



A Double Whammy of Mycotic Aneurysms and Acquired Dysfibrinogenemia in a Patient with Septicemia

*Chun Yang Sim¹ , *Ching Soong Khoo² , Ruslinda Mustafar² , Jia Ning Chai³

¹Faculty of Medicine and Health Sciences, Universiti Sarawak Malaysia, Sarawak, Malaysia

²Department of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia.

³Department of Radiology, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia

*Both authors contributed equally to this work.

A 65-year-old woman with immunoglobulin A nephropathy presented with a 2-day history of fever and right lower limb swelling. She had been treated with cyclophosphamide for a recent relapse 5 days before the current presentation. Apart from a body temperature of 38°C, her other vital signs were fairly stable. Her right lower limb, particularly the thigh, was swollen, erythematous, and mildly tender. Review of other systems was unremarkable. Blood tests revealed elevated levels of septic markers and presence of group A beta-hemolytic streptococci in the blood culture. She was treated with intravenous ampicillin/sulbactam for bacteremia. On the 12th day of hospitalization, her right thigh became more swollen and painful. The computed tomography showed a large heterogeneous retroperitoneal hematoma measuring 4.1 cm×14.2 cm×17.2 cm and another hematoma within the right gluteus medius muscle measuring 8.5 cm×14.6 cm×30 cm (Figure 1). The conventional angiogram showed an aneurysm from the right inferior pancreaticoduodenal artery with active bleeding, which was successfully secured

by arterial embolization (Figure 2. a, b). The right gluteal region was not intervened in the hope of achieving hemostasis through the tamponade effect of the hematoma. The embolization procedure was complicated with persistent oozing from the femoral puncture site. Repeated activated partial thromboplastin time (aPTT) was prolonged (>180 seconds), which was not corrected by the mixing test. Coagulopathy workup showed markedly prolonged thrombin time (TT) of 216.8 seconds, fibrinogen level of 2.14 g/L (normal range, 1.36-4.65 g/L), and elevated levels of factors VIII and IX of 384% and 185%, respectively. Acquired dysfibrinogenemia was highly suspected. The fibrinogen activity-to-antigen ratio was not calculated because the patient did not agree to perform the test. She was then treated with multiple transfusions of fresh frozen plasma, cryoprecipitate, packed cells, and desmopressin to achieve hemostasis. On the 16th day, she was found to have active bleeding from an aneurysm arising from a branch of the right inferior gluteal artery, which was then successfully treated with arterial embolization

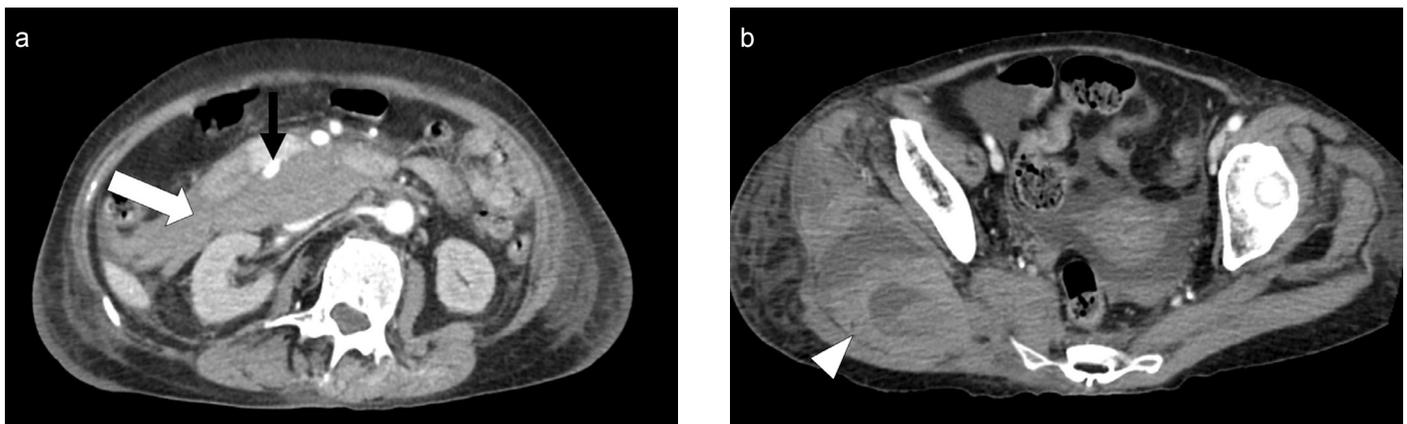


FIG. 1. a, b. Contrast computed tomography of the abdomen and pelvis in the portal venous phase. (A) A large retroperitoneal hematoma (white arrow) and aneurysm (black arrow) posterior to the uncinate process of the pancreas. (B) A large heterogeneous hematoma (white arrow head) within the right gluteus medius muscle

Address for Correspondence: Ching Soong Khoo, Department of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia
e-mail: chingsoongkhoo@gmail.com

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ORCID iDs of the authors: C.Y.S. 0000-0002-9797-6207; C.S.K. 0000-0002-6756-0411; R.M. 0000-0001-8423-707X; J.N.C. 0000-0001-8229-5276.

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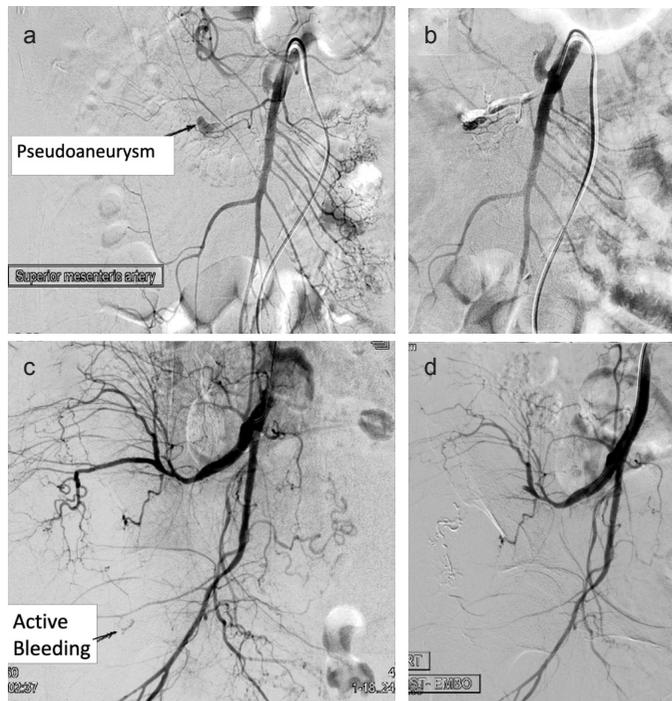


FIG. 2. a-d. (A) Digital subtraction angiogram (DSA) of the superior mesenteric artery shows a pseudoaneurysm arising from the right inferior pancreaticoduodenal artery (black arrow). (B) Post-embolization superior mesenteric angiogram shows no contrast flow into the pseudoaneurysm. (C) DSA of the right common iliac artery shows active contrast blush (black arrow) seen from a branch of the right inferior gluteal artery. (D) Post-embolization angiogram shows no active bleeding

(Figure 2. c, d). She was discharged on the 26th day of admission without any major complications. Written informed consent was obtained from the patient.

Staphylococci and streptococci are the most common causative pathogens for mycotic aneurysms (1). Aneurysms caused by group A streptococcal septicemia usually affect the large vessels, such as the aorta (2). Our patient developed 2 aneurysms along the right inferior pancreaticoduodenal and right inferior gluteal arteries, which were relatively uncommon. The management of an infected aneurysm is individualized and largely depends on the characteristics of the aneurysm and the patient. Treatment options include open surgery, endovascular stent placement, endovascular embolization, medical therapy, or a combination of any of these (3, 4). Any abnormality of fibrinogen can cause hemorrhage, thrombosis, or

both. Dysfibrinogenemia is a condition associated with prolonged TT or low fibrinogen level. If TT is prolonged, fibrinogen activity-to-antigen ratio test is performed to diagnose dysfibrinogenemia (5). Acquired dysfibrinogenemia occurs most often in patients with severe liver disorder, producing abnormal fibrinogen molecules. Dysfibrinogenemia may also be associated with cancer, most commonly being liver tumors. Auto-antibodies inhibiting specific functions of fibrinogen have been described in systemic lupus erythematosus, ulcerative colitis, and multiple myeloma (6). Our patient likely had auto-antibodies interfering with the fibrinogen activity owing to her recent relapsed immunoglobulin A nephropathy. This was further supported by her prolonged aPTT, which was not corrected with the mixing test. Concurrent occurrence of these 2 medical conditions in a patient is extremely rare and, to the best of our knowledge, has never been reported. This case highlights the importance of suspecting aneurysm-related bleeding in uncommon locations to avoid treatment delay in potentially reversible and life-saving conditions.

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