

Original Article

Intraoperative rupture of tuberculous pseudoaneurysm associated with spinal tuberculosis: A case report and literature reviewHaijun Tang^{1, #}, Yun Liu^{1, #}, Abu Moro¹, Zengming Xiao¹, Xinli Zhan¹¹ Department of Spine and Osteopathic Surgery, The First Affiliated Hospital of Guangxi Medical University, Nanning City, Guangxi Province, China

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Abstract

Introduction: The increase in the incidence of tuberculosis in developing countries has resulted in increased reporting on more related complications. We report the successful management of an intraoperative spontaneous rupture of a tuberculous pseudoaneurysm associated with spinal tuberculosis.

Methodology: We report the case of a 66 years old woman diagnosed with spinal tuberculosis of the T3/T4 vertebrae. During an anterior approach surgical reconstruction for the degenerative vertebrae, massive hemorrhage was observed after the removal of a portion of the prevertebral fascia of the affected vertebrae. This led to the exploration of the hilar fascia for the possibility of hilar hemorrhage, and when no hemorrhage was observed, aneurysm rupture was suspected. A portion of the hilar fascia was then sutured to the prevertebral fascia and the hemorrhage was partially controlled.

Results: On the 32nd month post-operative follow-up, the symptoms of chest and back pain had disappeared and the tuberculous lesion eradicated.

Conclusion: Presently, there are few reports on the successful treatment of intraoperative spontaneous rupture of tuberculous pseudoaneurysms. We therefore report on the successful management of such a case.

Key words: Spinal tuberculosis; tuberculous pseudo-aneurysm; hilar fascia; prevertebral fascia; hypothermia.*J Infect Dev Ctries* 2019; 13(2):174-178. doi:10.3855/jidc.10707

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Copyright © 2019 Tang *et al.* This is an open-access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.**Introduction**

Although the incidence of spinal tuberculosis is increasing in developing countries [1,2], thoracic aorta pseudoaneurysm associated with spinal tuberculosis is rare [3]. The consequence of the rupture of these pseudoaneurysms are often fatal. The success rate of a successful repair of a ruptured aneurysm is < 50% even among experienced cardiac surgeons [4,5], hence for spine surgeons who have little experience with aneurysm repair, they often do not know how to deal with such violent bleeding should there be a rupture during surgery [6].

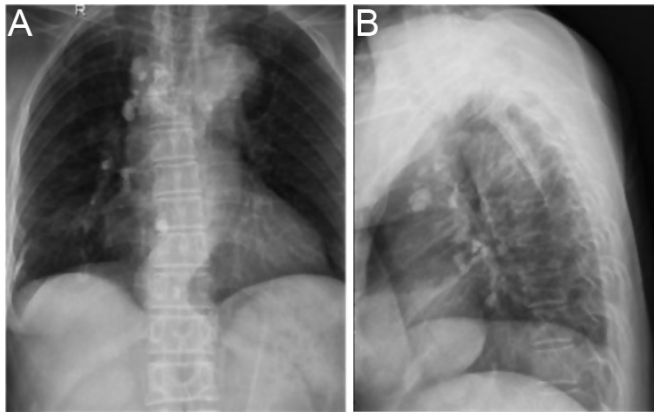
We therefore present a case on the successful intraoperative management of a ruptured tuberculous pseudoaneurysm of the aortic arch. The massive hemorrhage was successfully controlled, and hypothermia used to protect the brain and other vital organs of the patient. The patient survived and her prognosis is good without any complications 32 months after the operation.

Case report

A 66-year-old farmer, reported to our hospital with a history of chronic backache for 10 years. The pain was not intensified at night, and there was no prominent aggravating or alleviating factors. There were no accompanied fever, cough or expectoration symptoms. She was diagnosed with pulmonary tuberculosis at the local hospital where she was given the anti-TB drugs isoniazid + rifampicin + pyrazinamide + ethambutol for treatment. She reported that the pain gradually subsided and later disappeared after 12 months of therapy, although she also reported occasionally not taking her medications. One year later, the back pain resurfaced, accompanied with right arm pain and malaise, and the pain progressively radiating to the face and mouth. She therefore came to our hospital for consultation and treatment.

On admission in April 2015, her WBC was normal, ESR 42mm/hour, CRP 11.73 mg/L, Hb 132g/L,

Figure 1. Preoperative X-ray.



A: Preoperative thoracic X-ray AP view. B: Lateral view; showing T3/T4 vertebral damage consistent with spinal TB.

Albumin 37.5g/L, and serology for HIV, Hepatitis B and C, were all negative. Chest radiography showed multiple lung nodules, and kyphosis (Figure 1). Thoracic CT scan revealed T3 and T4 changes consistent with spinal tuberculosis (Figure 2). Physical examination revealed percussion tenderness of the T4-T5 spinous processes and paraspinous tissue. There were no abnormal neurological examination findings. The initial diagnosis was spinal tuberculosis.

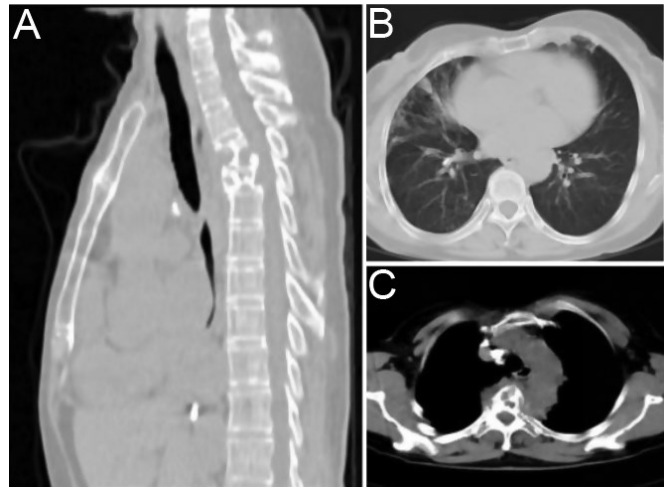
Four days after admission at 8:28 am, surgical treatment for the spinal TB via the right anterior approach was planned and performed. During the operation, 80 ml of abscess fluid was found adjacent to the T4/T5 vertebrae and drained successfully. However, while cleaning the prevertebral tissue with a pituitary rongeur, massive gush of blood was observed. At which point a possible damage to a pulmonary vessel was considered and a cardiac surgeon consulted. Upon exploring the hilum, no damaged vessels were

Figure 3. DSA of EVAR treatment.



A: Visible Contrast agent leakage before stent placement. B: Shows successful insertion of stent, without leakage of contrast agents.

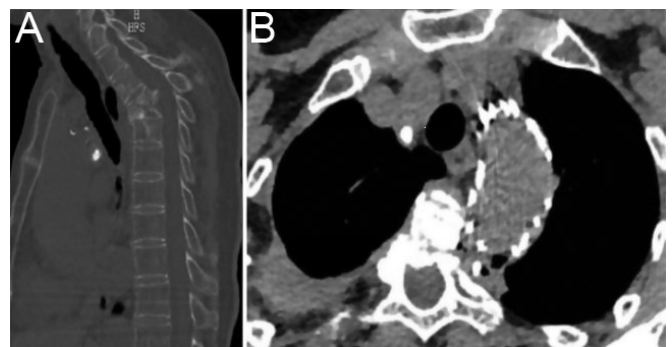
Figure 2. Preoperative CT.



A: Lateral view showing moth-eaten appearance of the T3, T4 vertebral bodies and sequestration around the vertebrae. B: increased lung markings. C: Paravertebral abscess indistinguishable from the thoracic aorta.

observed. It was at this point that a possible rupture of a pseudoaneurysm was suspected. At 10:30am, the patients’ vital signs suddenly dropped, with BP: 40/25mmHg, and HR:40 beats/min, soon after at 10:35am, the patient had a cardiac arrest due to hemorrhagic shock. The blood gas analysis at the time were pH = 7.146, pCO₂ = 44.7 mmHg, pO₂ = 176 mmHg, and Hb = 61g/L, the HR and pulse at this point could not be measured. Due to the shock and the worsening condition of the patient, it was impossible to perform direct suture closure, patch repair or endovascular aneurysm repair (EVAR) of the aneurysm. A portion of the hilar fascia was therefore sutured to the scraped prevertebral fascia and the hemorrhage partially controlled. At the same time, the anesthesiologist used hypothermia techniques to reduce

Figure 4. 32nd month Postoperative follow-up CT.



A and B: Improved spinal TB, with improved vertebral damage (vertebral bony has fused albeit limited kyphosis) and no aneurysm recurrence.

the body temperature to 31-33°C in order to protect vital organs. After the transfusion of 6 pints of blood and plasma, the patient BP improved (100/76 mmHg) and heart rate slightly recovered (50 beats/min), after being unable to be measured for 13 minutes. The patient was then transferred to the ICU after stabilizing the BP. At this point, the drainage volume from the pseudoaneurysm was around 450ml/h. An emergency angiography was therefore performed and a pseudoaneurysm leakage observed at the aortic arch. A thoracic aortic dissection with endovascular stent-graft was then performed by a vascular surgery specialist (Figure 3).

The patient was finally discharged after successful treatment of the pseudoaneurysm and the spinal TB treated with the anti-TB drugs isoniazid + rifampicin + pyrazinamide + ethambutol. The patient was also advised to avoid occasionally not taking the drugs, as she did previously, in order enhance complete recovery and prevent the development of drug resistance. Follow-up X-ray/CT were arranged for the 1st, 3rd, 6th, and 12th month postoperative respectively, after which followup was schedules once a year. On the 32nd month post-operative follow-up, the symptoms of chest and back pain had disappeared and the tuberculosis lesions was eradicated, without any obvious signs of relapse. The follow-up CT scan showed the tuberculous lesion and pseudoaneurysm were successfully treated (Figure 4).

Discussion

Tuberculous involvement of aortic vessels is a rare phenomenon, especially for spinal tuberculosis. Since the first report of tuberculous aneurysm by Kamen et al in 1895 [7], there have been 19 studies of spinal tuberculosis associated with tuberculous pseudoaneurysm involving 25 cases, among which 10 cases involved the thoracic vertebrae [3,5,7-23]. The underlying mechanism for the formation of tuberculous pseudoaneurysm is still unknown, but most academics believe that, it involves the infiltration of vascular walls by Mycobacterium via lymphatic or direct invasion through the paravertebral abscesses [3]. Also, since the majority of the available literature on tuberculous pseudoaneurysms are case reports, it's difficult to determine the possible risk factors associated with its development. Perhaps, more detailed research is warranted in order to understand this phenomenon.

Spinal tuberculosis is diagnosed by a combination of clinical manifestations as well as etiological confirmation of the presence of Tuberculous bacilli. Although X-rays have a 99% accuracy of diagnosing

spinal tuberculosis, it is difficult to assess the extent of vertebral damage, spinal cord compression, as well as abscess and pseudoaneurysm formations. The best diagnostic modality of tuberculous pseudoaneurysm are therefore CT/MRI scans [24]. CT scan can visualize spinal cord compression, paravertebral abscesses and the extent of vertebral damage, however, pseudoaneurysms are usually not clear since they are difficult to differentiate from paravertebral abscesses, which reduces it's diagnostic value [6,14]. MRIs produce better visualization of damages to surrounding soft tissues, spinal cord compression, and pseudoaneurysms, making it the better diagnostic modality [24]. In the case presented, the preoperative CT revealed paravertebral abscess, and since an MRI was not performed, the diagnosis of the pseudoaneurysm was missed (Figure 2). This led to the intraoperative complication of aneurysm rupture. We therefore believe that when a paravertebral abscess is very close to the aortic vessels on a CT, it is advisable to perform MRI or CT Angiography, to confirm the presence or absence of a pseudoaneurysm.

The treatment of tuberculous pseudoaneurysm includes the repair of the wall of the affected vessel and anti - TB drugs. In view of the unpredictability of aneurysm rupture, it should be timely operated regardless of the size of the aneurysm [8]. Presently, the treatment of aneurysm is by either open intervention: including direct suture closure, patch repair, vascular lesion removal and synthetic vascular replacement, and extra-anatomic reconstruction; Or Minimal invasive surgeries : such as embolization, endovascular stent-graft occlusion and endovascular aneurysm repair (EVAR) [5,7,23]. In a study conducted by Xue J. et al [6], the intraoperative spontaneous rupture of a tuberculous pseudoaneurysm resulted in excessive hemorrhage and loss of the patient. Liu W.C [14] et al on the other hand, reported that they successful used endovascular stent-graft to rescue a patient with a major rupture of a tuberculous aneurysm, resulting in limited bleeding and no shock.

The pulmonary hilar fascia, an extension of the pleura is often used by cardiothoracic surgeons as a marker for dissecting hilar vessel [25]. Due to its strong and thick nature, we therefore sutured it to the prevertebral fascia that had been scrapped off, and this partially controlled the bleeding and provided the prerequisite for successful cardiac resuscitation and EVAR. The patient recovered well, without any complications. We believe that in a case where there is tuberculous pseudoaneurysm and spinal tuberculosis,

perhaps, it will be beneficial to repair the pseudoaneurysm first, before proceeding to repair the vertebral damage.

Intraoperative hypothermia as a classic cerebral protection strategy is widely used in aortic repair surgeries [26]. Svensson LG *et al.* reported that intraoperative hypothermia resulted in neurological complications in some patients [27], however, Stein LH *et al.* reported that it provided a good protection against intraoperative brain damage [28]. Intraoperative hypothermia works by reducing metabolic rates of the nervous system, thereby providing protection and limiting the consequence of blocked cerebral circulation [29]. In the case presented, the operating room temperature was reduced to 18°C, and patient's core body temperature reduced to 30-34°C. The followup neurological examination did not reveal any long-term neurological or cardiovascular complications.

Conclusion

From the experience of this case, we believe that in the case of spinal tuberculosis with tuberculous pseudoaneurysms, the following should be carefully noted. 1. Paravertebral abscess observed close to thoracic aorta on preoperative CTs should be followed-up with either an MRI or CTA to exclude pseudoaneurysms. 2. Intraoperative hypothermia can be used to reduce the metabolic demands of the CNS and protect the brain and other vital organs in the case of excessive hemorrhage and hemorrhagic shock. 3. The hilar fascia can be used to effectively control excessive hemorrhage, and to suture close scrapped prevertebral fascia of a tuberculous lesion.

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