days and octreotide acetate for 24 days. The activated partial thromboplastin time was maintained at 38–42 s.

OUTCOME AND FOLLOW-UP

The clinical symptoms, imaging findings, and inflammatory reactions of acute pancreatitis were resolved, the MPPs shrunk, and the venous thrombi remained but shrunk. Table 1 summarizes the clinical course of the patient.

DISCUSSION

This is the first case report presenting a patient with brachiocephalic vein accompanying MPPS. MPPs are rarely found as a complication of acute pancreatitis, as first described by Clauss *et al*^[9]. Case reports about MPPs have been accumulated in recent years; however, to the best of our knowledge, there was no report on patients with a chief complaint of left arm tenderness derived from brachiocephalic to brachial vein thrombotic vasculitis. We could find a case report of a patient with ³multiple ascending aortic mural thrombi and acute necrotizing mediastinitis secondary to acute pancreatitis^[7]. The patient had acute pancreatitis with extended inflammation to the mediastinum, resulting in aortitis with thrombi. In contrast, in our case, the MPPs did

not reach the brachiocephalic vein, so a direct inflammatory infiltration from the MPPs cannot be assumed.

There were case series reports of patients with splanchnic vein thrombosis and acute pancreatitis^[10-12]. It was reported that the coagulopathy might have an effect on forming thrombi. In addition, the compression by MPPs to adjacent veins resulted in flow deficiency and coagulation^[13,14]. The possible causes of thrombosis in the brachiocephalic vein, in this case, were as follows: brachiocephalic vein stenosis due to compression of the brachiocephalic vein between the sternal body and aortic branches, resulting in systemic coagulopathy due to MPPs for an undetermined period, and edematous changes in the mediastinal tissues. Because the onset of pancreatitis in this patient is unknown, it is not clear whether systemic inflammation affected coagulopathy. Based on the presence of coagulopathy due to systemic inflammation, anatomical stenosis would be an additional cause of thrombotic vasculitis.

Panniculitis is a presenting type of pancreatic lesions derived from pancreatic enzyme migration into the systemic circulation that occur in systemic cutaneous inflammation^[15]. In patients with subcutaneous inflammatory changes and pancreatitis, panniculitis should be considered; however, because panniculitis usually occurs systemically, our case was suspected to have cellulitis on the left upper arm. The original symptoms of our case were associated with brachiocephalic to left brachial vein thrombosis, so the diagnosis of our case cannot be panniculitis derived from pancreatic enzyme leakage. The D-dimer level of the patient was elevated to 12.7 $\mu\text{g/mL}$. Thus, an evaluation for deep vein thrombosis might be considered, as the patient suffered from severe inflammatory disease with elevated D-dimer count.

In this case, the MPP was detected because the dermatologist who examined the patient considered the inflammatory change to be relatively worse than the severity of the left arm cellulitis. This is an instructive case demonstrating that it is essential to perform a workup study in patients with suspicious or undetermined unexplainable data.

CONCLUSION

Herein, we report a case of a patient with pancreatitis with an undetermined onset, respiratory syndrome for a few months, and thrombotic vasculitis in the brachiocephalic vein mimicking left arm cellulitis. Long-term pancreatitis might cause systemic coagulopathy that resulted in brachiocephalic vein thrombosis. Deep vein thrombosis should be kept in mind when treating patients with severe inflammatory disease.

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