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Asymptomatic splenic infarct and retroperitoneal sepsis in patient with Wegener's granulomatosis

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ABSTRACT

INTRODUCTION: We present a patient with an unusual combination of surgical findings, complicated by both a medical condition and the treatment there of.
PRESENTATION OF CASE: A 52 year-old man with Wegener's granulomatosis presented with groin pain and sepsis while on immunosuppressive therapy. Large retroperitoneal abscesses were found on CT scanning, together with a collapsed left lower lobe of the lung, a complete infarct of the spleen, and evidence of diverticulosis. At the exploratory laparotomy, the infarcted spleen was removed and the retroperitoneal abscesses were drained via a separate lateral incision. Uncomplicated diverticular disease were also confirmed.
DISCUSSION: The left lower lobe collapse, infarcted spleen and diverticular disease were all potential sources of the sepsis, but none of them could be clearly linked to the abscesses. The splenic infarct and a post-operative myocardial infarct were likely related to his vasculitic disease.
CONCLUSION: Patients with systemic vasculitis may present with unusual pathologies, and immunosuppressive treatment may also modify clinical presentation.

1. Introduction

We describe a case of incidental complete infarct of the spleen in a patient with Wegener's granulomatosis (WG), who presented with large retroperitoneal abscesses while on immunosuppressive therapy for his disease.

2. Case presentation

A 52 year-old man presented to the emergency department with a 2-day history of sudden onset left groin pain worsening over 48 h. There was no associated vomiting or altered bowel habit, and no fever or systemic symptoms.

He had a history of WG diagnosed two months earlier, which was treated with prednisolone (60 mg daily) and cyclophosphamide (150 mg daily). This had presented with epistaxis associated with a nasal mass and painless loss of vision. The nasal mass was biopsied and the diagnosis confirmed. In addition, a renal biopsy demonstrated focal sclerosing glomerulonephritis.

Physical examination revealed a temperature of 36.9 °C, pulse rate of 100 bpm, blood pressure of 155/95 mmHg and respiratory rate of 18 bpm. Examination of the abdomen demonstrated an erythematous discolouration in the left flank associated with tenderness and guarding in the left iliac fossa and a very tender, and elongated, mass along the line of the left inguinal canal. The testes and spermatic cords were normal. Decreased breath sounds at the left base were noted on chest examination.

A chest X-ray showed a collapsed left lower lobe and a subsequent CT of the chest and abdomen demonstrated a large retroperitoneal abscess with smaller adjacent abscesses on the left side extending from the flank and into the groin, as well as total infarction of the spleen (Fig. 1). Uncomplicated diverticulosis of the sigmoid colon was noted. There was no apparent communication between the abscesses and either the infarcted spleen or the sigmoid colon. No bony abnormality of the vertebral columns to suggest osteomyelitis was seen.

The patient was taken to the operating theatre for exploration. At laparotomy, the retroperitoneal abscesses were evident but there was no evidence of intraperitoneal sepsis. Total splenic infarction was confirmed. The spleen was encased by adherent omentum, but there was no evidence of infection of the spleen. Splenectomy was performed. The sigmoid and descending colons were mobilized. Although diverticular disease was confirmed, there was no evidence of diverticulitis or diverticular perforation.

The abscesses were confined to the retroperitoneal space, with an extension into the inguinal region. Drainage and placement of large calibre low-pressure drains was performed via a separate flank incision, avoiding contamination of the peritoneal space. Approximately 1 L of pus was drained. The post operative course was complicated by a myocardial infarct believed to be related to involvement of coronary vessels by WG vasculitis. The sepsis was

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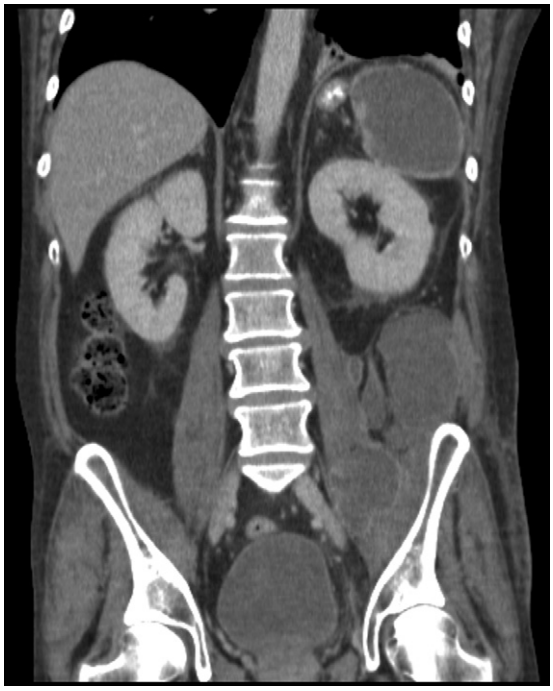

Fig. 1. Coronal image showing a complete infarct of the spleen and multiple retroperitoneal abscesses on the left side.

well controlled and resolved without further incident. Microbiological culture of the drained pus isolated Bacteroides and Prevotella suggesting gut as the likely source. No organisms were isolated from the infarcted spleen.

3. Discussion

This patient with WG had the classic triad of acute necrotising granulomas of the upper respiratory tract, vasculitis affecting small to medium-sized vessels (eye, spleen and heart), and glomerulonephritis. This pattern of organ involvement is seen in widespread disease. However, involvement of the spleen and heart is rarely seen, making this case unusual. In the recent literature, there had only been seven reports of symptomatic splenic involvement in WG. Although haemorrhagic and infective involvements are usually symptomatic, infarcts and splenomegaly can be asymptomatic, which may result in an underestimate of prevalence of splenic involvement in WG.[1] In WG related infarcts, the histological features of central arteritis, splenic trabeculitis, follicular arteriolitis and necrosis give clues to the pathogenesis.[1]

In this case, WG caused total splenic infarction. When symptomatic, patients with splenic infarcts typically present with left upper quadrant pain or left shoulder tip pain however up to 50% may be asymptomatic.[2] This patient's splenic infarct was apparently asymptomatic, though the collapsed left lower lobe may have been secondary to the infarct.

A conservative approach is possible in managing splenic infarcts even when complete, unless there are signs of imminent rupture, abscess or haemorrhage. Hafezi-Rachti et al.[3] illustrated a case of near complete splenic infarct from WG managed conservatively, with scarring of the infarcted spleen demonstrated on a repeat CT scan 2.5 years later. In our patient, splenectomy was performed because the concurrent sepsis posed increased risk of splenic abscess. Like post-splenectomy patients, those with large splenic infarcts are at increased risk of serious infection with encapsulated and intra-erythrocytic organisms; even more so if immuosuppresive drugs are used. Therefore it is important to administer appropriate prophylactic antibiotic therapy as well as immunisations.[2]

It is worthwhile noting that patients with WG may present with symptoms of primary disease, complications of the disease or complications of the treatment. Immunosuppression from treatment may modify the clinical presentation. This patient presented with extensive suppuration and relatively minor symptoms. No doubt his treatment with prednisolone and cyclophosphamide played a significant role in abscess formation as well as masking the extent of the disease and contributing to a late presentation. Although three possible sources for the sepsis were identified (spleen, colon and lung), no clear link could be found to support any of them as the primary source for retroperitoneal abscess by direct extension. It is likely that immunosuppressive treatment allowed bacterial seeding but the precise source and mechanism remains unclear.

4. Conclusion

Patients with systemic vasculitis such as WG may present with unusual abdominal pathology. This case report highlights the possibility of splenic involvement in WG. Such involvement is unusual but total infarction is possible. Furthermore, immunosuppressive treatment in these patients may mask the severity of abdominal or septic disease and thus modify clinical presentation.

Conflict of interest statement

None.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

James Lee—data collection, manuscript preparation and revision, study design.

Ingra Bringmann—surgeon who performed the operation.

Ahmad Aly—study design, senior surgeon, manuscript revision.

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