

## Case Report: Septic Pelvic Thrombophlebitis and wide spread extensive thrombosis in the background of Anti Phospholipid Antibody Syndrome

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### Abstract

Septic pelvic thrombophlebitis with wide spread thrombosis in the background of Anti phospholipid Antibody Syndrome has not been previously described. We aim to report such a case in a previously healthy young woman. Persistent fever with bacteremia in postpartum period along with vascular thrombosis at unusual sites was found in this patient, treatment was ensued with IV antibiotics and anticoagulation leading to full recovery.

**Keywords:** Septic pelvic thrombophlebitis; Anti phospholipid; Antibody syndrome; APLA

### Introduction

Septic pelvic thrombophlebitis affects 1 in 3000 deliveries worldwide [1]. In the past, high risk of mortality was associated with this disease as surgical exploration remained the only means of diagnosis. In the early nineteenth century, the disease entity concurred a mortality risk of 50% but recent advancements have shown improvements in survival rates. We aim to report a case of a 30-year-old female with possible APLA presenting with postpartum fever, abdominal pain and lower limb swelling. With a timely diagnostic approach, septic pelvic thrombophlebitis complicated with widespread extensive thrombosis of portal vein (pylphlebitis), superior mesenteric vein and pulmonary artery was diagnosed and managed with IV antibiotics and anticoagulation resulting in patient survival and complete resolution of symptoms.

### Case Description

30-year-old female presented with no prior comorbidities, gravida 7 parity 6+1 and recent twin delivery 40 days back. She lived in a rural area and the delivery took place at home with massive blood loss resulting in intrauterine demise of one fetus and neonatal death of the other. She developed fever 7 days postpartum

which continued for a month along with lower abdominal pain, persistent vomitings and left lower limb swelling. In her initial phase of illness she was evaluated at a secondary care facility with the probable diagnosis of puerperal sepsis along with left leg deep venous thrombosis (DVT) and portal vein thrombosis. She was hence treated at that time empirically with meropenem IV 1 gm q12 hourly, gentamicin IV 80 mg q12 hourly, metronidazole IV 500 mg q8 hourly along with therapeutic enoxaparin 60 mg SC q12 hourly. Despite an overzealous antibiotic treatment, she had to be referred to a tertiary care facility after 15 days due to worsening clinical condition and persistent fever. On arrival, she was found to be in sepsis, with initial vitals recorded as blood pressure (BP) of 90/65, heart rate (HR) of 110 beats/minute, temperature of 102 degree Celsius and respiratory rate (RR) of 42 breaths/minute with saturation of 93% on room air, sofa score 4 points. She was anemic with grossly increased bulk of both lower extremities and a distended tender abdomen with prominent superficial veins. Her cardiovascular and cranial nerve examination were unremarkable. Her pelvic examination revealed a closed cervical OS with anteverted uterus of normal size with no vaginal bleeding, and no fullness or tenderness in the fornices. Her laboratory work up revealed pancytopenia hemoglobin 6.9 mg/dl, total leukocyte count  $2.1 \times 10^9/L$  with low absolute neutrophil count, platelets 98,000/mm<sup>3</sup>, elevated C-reactive protein (CRP) of 135 mg/dl and D-dimer level of more than 4 mg FEU/L. Disseminated intravascular coagulation (DIC) was excluded as coagulation profile was normal, with high fibrinogen level and negative schistocytes on peripheral smear. Retained products of conception were excluded by transvaginal ultrasound which revealed postpartum bulky uterus with multiple tortuous congested venous channels in bilateral adnexal region representing pelvic venous congestion. Considering septic pelvic thrombophlebitis (SPT) and pulmonary embolism as patient was hypoxic (A-a gradient 38 mm Hg) with a high D-dimer, computed tomography (CT) of chest, abdomen and pelvis was done which revealed extensive thrombosis completely occluding the infrarenal inferior vena cava (IVC), partial occlusion of intrahepatic IVC with extension of thrombus into the left renal vein, left internal iliac vein, bilateral external iliac vein, left com-

mon femoral vein, bilateral gonadal veins, right and left portal vein, superior mesenteric vein and splenic vein. Furthermore, there was thrombosis in bilateral lower lobe pulmonary artery branch.

Her autoimmune profile was significant for a single positive speckled pattern for ANA, strongly positive lupus anticoagulant, and a positive anticardiolipin IgM with negative anti-double stranded DNA and normal complement levels. As she fulfilled both clinical (history of fetal demise with vascular thrombosis at unusual sites) and laboratory domains of the Sapporo criteria, preliminary diagnosis of APLA was established.

As upper gastrointestinal scopes were negative for varices in the presence of non-cirrhotic portal hypertension hence anticoagulation with warfarin and enoxaparin were continued with target INR of 2-3 keeping APLA in consideration. Blood cultures yielded acinetobacter and enterococcus species sensitive to first line antibiotics while anaerobic cultures were negative. She was therefore de-escalated to IV ampicillin 3 gm q6 hourly and IV gentamicin 250 mg once daily. Steroids initially given considering autoimmune phenomena were tapered off after workup. Patient recuperated on the above treatment with improvement in laboratory parameters and was discharged after a two week hospital stay in a stable condition. Outpatient visit one week post discharge showed clinical recovery of symptoms along with resolution of lower extremity edema (Figures 1-3).



Figure 1 Inferior vena cava thrombus



Figure 2 Portal vein and inferior vena cava thrombi

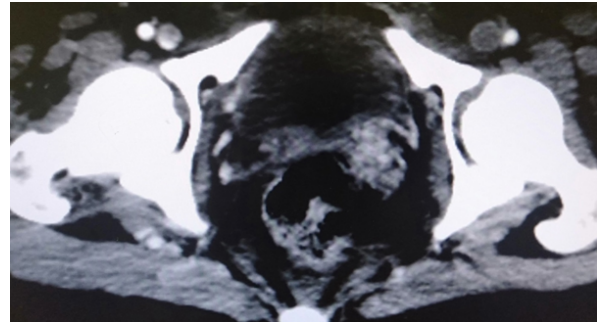


Figure 3 Bilateral iliac vein thrombi

## Discussion

Evaluation of post-partum fever unresponsive to standard broad spectrum antibiotics should implore clinicians to consider the possibility of SPT, an entity first described by Von Recklinghausen in the 18th century, characterized by pelvic infection with thrombosis of one or both ovarian veins. In a randomized control trial, the incidence of SPT was found to be of 20% in patients with prolonged febrile morbidity, defined as more than five days of fever regardless of appropriate antimicrobial treatment [1].

SPT has been described a cause of 'enigmatic fever' [2] in the puerperium period without any notable physical findings and an unrelenting fever. Physical findings, rarely if present, are lower abdominal pain and tenderness, fever and a palpable sausage shaped mass [3]. Maternal age <20, black race, multiple gestation, and preeclampsia are all significantly associated with increased odds of SPT [4].

SPT encompasses the criteria of Virchow triad, namely venous stasis, hypercoagulability and vascular injury and for this reason puerperal period is considered a major risk factor for the development of the disease. SPT has been associated with Lemierre syndrome (internal jugular vein septic thrombophlebitis), pylephlebitis (portal vein septic thrombophlebitis), and septic thrombophlebitis of the dural sinuses and the pelvic veins with incidence of pulmonary embolization of about 2.7% [5]. With widespread thrombosis, SPT entails a high mortality if not timely diagnosed.

Management of SPT has changed considerably in the last few decades, surgical approach by venous ligation once being the only preference antibiotics and anticoagulation are now considered the focal point of the disease management with reliance on imaging such as CT and magnetic resonance imaging (MRI) as diagnostic modalities. Selection of antibiotics for management of SPT must entail postpartum endometritis and should include but not be limited against enterobacteriaceae, anaerobes, and streptococci. Pertaining to anticoagulation, in a case series published, subcutaneous enoxaparin was continued for two weeks if imaging showed thrombosed pelvic vessel and for one week if imaging was negative for thrombosis. Warfarin was given only in cases of ovarian thrombosis [3]. On an average 5 days of heparin therapy are required before defervescence [6]. To our

knowledge, this is the first case report of SPT complicated with possible APLA. As a revised Sapporo criterion requires antibody titers to be repeated at 12 weeks, the patient is scheduled for repeat titers. Due to evidence of thrombosis in multiple sites, we continued warfarin in our patient for three weeks until her symptoms resolved. Total antibiotic regimen was of three weeks which was later de-escalated according to reported sensitivities on blood cultures. A high clinical suspicion allowed us to associate her symptoms to this under-diagnosed condition and consequently to treat her.

## Conclusion

In summary, SPI is a difficult diagnosis that must be presumed in the setting of unexplained persistent fever during the first week of puerperium. Antibiotics and anticoagulation must be initiated if the diagnosis is suspected with duration of treatment varying case to case.

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