

## CASE REPORT

**Non-recognized cause of intrathoracic bleeding**

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The authors present a case report of bleeding from the small dissection of descending aorta. Patient, 71 year old woman, with severe comorbidities (nephrotic syndrome based on membranous glomerulonephritis, diabetes mellitus, lung emphysema, hepatopathy, polyneuropathy and others). One month after last stay in hospital chemotherapy et corticotherapy.

In while on heparin during hospitalization at nephrologic department, patient developed right side haemothorax and haemomediastinum. An urgent transfer to surgical department with cardiopulmonary resuscitation was performed. The suspicion of aneurysm on CT angioscan was non confirmed. The damage of oesophagus due to previous endoscopy (for chest pain) was non confirmed, too.

During surgery masive sanquine effusion of mediastinum and right side haemothorax was found. The drainage and redrainage of the chest was performed. The cause of bleeding was not found.

Critically ill patient had died and on autopsy a small dissection of thoracic aorta was found as a cause of bleeding. The problems of diagnosis and therapy are discussed. (Fig. 2, Ref. 7.)

**Key words:** aortic dissection, bleeding.

A female patient, 71 year old, with several severe diseases, has been repeatedly hospitalized and treated in various departments of hospital. The main problem of the patient is nephrotic syndrome caused by membranous glomerulonephritis, evaluated as primary (per exclusionem). Other diagnoses were following: hypertension, hypercholesterolemia, diabetes mellitus, pulmonary emphysema, chronic venous insufficiency, epilepsy, alcoholic hepatic disease and polyneuropathy. The patient underwent an appendectomy, surgical intervention due to ectopic pregnancy and fracture of the L1 vertebra and serial costal fracture 1 year ago. She was treated with 5 pulses of chlorambucil and methylprednisolone at the department of nephrology 1 month before hospitalization at the surgical department. This treatment did not have any significant effect and cycle 6 was not introduced due to leukopenia. Moreover, a corticoid-induced psychosis developed. The patient was discharged from the hospital after stabilization of medical conditions; however, she was immediately admitted to another department of hospital due to fever, loss of appetite, dehydration and malnutrition. Nephrotic syndrome had progressed and therefore was the patient transferred to nephrology department, where was *Staphylococcus aureus* found in blood culture. The treatment resulted in a decrease of fever; however, the renal function did not improve. The patient reported chest

and back pain and associated them with previous spine and costal injury. The ECG and cardiac enzyme evaluation was negative. Fibroscopy of the upper GIT was performed, and a bluish strip, several centimeters long, was described along the distal esophagus (on the lateral part). The procedure did not reveal any further findings and was performed without complications. Treatment with painkillers resulted in improved condition. Low-molecular-weight heparin was introduced for three days; however, anemia was observed simultaneously – hemoglobin decreased by 20 g to 80 g/L! The change on auscultation of the right thorax was the indication for performing a chest X-ray, where a fluid extending to the aortal arch was found. A small amount of hemorrhagic fluid was drained by the puncture. Ultrasonography and CT follow-up examinations were performed and confirmed the

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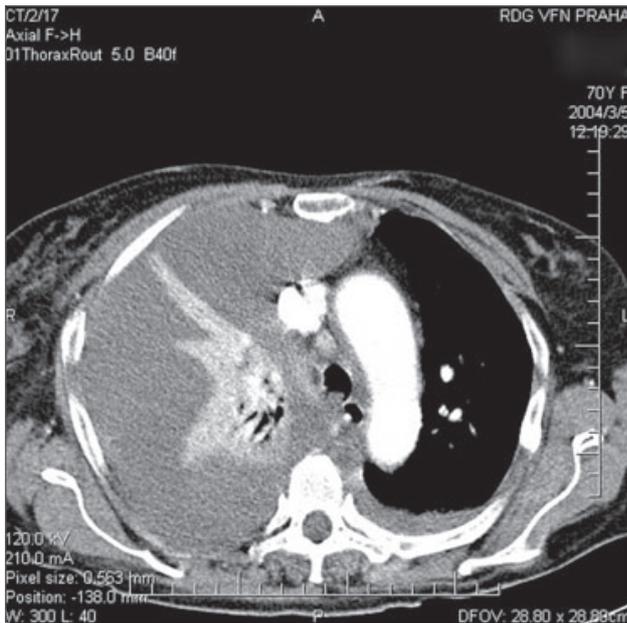


Fig. 1. CT examination again excluded an aortal lesion.

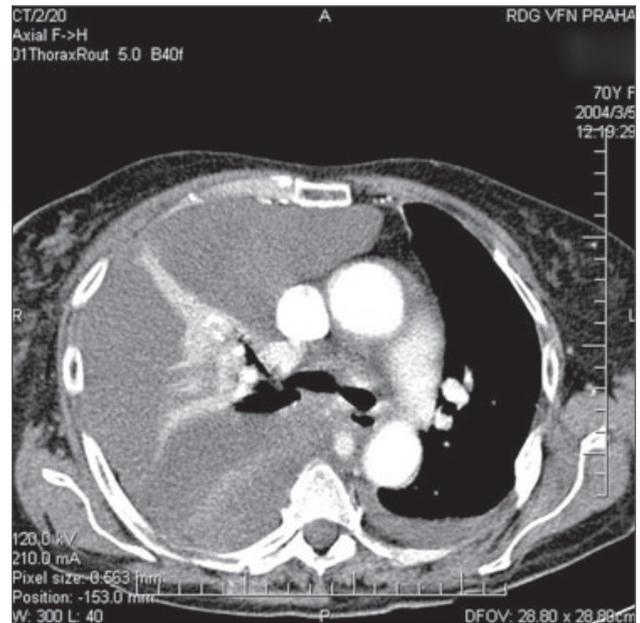


Fig. 2. CT examination again excluded an aortal lesion.

diagnosis of hemothorax. The patient was immediately transferred to the surgical department.

## Methods

During the transfer a circulatory collapse occurred and the patient underwent cardiopulmonary resuscitation. A thick thoracic drain was inserted with a marked blood outflow. The condition was complicated by unfavorable hemocoagulation results (INR 1.2, APTT 91.7, thrombin time 22, D-dimers 439, and anti-Xa 0.94). After stabilization the patient's condition (using protamine sulphate and plasma due to suspicion of heparin overdose), an angio CT was performed with negative findings. Surgical revision was indicated because of continual marked blood outflow from the right hemothorax. Approximately 1,500 to 1,700mL of blood (mainly older and new coagula) were removed from the hemothorax by right-sided posterolateral thoracotomy. The whole pulmonary ala released, as the lower lobe was fixed to the costal fracture. The apparent source of bleeding was not found. The mediastinum was significantly and sharply dilated from the level of lower pulmonary vein; it had normal width above this level. The hematoma, at the highest extent, reaches the esophagus. Both air and methylthionium chloride tests were performed with negative findings. Since the angioscan finding was negative, the esophageal lesion caused by previous fibroscopy was considered as the probable source of bleeding. The hematoma extends to the level of diaphragma. Drainage of the chest with two thick drains was performed.

Unfavorable APTT values were found after 24 hours, therefore plasma and protamine sulfate were administered. Bleeding, mainly from the surgical wound, occurred simultaneously. After the stabilization of coagulation status (a subsequent tendency to

hypercoagulation was managed with Clexane), a follow-up CT examination was performed that confirmed leakage to mediastinum with minimal coagula in the right hemothorax. Only minimal exudates were found on the left basal side. CT examination (+ angioscan) again excluded an aortal lesion (Figs 1 and 2).

Bleeding from the surgical wound continued, therefore revision of hemostasis and inspection of the drain position were performed.

A gradual reduction of blood loss from the chest was noticed one day after surgery. The patient was artificially ventilated and communication was impossible. The renal function was borderline (during forced diuresis) and circulation was relatively stable with an occasional tendency to tachycardia.

Repeated CT examinations confirmed an "expansive" process in posterior lower mediastinum, whose size was constant. The blood count was stable.

On day 7 after admission and first surgery, sudden ventricular fibrillation occurred. Cardiopulmonary resuscitation, including repeated defibrillation, was unsuccessful.

## Result

The autopsy revealed only a small volume of blood in the right pleural cavity and only a minimal volume of exudates in the left basal part of the pleural cavity. The mediastinum was filled with a hematoma upwards from the level of lower pulmonary vein. Atheromatic and calcified plaque were found in the aorta. Dissection of the aortal wall with a 15mm rupture was found in the aortal arch. This finding clearly caused the above-mentioned condition. Other autopsy findings corresponded to the diagnoses made during the patient's life and to the status after resuscitation.

## Discussion

Aortic dissection is usually caused by a fissure in the aortal intima and media (entry). Intramural blood leakage under systolic pressure results in the wall dividing and subsequently in the formation of false lumen that compresses the aortal lumen and the bases of its branches. The dissection can spread both proximally and distally and cover a large portion of the aortal diameter and length. A further fissure often forms in the peripheral part of the thoracic and/or abdominal aorta (reentry) and the false canal communicates through this fissure with the original aortal lumen. Dissection is caused by changes in media that result in reduction of the vascular wall resistance – cystic medial degeneration, congenital anomaly in various syndromes (Marfan's syndrome, Turner syndrome and Ehler–Danlos syndrome). The bicuspidal aortal valve and aortal coarctation predispose to dissection as well. Hypertension is a significant risk factor and it is associated with aortic dissection in 70 % to 90 % of patients (1). Pregnancy is associated with one half of the dissections in women aged up to 40. Approximately 20% of all dissections are located in the descendent aorta (Type B according to the Stanford classification, or DeBakey Type III). This condition is present commonly in males (3:1) and in the elderly population (aged 60 to 70) (2). Acute mortality rate ranges from 15 % to 40 %.

Pain, often shocking, localized interscapularly, is the most common clinical manifestation of aortic dissection. The pain can move to the distal parts as the dissection spreads. Concomitant hypotension may be caused by bleeding into the pleural cavity (most frequently the left cavity). The most frequent cause of death in Type B dissection is the intrathoracic aortal rupture (3). Rupture into the retroperitoneum or into the peritoneal cavity may occur as well. The closure of the lumen of the arteries by false lumen may cause intestinal ischemia and renal ischemia that can result in renal failure, paraplegia and/or ischemia of the lower limbs. Transesophageal echocardiography examination is the precise diagnostic method that may be accompanied with aortography or CT imaging, when needed.

The risk of surgical interventio overrides the risk of spontaneous course of disease in acute Type B dissection; therefore, non-surgical management (BP controls and reduction of myocardial contractility) is the most suitable method of treatment. Surgery is indicated in life-threatening complications resulting from the spreading of dissection (the clinical signs of a threatening rupture are persistent pain, uncontrolled hypertension, left-sided hemothorax and/or visceral, renal and limb ischemia or paraplegia). Recently, the less invasive implantation of endoluminal stent-graft represents an alternative for surgical treatment of descending aorta dissection (5).

This overview emphasizes the complexity of this clinical condition and management of thoracic aorta affection. It is highlighted (mainly in diagnostics) in patients in poor condition.

This case report describes an extraordinarily complication in patient with several severe diseases who was transferred to the department of surgery in a critical condition (cardiopulmonary resuscitation during admission), with severe multiorgan comorbi-

dity. Evaluation of the origin of the hemothorax involved several options – aneurysm, esophageal injury during recent fibroscopy, intrathoracic bleeding as another manifestation of a bleeding disorder (the patient suffered from skin and mucosal bleeding) associated with an unfavorable coagulation condition. The patient was in critical condition during her entire stay in the hospital and her prognosis was not favorable because of other illnesses present. Surgical revisions did not explain the condition. The hemothorax was present only on the right side and mediastinum, markedly filled by a hematoma, might be caused by several etiological factors. Repeated CT angioscans were always negative. The patient's health condition during the stay at the surgical department did not allow any major surgical procedures and she did not tolerate thoracotomy optimally.

Bleeding dissection of the thoracic aorta was found to be the cause of both the hemothorax (atypically right-sided) and the hemomediastinum. The etiology of the dissection is speculative, and a possible coincidence of the first bleeding manifestations and cytostatic agents and corticoid administration, as well as the progression of other chronic illnesses should be considered into account. Bleeding was intermittent, based on the change of blood pressure and coagulation. We can conclude that the consumption of coagulation factors during intermittent bleeding episodes could result in further bleeding and unfavorable laboratory parameters (6). The presence of dissection, and mainly its small size, was surprising, as there are no mentions in literature. However, this finding was explaining the relatively long-term course and was associated with leakage or bleeding during the pressure increase and coagulation changes. The diagnostic procedure was complicated by both the CT and angioscan failure. Transesophageal echocardiography could be beneficial in this case.

The ideal solution in case of early diagnostics – however, vague, due to patient's severe condition – would be stent-graft implantation (5, 7).

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