



Case Report

Pulmonary artery rupture during Swan-Ganz catheterisation: A case report



Raquel Vilariño Villaverde, Jessica Vanhaebost, Silke Grabherr, Cristian Palmiere*

University Centre of Legal Medicine, Lausanne, Switzerland

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ABSTRACT

Catheter-induced pulmonary artery rupture is an infrequent complication that may occur during invasive cardiopulmonary monitoring. Fatal cases are uncommon and result from hemoptysis and flooding of the opposite lung with resulting hypoxia. Alpha-1-antitrypsin deficiency is a rare genetic disorder characterised by low serum levels of alpha-1-antitrypsin, critical in maintaining connective tissue integrity. Besides pulmonary emphysema, recent observations suggest that alpha-1-antitrypsin deficiency may also be involved in vascular wall weakening, thereby predisposing arteries to dissection and aneurysm formation. In this article, we describe an autopsy case of pulmonary artery iatrogenic rupture due to insertion of a Swan-Ganz catheter in an 82-year-old woman suffering from pulmonary hypertension and alpha-1-antitrypsin deficiency. The exact source of bleeding could not be precisely identified during autopsy due to the extent of tissue hemorrhage, though postmortem angiography revealed a contrast medium extravasation from a branch of the left pulmonary lower lobar artery. The case herein emphasises the importance of postmortem angiography in facilitating the detection of vascular injuries, the importance of familiarity with intensive care techniques and procedures on behalf of forensic pathologists as well as in-depth knowledge of all possible contributing conditions and predisposing disorders in the pathogenesis of death.

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1. Introduction

Pulmonary artery rupture is an uncommon, though often lethal, complication of catheterisation during invasive cardiopulmonary monitoring. Comprehensive review of the literature has reported incidence rates averaging 0.01–0.47%, with a mortality of 50% that can reach as high as 75% in anticoagulated patients [1]. Vessel rupture usually occurs on the right side and is most common in elderly women undergoing cardiopulmonary bypass. Other predisposing factors are anticoagulation therapy and pulmonary artery hypertension. In most cases, the mechanism of injury is arterial wall damage caused by the advancing catheter tip or by normal or eccentric balloon inflation, especially when the catheter is wedged [2]. If death occurs, it is usually secondary to asphyxia rather than hypovolemic shock. Indeed, most fatal cases result from consequences of hemoptysis and opposite lung flooding with resulting hypoxia and asphyxiation [1,3].

Alpha-1-antitrypsin (AAT) deficiency is a rare genetic disorder characterised by hepatitis in neonates, childhood and adulthood and pulmonary emphysema with or without hepatitis in adulthood [4]. The disease is characterised by low serum levels of AAT, the main protease inhibitor in human serum, with clinical manifestations that may vary widely among patients, ranging from the absence of symptoms to fatal liver or lung disease [5]. Due to its anti-protease activity, AAT is critical in maintaining connective tissue integrity. Recent observations have suggested that AAT deficiency (AATD) may be responsible for nonarteriosclerotic vessel disease. AATD might predispose the arterial walls to dissection or aneurysm formation because the extracellular matrix has been inherently compromised. To date, AATD has been linked to the dissection of several arteries including the aorta, internal carotid, cervical, common iliac and coronary arteries as well as aneurysms of the splenic and mesenteric arteries [6].

In this article, we describe a case of fatal iatrogenic rupture of a branch of the left pulmonary lower lobar artery due to insertion of a balloon-tipped, flow-directed (Swan-Ganz) catheter in an 82-year-old woman suffering from pulmonary hypertension and AATD with severe pulmonary emphysema. The patient developed massive hemoptysis and died intraoperatively. Postmortem angiography allowed the source of bleeding to be detected while

* Corresponding author. Address: University Center of Legal Medicine, Rue du Bugnon 21, 1011 Lausanne, Switzerland. Tel.: +41 021 314 49 61; fax: +41 021 341 70 90.

E-mail addresses: cristian.palmiere@chuv.ch, palmierecristian@hotmail.com (C. Palmiere).

histology showed the presence of periodic acid-Schiff (PAS) positive granules in the hepatocytes, thus confirming the AATD diagnosis.

2. Case report

An 82-year-old woman with past medical history significant for pulmonary hypertension and AATD with severe pulmonary emphysema was admitted to the hospital due to progressive, worsening dyspnea at rest and shortness of breath, associated with fatigue. The patient also suffered from hypercholesterolemia, arterial hypertension and refractory atrial fibrillation, which had been treated by ablation of focal triggers (ablation of the atrioventricular node and pacemaker implantation).

Increased blood pressure was noted from the physical examination on admission. The chest roentgenogram was unremarkable, though the 2-dimensional echocardiogram showed severe tricuspid regurgitation without regional wall motion abnormalities. The right cardiac chambers were dilated with mild hypertrophy of the right ventricle.

The patient was transferred to the cardiac intensive care unit for hemodynamic monitoring. The following day, with no improvement in the patient's dyspnea, she was scheduled for pulmonary catheterisation using a right femoral approach. Catheterisation was performed under combined radioscopy and echocardiographic guidance with a Swan-Ganz catheter. The catheter was advanced and the balloon wedged into the pulmonary artery. Pulmonary capillary wedge pressure was found high after Swan-Ganz balloon inflation in a branch of the left pulmonary artery. Suddenly after Swan-Ganz deflation and retrieval, the catheter tip lunged forward, resulting in laceration of the artery. The patient started coughing and soon afterward experienced massive hemoptysis with loss of at least 200 ml of fresh blood.

Urgent anesthesiologist assistance was required. The patient was sedated and an endotracheal tube was immediately placed for airway protection. The culprit vessel was not identified and the massive bleeding could not properly be controlled. The patient died minutes thereafter despite resuscitation attempts.

A medico-legal autopsy was requested by the public prosecutor since the death occurred in the hospital and a correlation between pulmonary catheterisation, hemorrhage and fatal outcome could not be formally excluded.

A multi-phase postmortem computed tomography angiography was performed prior to autopsy and revealed an evident, intrapulmonary extravasation of contrast medium from a branch of the left pulmonary lower lobar artery. The precise localisation of the source of bleeding was therefore unidentified (Fig. 1).

External examination was unremarkable except for fresh injection marks due to medical intervention on the upper and lower limbs and cardio-pulmonary resuscitation marks on the chest.

Internal examination revealed rib fractures and intercostal space hemorrhagic infiltrations consistent with cardio-pulmonary resuscitation marks. On autopsy, the lungs were obtained en bloc with mainstem bronchi and trachea. The pericardial sac showed no adhesions and contained 50 ml of clear fluid. The heart weighed 400 g. Heart examination revealed right cardiac chamber dilatation and mild right ventricle hypertrophy. The coronary arteries had a normal anatomic course and revealed mild atheromatous disease without significant stenosis. Examination of the pleural cavities showed bilateral hemothorax (approximately 100 ml blood in each cavity). Large amounts of blood were found in the upper and lower respiratory tracts. Gross examination of the lungs revealed diffuse bilateral hemorrhages. On cut section, both lungs (right 930 g, left 900 g) showed diffuse emphysema and hemorrhagic infiltrations, especially in the left inferior lobe. The exact source of bleeding



Fig. 1. Visualisation of the hemorrhage originating from a branch of the left pulmonary lower lobar artery after the venous phase of the multi-phase postmortem computed tomography angiography. Left sagittal-oblique maximum intensity projection reconstruction showing the intra-pulmonary extravasation of contrast medium from a branch of the left pulmonary lower lobar artery (arrow) and the left pulmonary artery (star).

could not be precisely identified due to the extent of tissue hemorrhage. Pulmonary embolism was not observed. The spleen, liver, kidney and brain did not show any significant, macroscopic changes.

Sections of most of the organs were examined microscopically. Neuropathology was unremarkable. The heart revealed right ventricular hypertrophy with no evidence of subendocardial hemorrhage or acute myocardial infarction. The lungs showed generalised alveolar septal wall destruction and diffuse hemorrhagic infiltrations. Pulmonary artery examination (hematoxyline eosine stain) failed to reveal structural abnormalities. Periportal round to ovale eosinophilic globules, characteristic of AATD, were identified in the liver with PAS stain (Fig. 2).

Femoral blood, vitreous humor and cerebrospinal fluid as well as gastric content, hair and tissue samples were recovered for toxicology and biochemistry. Toxicological analyses were performed on blood. These analyses included ethanol determination as well as screening for common drugs and illegal substances by gas chromatography–mass spectrometry (GC–MS), high-performance liquid chromatography with diode-array detection (HPLC–DAD) and headspace-gas chromatography flame ionisation detection (HS–GC–FID). The results of the toxicological analysis were negative for ethanol and all screened drug substances. Postmortem biochemical investigation results were not contributory.

Based on the clinical information as well as postmortem investigation findings, the cause of death was determined to be hemorrhagic shock and massive aspiration of blood into the airways due to the rupture of a branch of the left pulmonary lower lobar artery following pulmonary catheterisation. The death was classified as a therapeutic accident resulting from pulmonary artery laceration during the Swan-Ganz catheterisation.

Although Swan-Ganz catheter manipulation was estimated to be the main factor leading to the vascular rupture, AATD was postulated to have played a role in the pathogenesis of death by possibly compromising the extracellular matrix and weakening the arterial wall, thereby predisposing or facilitating the rupture.

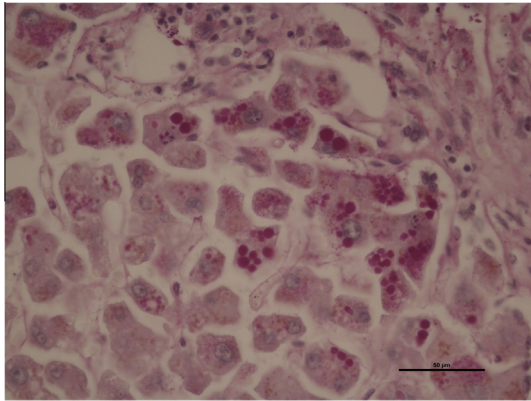


Fig. 2. Periodic acid-Schiff (PAS) stain of the liver showing intracytoplasmic pink globules (original magnification 40 $\times$).

3. Discussion

The concept of right heart catheterisation was first introduced by Dr. Warner Forrsmann in 1929 though it was in 1970 that Dr. H.J. Swan and Dr. W. Ganz introduced the flow-directed balloon-tipped catheter that led to a paradigm shift in the way right heart catheterisations are performed at the bedside using intracardiac pressure tracings, without utilizing fluoroscopic guidance. Since then, the pulmonary artery catheter, also called a Swan-Ganz catheter, has been utilised in the management of intensive care unit patients for the past 42 years and has become integrated with the diagnostic and therapeutic modalities for patients with severe cardiopulmonary diseases in adult and pediatric intensive care units [7–11].

The direct measured hemodynamic data available from the pulmonary artery catheter include the pressures in the right atrium, right ventricle, pulmonary artery and pulmonary artery occlusion pressure as well as mixed venous oxygenation and temperature. A large array of calculated information is available from these variables and three other measured variables (namely systemic arterial pressure, heart rate and cardiac output) including pulmonary and systemic vascular resistance, right and left ventricular stroke work indices, right and left ventricular end-systolic and end-diastolic indices, right ventricular ejection fraction, arterial and venous oxygen content, oxygen consumption, oxygen delivery and oxygen extraction ratio. These measured data are used to guide the treatment of critically ill patients [7,9].

In pulmonary artery catheterisation, the balloon-tip catheter is floated through a central venous access, through the right atrium and right ventricle to the pulmonary artery and left in position to measure the filling pressures of the heart. When the balloon is inflated, it measures pulmonary capillary wedge pressure or occlusion pressure, which is an indirect measure of left ventricular end-diastolic pressure. Despite current controversies regarding the use of pulmonary artery catheters in the intensive care unit, hemodynamic data are valuable in the care of critically ill patients with pulmonary hypertension. Severe tricuspid regurgitation and elevated pulmonary artery pressure often make placing a pulmonary artery catheter challenging, and may necessitate the use of fluoroscopy [7,9].

Insertion of a pulmonary artery catheter requires a central venous access with complications mainly related to line placement. Additional risks include possible pulmonary artery rupture and subsequent bleeding or pulmonary infarction. Though major morbidity related to pulmonary artery catheter seems uncommon, minor atrial and ventricular arrhythmias are frequent during catheter insertion [7,12].

Complications from pulmonary artery catheter can be classified as:

1. Those from central venous access (arterial puncture, postoperative pain and sensation deficit, air embolism and pneumothorax), reported in less than 3.6%.
2. Those arising from catheterisation (severe dysrhythmias, right bundle branch block and complete heart block), seen in 0.3–3.8%.
3. Those due to prolonged catheter residence (pulmonary artery rupture, pulmonary infarction, venous thrombosis), seen in 0.03–3%.

The overall death attributed to a pulmonary artery catheter is estimated from 0.02% to 1.5%. However, mortality as a result of pulmonary artery rupture can be as high as 50% [3,7].

The clinical presentation of pulmonary artery rupture varies from being asymptomatic to the development of minimal hemoptysis to massive pulmonary hemorrhage with ensuing hemodynamic instability. Hemothorax, although extremely rare, can also occur as a complication of pulmonary artery rupture if blood enters the pleural space.

The pathophysiologic mechanisms of iatrogenic pulmonary artery rupture include the following:

- Lodgment of the catheter tip in the arterial wall.
- Eccentric balloon inflation with inadvertent insertion of the catheter tip in the vascular wall.
- Catheter tip migration into small arterioles.
- Wedged balloon retraction.
- Wedged balloon flushing.
- Transmission of cardiac forces to the arterial walls through the catheter tip causing rupture [8,13,14].

Few reports are available in medico-legal literature describing fatal complications of pulmonary artery catheterisation. Lieske et al. [15] reported two fatal cases of pulmonary artery perforation in association with the use of Swan-Ganz catheters in 71 and 95 year-old women who were monitored by a flow-directed catheter pre- and intraoperatively. Resnick et al. [16] described one of the first cases in forensic pathology literature in which a postmortem angiography was performed and allowed the exact source of bleeding to be identified. The authors obtained the lungs en bloc during autopsy with mainstem bronchi and trachea. A Council catheter was inserted into the trachea and the lungs were inflated with air. A second Council catheter was placed in the right pulmonary artery and the iodinated water-soluble contrast material was injected for postmortem angiography, which allowed a perforation of the proximal medial segmental artery of the right middle lobe to be demonstrated. Lastly, De Leeuw and Jacobs [17] reported the unique case of a through-and-through perforation of the left carotid artery with a pulmonary artery catheter due to the incorrect positioning of the Swan-Ganz introducer. According to the reconstruction proposed by the authors, the introducer entered the skin anterior of the left sternocleidomastoid muscle, perforated the left carotid artery and was deflected on the lateral side of the cervical spine, with the tip of the introducer ending 5 mm from the left subclavian vein. The pulmonary artery catheter was then inserted through the introducer, passed by chance in the left subclavian vein and further followed its normal route with the catheter tip in situ in a branch of the left pulmonary artery. The authors concluded their report by emphasizing the importance of leaving all invasive medical devices in situ on a deceased person when a medico-legal autopsy is to be expected as well as the usefulness for forensic pathologists to be acquainted with intensive care techniques and procedures and emergency medicine. The rele-

vance of mutual interaction between pathologists and practitioners in documenting all significant medico-legal information for event reconstruction was also stressed.

In the case herein presented, a further important aspect to consider was the role that may have been played by preexisting diseases, particularly AATD, in the development of vascular damage and arterial wall rupture.

AATD is an under-recognised hereditary disorder first described by Laurell and Eriksson in 1963, after they linked the absence of an α_1 -globulin band on serum protein electrophoresis to the presence of premature emphysema. AAT is a 52 kDa, single-chain glycoprotein with a 394 amino acid sequence, which is synthesised predominantly in the liver. The protein functions as a serine proteinase inhibitor or serpin, providing essential protection to lung tissue against the actions of proteolytic enzymes such as neutrophil elastase and proteinase 3. Matrix metalloproteinases and cysteine proteinases have also been implicated as having a direct role in proteolytic alveolar destruction. The premature onset of pulmonary emphysema was the first identified clinical manifestation of AATD, which can also be associated with varying degrees of airflow obstruction, even varying within individual patients. Emphysema and airflow obstruction often coexist. However, subsets of patients with marked emphysema and preserved spirometry or severe airflow obstruction with relatively little parenchymal destruction are identifiable [18–20].

AAT achieves protease inhibition by forming a 1:1 complex with multiple different proteolytic enzymes. In the setting of AAT deficiency, a disequilibrium exists between protease and antiprotease activity, which may precipitate pathological consequences. Investigators have postulated that, besides promoting connective tissue degeneration in the lungs, excessive proteolytic activity may affect arterial wall integrity. Indeed, elastin is a major component of the elastic lamina that sustains blood vessel integrity, closely influenced by elastase levels. The breakdown and reduction of elastic fibres could therefore play a role in vessel tone loss and consequently be implicated in the development of vascular damage. Evidence both for and against this hypothesis is present in the literature. Abdominal aortic aneurysm, spontaneous dissection of the thoracic aorta, coronary artery dissection, intracerebral aneurysms and cervical artery dissection have all been reported to occur more commonly in patients with AAT deficiency. Other authors have discounted this association, although elevated levels of plasma elastase not associated with AATD may correlate with aneurysm formation [21–25]. In an animal model, Tsuji et al. [21] observed that the circulating AAT–elastase complex was incorporated into the elastic lamina of the arterioles through the endothelial layer, resulting in liquefaction of the lamina, desquamation of endothelial cells and leakage of the complex into perivascular tissues via the vascular walls. Vizzardi et al. [22] described enlarged aortic diameters, reduced aorta distensibility and increased aortic wall stiffness in subjects suffering from AATD. Schachner et al. [26] observed that AAT amounts were reduced in the vascular wall of ascending aortic dissections compared to healthy aortas, suggesting that local AATD in the human ascending aorta might lead to proteolytic damage thus facilitating aortic dissection.

In the case herein discussed, histology of the aorta and pulmonary artery failed to reveal particular features suggesting structural abnormalities that could have predisposed vessel wall weakening and vascular rupture, such as elastic tissue destruction with inflammatory cell infiltration. However, circulating elastase and AAT were not assayed. We could therefore not formulate conclusive hypothesis concerning the specific role of AATD in the pathogenesis of vascular rupture and patient death. It must be emphasised, however, that pulmonary artery ruptures during pulmonary artery catheterisation have been reported, to present, in individuals who did not suffer from any underlying enzymatic

deficiency affecting the vascular wall, which would not be necessary to explain the sequence of events or the fatal outcome following the vascular injury.

Lastly, the case herein reported emphasises the usefulness of postmortem angiography in order to detect bleeding sources with precision. This is paramount in cases of acute hemorrhage with fatal outcomes, when the affected vessels are of small caliber, when the injury site cannot be identified on gross examination alone or when the injured vessels are located in anatomical areas of the body difficult to access. Indeed, the detection and precise identification of the vessel responsible for bleeding are of utmost importance in the practice of forensic pathology, especially when procedural mishap is a concern. In the case presented, the value of postmortem angiography in the diagnosis of catheter-induced pulmonary artery perforation and the precise localisation of the perforation was evident.

To conclude, pulmonary artery rupture is a life-threatening and potentially lethal complication following pulmonary artery catheterisation. Some of these deaths may fall under the purview of the inquiring authorities and forensic pathologists due to their in-hospital outcome. By facilitating the detection of the vascular injury, postmortem angiography may assist pathologists in documenting the manner and cause of death. Knowledge of intensive care techniques and procedures as well as emergency medicine can also assist forensic pathologists in understanding and reconstructing the exact sequence of events. Lastly, all possible contributing conditions and predisposing disorders must be carefully considered in order to formulate appropriate hypotheses concerning the pathogenesis of death.

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