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Tubercular pseudoaneurysm of aorta: a rare association with vertebral tuberculosis

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Abstract

BACKGROUND CONTEXT: Pseudoaneurysm of the aorta in association with vertebral tuberculosis is a rare phenomenon. With the resurgence of human immunodeficiency virus (HIV) and associated resistant tuberculosis, this life-threatening complication requires greater awareness.

PURPOSE: Our purpose is to report the rare presentation and successful management of tubercular pseudoaneurysm of the aorta in association with vertebral tuberculosis, and to highlight the clinicoradiological features for early and prompt diagnosis of this potentially fatal, but treatable, disease.

STUDY DESIGN: A single case report and overview of the disease comprises the design of this study. **PATIENT SAMPLE:** The patient, already surgically intervened, is a 27-year-old male with increasing abdominal and back pain, upper motor neuron signs, and constitutional signs and symptoms.

OUTCOME MEASURES: At 33 months follow-up, there is complete resolution of the signs and symptoms, and the patient is back to his previous vocation.

METHODS: The diagnosis was confirmed by magnetic resonance imaging and contrast computed tomography. Endoaneurysmorrhaphy of the pseudoaneurysm along with a complete course of antitubercular treatment was given to the patient, and he has presently been followed up for 33 months.

RESULTS: The patient's signs and symptoms have been completely resolved without any recurrence.

CONCLUSION: Despite the use of modern chemotherapy and imaging techniques, this disastrous complication still occurs and reinforces the need for early suspicion, diagnosis, surgical resection, and antitubercular therapy along with close postoperative follow-up to prevent recurrence. With the resurgence of HIV (and other immunocompromised states) associated and resistant tuberculosis, we should be more alert than ever to this life-threatening complication. © 2007 Elsevier Inc. All rights reserved.

Keywords:

Tubercular pseudoaneurysm of aorta; Vertebral tuberculosis; Tubercular aortitis; Mycotic aneurysm of aorta

Introduction

Tubercular pseudoaneurysm is a potentially fatal, but rare, complication of tuberculosis. Out of 96 cases of tubercular aortic aneurysm described in English literature, only 2 had an aneurysm of the thoracoabdominal aorta [1,2] and 6

had pseudoaneurysm associated with vertebral tuberculosis [2–5].

We present a case of tubercular pseudoaneurysm of thoracoabdominal aorta in association with vertebral tuberculosis to highlight the clinicoradiological features for early and prompt diagnosis of this potentially fatal, but treatable, disease.

Case report

A 27-year-old male army personnel presented in June 2003 with anorexia, weight loss, low-grade fever, and increasing abdominal and back pain over the past month. This patient was being treated elsewhere for tuberculosis of dorsolumbar spine since September 2002. Drainage of a psoas

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abscess had been performed in November 2002, and the pus had shown presence of acid-fast bacilli. A costotransversectomy had been performed through the right eleventh rib in February 2003, but it did not relieve his symptoms. An endoscopy, which had been done in May 2003 for epigastric pain, had shown erosive gastritis and a gastric ulcer, resulting in the cessation of antitubercular treatment.

On examination, the patient had two healed surgical scar marks-one measuring 6 cm in length on the left iliac fossa, and the other on the right paraspinal region over the eleventh rib measuring 8 cm in length. Paraspinal tenderness was present from the mid dorsal to the upper lumbar spine with a mild kyphotic deformity. A slight fullness in the left paraspinal area was noted at the D12-L1 level. Neurologically, the patient showed increased tone in his lower limbs, brisk deep tendon reflexes, and upgoing plantars without gross motor or sensory loss and without bladder or bowel involvement. Hematological investigations were essentially within normal limits (including a negative human immunodeficiency virus status), except for a low hemoglobin level of 8 gm % (normal 12-18 gm %). X-rays and computed tomography (CT) of the dorsolumbar spine showed destruction of the D9-L1 vertebral bodies with scalloping of the anterior part of D7-L2 vertebral bodies (Fig. 1). Previously obtained magnetic resonance imaging films showed a low signal on T1-weighted imaging and a high signal on T2-weighted imaging in front of the D7-D11 vertebral bodies, suggestive of infective pathology (Fig. 2). However in front of D11-L1, a low signal lesion on T1-weighted imaging and T2-weighted imaging was observed suggestive of a vascular lesion. Hence a contrast CT was done which revealed a large (11.9 cm×5.1 cm) aortic pseudoaneurysm arising from the descending thoracic and proximal abdominal aorta with bilateral psoas abscesses (Fig. 3).

The patient was referred to a vascular surgeon and was operated on an emergency basis. Peroperatively, there was a large pseudoaneurysm occupying the whole upper abdomen displacing spleen and liver anteriorly and eroding D11-L1 vertebral bodies. Calcified walls were present extending into the bilateral paravertebral region. Endoaneurysmorrhaphy (consisting of proximal control of bleeding by clamping, entering the aneurysm, evacuation of blood clot, aspiration of blood, and oversewing the distal uninvolved segment from within the aneurysm, which was then obliterated by multiple layer suturing) of thoracoabdominal aneurysm was carried out at a specialized cardiovascular center, and a full course of antitubercular treatment was continued postoperatively for 9 months. Both the acid-fast stain and the culture were positive for Mycobacterium tuberculosis. The histopathological examination of the aneurysm wall revealed features of tuberculosis (Fig. 4). The patient made an uneventful recovery. The follow-up magnetic resonance imaging at 4 months showed resolution of the low-intensity area (Fig. 5). At 2 years postoperative, the patient had complete resolution of the signs and symptoms and was back to his previous vocation. At his last follow-up, 33 months postoperative, there were no clinicoradiological signs of disease activity.

Discussion

Despite the fact that tuberculosis is still prevalent in the developing countries, tubercular involvement of the aorta is still exceedingly rare. Thus, establishing an accurate

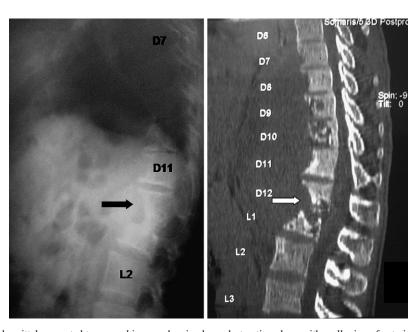


Fig. 1. X-ray lateral view and sagittal computed tomographic scan showing bone destruction along with scalloping of anterior vertebral bodies in dorsolumbar region (D7–L2).

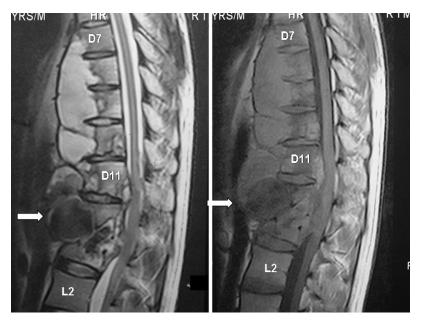


Fig. 2. Magnetic resonance imaging (sagittal sections, T2-weighted imaging and T1-weighted imaging) showing a signal void lesion in front of D11–L1 vertebral bodies in both films. Multiple outpouchings suggest breach of intima and hence the formation of a pseudoaneurysm.

diagnosis is essential, as approximately half of these cases can develop aneurysm formation and subsequent fatal perforation if untreated [6]. Before 1950, most of the reported cases were from autopsies [1,7,8]; in later years, antemortem diagnosis and successful treatment became more common with the availability of better diagnostic facilities and enhanced awareness [2,9,10].

Tubercle bacilli may reach the aortic wall in one of four ways [2]: 1) the bacilli may implant directly on the internal surface of the vessel wall in patients suffering from miliary tuberculosis; 2) the bacilli may be carried to the adventitia or media by the vasa vasorum; 3) infection may reach the

vessel wall by the lymphatics of the vasa vasorum; 4) the outside of the vessel wall may be affected by direct extension from a neighboring tuberculous lymph node, abscess, or bone. This has been reported as the most common cause (75%) in literature [2]. In our case too, contiguous focus from vertebral tuberculosis and a paraspinal abscess probably produced tubercular aortitis. The arterial system may be affected by miliary tuberculosis of the intima, tubercular polyps attached to the intima, tuberculosis involving several layers of the arterial wall, aneurysm formation, or even by hypersensitivity reaction to the tubercular antigens [2,9]. Tubercular aneurysms are mostly pseudoaneurysm and

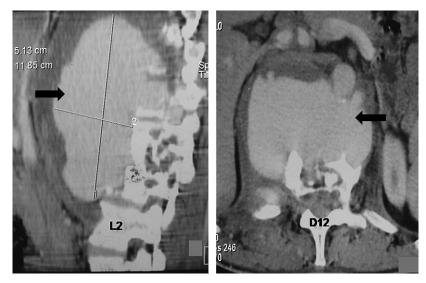


Fig. 3. Contrast computed tomographic scan (sagittal and transverse sections) showing an enhanced large pseudoaneurysm mass $(11.9 \times 5.1 \text{ cm})$ arising from the thoracoabdominal aorta at the level of D11–L1 vertebral bodies.

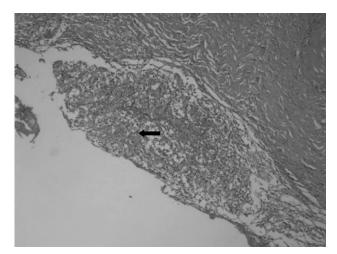


Fig. 4. Histopathological slide (hematoxylin-eosin stain ×40) showing multiple epitheloid cell granuloma consistent with tuberculosis in the aortic wall.

rarely true or dissecting [9]. The ensuing complications depend largely on the extent and the speed of the inflammatory process [10]. Caseating necrosis, involving the entire thickness of the aortic wall, results in perforation, either with massive hemorrhage or with the formation of a perivascular hematoma. The latter may become encapsulated and retain communication with the lumen, in which case it is referred to as a false aneurysm or pseudoaneurysm [10]. Most tuberculous aneurysms, whether true or false, involve the thoracic or the abdominal aorta; however, they may occur anywhere along the arterial system [9]. Thus far there are only three reports (including this article) of tubercular pseudoaneurysm arising from the thoracoabdominal aorta, but this is the first such case report of the lesion at the thoracoabdominal aorta in association with vertebral tuberculosis [2]. This rarity is perhaps a result of the low incidence of atheroma at this level, the small number of lymph nodes around this segment of the aorta, or the separation of the aorta from its neighboring structures by the diaphragm [2].

Clinically, tuberculosis of the aorta manifests only after the onset of its major complications [11]. One should have a high index of suspicion, and an aneurysm should be suspected in patients with active tuberculosis if they deteriorate suddenly or if a mass lesion is present [9]. The patient may present initially with persistent pain related to the location of the aneurysm. Other symptoms include fever, weight loss, hoarseness, dysphasia, palpable paraaortic mass (especially if pulsatile and expanding), hemoptysis, and hypovolemic shock or other evidence of massive bleeding, particularly into the lungs, gastrointestinal tract, peritoneal cavity, retroperitoneum, or even pericardial space. Short periods of "herald" bleeding often warn of subsequent exsanguinating hemorrhage. In the present case, the patient presented with acute abdominal pain and persistent back pain despite being treated for tuberculosis. In the past, most cases were diagnosed by chest radiography and

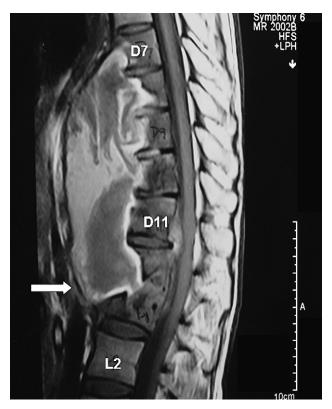


Fig. 5. Four months follow-up magnetic resonance imaging (T1-weighted imaging) showing resolution of signal void lesion seen in Fig. 2.

aortography. Over the last 20 years, contrast-enhanced computed tomography and magnetic resonance angiography have proved effective methods in the early detection of tuberculous aortic aneurysm [10]. The diagnosis should always be considered if the aneurysm is false, if it does not involve the ascending aorta, and if it is associated with a contiguous focus of disease on CT scan [2].

In our case, the patient was a bacteriologically proven case of the tubercular spine with psoas abscess. Usually with antitubercular therapy (ATT), such lesions heal and the patient continues to obtain relief of symptoms. However, this patient's symptoms remained unabated, and he continued to have low hemoglobin and increasing back and abdominal pain. The scalloping of vertebrae became more prominent, and this should have been a warning sign. Once a bacteriological diagnosis was available and the patient had no neural deficit, there was no justification for costotransversectomy. The patient had a pseudoaneurysmal mass in the left paraspinal region, and luckily the costotransversectomy was done from the right side and the pseudoaneurysm was not inadvertently entered. Had this procedure been performed from the left side, which is usually the case, then he might have collapsed on the operating table with rupture of the pseudoaneurysm. Even the magnetic resonance imaging findings had been missed previously. Furthermore, the endoscopic diagnosis of gastritis added to the confusion of diagnosis.

The management of tuberculous aortitis is both surgical and medical. There is no evidence that either of these alone will cure the disease [2,9,11]. All the survivors in literature have received both [9]. Antitubercular treatment must be instituted the moment tuberculosis is suspected [2]. It must be recognized, however, that although the presence of an aneurysm and its mycotic nature might be predicted preoperatively, the likelihood of its being tubercular might not be anticipated. Review of literature suggests that over onethird of these patients were not diagnosed at presentation [2]. Even at surgery, determining the tubercular nature of the lesion is problematic. Once symptomatic pseudoaneurysm is diagnosed, surgery should not be delayed. Size does not correlate to the need for surgery, as even a pseudoaneurysm of 1 cm in diameter can rupture [2]. Surgical options include resection and graft replacement, aneurysmorrhaphy, exclusion and bypass (extra-anatomic), and direct or patch closure. The extent of procedure depends on the size of the aortic rent and the condition of the neighboring aortic wall. Because of the clamp-related ischemia, the surgeon should rely on visual inspection of the aortic wall to decide on the extent of resection, rather than on time-consuming frozen section and histology reports [9]. However, all pseudoaneurysms should undergo histologic and microbial assessment with special consideration to Mycobacterium infection [9]. Perioperative and postoperative chemotherapy with standard antitubercular drugs will take care of the contiguous lesion and will prevent dissemination of the lesion in the arterial system as well. The surviving patients should be checked regularly for healing of the contiguous lesion in case of possible reactivation of the tuberculous process, which could occur near the site of the resection and result in an anastomic aneurysm or aortoenteric fistula or a fistula into another vessel [10].

Conclusion

Despite the use of modern chemotherapy and imaging techniques, this disastrous complication still occurs and reinforces the need for early suspicion, diagnosis, surgical resection, and antitubercular therapy along with close post-operative follow-up to prevent recurrence. With the resurgence of human immunodeficiency virus (and other immunocompromised states) associated and resistant tuberculosis, we should be more alert than ever to this life-threatening complication.

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