Unusual Complications in an Inflammatory Abdominal Aortic Aneurysm

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An unusual case of an inflammatory abdominal aortic aneurysm (IAAA) associated with coronary aneurysms and pathological fracture of the adjacent lumbar vertebrae. The associated coronary lesions in cases of IAAA are usually occlusions. In the present case, it was concluded that a possible cause of the coronary aneurysm was coronary arteritis and the etiology of the pathological fracture of the lumbar vertebrae was occlusion of the lumbar penetrating arteries due to vasculitis resulting in aseptic necrosis. Inflammatory AAA can be associated with aneurysms in addition to occlusive disease in systemic arteries. The preoperative evaluation of systemic arterial lesions and the function of systemic organs is essential. (Jpn Circ J 1999; 63: 914–916)

Key Words: Coronary artery aneurysm; Inflammatory abdominal aortic aneurysm; Pathological fracture; Vertebrae

Inflammatory abdominal aortic aneurysm (IAAA) is characterized by peri-aneurysmal fibrosis with adhesions and encasement to adjacent structures. Concomitant arterial lesions are usually obstruction in the smaller diameter arteries and aneurysmal dilatation in the larger ones. The cause of IAAA is usually thought to be an immunological response to atherosclerosis. We present an unusual case of IAAA associated with coronary artery aneurysm (CAA) and pathological fracture of the adjacent lumbar vertebra.

Case Report

A 49-year-old man was admitted to hospital with the chief complaint of lumbago. He had suffered a sudden onset of lumbago when he picked up a heavy stone. He had a past history of habitual smoking, hyperlipidemia, and hypertension, but no history of Kawasaki disease, angina pectoris or connective tissue disease. Laboratory examination showed an elevated erythrocyte sedimentation rate (ESR) of 33 mm/h (57 mm per 2h) and negative blood culture. C-reactive protein was 0.27 mg/dl and the serological test for syphilis was negative. There was no evidence of malignancy or bone metabolic disorder. The electrocardiogram showed regular sinus rhythm, no abnormal Q wave and no ST–T change. Magnetic resonance imaging (MRI) showed a pathological compression fracture of the third lumbar vertebrae and aneurysmal dilatation of the adjacent abdominal aorta (Fig 1). Computerized tomography (CT) showed infrarenal AAA with a diameter of 50 mm and thick peri-aneurysmal fibrosis on the antero-left lateral wall (Fig 2). Coronary angiography showed an aneurysm in the left coronary-pulmonary artery fistula and another aneurysm in the midportion of the left circumflex artery (Fig 3).
At operation, there was no adhesion of the small intestine or ureters onto the aneurysmal wall. The infrarenal AAA, with a thick and hypervascular anterior wall of 8 mm in thickness, was incised and a small amount of mural thrombus on the posterior wall was removed. The lumbar arteries were occluded and the AAA and the bilateral common iliac arteries were replaced with a sealed, knitted Dacron bifurcated graft (18 × 9 mm).

Pathological findings of the resected aneurysmal wall showed the typical findings of IAAA, consisting of thick adventitia and inflammatory infiltrates in the media and adventitia (Fig 4). A specimen of the third lumbar vertebral body, resected at the same time as the AAA, showed aseptic necrosis and fibrosis. Histopathology of the anterior longitudinal ligament showed perivasculitis of the penetrating arteries (Fig 5). There was no evidence of malignancy or other inflammation. Gram stain and culture of the aneurysmal wall was negative.

The postoperative course was uneventful and the patient remained well without anginal pain under treatment with antiplatelet agents for 3 years after the operation. His ESR measured at 2 years and 3 years after operation was 17 mm/h (34 mm per 2 h) and 10 mm/h (25 mm per 2 h), respectively.

**Discussion**

IAAA is more commonly associated with aneurysmal and/or obstructive lesions in other arteries than is the standard atherosclerotic AAA. The usual sites of the associated arterial lesions are the thoracic aorta, the femoral and popliteal arteries for aneurysmal dilatations, and the coronary, femoropopliteal, aortoiliac, carotid, and renal arteries for occlusive disease. In all previous reports of IAAAs, the associated lesions of the coronary artery have been occlusive. It has been established by postmortem examination that some cases of IAAAs have shown pathological evidence of pericoronary arteritis manifesting as stenosis. In those reports, the pathological findings in the AAA were adventitial fibrosis with inflammatory infiltrates in the media, whereas the coronary artery had intimal fibrosis, in addition to the same findings shown in the AAA, which resulted in the stenotic lesions. Some authors suggest that IAAA are simply a variant manifestation of atherosclerotic change, and have no special significance. Schwartz and Mitchell suggested that inflammatory changes were...
involved in the pathogenesis rather than the etiology of atherosclerosis and were due to some change in the immunological tolerance to a component of the atheromatous plaque. However, IAAA often presents with systemic inflammatory reactions such as fever, weight loss, anemia, and elevated ESR in addition to local symptoms. Therefore, similar histopathology findings between the IAAA and coronary periarteritis could suggest systemic vasculitis. Coronary periarteritis has also been reported in idiopathic retroperitoneal fibrosis, which has identical clinicopathological findings to IAAA without aneurysmal dilatation of the abdominal aorta. A case with CAA and IAAA has not been reported previously and the CAA in the present case was not examined histopathologically; however, it is likely that the same inflammatory mechanisms of the AAA play an important role in the development of CAA. Fibrotic changes only in the adventitia with scant fibrous intimal proliferation plus predominant medial disruption by inflammatory infiltrates, as well as degeneration such as that seen in the abdominal aorta, are considered to enable aneurysmal dilatation of the coronary artery to develop.

The precise cause of the pathological fracture of the lumbar vertebrae is obscure. It usually occurs in metastatic tumors, infections or metabolic disorders, but the pathology and bone mineral analysis did not show any related evidence. A huge aneurysm can erode through the body of the vertebrae causing fracture, but in the present case the size of the AAA was not large enough to cause erosion. There are some reports of the relationship between arteritis and lesions of the spine. Systemic vasculitis associated with ankylosing spondylitis is reported to involve fibrous intimal proliferation of the artery in the lumbar vertebrae and spinal fractures following trivial trauma are well known. The pathological findings of the fractured vertebrae in the present case showed fibrotic degeneration due to aseptic necrosis, and perivasculitis resulted in occlusions of the artery in the anterior longitudinal ligament. In IAAA, the amount of posterior peri-aneurysmal fibrosis is smaller than that of the anterior wall so we speculate that the posterior extension and involvement of the peri-aneurysmal fibrosis by peri-aortic inflammation did not result in the pathological fracture of the adjacent lumbar vertebrae. Occasional occlusion of the lumbar arteries by endoarteritis has been reported in IAAA. The vertebral body receives its blood supply from the numerous branches of the internal and external plexus of the lumbar artery, which make terminal arteries. Evidence of perivasculitis in the anterior longitudinal ligament, which has foramina for arteries and veins passing to and from the vertebral body, and the presence of the intervertebral disk without a blood supply could suggest ischemic necrosis of the vertebral body due to occlusion of the feeding arteries, and this is considered to be the most reliable mechanism of the pathological fracture in the present case.

It has been reported that there is a high incidence of operative complications and increased operative mortality with IAAA! The cause of the unusual complications in the present case is suggested to be systemic vasculitis. Although patients with the clinical manifestations of so-called IAAA do not belong to only one clinical entity, IAAA can be complicated with various arterial lesions and organ malfunctions due to the systemic vasculitis. Preoperative evaluation of systemic arteries, including the function of vital organs, and close postoperative follow-up is mandatory in a patient with IAAA.

References