SPONTANEOUS ISOLATED CELIAC AND SUPERIOR MESENTERIC ARTERY DISSECTION: CASE REPORT WITH LITERATURE REVIEW

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ABSTRACT

Introduction: Spontaneous isolated celiac and superior mesenteric artery dissection is rare, but it might have been underestimated due to the possibly occult presenting symptoms and good outcomes. The natural history is unpredictable due to its large variations of the clinical features.

Case presentation: A 50-year-old patient had complained of abdominal pain. Abdominal computed tomography revealed spontaneous isolated celiac and superior mesenteric artery dissection. He was conservatively managed with nitroglycerin infusion, atropine injection and warfarin intake and his pain was relieved soon.

Conclusions: There have been no established therapeutic algorithms for the management of the spontaneous isolated splanchnic artery dissection. Conservative treatment is preserved for most patients with stable hemodynamics. In the condition of unstable hemodynamics, progression of dissection and signs of splanchnic ischemia, a surgical, interventional, or hybrid treatment is warranted.

Key words: celiac artery; dissection; superior mesenteric artery.

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Introduction

Spontaneous isolated splanchnic artery dissection is uncommon with superior mesenteric artery (SMA) being the most commonly dissection-affected artery. Spontaneous celiac artery dissection is rare, but underestimation is anticipated due to variable clinical manifestations. Only 11% of celiac artery dissections are associated with SMA dissections. Spontaneous isolated splanchnic artery dissection has been sporadically but increasingly reported, however, the clinical features and management strategies were different case by case. In order to obtain an optimal therapeutic effect, a consensus in decision-making of the dissection management is necessary.

This article reports a particular case of spontaneous isolated celiac and SMA dissection, outstanding the importance of conservative treatment in the patients with stable hemodynamics and good responses to the therapy.

Case presentation

While doing push-ups, a 50-year-old male developed acute upper abdominal pain, which was dull and persistent. His pain did not relieve or aggregate overnight and he thus referred to our hospital the next morning. He was significant for hypertension in the past 4 years with a maximal blood pressure of 170/110 mmHg with no regular intake of hypotensive agents.
Physical examination revealed no peritoneal signs. An urgent abdominal computed tomography (CT) revealed celiac and SMA dissection with no obvious dilation of the dissected arteries (Figure 1). The patient had leukocytosis and elevated C-reactive protein. The laboratory findings on admission were shown in Table 1.

He was conservatively managed with nitroglycerin infusion (5 mg/50 ml, i.v. through syringe pump at 3 ml/h), atropine injection (1 mg, s.c., once daily) and warfarin intake (2.5 mg, once daily). He was hemodynamically stable with blood pressure within normal ranges and symptoms were relieved soon. Two days later, blood tests turned to be normal. He was discharged shortly. He was advised regular intake of hypotensive agents and blood pressure monitoring.

Discussion

Literature review

Spontaneous isolated celiac and SMA dissection is extremely rare. By extensive literature search, only 15 such patients were reported from 9 reports\(^1\)\(^-\)\(^9\). One of them was a repetitious publication\(^9\). As a result, there were totally 15 patients including the present patient. Their age was 55.7 ± 9.6 (range, 38-66; median, 62) years (n = 9). Six patients were males and one patient was female with a significant male predominance. All five patients whose presentation symptoms were given were abdominal pains. The diagnosis was established based on CT in 6 (85.7%) patients and on abdominal ultrasonography in 1 (14.3%) patient. The imaging presentations of the dissections were a false lumen in 4 (66.7%) patients, one of whom had a false lumen dilation, intimal laceration in 4 (66.7%) patients, one or two intimal flaps in 3 (50%) patients and thrombus formation in the false lumen in 2 (33.3%) patients. Treatment strategies were described for 5 (33.3%) patients, 4 (80%) patients were conservatively and 1 (20%) patient was interventionally treated. All patients were symptom-free and doing well during follow-up.

Mechanisms

The mechanisms of spontaneous splanchnic artery dissection remain unknown\(^1\)\(^0\). However, inflammatory responses in the vascular wall initiated by the immune mediators with subsequent intimal hyperplasia and eventual luminal narrowing and occlusion were considered the underlying etiologies\(^1\)\(^1\). The predisposing risk factors of spontaneous splanchnic artery dissection may be iatrogenic, or secondary to atherosclerosis, trauma, pregnancy, fibromuscular dysplasia, inflammatory or infectious diseases, or congenital vasculopathies\(^1\)\(^,\)\(^2\)\(^,\)\(^1\)\(^2\). Medial necrosis or degeneration of the arterial wall\(^1\)\(^,\)\(^1\)\(^2\), fibromuscular dysplasia\(^1\)\(^,\)\(^1\)\(^3\) vasculitides including giant-cell arteritis, Takayasu arteritis and polyarteritis nodosa\(^1\)\(^\text{h}\) and connective tissue disorders, such as Ehlers-Danlos syndrome\(^2\)\(^,\)\(^1\)\(^\text{i}\), were

<table>
<thead>
<tr>
<th>Item</th>
<th>Result</th>
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<tr>
<td>Leukocyte (×10^9/L)</td>
<td>10.28</td>
<td>3.5-9.5</td>
</tr>
<tr>
<td>Neutrophil (%)</td>
<td>84.1</td>
<td>40-75</td>
</tr>
<tr>
<td>lymphocyte (%)</td>
<td>10.92</td>
<td>20-50</td>
</tr>
<tr>
<td>C-reactive protein (mg/L)</td>
<td>25.93</td>
<td>0-8</td>
</tr>
<tr>
<td>Amylase (U/L)</td>
<td>84</td>
<td>30-110</td>
</tr>
<tr>
<td>Lipase (U/L)</td>
<td>120</td>
<td>23-300</td>
</tr>
<tr>
<td>Troponin I (µg/L)</td>
<td>0</td>
<td>0-0.06</td>
</tr>
<tr>
<td>pro-brain natriuretic peptide (pg/mL)</td>
<td>80</td>
<td>0-900</td>
</tr>
</tbody>
</table>

Table 1: Laboratory findings on admission.
also reported to be the underlying causes.

**Clinical presentation**

More than half (66%) of the patients were asymptomatic at presentation\(^{(2)}\). Of the symptomatic patients, 69% had pains only and 15% had pains, nausea and vomiting. Moreover, the presenting symptoms due to the dissection was in only 23% of the symptomatic patients\(^{(2)}\). Yun et al.\(^{(3)}\) stated that the severity of abdominal pain correlated positively with the length of the dissection, i.e., the more the extent of inflammation, the severer the irritation to the nerves. The risk of bowel ischemia associated with celiac artery dissection is less than that associated with SMA dissection\(^{(16)}\). In addition, 53% of the patients had a history of hypertension\(^{(17)}\).

**Diagnosis**

Work-up may show mild elevation of C-reactive protein. An elevated C-reactive protein of 6.6 mg/L and negative anti-neutrophil cytoplasmic antibodies (ANCA) were reported by Kang et al.\(^{(3)}\), indicating the possibility of a non-specific vasculitis. Seidita et al.\(^{(17)}\) reported a patient with isolated SMA dissection had an elevated C-reactive protein and leukocytosis as in the present patient. Slightly elevated serum amylase can be noted in the patients with occlusion of duodeno-pancreatic arcades\(^{(18)}\).

Before the era of aortography and CT, celiac artery dissection was incidentally found at autopsy\(^{(18)}\). Nowadays, abdominal ultrasonography is the first choice of diagnostic modality, it can demonstrate the hemodynamic changes within the SMA, bowel movement and signs of bowel ischemia\(^{(17)}\), but it might poorly display the SMA with a thrombus inside the false lumen\(^{(19)}\). Digital subtraction angiography was the gold standard for the diagnosis of splanchic artery dissection, and a contrast agent can display lumen filling. However, a false lumen might be overlooked when lumen filling failed. Magnetic resonance imaging is helpful in the diagnosis of vascular dissection, but slower scanning speed, low spatial resolution and poor sensitivity of displaying the mesenteric vascular lesions have limited its use\(^{(20)}\). Helical CT permits the diagnosis of acute aortic dissection with high sensitivity and specificity and with the advantages of non-trauma, high speed and high spatial and temporal resolutions\(^{(21)}\).

In comparison with digital subtraction angiography, CT angiography can display the mesenterium and intestinal structures more comprehensively in etiological diagnosis and display of the extent of the dissection\(^{(22)}\).

CT presentation of the dissections may display a linear or a curved dissection flap, the relative positions between the true and false lumens, the location of the entry and reentry and its distance from the arterial origin, thrombus formation in the false lumen, patency or occlusion of the true lumen, aneurysmal dilation, branch involvement, fuzzy fat space around the dissected artery and signs of intestinal ischemia or necrosis\(^{(20,23)}\). Solis et al.\(^{(24)}\) found that most of the intimal lacerations were located about 1.5-3.0 cm from the origin of the SMA, just a transitional zone of the fixed and free portions of the artery and therefore the intima was susceptible to shear stress.

Isolated SMA dissection should be differentiated from acute SMA thrombosis, non-occlusive mesenteric ischemia, mesenteric vein thrombosis and acute aortic dissection\(^{(19)}\). Angiography and CTA can display the specific of these lesions including filling defect in the SMA, bowel wall thickening and pneumatosis, venous occlusion or intraluminal thrombosis, and narrowed true lumen with aneurysmal dilated false lumen\(^{(17)}\).

**Treatment**

The favorable effect of conservative treatment in such patients has been emphasized\(^{(3)}\). Stable hemodynamic patients with vague abdominal pain, free of peritoneal signs are indicated for anticoagulation\(^{(5)}\). Takayama et al.\(^{(3)}\) suggested that medical treatments include antihypertensive drugs, anti-inflammatory drugs, steroids (in case of vasculitis) and anticoagulation to prevent thromboembolic complications\(^{(1,23)}\), where anticoagulant or antiplatelet agents for 3-6 months with a target International Normalized Ratio of 2.0-3.0 with strict blood pressure control are recommended\(^{(5,18)}\). Surgical treatment of the dissection is to prevent acute complications, such as progressive aneurysmal dilatation\(^{(18)}\), aneurysm rupture, intestinal ischemia, or chronic arterial stenosis\(^{(16,26)}\). Surgery and endovascular procedures may be considered in the condition of unstable hemodynamics, persistent abdominal pain, failure of blood pressure control by medical therapy and dissection progression\(^{(18)}\).

Glehen et al.\(^{(26)}\) recommended surgical repair in celiac artery dissection in patients with occlusive lesions, aneurysm formation, arterial rupture, or extension of the celiac artery dissection into the hepatic arteries. Open repair include thrombectomy,
aneurysmorrhaphy, intemectomy with patch angio-
plasty, ligation, resection, venous graft bypass and
arterial bypass graft(3). Endovascular bare stent
deployment may be helpful in an arterial dissection
to cover the entry points(29). A hybrid laparotomic
retrograde ileal and endovascular right femoral
realignment approach was successfully applied in a
48-year-old male with SMA dissection in whom
sole endovascular approach failed due to the total
obstruction of the true lumen caused by severely
dilated false lumen(29). In the present patient, con-
servative treatment is opted as for his stable hydrom-
dynamics, good response to the treatment, quick sym-
tom relief and free of dissection progressions.

Clinical outcome

Treatment was required in 18% of the patients, with
a near-even distribution of anticoagulation, stenting and surgery(28). Most of the patients who did not undergo an intervention at presentation and no progression of dissection were observed(28). The success rates were 55.4% (31/56) for expectant ther-
apy, 65.2% (15/23) for anticoagulation, 100% (22/22) for open surgery and 80% (4/5) for endovascular stent placement(3,4). Endovascular management of celiac artery dissection has been infrequently reported, however endovascular stent
placement or fenestration has been reported in spontaneous SMA dissection(4). Endovascular treat-
ment is an attractive option in patients with high surgical risks(20). Patients with endovascular stent
placement had a significantly greater reduction in the diameter of the true lumen. Gobble et al.(4)
reviewed 15 cases of SMA dissections treated with endovascular stent placement and found all patients had pain resolution at a mean follow-up of 11.4 months. However, Kim et al.(30) reported recurrence of the aneurysmal dilated false lumen in a 54-year-
old man 4 months after endovascular stent place-
ment. In addition, bowel resection is required with progressive symptoms of bowel ischemia(4).

Conclusions

Spontaneous isolated celiac and SMA dissec-
tion is rare. The clinical features include male predomina-
ce, hypertensive background, and acute onset of abdominal pain. CT angiography is a non-
innvasive and accurate diagnostic method. Conservative treatment is preserved for stable hemodynamic patients. In patients with unstable hemodynamics, progression of dissection and signs
of splanchnic ischemia, a surgical, interventional, or hybrid treatment is warranted.

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