Sudden Death with Massive Hemothorax from Rupture of a Thoracic Inflammatory Aortic Aneurysm: An Autopsy Case Report

Hitoshi Ando, Osamu Suzuki, Fumitaka Sakuma, Hitoshi Oyama, Yoshihiro Kazuta, Masahito Kuroda, Kumiko Terashima and Masayuki Miyata

Abstract

An 84-year-old man was admitted to the Department of Neurosurgery for a sudden episode of fainting. Brain computed tomography and magnetic resonance imaging demonstrated no fresh lesions. Anorexia, fever and elevation of C-reactive protein and creatine phosphokinase were observed, and the patient was transferred to the Department of Internal Medicine for further examination and treatment. High-dose steroids and antibiotics were administered, and his fever subsided. However, massive hemothorax suddenly developed and the patient died. A thoracic aortic aneurysm that had coalesced and ruptured a left lung bronchus was detected at autopsy. Pathological examination revealed an inflammatory aortic aneurysm.

Key words: inflammatory aortic aneurysm, thoracic aorta, fever of unknown origin, hemothorax


Introduction

A total of 4 to 5% of abdominal aortic aneurysms (AAA) are inflammatory aortic aneurysms (IAA) (1-3). Symptoms of IAA include fever and weight loss, and mantle sign is a characteristic finding of IAA on computed tomography (CT). However, IAA of the thoracic aorta is seen only rarely, and few cases have been reported in the literature (4, 5). We report herein a patient with fever of unknown origin who died suddenly of massive hemothorax. Upon autopsy, a thoracic IAA that adhered solidly to the left lung and ruptured a left lung bronchus, was found.

Case Report

An 84-year-old man was admitted to the Department of Internal Medicine at Fukushima Red Cross Hospital because of fever and anorexia from the end of March to early April 2006. Fatty liver and splenomegaly were detected by abdominal ultrasonography at that time. He was later admitted again because of fever and diarrhea from the middle of April to the middle of May 2006. Close investigation was performed, including a variety of blood tests, thoracic and abdominal CT, upper gastrointestinal fiberscopy, lower gastrointestinal contrast radiography and gallium-scintiscan. Other than fatty liver and splenomegaly, there were no remarkable findings. At that time, after discharge, he reported no serious problems except for a slight fever. However, he suddenly fainted while defecating at home and was urgently admitted to the Department of the Neurosurgery in early October 2006. Brain CT and MRI revealed no fresh lesions, but blood tests revealed elevated creatine phosphokinase (CK) at 5,656 IU/L and myoglobinuria. The patient was diagnosed with rhabdomyolysis due to a side effect of fenofibrate and was treated with an intravenous drip infusion of Ringer solution. Although the CK level became normalized to 69 IU/L by the middle of October 2006, he still had a fever over 38°C, anorexia and high C-reactive protein (CRP) at 13.11 mg/dl. Therefore, he was transferred to the Department of Internal Medicine for further investigations in mid-October 2006. His past medical history included the follow-
The patient was transferred to the Department of Internal Medicine, his fever (37.5-38°C) continued and his oxygen saturation sometimes went down to 89% on room air. He suffered from a severe cough at night. Methylprednisolone at a dose of 125 mg/day was administered for fever of unknown origin beginning in early November 2006. The second CT performed on the following day is shown in Fig. 2. Although bilateral pleural effusions disappeared, undefined consolidated tissue around the thoracic aorta seemed to have expanded. Considering his severe cough as well as fever, it was speculated that the lesion around the thoracic aorta seen on the CT was pneumonia and antibiotics (biapenem 0.6 g/day and minocycline 200 mg/day) were co-administered. Two days after the administration of antibiotics, his body temperature dropped to 36°C and his cough improved. However, 4 days after the administration of antibiotics, the patient suddenly coughed up a large volume of blood, fell into shock, and died soon thereafter. Body temperature and CRP levels remained elevated even though he received two courses of 4th generation cefem.

### Pathological findings

Autopsy of the lung, a part of the thoracic aorta and the spleen was performed with permission of the bereaved family. Gross pathological findings are shown in Fig. 4a, b. As shown in Fig. 4a, a ping-pong ball-sized (Ø30 mm x 30 mm) aneurysm was tightly adhered to the left lung. This aneu-

### Table 1. Laboratory Data on Transfer to the Department of Internal Medicine

<table>
<thead>
<tr>
<th>Hematologic test</th>
<th>Biochemical test</th>
<th>Infectious disease test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Red blood cells</td>
<td>Total bilirubin 1mg/dL</td>
<td>HBsAg (-)</td>
</tr>
<tr>
<td>White blood cells</td>
<td>Direct bilirubin 0.4mg/dL</td>
<td>HCVAb (-)</td>
</tr>
<tr>
<td>Neutrophils 73.7%</td>
<td>Total protein 6.7g/dL</td>
<td>VD (-)</td>
</tr>
<tr>
<td>Lymphocytes 14.2%</td>
<td>Albumin 3.11g/dL</td>
<td></td>
</tr>
<tr>
<td>Monocytes 8.9%</td>
<td>α1 0.35g/dL</td>
<td></td>
</tr>
<tr>
<td>Basophils 0.7%</td>
<td>α2 0.78g/dL</td>
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</tr>
<tr>
<td>Eosinophils 2.5%</td>
<td>β 0.67g/dL</td>
<td></td>
</tr>
<tr>
<td>Hemoglobin 7.7g/dL</td>
<td>γ 1.95g/dL</td>
<td></td>
</tr>
<tr>
<td>Hematocrit 22.60%</td>
<td>TTT 5.8U</td>
<td></td>
</tr>
<tr>
<td>Platelets 168×10^3/μL</td>
<td>ZTT 8.7U</td>
<td></td>
</tr>
<tr>
<td>AST 23IU/L</td>
<td>BUN 23.9mg/dL</td>
<td></td>
</tr>
<tr>
<td>ALT 14IU/L</td>
<td>Ch-E 66IU/L</td>
<td>Anti-nuclear antibody (-)</td>
</tr>
<tr>
<td>ALP 257IU/L</td>
<td>CK 62IU/L</td>
<td>Anti-DNA antibody (-)</td>
</tr>
<tr>
<td>LDH 160IU/L</td>
<td>BUN 23.9mg/dL</td>
<td></td>
</tr>
<tr>
<td>γ-GTP 31IU/L</td>
<td>Crea 1.5mg/dL</td>
<td></td>
</tr>
<tr>
<td>LAP 120IU/L</td>
<td>UA 5.8mg/dL</td>
<td></td>
</tr>
<tr>
<td>Ch-E 66IU/L</td>
<td>CRP 11.85mg/dL</td>
<td></td>
</tr>
<tr>
<td>CK 62IU/L</td>
<td>ESR1h/2h 101nm/112mm</td>
<td></td>
</tr>
<tr>
<td>BUN 23.9mg/dL</td>
<td>Anti-nuclear antibody (-)</td>
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</tbody>
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TTT: thymol turbidity test; ZTT: zinc sulfate turbidity test; AST: aspartate aminotransferase; ALT: alanine aminotransferase; ALP: alkaline phosphatase; LDH: lactate dehydrogenase, γ-GTP: γ-glutamyl transpeptidase; LAP: alkaline phosphatase; CH-E: cholinesterase; CK: creatine kinase; BUN: blood urea nitrogen; Crea: creatinine; U-A: uric acid; CRP: C-reactive protein; ESR: erythrocyte sedimentation rate; HBs-Ag: hepatitis B surface antigen; HCV-Ab: hepatitis C virus antibody; VD: venereal disease; and ANCA: antineutrophil cytoplasmic antibodies.
rhythm penetrated the left lung bronchus as shown by a probe. This penetration resulted in massive bleeding to the left upper lobe of the lung, as seen in a sagittal section of the severely thickened wall of aneurysm (Fig. 4b).

Microscopic findings are shown in Fig. 5. The wall of the aneurysm with Elastica-Masson (EM) staining is shown in Fig. 5a. The labels “A” and “B” indicate the vascular lumen and adhering sides, respectively. The normal structure of the tunica media was replaced by atheroma as indicated by a sharp (#). Atheroma of the tunica media had torn a part of the tunica adventitia as indicated by the arrow (→) and leaked into the tunica adventitia, resulting in formation of cholesterol crystallization as indicated by the asterisk (*). A specimen of left lung that had adhered to the aneurysm stained by EM is shown in Fig. 5b. The labels “A” and “B” indicate the adhesion site and organized lung tissue, respectively. Fibrosing tissue of the lung adhering to the aneurysm as indicated by an asterisk (*) demonstrates chronicity of the inflammation. Specimens of tunica adventitia of the aneurysmal wall and lung stained by Hematoxylin-Eosin (HE) are shown in Fig. 5c, d, respectively. Both specimens demonstrate typical inflammatory cell infiltration composed mainly of lymphocytes and fibroblasts. These findings demonstrate that there was long-standing inflammation. CD68 and muscle actin immunostaining of the aneurysmal wall are shown in Fig. 6a, b, respectively. As shown in Fig. 6a, CD68-positive macrophage accumulation is remarkable in the tunica adventitia of the aneurysmal wall, suggesting an immune reaction such as phagocytosis to ectopic atheroma. Furthermore, smooth muscle actin (SMA) positive fibroblasts were increased and formed a layer of fibrosing lesion in the tunica adventitia of the aneurysmal wall as shown in Fig. 6b.

**Discussion**

The present patient had suffered from a longstanding fever of unknown origin, and suddenly coughed up a large amount of blood, and died shortly thereafter. Autopsy revealed communication between a thoracic aneurysm and a bronchus of the left lung. The aneurysm was tightly adhered and ruptured a bronchus through severe inflammation. This
Figure 3. Body temperature (BT) and CRP levels after admission to the Department of Neurosurgery to death of the patient are shown. During the stay at the Department of Neurosurgery, his body temperature ranged from 37-39°C almost all of the time even though he had received 2 courses of 4th generation cefem. Likewise CRP stayed over 4 mg/dL all of the time.

Figure 4. As shown in Fig. 4a, a ping-pong ball-sized (Ø 30 mm × 30 mm) aneurysm (*) severely adhered to the left lung (arrows). This aneurysm penetrated the left lung bronchus as shown by a probe. Fig. 4b Sagittal section of the aneurysm wall, which is severely thickened and ruptured as indicated by the probe.

aneurysm was diagnosed as IAA due to clinical features and pathological examination. Epidemiologically, IAA is more common in men who are over 60 years old. Its incidence has been reported to be from 4 to 5% of all abdominal aortic aneurysms (1-4). Clinical symptoms and laboratory data of patients with abdominal IAA are abdominal pain, lumber, body weight loss, occlusion of the urinary tract or intestinal canal, elevation of the erythrocyte sedimentation rate and fever. Pathological findings are fibrosis of periaortic tissue, a markedly thickened aneurysmal wall and adhesion of the aneurysmal wall to surrounding tissues. IAA of the thoracic aorta has been reported only rarely; only 2 reports in
Figure 5. (a) The labels “A” and “B” indicate the vascular lumen and adhering sides, respectively. The normal structure of the tunica media is replaced by atheroma (#). Atheroma of the tunica media tore a part of the tunica adventitia as indicated by arrow (→) and leaked into the tunica adventitia, resulting in formation of cholesterol crystallization as indicated by asterisk (*). (b) The labels “A” and “B” indicate the adhesion site of the aneurysm and organized lung tissue, respectively. Organized lung tissue adhering to the aneurysm demonstrates inflammatory cell accumulation (c), and typical inflammatory cell infiltration composed mainly of lymphocytes and fibroblasts (d). These findings indicate a long-standing inflammation.

Figure 6. (a) CD68-positive macrophages were markedly increased in the tunica adventitia of the aneurysmal wall. (b) SMA-positive muscle fibroblasts were markedly increased in the tunica adventitia of the aneurysmal wall.
Autopsy was performed in this case of sudden death with massive hemoptysis. The cause of death was considered to be rupture of a thoracic aneurysm to an adhering bronchus mass. Hemoptysis was massive. The cause of death was considered to be due to rupture of a thoracic aneurysm. To the best of our knowledge, this is the first report in this respect.

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