A Case of Inflammatory Solitary Internal Iliac Artery Aneurysm Successfully Treated by an Endovascular Procedure

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An inflammatory aneurysm localized in the iliac artery is very rare. Aneurysms of the common iliac artery have been reported sporadically, but there has been no report of internal iliac artery aneurysm. The patient was a 64-year-old male presenting with left lower abdominal pain and fever. Abdominal computed tomography (CT) revealed an aneurysm of $42 \times 52$ mm in the left internal iliac artery. The aneurysmal wall had thickened and mantle sign was positive. Since the aneurysm was expected to adhere tightly to surrounding tissues, the risk of open surgery was judged to be high, and coil embolization of the aneurysm and the peripheral branches of the internal iliac artery was performed. Postoperatively, the left lower abdominal pain disappeared, and signs of inflammation were mitigated. The endovascular procedure was extremely effective for the treatment of this inflammatory iliac artery aneurysm.

Key words: inflammatory aneurysm, solitary iliac artery aneurysm, endovascular surgery

INTRODUCTION

An inflammatory aneurysm localized in the iliac artery is rare. There have been sporadic reports of solitary inflammatory aneurysms of the common iliac artery, but none of the internal iliac artery. We encountered a case of a solitary inflammatory aneurysm of the internal iliac artery, and we successfully treated it by employing endovascular procedures.

CASE REPORT

The patient was a 64-year-old male with left lower abdominal pain that had gradually intensified, and a fever above 37°C. As computed tomography (CT) examination suggested the impending rupture of a left internal iliac artery aneurysm, the patient was referred to our department. His history included hypertension and smoking. On admission, the blood pressure was 112/66 mmHg, heart rate was regular at 68 bpm, body temperature was 37.2°C, and the abdomen was soft and flat, but tenderness was noted in the left lower abdominal region. The ankle brachial index (ABI) was 1.20 on the left and right, showing no abnormality. On blood tests, the white blood cell (WBC) was normal at 7,300, but c-reactive protein (CRP) was elevated at 10.4 mg/dl, and the erythrocyte sedimentation rate (ESR) at 60 minutes was increased at 97 mm. There was no sign of anemia, and no abnormality was noted in the liver or kidney function. Abdominal CT revealed no aneurysmal change in the abdominal aorta, common iliac artery, or external iliac artery, but an aneurysm of $42 \times 52$ mm was noted in the left internal iliac artery. The aneurysmal wall had thickened to 11 mm, contrast enhancement was observed in the late phase, and mantle sign was noted, facilitating a diagnosis of inflammatory solitary internal iliac artery aneurysm (Fig. 1).

Since marked adhesion was expected around the aneurysm, open surgery was considered difficult, and endovascular surgery was selected as a less invasive treatment. As the neck of the internal iliac artery is not wide, we
considered coil embolization. The inferior mesenteric artery and right internal iliac artery were patent, and it seemed that the blood supply to the sigmoid colon was maintained after occlusion of the left internal iliac artery. We placed a guiding catheter in the left internal iliac artery, by employing a contralateral approach via the right femoral artery, and placed 23 coils from two peripheral branches flowing into the internal iliac artery to the aneurysm by using a child catheter and microcatheter. Five of the 23 coils were used for branches and 18 were used for the aneurysm itself. Contrast enhancement of the internal iliac artery subsequently disappeared (Fig. 2). Postoperatively, left lower abdominal pain resolved, and CRP became negative. A postoperative CT scan revealed that the contrast enhancement of the internal iliac artery and mantle sign of aneurysmal wall had disappeared.

**DISCUSSION**

Inflammatory aneurysm as a disease concept was proposed by Walker et al. in 1972 as an abdominal aortic aneurysm with marked wall thickening and adhesion to surrounding organs, but its etiology remains unclear. It is considered to be a subtype of atherosclerotic aneurysm, and the involvement of inflammation following atherosclerosis, an autoimmune mechanism, and cytomegalovirus infection has been speculated. Symptoms such as abdominal and back pain are frequently observed. Abdominal CT is useful for the diagnosis; characteristic findings are homogeneous and dense staining of the aneurysmal wall and retroperitoneum around the aneurysm with a slight delay compared with the aortic lumen, which is known as the mantle sign. This type of aneurysm may also involve the ureter due to the extension of inflammation, and its obstruction may lead to hydronephrosis.

There have been few reports of solitary inflammatory iliac artery aneurysms, with only 6 known cases (Table 1). In all of these cases, aneurysms were located in the common iliac artery, and there has been no report, to our knowledge, of a solitary inflammatory aneurysm confined to the internal iliac artery. The seven patients with inflammatory solitary iliac artery aneurysms, including our patient, consisted of 6 males and 1 female aged 43–83 years, with a mean of 62.6 years. The aneurysms were between 2.5 and 10.0 cm, with a mean of 4.6 cm. Symptoms such as abdominal and lumbar pain were noted in all patients, and 5 had hydronephrosis. The six patients with common iliac artery aneurysms were treated with artificial vascular graft replacement, and only our patient with an internal iliac artery aneurysm underwent endovascular treatment, with a favorable outcome.
Rupture (1), an aneurysm size exceeding 5 cm including the mantle sign (2), and medial displacement and marked stenosis of the ureter (3) are considered to be surgical indications of inflammatory abdominal aortic aneurysm. However, as there are no clear surgical indications concerning inflammatory iliac artery aneurysm because of its rarity, we adopted those for non-inflammatory solitary iliac artery aneurysm. Many solitary iliac artery aneurysms are 3 cm or less in diameter, and the prognosis of a patient having a ruptured aneurysm is poor. Early surgery is thus recommended, particularly for saccular aneurysms. We judged our patient to have these surgical indications. Resection and artificial vascular graft replacement are basic treatments for inflammatory as well as non-inflammatory abdominal aortic aneurysms, but open surgery often causes hemorrhage and multiple organ damage during detachment of the aneurysm adhering to the surrounding structures. Stent graft insertion is a promising treatment that may overcome many problems associated with artificial vascular graft replacement. Surgical resection is also the optimal treatment for solitary internal iliac artery aneurysms, but exclusion of the aneurysm is regarded as an acceptable alternative if it is large and it tightly adheres to the surrounding tissues. However, there is a possibility of enlargement of the aneurysm after this treatment if its blood supply is maintained, so complete blockage of blood flow into the aneurysm is necessary. Endoaneurysmorrhaphy; carried out by incision of the aneurysm after proximal clamping, obstruction of branches from the aneurysmal lumen, and constrictive suturing of the aneurysmal wall; is also useful. Endovascular surgery is very useful when open surgical treatment seems to be impractical for adhesion to surrounding tissues including the ureter, difficulty of detachment of the aneurysm and ligation of distal branches and risk of multiple organ injury. Embolization can also be performed as an endovascular treatment. Coil embolization is usually carried out on distal and proximal sides of the aneurysm, but proximal embolization may be difficult depending on the morphology of the aneurysmal neck. For such lesions, intra-aneurysmal coil packing may be performed. Recently, as stent graft implantation is being applied more often, favorable results have been reported for stent graft placement following coil embolization of the peripheral branches of the internal iliac artery. Although we had prepared a stent graft for our patient, we could complete the operation without its use because the blood flow could be successfully blocked through coil embolization of the distal

<table>
<thead>
<tr>
<th>Age/Sex</th>
<th>Site of aneurysm</th>
<th>Mantle sign</th>
<th>Treatment</th>
<th>Hydronephrosis</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>83/F</td>
<td>L.c. iliac artery</td>
<td>+</td>
<td>tube graft</td>
<td>+</td>
<td>alive</td>
</tr>
<tr>
<td>80/M</td>
<td>R.c. iliac artery</td>
<td>+</td>
<td>tube graft</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>66/M</td>
<td>L.c. iliac artery</td>
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<td>tube graft</td>
<td>+</td>
<td>alive</td>
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<tr>
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<td>unknown</td>
<td>unknown</td>
<td>unknown</td>
</tr>
<tr>
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<td>Y graft, IIA reconstruction</td>
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<tr>
<td>43/M</td>
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<td>-</td>
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<td>-</td>
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</tr>
<tr>
<td>64/M</td>
<td>L.int. iliac artery</td>
<td>+</td>
<td>coil embolization</td>
<td>-</td>
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</tr>
</tbody>
</table>

Table 1 Summary of reported cases of inflammatory solitary iliac artery aneurysm
branches and intra-aneurysmal and proximal coil embolization. If the blood supply from connecting branches to an aneurysm is maintained, there is the possibility of enlargement of the aneurysm after coil embolization, and complete blocking of blood flow into the aneurysm is important. The artifact due to the coil is prominently visible in the CT scan, making it difficult to measure the correct size of aneurysm. Since there is the possibility of rupture or consumption coagulopathy due to the development of collateral circulation to the internal iliac artery aneurysm, careful follow up is necessary.

**CONCLUSION**

A patient with an inflammatory solitary internal iliac artery aneurysm could be successfully treated by employing endovascular procedures.

**REFERENCES**