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**Unusual cause of hemorrhagic pleural effusion: A case report of nontyphoidal
Salmonella infected aortic aneurysm and literature review**

KT Lee *et al.* Salmonella mycortic aneurysm

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Abstract

BACKGROUND

Infected aortic aneurysms are uncommon and difficult to treat. We present a case of infected aortic aneurysm with recurrent nontyphoidal *Salmonella* bacteremia.

CASE SUMMARY

A 68 years old gentleman presented with non-specific symptoms and was found to have nontyphoidal *Salmonella* bacteremia and was treated with intravenous (IV) ceftriaxone. However his condition didn't improve and he developed a multiloculated right pleural effusion. Thoracocentesis was done drained out hemorrhagic pleural fluid. Chest computed tomography demonstrated descending thoracic aorta saccular aneurysm with periaortic hematoma likely due to recent bleed and extending to the right pleural cavity. He was referred to cardiothoracic surgery team and was planned for medical therapy in view of hemodynamically stable and no evidence of active leakage. He completed IV antibiotic for five weeks and refused for surgical intervention. Unfortunately, he had another two admissions for recurrent nontyphoidal *Salmonella* bacteremia. Finally, he agreed for surgical intervention and underwent endovascular aortic repair three months later. Postoperatively, his condition remained stable with no recurrence of infection.

CONCLUSION

Our case highlights the importance of high index of suspicion of infected aortic aneurysm in patient with *Salmonella* bacteremia with high-risk factors such as atherosclerosis.

Key Words: Infected aneurysm; aorta; nontyphoidal *Salmonella*; pleural effusion

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Core Tip: Infected aortic aneurysm is a rare condition with high mortality. Our aim of this case report is to highlight the importance of high index of suspicion of infected aortic aneurysm in patient with *Salmonella* bacteremia with additional literature review to help clinician in the management of this disease. Medical therapy alone in this condition is associated with poor outcome.

INTRODUCTION

Infected aneurysm is a rare disease associated with significant morbidity and mortality. Initial symptoms are non-specific and due to lack of conclusive signs and symptoms, patients are often subjected to various investigations until a diagnosis is made. Standard treatment consists of antibiotic therapy and open surgery with surgical debridement and vascular reconstruction. However, there are no clear guidelines or randomized controlled trial on the best approach for the management of this condition. We report a patient with nontyphoidal *Salmonella* infected aortic aneurysm who was treated with medical therapy initially and complicated with recurrence. He was successfully treated with endovascular aortic repair and was well during follow up with no recurrence of infection. Relevant literature is reviewed.

CASE PRESENTATION

Chief complaints

A 68 years old man presented to our hospital with one week duration of fever associated with lethargy, reduced oral intake and dyspnoea.

History of present illness

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History of past illness

His medical illness includes diabetes mellitus, hypertension, ischemic heart disease and chronic kidney disease.

Personal and family history

No relevant family history.

Physical examination

He was febrile and required the support of facemask oxygen during presentation. Examination revealed reduced air entry over right lower zone with bilateral lower zone crepitations.

Laboratory examinations

Laboratory results showed ⁷ white blood cell count of $16 \times 10^9/\text{L}$, hemoglobin level of 8.3g/dL, urea 16.9mmol/L, creatinine 301 $\mu\text{mol/L}$.

Imaging examinations

Chest x-ray on admission showed blunted right costophrenic angle (Figure 1).

PROGRESS

He was admitted to the medical ward with the diagnosis of pneumonia and acute on chronic kidney disease. He was started on IV ceftriaxone 2g daily. Blood culture on admission was positive for nontyphoidal *Salmonella* spp. During the course of admission, he became more tachypnoeic and right thoracocentesis was done in view of worsening pleural effusion, which drained out 250 mL of blood stained fluid. Lung ultrasound showed multiseptated right pleural effusion. Despite ultrasound guided pigtail drainage of right pleural effusion, his condition did not improve. Chest computed tomography (CT) demonstrated descending thoracic aorta saccular

aneurysm with periaortic hematoma (Figure 2), likely due to recent bleed and extending to the right pleural cavity. Pleural fluid culture was negative.

FINAL DIAGNOSIS

Salmonella infected aortic aneurysm

TREATMENT

He was referred to cardiothoracic surgery team and was planned for medical therapy in view of hemodynamically stable and no evidence of active leakage. There were multiple changes in his antibiotics regime due to persistent fever (which included ceftazidime, piperacillin-tazobactam, meropenem and cefepime). He completed 5 wk of antibiotics and was planned for a CT angiography of aorta at a later date to decide on surgical intervention.

OUTCOME AND FOLLOW-UP

Unfortunately, the patient was readmitted two months later with abdominal discomfort and unable to pass motion for one week. Abdominal X-ray demonstrated dilated large intestine. CT abdomen and pelvis showed descending thoracic aorta saccular aneurysm (slightly larger) with features suggestive of superimposed infection of periaortic hematoma; size of aneurysm: 2.4 x 3.6cm and fecal laden bowels with features of impending intestinal obstruction. No obvious bowel related mass was seen. He was treated conservatively for the ileus which resolved after one day. Blood culture on this admission was positive again for nontyphoidal *Salmonella* spp. Echocardiogram did not show any vegetations. He completed 6 wk of IV ampicillin and was discharged well. A repeated CT scan after two months showed resolved periaortic hematoma, however patient refused for surgical intervention and was given lifelong prophylactic oral antibiotic. Unfortunately, patient presented again with second recurrence after one month and blood culture positive again for non-typhoidal *Salmonella*. He completed IV antibiotic and finally agreed for surgical intervention. He underwent endovascular

aortic repair and his condition remained stable with no recurrence of infection during his last follow up after 2 years.

DISCUSSION

Infected aneurysm is a serious clinical condition. The term mycotic aneurysm was first described by William Osler in 1885¹, however the nomenclature of mycotic aneurysm *vs* infected aneurysm remains controversial as majority of infected aneurysm are due to bacterial infection. Etiology of infected aneurysm includes direct bacterial inoculation, bacteremic seeding of existing intimal injury, atherosclerotic plaque, or preexisting aneurysm, contiguous infection or septic emboli from heart can occlude vasa vasorum of blood vessel and leading to infected aneurysm.

Infected aortic aneurysm is a rare but life-threatening condition with devastating outcomes. Initial clinical presentations are often non-specific; therefore, misdiagnosis is common. Infected aortic aneurysms may manifest as vague pain at the back, chest or abdomen. Some patients may present as pyrexia of unknown origin and remain undiagnosed until rupture of aneurysm. In our patient, he was only diagnosed with infected aortic aneurysm after three weeks of admission.

Salmonella species and *Staphylococcus aureus* are the most common pathogens of infected aortic aneurysm, followed by other organisms such as *Streptococci* species, *Treponema pallidum* and *Mycobacterium spp*²⁻⁴. In East Asia, gram negative bacteria related infected aortic aneurysm is more prevalent, where *Salmonella* species are the most common organisms ^{2,3,5,6}. ¹ It is important to obtain a microbiological diagnosis given the need for protracted courses of antimicrobial therapy. However, blood cultures are negative in around 50% of cases^{5,7,8}. Initial empirical treatment is often required and guided by the most likely infecting organism according to the individual and clinical circumstances. It is also prudent to use antibiotics judiciously as prolonged broad spectrum antibiotics may lead to development of antimicrobial resistance. ² *Salmonella* has a strong affinity for large blood vessels, and can easily adhere to the damaged vascular wall, strongly affecting the natural course of the disease.

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However studies showed that non- *Salmonella* infections are associated with higher aneurysm- related complications and mortality after treatment^{9,13,17}.

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Study by Oderich *et al*, with 43 patients with infected aortic aneurysm found that the risk factors of aneurysm-related death were extensive periaortic infection, female gender, *Staphylococcus aureus* infection, aneurysm rupture and suprarenal aneurysm location¹⁰. Another larger series done in Taiwan reported that the independent predictors of aneurysm-related death were advanced age, non-*Salmonella* infection and non-surgical treatment².

Until now, there are no randomized clinical trials to guide the management of infected aneurysm. Therapy mainly includes the control of infections by antibiotic and surgical debridement with or without reconstruction of arterial circulation. 5
Management strategies are primarily based upon clinical experience guided by case series. The optimal duration of antibiotic therapy remains inconclusive. Most studies recommend at least six week of antibiotic and stop only when there is no longer clinical and laboratory evidence of ongoing sepsis¹¹⁻¹².

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Medical therapy alone is associated with devastating outcomes, with in-hospital mortality of 50%¹³. Since infected aortic aneurysm is at high risk of rupture, surgical treatment is encouraged in the absence of absolute contraindications. Open surgery with extensive debridement of infected tissue and in situ or extra-anatomical reconstruction has been the gold standard treatment. However, it is associated with high risk of morbidity and mortality^{3,14-16}. Recently, endovascular techniques has become an emerging treatment alternative especially in the treatment of patients at prohibitive risk for open surgery. Few studies have showed that endovascular treatment of infected aortic aneurysm is feasible and has been described as a durable treatment option for high risk patients ^{17,18}. Luo *et al* reported that survival at 1 mo, 6 mo, 1 year and 5 year was 90%, 82%, 71% and 53% respectively¹⁸. In addition, a European multicenter study of endovascular treatment for infected aortic aneurysm also showed similar results ¹⁷.

CONCLUSION

Infected aortic aneurysm is a rare clinical entity with high mortality. Due to non-specificity of the early symptoms, misdiagnosis is common. Our case highlights the importance of high index of suspicion of infected aortic aneurysm in patient with recurrent nontyphoidal *Salmonella* bacteremia. Endovascular technique has become an emerging treatment option.

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SIMILARITY INDEX

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