Case report

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Lessons and insights on diagnostic challenges in mycotic aneurysm: A case report of a rare and lethal disease

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Abstract

The diverse clinical presentations of mycotic aneurysms often present challenges in achieving early clinical diagnosis. Clinically, evident mycotic aneurysms frequently manifest at an advanced stage of progression or are concurrent with complications, such as rupture. In this report, we aim to discuss the mortality of a 68-year-old woman who had a rare ruptured mycotic aneurysm. Initially, the patient presented to the emergency department (ED) due to a sudden, sharp, stabbing sensation in the middle chest area, radiating to her back. An electrocardiogram (ECG) revealed sinus rhythm without ST-T changes. Chest X-ray (CXR) showed an enlarged aorta, which led to a diagnosis of suspected aortic dissection. During close monitoring in the Intensive Care Unit (ICU), the patient's blood pressure dropped, and peripheral oxygen saturation was 70%. After urgent intubation and resuscitation, the patient was finally stable. Another CXR was performed and found massive pleural effusion; prompt pleural aspiration found the presence of blood. It was suspected that the suspected aortic dissection had ruptured causing massive left hemothorax. Computed tomographic angiography (CTA) was done but did not find evidence of aortic dissection. After a thorough discussion, the attending thoracic surgeon was to perform an exploratory thoracotomy. During preparation, the patient went into cardiac arrest. Adequate resuscitation was performed. Unfortunately, the patient was pronounced dead soon after. It was determined that mycotic aneurysm rupture was the underlying cause. This study showed a significant occurrence of aneurysm ruptures, underscoring the critical need for achieving early diagnosis of mycotic aneurysm.

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Introduction

Mycotic aneurysm is a very uncommon but highly consequential disease characterized by the development of aneurysmal dilation in the aorta as a consequence of infection. Mycotic aneurysms account for only 1% to 2.6% of all aortic aneurysms, while the occurrence of spontaneous rupture of mycotic aneurysms ranges from 7% to 24% of all mycotic aneurysms, with 47% to 61% of cases presenting as either a confined or imminent rupture. Ruptured mycotic aneurysms carry a significant risk of mortality (63% to 100%). (1,2) The myriad presentations exhibited of mycotic aneurysms present a formidable challenge when it comes to achieving early clinical diagnosis. Clinically evident mycotic aneurysms often manifest themselves in an advanced stage of progression or are concomitant with complications, such as rupture. However, as mycotic aneurysms are rarely encountered and few literatures are available on the topic of mycotic aneurysms, we would like to report on our difficulties and experiences in diagnosing and managing mycotic aneurysms to add to the pool of available literature. In this report, we provide a case study involving the death of a 68-year-old woman who had a ruptured mycotic aneurysm.

Case report

A 68-year-old woman presented to the emergency department with a complaint of chest pain that had onset three hours prior to her admission to the hospital. The chest pain was characterized by a sudden, sharp, stabbing sensation, primarily located in the middle area and radiating to her back, and it progressed gradually. She had a history of hypertension and no relevant family history. Upon admission, the chest pain persisted, and her vital signs included blood pressure (BP) of 190/100 mmHg, heart rate (HR) of 98 beats per minute (bpm), respiration rate of 18 times per minute, and a peripheral oxygen level of 99%. Additionally, she had a marked fever with a temperature of 38.5 °C. On examination, a bruit was noted on auscultation, with normal pulmonary sounds. The electrocardiogram (ECG) revealed sinus rhythm without ST-T changes. The chest X-ray (CXR) showed an enlarged aorta and mediastinum, leading to a working diagnosis of a suspected aortic dissection (**Figure 1**). Initial laboratory findings included leukocytosis with a white blood cell (WBC) count of 22,650 cells/mm³ and a normal hemoglobin level of 12 g/dl. The patient was prescribed amlodipine 10 mg once daily and bisoprolol 2.5 mg twice daily, as well as an injection of meropenem 1g three times daily. Computed tomography angiography (CTA) could not be performed at that moment as there was ongoing chest pain and high BP. Therefore, the patient was transferred to the intensive care unit (ICU) for further monitoring.

In the ICU, the patient complained of escalating intense sharp chest pain with a visual analog scale (VAS) of 8, BP of 124/85 mmHg, and HR of 87 bpm. Five mg of nitrate was given sublingually, followed by an intravenous drip of nitroglycerin. Nevertheless, it was ineffective in mitigating the chest pain. We contemplated administering morphine to relieve the pain, but the patient's BP dropped to 77/55 mmHg, followed by desaturation, with peripheral oxygen levels dropping to 70%. On auscultation, bruit was detected, along with decreased lung sounds in the left hemithorax. Immediately, the emergency protocol was initiated, the patient was intubated, connected to a ventilator within 5 minutes, and given intravenous inotropic support. Immediate CXR evaluation revealed a massive left pleural effusion (**Figure 2**). The hemoglobin level had decreased to 7.9 g/dl. Subsequently, diagnostic pleural puncture aspiration was promptly performed, revealing gross blood during the aspiration, confirming the presence of massive left hemothorax likely caused by suspected aortic dissection rupture. To address this critical situation, the patient was resuscitated with a bolus of 500 cc of gelofusine infusion, 2 units of packed red cells, and 3 units of fresh frozen plasma. After close monitoring, the resuscitation was successful, and the patient was hemodynamically stable, eliminating the need for inotropic support. CTA of the aorta revealed the presence of a saccular aneurysm in the superior aspect of the aorta bifurcation, with the presence of mural calcification (**Figure 3**). Intriguingly, the imaging did not manifest any indications of aortic dissection, thereby engendering a discordance between the patient's clinical presentation of a suspected aortic dissection and the radiological diagnostic findings. We decided to do a clinical pathological conference between the cardiologist, thoracic, vascular surgeon, intensivist, pulmonologist, and radiologist with the objective of formulating an appropriate treatment plan in light of the inconclusive diagnosis. Subsequently, a collective decision was reached to proceed with an exploratory thoracotomy for the purpose of identifying the precise source of hemorrhage. Subsequent to this identification, aortic repair would be undertaken, with a consideration of additional pleural water shield drainage placement if the patient's hemodynamic condition was unstable.

During the preparation for surgery on the same day, the patient encountered an abrupt episode of lost

consciousness and a gradually worsening bradycardia from 50 bpm to 30 bpm, followed rapidly by a pulseless electrical activity (PEA). Cardiopulmonary resuscitation (CPR) and cardiac arrest algorithm were immediately initiated, leading to the return of spontaneous circulation (ROSC). However, ten minutes later, the ECG showed PEA again. Despite resuscitation efforts for over half an hour, the patient remained unresponsive. ECG showed asystole and the presence of completely dilated pupils, and the patient was pronounced dead. This critical event prompted a meticulous assessment of the contributing factors underlying the patient's death. It was determined that the rare event of a mycotic aneurysm rupture was the underlying cause, as elucidated in the subsequent section for further elaboration. Informed consent was obtained from the patient's family.

Discussion

The exceedingly rare occurrence of mycotic aortic aneurysms complicates efforts to identify the condition and investigate it statistically. Moreover, its management is challenging and entails a high mortality rate. Mycotic aneurysms are localized, irreversible arterial dilatations brought on by the invading organism weakening and destroying the artery wall and causing infective arteritis. The occurrence rate of mycotic aneurysms is exceptionally low, ranging from 1% to 2.6%. However, it can be life-threatening. The term 'mycotic' refers to the mushroom-like appearance of the aneurysms that were first described, rather than their underlying microbiological etiology. (3)

There is no universally accepted diagnostic procedure or set of criteria for mycotic aneurysm. It can be suspected based on a combination of clinical, laboratory, imaging, and intraoperative data. The clinical presentation of mycotic aneurysms is typically vague and depends on the location and intensity of infection, comorbidities, and aneurysm size. (4) An individual may present with a variety of symptoms, although fever (75%), chest and back pain (60%), stomach discomfort (20%), and chills (16%) are the most prevalent. An infected thoracic aortic aneurysm is associated with chest and interscapular pain, and abdominal aortic aneurysms are associated with abdominal pain. (5) However, in certain cases, symptoms may not be present at all. (6) Furthermore, expansion of the mycotic aneurysm may cause compression to surrounding structures. If the aneurysm compresses the esophagus or trachea, it may cause dysphagia, dyspnoea, hoarseness, cough, or superior vena cava syndrome. An aneurysm may also cause Ortner's syndrome (cardio-vocal syndrome) if compression occurs to the recurrent laryngeal nerve. Thus, presenting symptoms are generally unspecific and a high index of suspicion is needed especially in high-risk populations, especially when accompanied by relevant physical findings. (7)

Physical examination may reveal signs of localized inflammation, characterized by a painful and hardened mass, as well as the presence of a bruit during the examination, which is observed in approximately 50% of cases. (8) Leucocytosis along with elevated levels of erythrocyte sedimentation rate and C-reactive protein (CRP), are often detected. Typically, the initial investigation to identify the source of infection yields negative results. Consequently, mycotic aneurysms become significant as a potential differential diagnosis in patients presenting with pyrexia of undetermined origin or chronic bacteremia without an apparent cause. (3,6)

In the event of a delayed diagnosis, a mycotic aneurysm has the potential to present with severe septicemia or serious complications resulting from the rapid enlargement, rupture, and bleeding of the aneurysm. Infection may spread to the surrounding structures. In a study by Lin et al., 8 patients (7%) with infrarenal mycotic aneurysms had concomitant psoas abscesses. (5) While dissections of the abdominal or descending aorta may result in renal failure if the renal arteries are blocked. Dissections of the ascending thoracic aorta may result in abrupt acute aortic regurgitation, myocardial ischemia or infarction, or cardiac tamponade. Rupture leads to significant internal bleeding and subsequent hypovolemic shock. (9)

When diagnosing mycotic aneurysms, radiologic evidence offers the best sensitivity and specificity. CTA remains the preferred imaging technique for evaluating mycotic aortic aneurysms, with a sensitivity ranging from 92% to 96% and a specificity ranging from 93% to 100%. (10) Other advantages include the detection of simultaneous or source lesions and the use of three-dimensional modeling for intervention planning. An infected aortic aneurysm can be identified as a focal, contrast-enhancing dilatation that is usually saccular. This is one of the main differences compared to atherosclerotic aneurysms which are usually more fusiform. Other strong diagnostic signs suggesting mycotic aneurysms include multi-lobulated appearance, soft tissue inflammation around the vessel wall, which appears as perivascular contrast enhancement, intramural air, accumulation of air around the blood vessel, and perianeurysmal fluid collection. Extravasation of high attenuating fluid suggests hematoma, suggesting that a rupture has already occurred. This

may be more evident in abdominal aortic aneurysms as fluid may spread to the pararenal space, perirenal space, and peritoneal cavity. The risk of rupture is related to the aneurysm diameter and rate of expansion; no other imaging characteristics can accurately predict the outcome. (11)

Magnetic resonance angiography (MRA) is a potential alternative modality; however, its use is currently limited due to longer examination durations and a higher susceptibility to motion artifacts, as flow-related artifacts frequently appear in dilated lesions. Several techniques are developed to provide a better temporal view and reduce motion artifacts such as cardiac-gating and respiratory-gating; these techniques involve timing the image acquisition to specific times in the cardiac contraction cycle or the breathing cycle. Nevertheless, it is notably advantageous in the context of cerebral lesions, with a sensitivity ranging from 95% to 100% and a specificity ranging from 82% to 96%. It also provides several advantages such as no radiation exposure, low contrast medium adverse effect, and easy image post-processing. (12) T1-weighted, fat suppression MRA may increase the conspicuity of hemorrhagic lesions, making acute hemorrhages appear hypointense (dark). Findings suggestive of the presence of an aneurysm are hypointense lesions in the pleural cavity, mediastinum, retroperitoneal, or intramural. (13) Invasive aortography can only image the vessel lumen and not extravascular changes; therefore, it is reserved for patients in whom mycotic aneurysms cannot be ruled out by non-invasive tests. It also carries risks of distal embolization and rupture of the already fragile inflamed arterial wall. (14)

The management of unruptured mycotic aneurysms remains contentious due to the absence of randomized controlled trials and consensus guidelines. The range of strategies that may be used includes conservative care which involves the use of antimicrobial medication along with regular monitoring, as well as endovascular intervention and surgical intervention. Expert opinion leans towards surgical or endovascular procedures, combined with intensive antibiotic therapy. (15) Antibiotic therapy given is based on antibiotic sensitivity of the causative organism. (11) Broad-spectrum antibiotics should be initiated early, using agents that may combat Salmonella species such as chloramphenicol, ampicillin, quinolone, or a third-generation cephalosporin. Recommendations for the duration of antibiotics vary from 6 months to 12 months and even lifelong treatment. However, such considerations are made on an individual basis. (16) Lee et al. suggested the use of 4-6 weeks of initial antibiotic therapy when non-emergent surgery is needed, as pre-operative antibiotic treatment may decrease morbidity and mortality. (11,16) Surgical management remains the mainstay treatment using either open surgical repair or endovascular repair accompanied by intensive antibiotic therapy, and only a selected few may resolve using just antibiotic therapy. (7,16)

This case underscores the critical importance of early consideration of mycotic aneurysms in patients with relevant clinical profiles, as delayed diagnosis elevates the risks of rupture and mortality. The absence of distinctive presenting features, the inability to palpate the aneurysm, and the limited sensitivity of chest X-rays often lead to the underdiagnosis of mycotic aneurysms. Currently, there exists no singular diagnostic test or algorithm for mycotic aneurysms. Consequently, the diagnosis relies on a combination of criteria encompassing clinical presentation, laboratory findings, and radiological findings. Clinical presentation such as the presence of symptoms like pain, fever, or signs of infection, particularly in elderly individuals with cardiovascular ailments or immunosuppressive conditions, Laboratory assessments play a crucial role, with elevated inflammatory markers such as C-reactive protein, leucocytosis, and positive blood cultures providing valuable insights, and radiological examinations, notably contrast-enhanced CTA or MRA, serve as essential components of the diagnostic process. (7)

Conclusion

The diagnosis of mycotic aneurysms presents continual clinical challenges, with CTA serving as the preferred diagnostic modality in conjunction with clinical and laboratory features. Identifying mycotic aneurysms can be intricate, as their symptoms can mimic those of other conditions, like atherosclerotic aneurysms. Therefore, it is important to always maintain a high index of suspicion for high-risk populations such as intravenous drug users or immunodeficient patients presenting with fever and other signs and symptoms that may lead to mycotic aneurysm. This heightened awareness is pivotal in preventing misdiagnosis and ensuring early and appropriate initiation of antibiotic therapy or necessary surgical management.

Figure 1. Initial chest X-ray showed an enlarged aorta and mediastinum

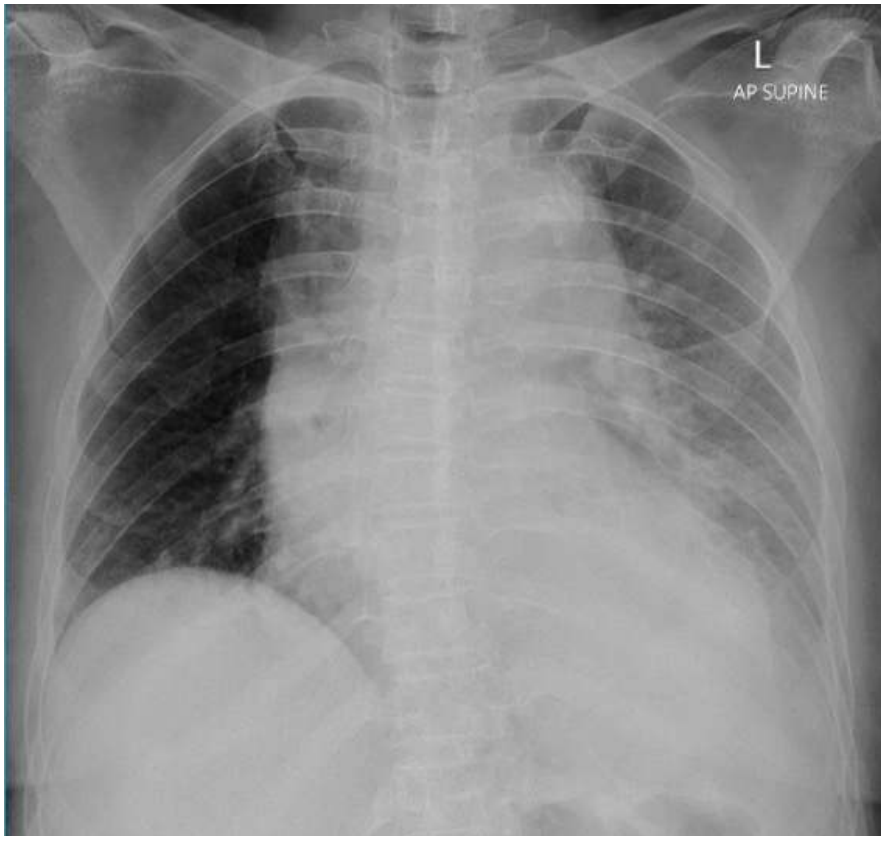


Figure 2. Chest X-ray evaluation revealed a massive left pleural effusion

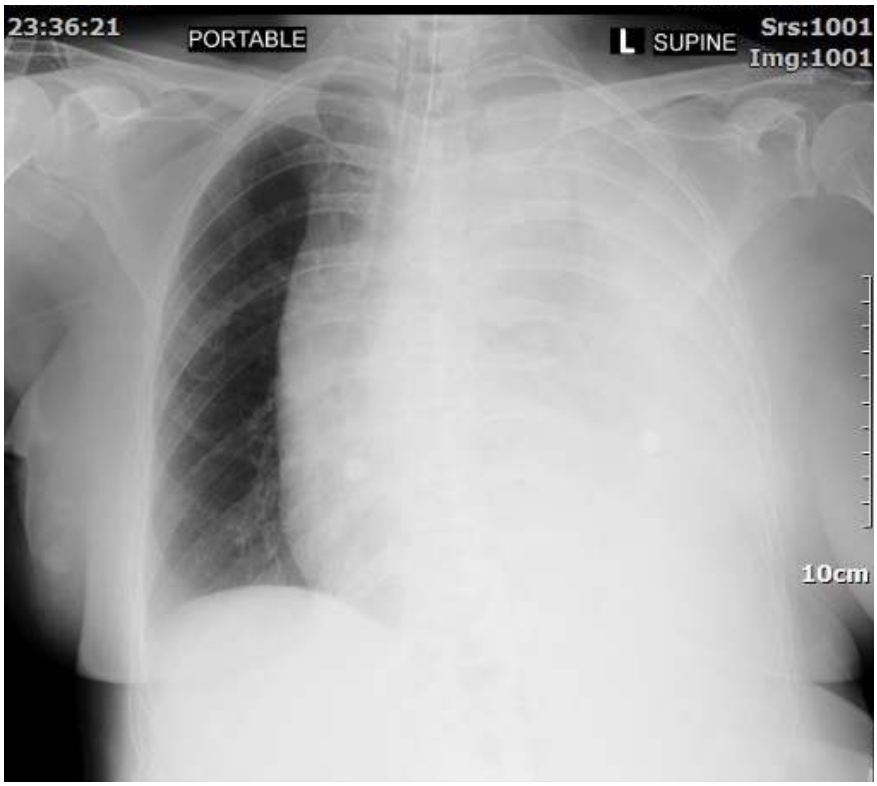
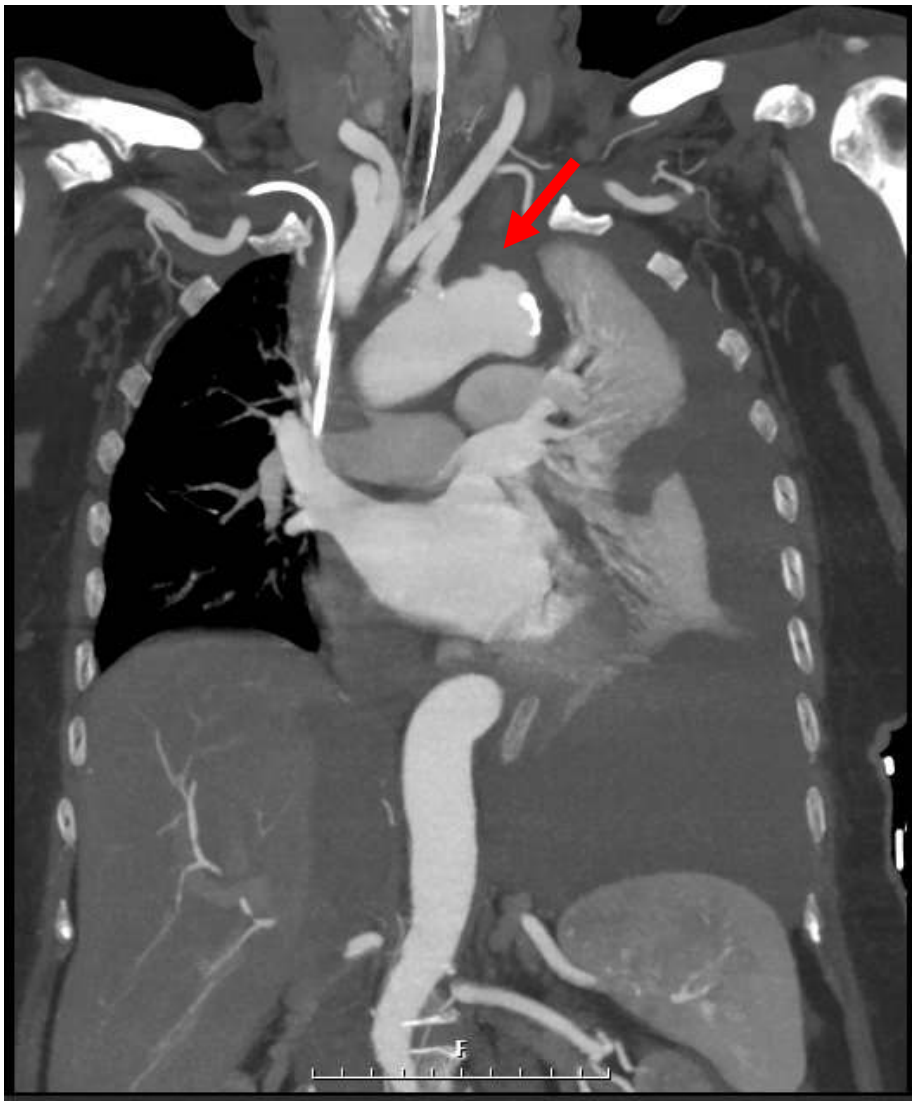


Figure 3. Computed tomographic angiography



Legend: A mycotic aneurysm (red arrow) on the superior aspect of the abdominal aorta with a diameter of 1.9 cm, length of 2.5 cm, diameter of inlet of 1.3 cm, and outlet of 1.3 cm. Note the presence of calcification and left massive pleural effusion.

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