Beware the pseudo-IBD flare

An atypical cause of colitis, pancytopaenia and splenic abscesses in a young patient on anti-TNF therapy for ulcerative colitis

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Background

A 23 year old male with a ten year history of ulcerative colitis and recurrent *Clostridioides difficile*, presented with six weeks of lethargy, weight loss, PR bleeding and fever. He was commenced on empiric Piperacillin-tazobactam and Gentamicin for intra-abdominal sepsis and proceeded to CT imaging of the abdomen and pelvis.

This demonstrated recto-sigmoid colitis and multi-focal areas of splenic hypoattenuation in keeping with abscesses. Fluconazole was added empirically given splenic abscesses and Adalimumab and Mesalazine held.

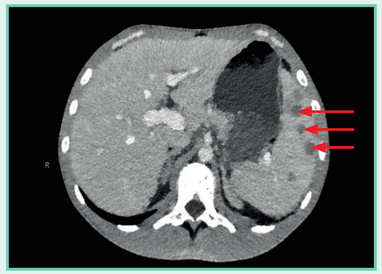
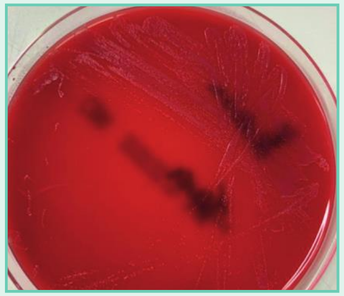


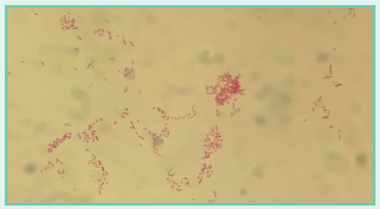
Figure 1.CT abdomen demonstrating splenic abscesses

Progress

3 sets of blood cultures yielded *Yersinia pseudotuberculosis. Streptococcus gordonii* and *Staphylococcus epidermidis* were also present in one isolate, with the latter a presumed contaminant. The patient developed pancytopaenia and deteriorated clinically despite escalation of antimicrobial therapy. He proceeded to have a laparotomy with subtotal colectomy, splenic biopsy and bone marrow aspiration.

Haemophagocytic lymphohistiocytosis (HLH) was confirmed, based on blood markers, bone marrow analysis and 'H' score (see figure 4). The patient was discussed at a national HLH forum and treated with Intravenous methylprednisolone, then Anakinra and Intravenous immunoglobulin.





Figures 2 & 3. Blood agar and Gram stain demonstrating *Yersinia pseudotuberculosis*

Figure 4. H score – HLH diagnostic aid

Feature	Ref. range and score	Patient value and score	
Immunosuppression	Yes (18) No (0)	**Yes (18)**	
Temperature	38.5 -39.4 (33)	**38.5 – 39.4C (33)**	
Hepatomegaly	Yes (23) No/unknown (0)	**Unknown (0)**	
Splenomegaly	Yes (23) No/unknown (0)	**No (0)**	
Haemoglobin	<9.2g/L	**6.8**	**(34) – 3 lineages affected**
Leucocytes	</= 5000/mm3	**1.4**	
Platelets	</= 110000/mm3	**32**	
Ferritin	>6000ng/ml (50)	**9280 (50)**	
Triglycerides	1.5-4 mmol/L (44)	**1.53 (44)**	
Fibrinogen	</= 2.5g/L (30)	**1.1 (30)**	
AST	>/= 30 U/L (19)	**(143)**	
Haemophagocytic features onbone marrow aspirate	Yes (35) No (0)	**Yes (35)**	
Score = 264 = 99.4% probability of having HLH			

Outcome

Post-operatively, this patient clinically responded to therapy and was transitioned to Intravenous Ceftriaxone and Metronidazole to facilitate treatment in the community. After continued clinical response and repeat imaging demonstrating resolution of abscesses, antimicrobial therapy was stopped 11 weeks from admission. On follow-up, the patient remains asymptomatic and inflammatory markers, blood cell counts and ferritin have normalised. He is being investigated by Immunology as an outpatient but thus far there is no evidence of underlying primary immunodeficiency.

Discussion

Yersinia pseudotuberculosis is a gram negative coccobacillus that rarely causes bacteraemia in humans. Infection commonly occurs due to consumption of contaminated food or water.

Typically, patients infected with *Y.pseudotuberculosis* present with fever, abdominal pain, diarrhoea and vomiting. Mild cases of intestinal disease may not require antimicrobial treatment, however the risks and benefits of therapy should be examined on a case by case basis.

Extra-intestinal sequalae include metastatic abscess formation, septic arthritis and infective endocarditis. Overall mortality is relatively low with an American study reporting 18 deaths from 1373 cases of Yersiniosis (1.3% mortality).

Conclusions

- Inflammatory bowel disease (IBD) exacerbations can be difficult to distinguish from invasive infection and a wide differential should be entertained in immunosuppressed patients.
- History of *Clostridioides difficile* infection complicates management of acute IBD flares.
- Yersiniosis may masquerade as colitis or appendicitis.
- Infection with Yersinia is associated with metastatic abscess formation.
- Secondary HLH requires multidisciplinary input with urgent treatment of the underlying trigger.

References
Yersinia pseudotuberculosis and *Y. enterocolitic*a infections, FoodNet, 1996-2007. LC, Jones et al Emerg Infect Dis. 2010;16(3):566
Development and validation of a score for the diagnosis of reactive hemophagocytic syndrome (HScore) Fardet L et al., Arthritis & Rheumatology 2014 - as accessed at http://saintantoine.aphp.fr/score/ on 18/05/2021 – Figure 4 adapted from this
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The patient has consented to this case being presented and discussed