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RESEARCH ARTICLE

MYCOTIC ANEURYSM OF INTERNAL ILIAC ARTERY CAUSED BY *Salmonella* spp. INFECTION: A CASE REPORT

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Abstract

Mycotic aneurysm (MA) is an extremely rare disease caused by secondary infection, one of the most common causes is *Salmonella* spp. A male patient with type 2 diabetes mellitus presented with edema on the left lower limb, fever, abdominal pain radiating to the back, and was suspicious of Deep Vein Thrombosis (DVT). Laboratory examination showed leukocytosis and elevated inflammatory markers such as C-reactive protein and procalcitonin, with *Salmonella* spp. detected in the blood culture, and the image of MA with thrombus presented on the run-off CT scan with contrast. The patient was scheduled to undergo Endovascular Aneurysm Repair (EVAR) procedure. However, he suddenly had a cardiac arrest on the sixth day of treatment before surgery, caused by suspected pulmonary embolism with suspected DVT. MA has a high mortality rate (around 40%) with a high risk of aneurysm rupture, however, pulmonary embolism has been reported as a cause of death in MA patients with DVT. In addition to rapid diagnosis, aggressive yet adequate therapy, namely surgery and long-term intravenous antibiotics, is crucial.

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Introduction:-

Infected/Mycotic Aneurysm (MA) is a condition where an artery dilates caused by infection. It is called "mycotic" for its appearance which resembles a fleshy fungus. It can be caused commonly by bacteria, but it can also be caused by fungi or viruses. The most common etiology of MA in East Asia is *Staphylococcus aureus* and *Salmonella* spp. infection (Guo et al., 2018). *Salmonella* spp. infection can develop bacteremia in 5% of the cases (Sörelus et al., 2019). The risk factor for MA is immune-compromised host, which can be caused by several diseases, such as diabetes, patients receiving chemotherapy/high dose glucocorticoids, and HIV (Majeed and Ahmad, 2023).

Patients can come with various symptoms, mostly related to non-specific gastrointestinal complaints, such as nausea and vomiting, hemoptysis, gastrointestinal bleeding symptoms, or even heart failure symptoms (Ram, Deslouches and Punnapuzha, 2022). Gastrointestinal symptoms are usually associated with *Salmonella* infection (Guo et al., 2018). Some patients also experience chills, fever, and pain at the aneurysm location (Guo et al., 2018). Tender indurated abdominal mass and bruit may be found during physical examination. Laboratory results may vary, elevated white blood cell count, especially neutrophils, and inflammatory markers, such as C-reactive protein and dynamic erythrocyte sedimentation rate may be found (Guo et al., 2018; Ram, Deslouches and Punnapuzha, 2022). Positive

culture results from the aneurysm wall may show the bacteria causing the MA (Guo et al., 2018). The most sensitive and specific diagnostic resource is CT Angiography (CTA) (Ram, Deslouches and Punnapuzha, 2022).

MA has a poor prognosis, for its high risk of rupture during the peri-operative period (Sörelus et al., 2019). It is because the wall of MA consists only of hematoma, inflammatory material, and inflamed/thickened perivascular soft tissue (Raman and Fishman, 2014). The mortality rate after Open Surgical Repair (OSR) is 21,43%, and 7,14% for Endovascular Aneurysm Repair (EVAR). There is also a 17,85% recurrence rate of infection after EVAR. Specifically, the mortality rate of MA caused by *Salmonella spp.* infection is high in middle-aged males with hypertension, diabetes, and/or atherosclerosis (Guo et al., 2018).

In this case report, we present a patient with MA in the internal iliac artery caused by *Salmonella spp.* infection.

Case Report

A 50-year-old male came to the ER with swollen left leg for two weeks, a fever, and abdominal pain radiating to the back. The patient had a history of naïve diabetes mellitus. The vital signs were obtained: blood pressure of 129/83 mmHg, heart rate of 89 bpm, temperature of 37,2°C, and oxygen saturation of 94% breathing room air. There were some significant findings during physical examination, such as pulsating mass at the umbilical region with no sign of peritoneal irritation, and strong pulsation of the dors pedal artery and posterior tibial artery.

Several laboratory examinations were conducted, with the results of leukocytosis (15.770 leukocytes/uL), high C-reactive protein (> 200 mg/L), high procalcitonin with a high risk of severe sepsis (6,48 nG/mL), and high level of D-dimer (4372). The blood culture showed positive for *Salmonella spp.* The patient was assessed with suspected typhoid fever and Deep Vein Thrombosis (DVT), then received initial therapy with double antibiotics, cefoperazone-sulbactam and levofloxacin, which were sensitive to *Salmonella spp.*, and enoxaparin sodium 0,6 mg subcutaneously every 12 hours for the suspected DVT.

The patient then underwent Run-off Computed Tomography Angiography (CTA) which showed an infected mycotic fusiform aneurysm starting 1 cm from the origin of the internal left iliac artery with a mural thrombus of 1 cm thick (see Figure 1). The patient was planned for Endovascular Aneurysm Repair (EVAR) surgery, however, on the sixth day of treatment, he experienced cardiac arrest before surgery with the cause of death suspected of pulmonary embolism as a complication of the suspected DVT.



Figure 1:- Infected mycotic fusiform aneurysm sized 8 x 5,5 x 4 cm (Run-off CTA – Artery).

Discussion:-

Mycotic aneurysm is a disorder that arises from damage to the walls of the arteries caused by secondary infection. The most common cause is a hematogenous infection, one of which is by *Salmonella spp.* (Nídia et al., 2018). The patient in this study was a 50-year-old male with diabetes mellitus and suspected DVT. A study by Guo et al. (2018) showed that the mortality rate of MA caused by *Salmonella spp.* infection is high in middle-aged males with diabetes and/or atherosclerosis. Patients with diabetes are at risk of developing MA because they are considered immune-compromised (Majeed and Ahmad, 2023). Therefore, *Salmonella spp.* infection, which normally only causes mild self-limiting gastroenteritis, may develop bacteremia that can lead to endovascular infection. It is because *Salmonella* has a strong affinity to attach to damaged vessel walls, causing inflammation, destruction, localized abnormal dilation, and finally mycotic aneurysms. This patient was at high risk of having damaged vessel walls since he was diabetic (Suppamutharwyam and Radhakrishnan, 2022). It is important for clinicians to notice the patients with enhanced risk factors to develop *Salmonella* bacteremia, as shown in Table 1.

Table 1: Patients with enhanced risk factors for developing *Salmonella* bacteremia (Suppamutharwyam and Radhakrishnan, 2022).

Category	Risk Factor
Non-modifiable	Extreme age group (Aged > 65 years old and neonates)
Cardiovascular	Chronic smoking Hypertension Poorly controlled diabetes mellitus Stroke Hyperlipidaemia Congestive heart failure
Immunocompromised State	Long-term steroid usage Rheumatological disease Liver cirrhosis End-stage renal failure Acquired immunodeficiency syndrome Active malignancy Recent chemotherapy

DVT of the patient can also be a manifestation of the *Salmonella* bacteremia. *Salmonella* bacteremia can cause hypercoagulability in the patient's body, and furtherly causes thrombosis, which may be seen as DVT. Another mechanism that may cause this condition is the compression of the iliac vein by the MA of the same side, which can induce venous stasis and thrombosis (Suppamutharwyam and Radhakrishnan, 2022).

The patient showed symptoms of swollen unilateral leg, fever, and abdominal pain radiating to the back. A swollen leg is a symptom of DVT (Rajah, Mustafa and Payus, 2020). Fever, especially long high-grade fever with associated chills and rigor is often found in MA patients. Furthermore, abdominal and/or back pain is also associated with MA (Kordzadeh, Watson and Panayiotopolous, 2016; Kumar et al., 2023). The abdominal pain is often located in the epigastrium or lower quadrant area. In this case, the abdominal pain can be a sign of the MA, since the MA is located in the left iliac artery, which is located in the abdominal area. Other symptoms that may be associated with MA are malaise, weight loss, and nausea (Kordzadeh, Watson and Panayiotopolous, 2016; Kumar et al., 2023). A pulsatile mass in the umbilical region was found during the physical examination of the patient. Pulsatile mass along with abdominal pain is a manifestation of MA (Dwivedi, Srinivasan and Jain, 2015).

All the laboratory results supported the clinical signs of infection, such as leukocytosis, fever, and elevated C-reactive protein (Huang et al., 2014). The patient had a high level of procalcitonin which was categorized as at high risk of severe sepsis. It is consistent with a study suggesting that Gram-negative bacteria have a greater risk of developing sepsis (Kang et al., 2021). Elevated D-dimer in this patient is associated with suspected DVT (Kearon et al., 2022). The blood culture showed a positive result of *Salmonella spp.*, which aligns with other clinical and laboratory presentations, and is also the most common pathogen in Asian patients with MA (Huang et al., 2014).

The patient underwent Run-off CTA to diagnose the abnormalities in the vascular system more precisely. According to studies related to MA, CTA is indeed the most sensitive and specific diagnostic resource for MA (Ram,

Deslouches and Punnapuzha, 2022). The CTA result showed an infected mycotic fusiform aneurysm starting 1 cm from the origin of the internal left iliac artery with a mural thrombus 1 cm thick. There are several types of aneurysms, and men tend to have the fusiform type (Barletta et al., 2019). A fusiform aneurysm is a type of aneurysm that is described as circumferential dilations of an artery without an ostium/neck (Awad et al., 2017). In terms of location, the iliac artery is considered to be a rare spot of MA, which is only found in 0,03% of the population in autopsy studies. However, the mortality rate of mycotic iliac aneurysms is very high, especially following a sudden rupture. This type of MA is also associated with a high risk of systemic bacterial embolization (Aziz et al., 2013). A mural thrombus is a thrombus attached to the wall of a blood vessel. It can be induced by an aneurysm and can cause restriction of blood flow (Singh et al., 2023). This patient's aneurysm size is considered large, which was 8 x 5,5 x 4 cm. Size larger than 4 cm is at higher risk of rupture (Laine et al., 2017).

The patient received initial therapy with double antibiotics, cefoperazone-sulbactam and levofloxacin. Levofloxacin is a fluoroquinolone antibiotic, which is the most used regimen for *Salmonella spp.* bacteremia, whereas cefoperazone is a third-generation cephalosporin antibiotic, combined with sulbactam, a beta-lactamase inhibitor. Cefoperazone alone has 75,2% sensitivity towards *Salmonella spp.*, while cefoperazone-sulbactam has 99,3%, and is also another regimen that is commonly used for *Salmonella spp.* bacteremia (Chiang et al., 2016; Guo et al., 2018; Sörelus et al., 2019). The patient is also treated with enoxaparin sodium for his DVT, which is indeed a Low Molecular Weight Heparin (LMWH) that is indicated for DVT treatment (Padayachee, Schoeman and Schellack, 2022).

The patient was then planned to undergo EVAR. There are several surgery methods for MA, such as Open Surgical Repair (OSR) which consists of aneurysm resection with tissue debridement and revascularisation process, and Endovascular Aneurysm Repair (EVAR). EVAR is usually chosen for elderly patients with comorbidities since it is minimally invasive. It is also associated with a higher survival rate and less risk for Infected Related Complications (IRCs) after surgery. The decision to combine antibiotics and surgery is based on literature that the two treatments must be combined since conservative treatment with only antibiotics is associated with poor outcomes, with 50% mortality (Islam, Harnarayan and Naraynsingh, 2022).

However, the patient experienced cardiac arrest before surgery. MA indeed has a poor prognosis, for its high risk of rupture during the peri-operative period (Sörelus et al., 2019). The cause of death was suspected pulmonary embolism. A study reports that pulmonary embolism can also be associated with MA, since there was a case where a patient with mycotic pseudoaneurysm and suspected DVT suddenly experienced acute respiratory distress, coughed with pink sputum, then had sudden cardiac arrest. During the post-mortem examination, a massive embolism was found in the patient's lung (Karkos et al., 2014). Moreover, pulmonary embolism is known to be a consequence of DVT (Torbicki, Kurzyna and Stavros, 2018).

Strength and limitations

This case report can become a reference for clinicians about mycotic aneurysms since this condition is extremely rare yet has high mortality. This study confirms that one of the most common pathogens causing MA is *Salmonella spp.* The finding of this study also shows that adequate diagnosis and treatment is crucial for this condition. However, this study does not show the positive result of the treatment, therefore it is unable to demonstrate a reference for MA treatment.

Conclusion:

Mycotic aneurysm is caused by infection, mostly bacterial, with *Salmonella spp.* being one of the most common pathogens. Diabetes mellitus is a risk factor for MA. Signs and symptoms of MA can be varied. Increase in leukocytes, CRP, procalcitonin, and positive blood culture results, supported by CTA are the main modalities. Several studies have shown aneurysm rupture as the most common complication, while pulmonary embolism has been reported as a cause of death. Definitive therapy for MA is surgery combined with long-term intravenous antibiotic therapy according to culture results. This disease has a high mortality, therefore prompt diagnosis and adequate therapy must be established immediately.

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Conflict of interest

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Author contribution

MH and ZFT contributed equally to the design of this study, manuscript preparation and drafting, proposed the main idea, methodology, and formal analysis. RL gave final approval for the manuscript to be published, and agreed to be accountable for all aspects of the work.

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