Aortobronchial Fistula Complicated with an Aortic Aneurysm in Hemodialysis Patient

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Dear Sir,

This is the first report of aortobronchial fistula complication in a hemodialysis patient. The patient was 56-year-old male who underwent hemodialysis for 10 years, and complicated aortobronchial fistula with an aortic aneurysm. He died due to massive hemoptysis on admission. It was difficult to make the diagnosis because of a small amount of intermittent hemoptysis before admission and lack of evidence of an aneurysm on the chest X ray. The purpose of this report is to draw attention to the little known disorder, and to stress the importance of differential diagnosis of hemoptysis in hemodialysis patients.

Case Report

A 56-year-old male was admitted to the emergency center in Tokai University Hospital because of massive hemoptysis. He had a long history of hypertensive renal disease, and hemodialysis was started 10 years ago due to end-stage renal failure. Hemodialysis was performed 3 times a week. Three weeks before admission he noticed left back pain and a small amount of intermittent hemoptysis. Chest X-ray films disclosed no abnormalities. The left back pain worsened 2 days before admission. He suddenly developed hemoptysis with 300 ml of fresh blood in the morning of admission, and was immediately transferred to our hospital.

Physical examination revealed anemic conjunctive palpebrae, and moist rale was auscultated in the left lower lung field. Blood pressure was 168/90 mm Hg. No abnormalities in the abdomen nor peripheral edema were found. Laboratory examination disclosed leukocyte count of 12,100/mm3 with normal differentiation, and hemoglobin 6.3 g/dl with normochromic normocyte. Blood urea nitrogen was 48 mg/dl and serum creatinine 6.8 mg/dl. The value of potassium was 5.9 mEq/l and phosphate 4.7 mg/dl. Serum albumin was 3.2 mg/dl, and glucose,
bilirubin, lactic dehydrogenase, glutamic oxaloacetic and glutamic pyruvic transaminases were within normal limits. C-reactive protein was three positive. Chest X-ray film revealed an infiltrative shadow in the left lower lobe obscuring the line of the descending aorta (fig. 1). Shortly after admission, the patient became hypotensive, and died despite all-out cardiac resuscitation.

Autopsy findings revealed severe atherosclerotic changes in systemic vessels and an atherosclerotic aneurysm of 3 cm in diameter located in thoracic descending aorta, which eroded into the adjacent left lower bronchus (fig. 2). Fibrous tissue was developing around the aortobronchial fistula.

Discussion
The number of hemodialysis patients has been increasing with the progress of technique and the increasing availability of hemodialysis for the older patients with end-stage renal failure. Atherosclerotic change of the vessels increasing remarkably and atherosclerotic cardiovascular complications have become serious problem in patients. Our recent study disclosed that the frequency of aortic aneurysm was 20 (2.5%) out of 805 end-stage renal disease and 6% of 71 dialysis patients [1]. Aortobroncho-pulmonary fistula is invariably complicated with aortic aneurysm, itself a rare but lethal disorder. If the lesion is discovered earlier, the survival rate becomes greater than 80%. However, if not, the patient dies due to massive hemoptysis [2]. This disorder was reported in only seven cases in the literature since 1934 [3]. Most of the lesions result from thoracic aortic aneurysms [2]. The causes of aortic aneurysm were defined as mycotic, tuberculous and syphilitic in the past; however, atherosclerotic lesion has been increasingly revealed recently. Cobletz et al. [4] reported four cases of aortobronchial fistula, two of which derived from atherosclerosis.

The exact diagnosis of aortobronchial fistula is sometimes very difficult due to nonspecific clinical findings. Hemoptysis is the most frequent symptom and occurs in over 95% of the cases [2]. However, hemoptysis is sometimes only small in amount and intermittent, which can lead to the misdiagnosis as bronchitis [5, 6]. The aneurysm invades the lung by pressure or chronic inflammation to represent hemoptysis. The patient does not complain of hemoptysis until the formation of thrombus in fistula. The duration to the presentation with massive hemoptysis in the past cases varied, from 2 days to 1 year [6, 7]. Other symptoms include chest pain, cough and dyspnea. Back pain as in our case before admission might be associated with the development of the aneurysm. Chest X-ray film shows the consolidation of the lung, which demonstrates the existence of aortic aneurysm in 46% cases [2]. In our case, the initial hemoptysis was only small in amount and intermittent, and furthermore the chest X-ray films showed no abnormalities. After admission, the chest X-ray film also revealed only the consolidation in the left lower lobe. Therefore, we could not make the exact diagnosis before autopsy.

As to the examination of the patient with hemoptysis, some authors recommend CT and aortography, which are indispensable for the definitive diagnosis. Bronchos-copy is usually the
best tool to identify the source of hemoptysis. In aortobronchial fistula, bronchoscopy should be performed with caution, otherwise, thrombi within the fistula might be dislodged to the fatal hemorrhage [4]. In our case, neither CT nor aortography were performed because the shadow of the aneurysm and fistula did not show on the chest X-ray films.

Aortopulmonary fistula must be considered in long-term hemodialysis patients complaining of hemoptysis and with serious atherosclerotic lesions. An aggressive diagnostic approach including CT and aortography is warranted in all hemodialysis patients with hemoptysis, even if small in amount and intermittent.

References