

## CASE REPORT

# Emphysematous aortic arch aneurysm infected with *Salmonella*: A case report

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**Abstract**

Infected aortic aneurysm is a relatively rare disease that is extremely difficult to manage, resulting in a poor prognosis. We rescued a patient with *Salmonella*-infected aortic arch aneurysm surrounded with a specific and massive emphysema, despite experiencing aortic rupture, including delayed esophageal perforation after surgery.

**KEYWORDS**

emphysema, esophageal perforation, infected aortic aneurysm, *Salmonella*

## 1 | INTRODUCTION

Infected aortic aneurysm is a relatively rare disease with a high mortality.<sup>1</sup> We treated a patient with *Salmonella*-infected aortic arch aneurysm surrounded with a specific and massive emphysema.<sup>2</sup> Despite experiencing rupture, including delayed esophageal perforation after surgery, the patient was successfully treated with combined treatment involving open surgery, endovascular repair, and conservative antibiotic treatment. The most significant aspects of managing an infected aortic aneurysm include early diagnosis, infection control with adequate antibiotics, and a thorough resection of the infectious tissue for aortic reconstruction.

## 2 | CASE PRESENTATION

A 66-year-old man with a history of controlled hypertension and diabetes mellitus experienced high-grade fever and progressive hoarseness for approximately 1 month. Chest X-ray detected an enlarged aortic arch (Figure 1), and computed tomography (CT) revealed aortic arch aneurysm with specific and massive air-filled collection (Figure 2). Based on blood culture results, he was diagnosed with *Salmonella* aortitis with aneurysm. Prior to the result, a high dose of

intravenous tazobactam/piperacillin hydrate (18 g/day) was started to control the infection as an empiric therapy based on the guideline. However, he complained of sudden chest pain, and CT revealed an impending rupture of the infected aneurysm at 11 days after admission (Figure 3). Thus, he underwent emergency total arch replacement with rifampicin-soaked (0.2% solution) woven polyester graft (gelatin sealed and four branched; 22 mm in diameter), and greater omental flap coverage under circulatory arrest with selective cerebral perfusion (Figure 4). Intraoperatively, we found that the infected aorta spanned from the middle to distal portion of the arch, with tear on the back and upper side. Massive infectious tissue existed around the aorta and was removed as possible together with the aortic wall.

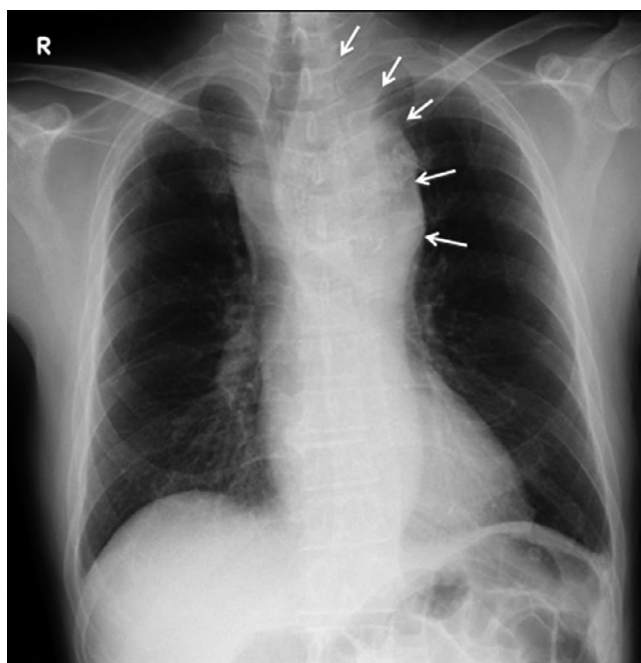
Postoperatively, his condition, including his brain function and oral intake, was uneventful. Additionally, his leukocyte count and C-reactive protein (CRP) decreased easily, owing to the switching of antibiotics to ceftazidime hydrate (4 g/day). However, after 22 days postoperatively, CT inadvertently revealed a small esophageal perforation (Figure 5A). Consequently, oral intake was stopped, and a feeding tube was inserted before performing gastrostomy. Infection was not completely controlled, but aortic shape deformity including anastomosis was not clearly recognized, and the inflammation did not remarkably worsen

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on the repeat blood test. Therefore, his intravenous antibiotics were switched to levofloxacin hydrate (500 mg/day), and vancomycin hydrochloride (2 g/day) was added for a limited period (approximately 3 weeks), with tube feeding through gastrostomy and total parenteral nutrition and without invasive strategies. Subsequently, the CRP level gradually decreased, and the infectious findings on imaging remarkably improved. Particularly, through conservative treatment, the esophageal perforation was barely visible roughly 2 months and a half later (Figure 5B). Then, his oral intake finally resumed.

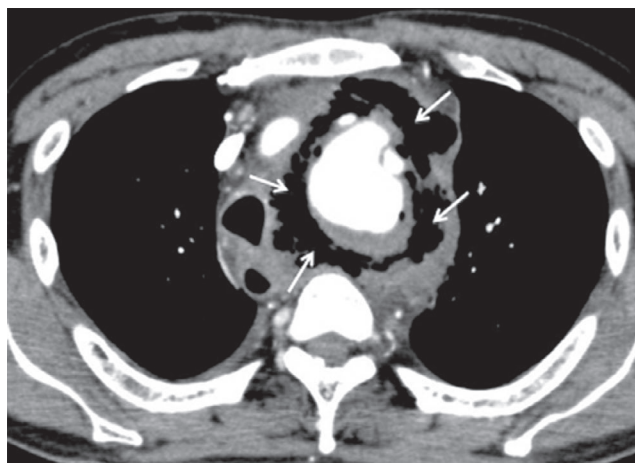
Four months postoperatively, a slight deformity as tiny bulging of the medial wall on the distal anastomosis of the graft was suspected on CT. Considering that patient's infection gradually alleviated with peroral levofloxacin hydrate (500 mg/day) and the CRP level was 0.5-3.0 mg/dL, after several examinations and discussions, the patient underwent scheduled endovascular stent-graft repair (ePTFE stent graft; 28 × 150 mm; Figure 6)<sup>3,4</sup> to reinforce anastomosis on the inside and to control bleeding if the anastomosis ruptured. That procedure was uneventfully performed except for initial type-1 endoleakage that was finally controlled. Fortunately, the site remained unchanged on an imaging study 6 weeks after the endovascular stent-graft procedure. The infection was also controlled, given that the CRP level was 0.4-2.5 mg/dL. He was finally discharged approximately 8 months after the initial surgery, continuing the same peroral antibiotics.



**FIGURE 1** Chest X-ray on admission. The aortic arch shadow is remarkably enlarged, resulting in tracheal deviation to the right side (arrows)

### 3 | DISCUSSION

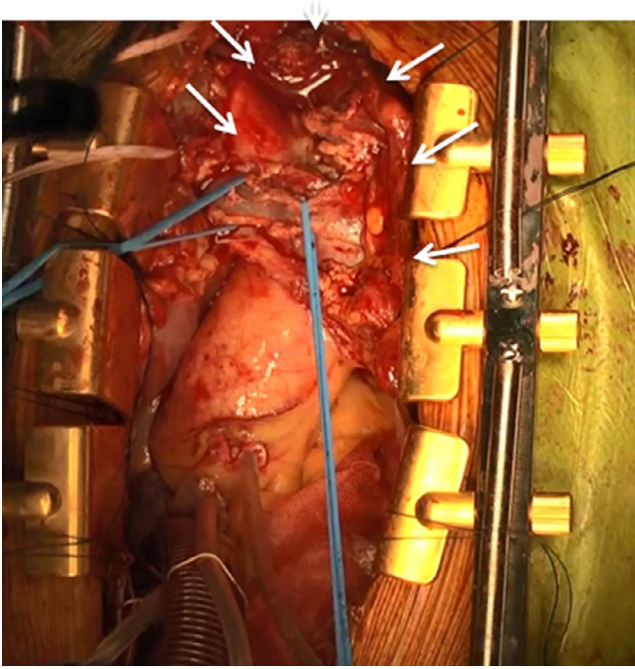
Infected aortic aneurysm, particularly aortic arch aneurysm, is a rare disease that is immensely difficult to manage.<sup>1,5,6</sup> Our patient with aortitis due to *Salmonella* infection was



**FIGURE 2** Computed tomography on admission reveals a specific and massive air-filled collection, indicating emphysema, around the infected aortic arch aneurysm (arrows)



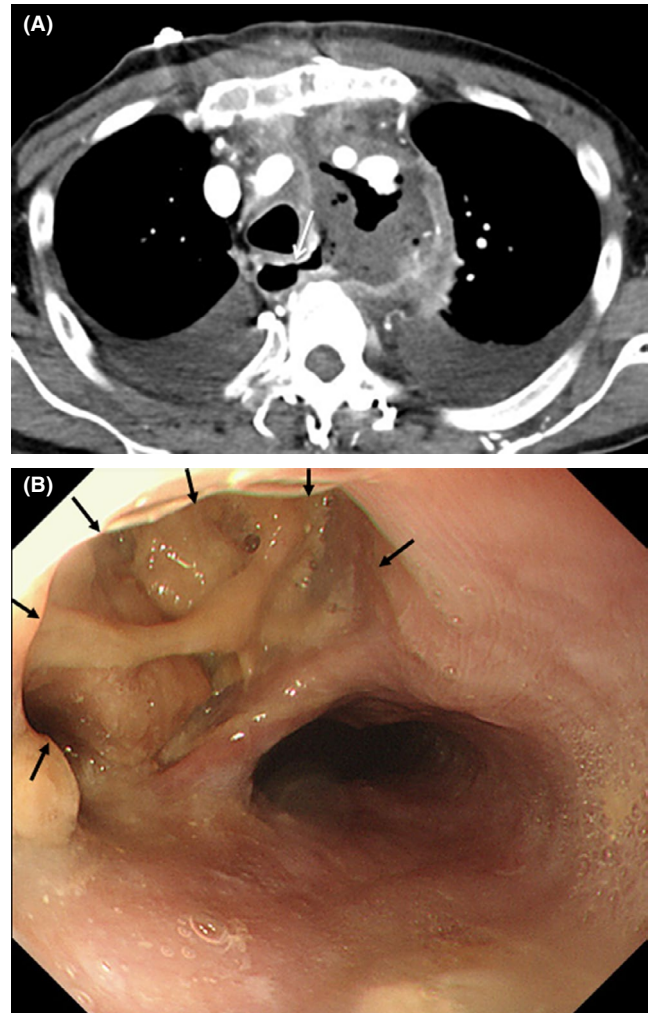
**FIGURE 3** Computed tomography reveals a ruptured but shielded aortic arch aneurysm at 11 d after admission (arrow)



**FIGURE 4** Intraoperative view. Infected aortic arch aneurysm involving the left common carotid and left subclavian arteries (arrows)

successfully treated despite experiencing aortic rupture, including delayed esophageal perforation after surgery. Upon admission, CT revealed an aneurysmal formation with specific and massive air-filled collection, suggesting unique features.<sup>2</sup> Therefore, severely infected aortic aneurysm was suspected, and high-dose intravenous antibiotics were started to be administered before the arrival of the blood culture results. The patient visited our institute approximately 1 month after the onset of symptoms; hence, the infectious invasion to the vascular wall was already progressive when the treatment was started. Ideally, initial antibiotic treatment should be started as soon as possible before vascular wall degeneration caused by the progression of infection. However, diagnosing an infection by *Salmonella* species in its early stages is difficult. In our case, the patient was under dental treatment for a few months when his fever developed. If the bloody dental procedure induced *Salmonella* infection in the aorta, then antibiotics should have been administered to prevent systemic infection.

This case has some points that need to be discussed. The first point was the decision to wait for the initial surgery to treat the infected aortic aneurysm until the infection was controlled. Upon admission, the patient had a high-grade fever, with a leukocyte count of 7000/ $\mu$ L (neutrophil: 83.2%) and a C-reacting protein value of 18.6 mg/dL. For infectious aortic disease, aortic replacement using a homograft or biological pericardial tube is recommended. However, homograft is generally limited in our country, and the technique of replacement using a biological pericardial tube is extremely difficult to design, especially for branched



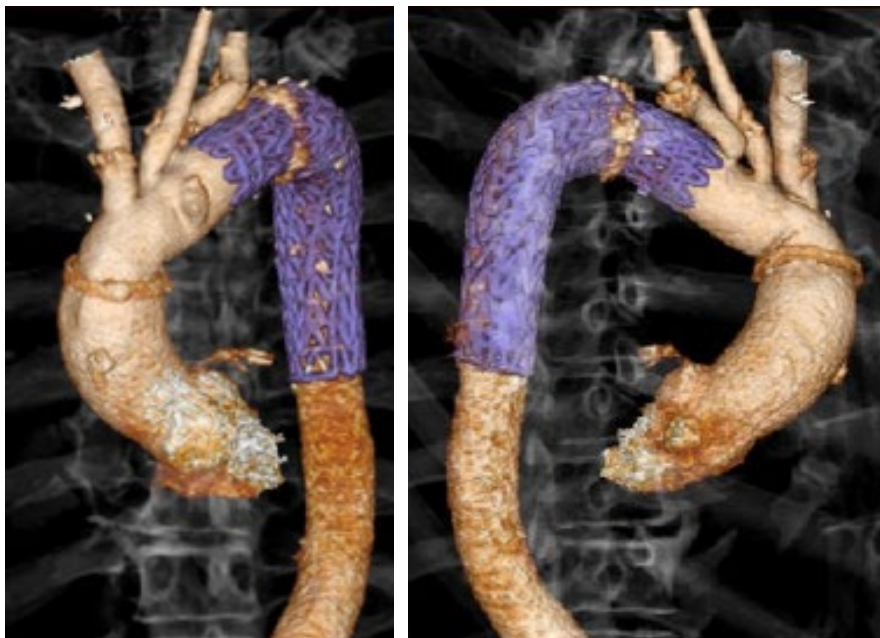
**FIGURE 5** A, Computed tomography reveals the delayed esophageal perforation after surgery (white arrow); B, After 2 mo and a half, upper gastrointestinal endoscopy reveals the closure of esophageal perforation such as a small diverticulum without inflammation (black arrows)

and curved total aortic arch; furthermore, its patency, durability, and endurance for bleeding is still controversial.<sup>7-9</sup> Therefore, we decided to perform a conservative method, that is, total arch replacement with rifampicin-soaked woven Dacron graft right after the infection was controlled as much as possible. This procedure was supposed to be performed within 2 weeks after admission. However, our patient's infected aortic aneurysm ruptured on day 11 after admission. Fortunately, we had time to prepare for an urgent open surgery because the rupture was abided as an impending rupture, but this surgery should have been performed as an emergency within a few days after admission.<sup>5,6</sup>

In our case, delayed esophageal perforation, which was not directly communicated as a fistula formation after surgery, was not recognized until CT was performed.

Intraoperatively, we found that the infectious tissue was widely spread to the cranial and posterior regions. We

**FIGURE 6** 3D computed tomography reveals the endovascular stent-graft indwelling at the distal anastomosis



resected these tissues as much as possible and performed greater omental flap coverage for the graft and anastomosis. However, these steps were supposed to be insufficient to eliminate the infection. Moreover, the infection might have become difficult to control by the leakage of the digestive tract contents via the perforation into the mediastinal space. Subsequently, we chose conservative treatment, which included tube feeding through gastrostomy and the addition and switch of antibiotics because no new infectious lesion of the graft, anastomosis, and native aorta were recognized. A second surgical reconstruction of the thoracic aortic arch with total esophagectomy should have been avoided as possible. Eventually, the esophageal perforation spontaneously closed after around 2 months and a half.

Four months after the graft replacement, CT revealed a slight deformity as tiny bulging of the medial wall on the distal anastomosis of the graft. However, this finding was not definitely diagnosed as acute aortic syndrome and the patient's CRP level was stable (0.5-3.0 mg/dL), with no fever. After a thorough discussion, we decided that the stent graft would be indwelled covering the distal anastomosis site, which might be deviated from absolute indication.<sup>5,10</sup> Although this step is highly controversial, it was performed mainly to avoid massive hemorrhage if the distal anastomosis ruptured. Fortunately, neither distal anastomosis rupture nor the recurrence of infection occurred 1 year after the initial surgery.

Infected aortic aneurysms including *Salmonella* are life-threatening, and diagnosis and treatment are still challenging.<sup>5,6</sup> These diseases are associated with high morbidity and mortality, and the mortality ranges from 23% to 37%.<sup>5,11</sup> In our institute, only two cases suffering from infected

thoracic or thoraco-abdominal aortic aneurysm were previously experienced in the recent 9 years, and neither of them could be discharged because of sepsis. Moreover, we treated two cases of the graft or the endovascular stent infection positioned at the descending thoracic aorta; however, one of them was not rescued because of sepsis. Therefore, in this reported case, severe management would be continuously necessary for a long term in the outpatient department.

Based on this case, we propose that in infected aortic aneurysms, physicians should focus on infection control. Early diagnosis and the immediate administration of sufficient high-dose antibiotics are the most important treatment measures; if surgery is performed, the infectious tissue should be thoroughly resected for aortic reconstruction.<sup>11,12</sup> In *Salmonella* aortitis, improved blood test results do not always indicate that the infection is controlled.<sup>1</sup> Even with successful treatment, we should consider the possibility that the infected aortic wall has been weakened due to its scarring and thinning.<sup>1,11</sup> Therefore, even after discharge, postoperative long-term antibiotic therapy with subsequent imaging studies is required for *Salmonella* aortitis.<sup>11,13</sup>

#### 4 | CONCLUSION

A patient with *Salmonella*-infected aortic arch aneurysm was rescued by a combined treatment involving open surgery, endovascular repair, and conservative antibiotic treatment despite experiencing aortic rupture, including delayed esophageal perforation after surgery. Not all of our medical strategies in this case can be justified, but we successfully treated it with trial and error despite being in an advanced phase, which revealed a remarkably specific and massive

emphysema around aneurysmal formation on imaging studies. This report aims to add vital information to all medical staff concerned with cardiovascular disease.

### CONFLICT OF INTEREST

None declared.

### AUTHOR CONTRIBUTION

SA: is sole author who fully contributed to the article.

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