

Antiphospholipid Syndrome in a Male Patient Presenting with Abdominal Pain

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Abstract

Introduction: Antiphospholipid syndrome (APS) is a systemic autoimmune disorder characterized by antiphospholipid antibodies (aPL), leading to a hypercoagulable state and an increased risk of thrombotic events. While abdominal complications have been reported as the initial presentation of APS in some cases, these instances are predominantly observed in female patients. Here, we present a case of a 49-year-old male patient who presented with complaints and a CT scan mimicking intestinal ileus. However, no mechanical or other evident cause of ileus could be found on the explorative laparotomy. After an uneventful postoperative hospitalization and being discharged in good condition, the patient was readmitted within two weeks, and this time, an extensive small bowel resection due to ischemia was done. After going home in a good and stable condition, he presents again, but this time with cerebral ischemia.

Further investigations led to the APS diagnosis. With this case, we want to emphasize the importance of being aware of and considering the diagnosis of APS, especially in cases with repeated, unexplained abdominal pain and non-typical complaints, even in male patients. An early diagnosis could prevent a more complex disease complication.

Conclusion: This case underscores the importance of considering APS in the differential diagnosis of unexplained abdominal pain, particularly in male patients with a history of thrombotic events or elevated aPL levels. Heightened awareness of APS in the emergency setting can facilitate timely diagnosis and appropriate management, ultimately optimizing patient care and outcomes.

Keywords: antiphospholipid syndrome, emergency laparotomy, abdominal pain, Mesenteric ischemia

Introduction

“The antiphospholipid syndrome (APS) is defined by the occurrence of venous and arterial thrombotic events and pregnancy-related morbidity (≥ 3 unexplained consecutive spontaneous abortions < 10 weeks with exclusion of chromosomal causes, fetal death or severe pre-eclampsia before 34th week of gestation), combined with the presence of circulating antiphospholipid antibodies (aPL) and a lupus anticoagulant (LAC).” [1]

While APS commonly manifests with arterial and venous thrombosis, its association with abdominal complications as an initial presentation is relatively uncommon.[2]

Previous reports have documented such cases mostly in female patients [3, 4, 5].

Here, we describe a male patient who presented with severe abdominal pain and mesenteric thrombosis as the primary presentation of the disease. This case highlights the importance of recognizing APS as a potential underlying cause of abdominal symptoms, even in male patients, thus ensuring timely intervention and appropriate management.

Case Report

A 49-year-old male patient presented to the emergency service with severe abdominal pain, distension, and inability to pass flatus and stool, mimicking intestinal ileus. He had a history of laparoscopic appendectomy one year before and laparotomy for intestinal ileus six months before. On

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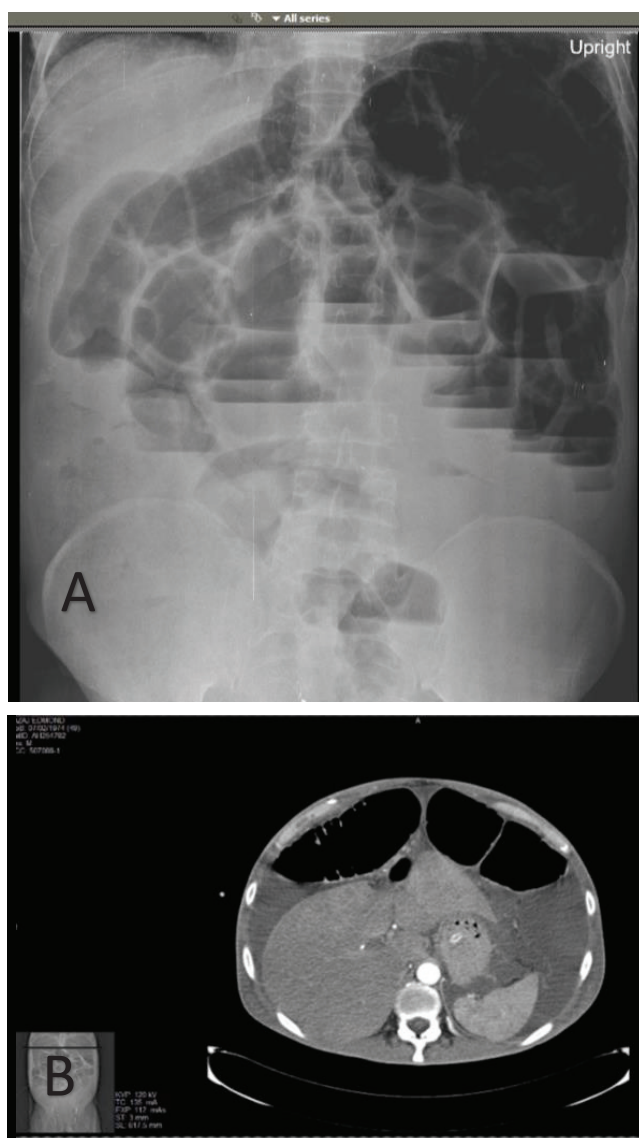


Figure 1. (A) Abdominal plain X-ray showing air-fluid levels and enlarged intestinal ansae. (B) CT scan showing the dilated intestine with free intraabdominal liquid.

the Abdominal CT scan, the small intestine, ascending, and transverse colon were enlarged, with a diameter up to 6.5 cm in the splenic flexure, with hydraulic levels. The free liquid was evident in the abdomen, and reactive character enlarged mesenteric lymph nodes up to 21 mm were seen. (Fig 1)

In this ileus situation, the patient went for an explorative laparotomy. Surgical exploration revealed a dilated small intestine and colon, albeit without any apparent mechanical obstruction. The patient had no electrolyte disbalance or other cause that could explain this ileus situation. Consequently, the decision was made to close the abdomen without further intervention.

The patient experienced a favorable postoperative recovery and was discharged home. However, approximately two weeks later, he returned with recurrent severe abdominal pain. A repeat CT scan demonstrated intestinal dilation, raising suspicions of mesenteric ischemia. (Fig 2)

During the subsequent surgical intervention, extensive small bowel resection due to mesenteric ischemia/necrosis was performed. The postoperative period was uneventful, and the patient was discharged satisfactorily.

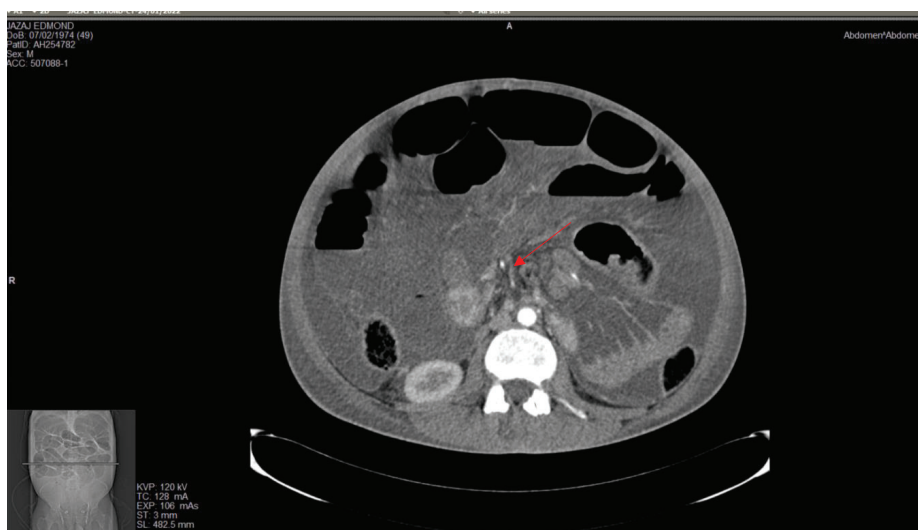
After finishing the postoperative DVT prophylaxis with enoxaparine, the patient presents again with severe cephalgia. On further evaluation, a head MRI showed acute ischemic areas on the left parietal and occipital zone. (Fig 3)

Considering the two episodes of arterial thrombosis, a deep investigation with clinical and laboratory work-up was conducted during the patient's management to understand the underlying cause.

The laboratory investigations revealed negative ANA 1:100 reference range 1:160 and double positivity of APS antibodies abnormal lupus anticoagulant (LA) test results: LA1 (54 sec, reference range: 31-44 sec), LA2 (41 sec, reference range: 30-38 sec), and LA1/LA2 ratio (1.31, reference range: 0.8-1.2) and positive IgM Beta2GPI 60 (reference range < 20 U/mL).

These findings led to the diagnosis of APS. The patient was subsequently referred to our rheumatologist for further

Figure 2 CT scan with IV contrast showing discontinuation of the contrast material on the mesenteric artery (red arrow) and also enlarged intestinal ansae with free intaabdominal liquid.



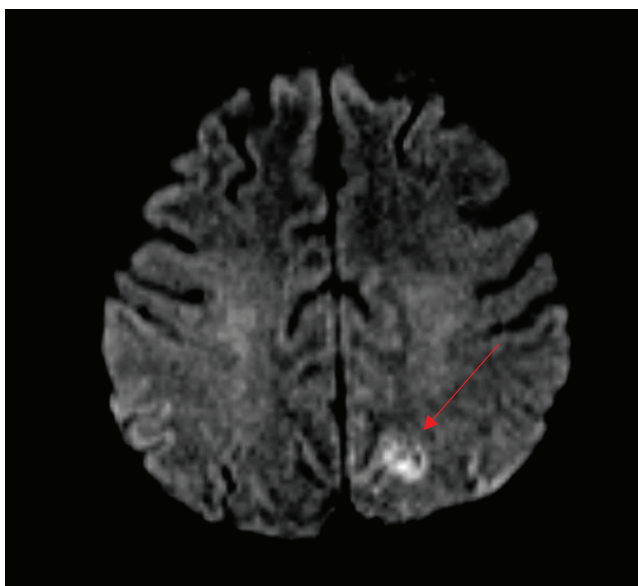


Figure 3. Head MRI showing ischemic lesions in the left basal occipital region (red arrow)

evaluation and is currently receiving warfarin with an international normalized ratio (INR) target of 3-4.

Discussion:

Antiphospholipid syndrome (APS) is a recognized autoimmune disorder characterized by recurrent thrombotic events and antiphospholipid antibodies. While APS commonly manifests as arterial and venous thrombosis, the involvement of abdominal organs as the primary presentation is rare and rarely reported [6].

Existing literature on abdominal complications of APS predominantly features female patients [3, 4, 5, 7]. Our case report presents a male patient who initially presented with severe abdominal pain, which was initially misdiagnosed as intestinal ileus. Subsequent surgical interventions led to the diagnosis of mesenteric ischemia, necessitating extensive small bowel resection.

The recognition of APS was made only after severe complications of the disease. We recommend and highlight the importance of recognizing APS as a potential underlying cause of abdominal symptoms, even in male patients, thus ensuring timely intervention and appropriate management. High clinical suspicion, timely diagnosis, and appropriate management, including anticoagulation therapy, are crucial in diagnosing and preventing further complications and improving patient outcomes.

Because Gastrointestinal symptoms of APS are not reported as frequently as they really are [6], reporting similar cases is important to increase our understanding of APS and its possible clinical presentations and maybe define more specific findings to help make a better and more timely diagnosis.

Conclusion

This case underscores the importance of considering APS in the differential diagnosis of unexplained abdominal pain, particularly in male patients with a history of thrombotic events or elevated aPL levels. Heightened awareness of APS in the emergency setting can facilitate timely diagnosis and appropriate management, ultimately optimizing patient care and outcomes.

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References:

1. Limper M, de Leeuw K, Lely AT, Westerink J, Teng YKO, Eikenboom J, Otter S, Jansen AJG, V D Ree M, Spierings J, Kruyt ND, van der Molen R, Middeldorp S, Leebeek FWG, Bijl M, Urbanus RT. Diagnosing and treating antiphospholipid syndrome: a consensus paper. *Neth J Med.* 2019 Apr;77(3):98-108. PMID: 31012427.
2. Pak H, Tajik A, Soltanian A. Abdominal pain in the patient with antiphospholipid antibody syndrome: A case report. *Clin Case Rep.* 2019 Oct 15;7(12):2327-2330. doi: 10.1002/ccr3.2488. PMID: 31893051; PMCID: PMC6935617.
3. Saponjski J, Stojanovich L, Saponjski J, Mirilovic M, Saponjski D. Abdominal pain in patient with antiphospholipid syndrome-the role of MDCT angiography on visceral blood vessels. *Immunol Res.* 2017 Dec;65(6):1150-1155. doi: 10.1007/s12026-017-8968-1. PMID: 29134567.
4. Pak H, Tajik A, Soltanian A. Abdominal pain in the patient with antiphospholipid antibody syndrome: A case report. *Clin Case Rep.* 2019 Oct 15;7(12):2327-2330. doi: 10.1002/ccr3.2488. PMID: 31893051; PMCID: PMC6935617.
5. Selby R, Abdulrehman J. Antiphospholipid Antibodies and the antiphospholipid syndrome: From Coagulation to the Clinic. *J Appl Lab Med.* 2022 Jan 5;7(1):373-378. doi: 10.1093/jalm/jfab134. PMID: 34996086; PMCID: PMC8741326.
6. Zhang J, Li C, Han X, Chen Z, Adhikari BK, Wang Y, Wang Y, Sun J. The digestive system involvement of antiphospholipid syndrome: pathophysiology, clinical characteristics, and treatment strategies. *Ann Med.* 2021 Dec;53(1):1328-1339. doi: 10.1080/07853890.2021.1962964. PMID: 34409894; PMCID: PMC8381908.
7. Srivastava V, Basu S, Ansari M, Gupta S, Kumar A. Massive gangrene of the stomach due to primary antiphospholipid syndrome: report of two cases. *Surg Today.* 2010;40(2):167-70. doi: 10.1007/s00595-009-4015-8. Epub 2010 Jan 28. PMID: 20107959.