

A Rare Case of Tissiarella praecuta in Gonadal Vein Thrombophlebitis

Ashley Thakur*, Leonardo Ramos, Angelica Matteo, Ucia Di Francesco, Dara Forrester, Aruna Mishra,

Magdy Mikhail

Department of Ob-Gyn, BronxCare Hospital, Affiliate of Mt. Sinai School of Medicine, Bronx, NY, USA

ABSTRACT

A 24-year-old female post-partum patient presented to the emergency department with right lower quadrant pain that was present since her vaginal delivery a week ago and undulating episodes of fever and chills. Lab workup was positive for *Tissiarella praecuta* blood cultures and elevated white blood cell count with a 92% neutrophil shift. CT scan confirmed the diagnosis of gonadal vein thrombophlebitis demonstrating expansion of the right gonadal vein with internal hypodensity suggesting a thrombus extending from the right adnexa to the IVC.

Collaborative, multi-disciplinary teamwork consisting of medicine, hematology, infectious disease, and surgery resulted in steady improvement of the patient and discharge within one week. Prompt administration of IV antibiotics and anti-coagulation were required for resolution of inflammation and steady recovery.

Keywords: Gonadal vein thrombophlebitis; Tiessiarella; CT

INTRODUCTION

Gonadal vein thrombophlebitis is a rare but serious condition that has been reported in less than 5% of scientific literature. It was first described in 1956 by Austin, et al but since then a vast amount of information and knowledge has been accrued [1]. The literature has shown that 80% of GVT cases have been reported in patients after delivery, hysterectomy, lymphadenectomy for gynecological neoplasms [2]. Specifically, 80-90% of cases showed that GVT occurs more commonly on the right side because the right ovarian vein is larger and more susceptible to retrograde flow caused by a lack of competent valves [3].

The development of this pathology is centered around combination of several factors including infection, thrombophilia, and inflammation. Development of idiopathic GVT has not been highlighted in literature as the association is more commonly linked in some form to pregnancy. Endothelial damage caused by either trauma of the delivery secondary to infection as well as venous stasis and the natural hypercoagulable state of pregnancy all contribute to the thrombogenic environment. The typical presentation varies from asymptomatic to non-specific abdominal pain [4].

CASE PRESENTATION

A 24-year-old female post-partum patient, day six since vaginal birth, presented to the emergency department with right lower quadrant pain that she states occurred the day after delivery. Initially she felt the pain bilaterally but then it localized to the right lower quadrant and was accompanied with nausea and vomiting. At home she states that she had a fever of 101.3 prompting her to come to the hospital. In the emergency department her temperature was 99.3.

On physical examination blood pressure was normotensive, 112/75, heart rate was 86 bpm, and temperature was 98. General examination was unremarkable. She had slight tenderness in the right lower quadrant upon palpation with no rebound, guarding or costovertebral tenderness. She denied

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Corresponding author: Ashley Thakur, Department of Ob-Gyn, BronxCare Hospital, Affiliate of Mt. Sinai School of Medicine, Bronx, NY, USA; Tel: 3612122400; E-mail: ashleyithakur@outlook.com; athakur@bronxcare.org

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any shortness of breath, headache, diarrhea, constipation, or blood in her rectum.

Investigations

Labs were immediately drawn which showed an elevated white count (15.6 ref range [4.8-10.8 k/ul]) with a 92% neutrophil shift. Lactic acid, ESR and C-reactive protein were all elevated indicating some underlying inflammatory condition. Amylase and lipase were normal. Liver enzymes and alkaline phosphatase were significantly elevated. Alanine aminotransferase 80 [ref range 5-40 unit/L], aspartate transaminase 142 [ref range 9-36 unit/L] and alkaline phosphatase 715 [ref range 42-98 unit/L].

Urinalysis revealed few bacteria and urine culture was normal. Blood culture was abnormal and was positive for *T. praeacuta*. Patient was then sent for CT of the abdomen and pelvis which showed expansion of the right gonadal vein with internal hypodensity suggest a thrombus extending from the right adnexa to the IVC, confirming the the diagnosis of gonadal vein thrombophlebitis (Figure 1).

Treatment

Vascular surgery was then consulted along with hematology and infectious disease for further management. Surgery recommended medical management per primary team and IV antibiotics as well as anticoagulation with no surgical intervention warranted. Hematology recommended starting the patient on Lovenox 60 mg BID. Patient was then admitted to the hospital for gonadal vein thrombophlebitis and started on Zosyn and Lovenox.

Outcome and follow-up

On day two of admission, patient spiked a fever of 103.1. She was then started on Vancomycin. CBC was still abnormal, white blood cell count was 14.5 but transaminitis had improved [AST 91, ALT 69]. Patient was then switched to clindamycin and meropenem per infectious disease. Repeat labs, UA, urine culture and blood culture were also sent. Labs showed an improvement in CBC with white blood count at a normal level of 10.7.

By day eight of admission patient's fevers had resolved, abdominal pain and tenderness were nearly gone, no leukocytosis was present and no regrowth of *T. praeacuta* was seen in repeat blood cultures. Per infectious disease team it was recommended to switch the patient after the last IV meropenem dose to Augmentin 875 mg PO every 8 hours for an additional 12 more days along with Flagyl 500 mg PO every 8 hours. Lovenox was continued as she was breast feeding and hematology recommended, she continue taking Lovenox for 6 months.

DISCUSSION

Tissierella praeacuta, also known as *Clostridium hastiforme*, is a gram-negative bacteria found both in humans and the environment. It was isolated in 1908 by P.H. Tissier but has very few documented cases in the literature linking it to human infection. Furthermore, of the clinical presentations it has been linked to none of those is gonadal vein thrombosis. The six limited ones have been septic pseudoarthrosis, fistula formation, bacteremia secondary to septic arthritis, brain abscess and eyelid gangrene [5].

In the latter cases all the infections were treated with a cocktail of beta lactams, Metronidazole and Rifampicin as *Tissierella praeacuta* exhibited antibiotic sensitivity to them [6]. In our patient antibiotic sensitivity wasn't assessed, instead infectious disease had started the patient initially on Vancomycin and then assessed how the patient responded. Since the patient still had an elevated blood count with undulating fever, it was then recommended to switch to clindamycin and meropenem which reduced inflammation. By the time the patient was discharged antibiotics were switched to oral metronidazole and augmentin which the patient successfully completed.

The diagnosis of gonadal vein thrombosis is challenging as

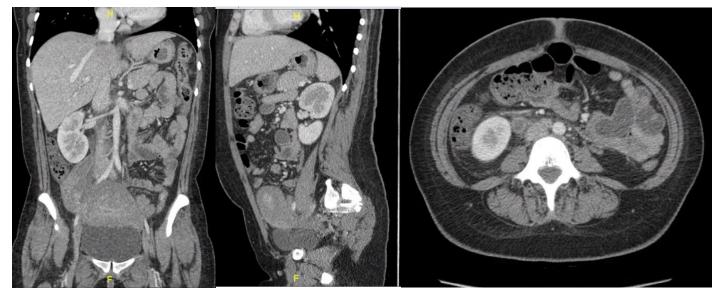


Figure 1: Main portal vein and proximal branches are patent. There is dilatation of the right gonadal vein with peripheral enhancement and surrounding inflammation extending to the level of the IVC. This extends to the level of the right adnexa. The uterus is enlarged and heterogeneous edematous compatible with postpartum state.

there are no laboratory tests that are specific to that detecting this condition. Positive blood cultures and leukocytosis as was seen in this patient are present in less than 1/3rd of cases [7]. While inflammation and leukocyte count improved after antibiotic therapy, this patient's clinical condition was unchanged. Given that GVT is often misdiagnosed (ex: endometritis, appendicitis, pelvic abscess) and there are no specific physical exam findings, its diagnosis requires a high level of clinical suspicion.

The role of anticoagulation in GVT has been well accepted as a mainstay treatment but antibiotic therapy is still questionable as the selections of certain antibiotics are often extrapolated from literature on postpartum endometritis. The duration of antibiotic therapy is yet another variable that is not well defined. Had blood cultures not been collected in this patient probably the appropriate antibiotics would not have been given and the patient may not have improved as quickly as she had.

In conclusion, this case highlights the need of both antibiotic and anticoagulation therapy as mainstay treatment for gonadal vein thrombophlebitis. Since this condition can have various presentations and is so rare diagnosis can be challenging and treatment is often delayed. In this context, we hope that this case report substantiates this diagnosis and helps physicians become more aware of this complication in order to diagnose and treat this condition effectively.

LEARNING POINTS

- This is a rare case *Tissierella praeacuta* linked to gonadal vein thrombophlebitis.
- Due to the rare and atypical presentation of gonadal vein thrombophlebitis low suspicion was suspected with the patient's presentation. A multidisciplinary team approach was required to achieve appropriate management.
- Although there is currently little evidence in the literature reporting such a case, *T. praecuta* should be considered a bacterial differential and appropriate antibiotic therapy should be used in cases of suspected GVT.

PATIENT'S PERSPECTIVE

"I first felt some abdominal pain after giving birth, but I figured it was just normal labor pains. I was reassured by Dr. Forrester that this was normal postpartum pain and it would subside, but it only got worse once I got home. I started having periods of fever and then chills and this pain wouldn't go away even after I took some ibuprofen.

I decided to come to the hospital and just wanted to feel better. After some tests were run and I was sent for imaging

I was told about the condition I had. I was shocked because it sounded serious and wasn't what I was expecting to hear. I couldn't believe that I would be staying at the hospital for so long, but I knew it was the only way to feel better and be in better spirits not only for me but for the baby.

After a couple of days of intense antibiotic therapy, I started feeling much better. I was able to walk around and breastfeed without feeling these intense cramp-like pains. About a week after I was admitted I felt like a new human. I knew I had to continue the antibiotics and the anticoagulation therapy that I was started on but given the way I had felt before compared to now, I didn't mind.

I was so happy to get back to a normal routine and spend quality time with my baby without feeling so ill. My partner was also so happy to see me healthy and pain-free. I can't thank you all for taking such good care of me while I was admitted.

I was seen about a month after being discharged and my scans showed that the inflammation and clot had disappeared. I couldn't believe how complex this diagnosis was. I still think about it from time to time. I remember when I first came in how the doctor in the emergency department figured it was appendicitis or something but to see that it was a clot like condition is mind-blowing. I hope that by you sharing my story this provides some light for other patients like me who may have the same condition."

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