An unusual case of abdominal pain and splenomegaly in a paediatric patient

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Abstract

Fusobacterium species are members of the oral microbiota and have been found to cause a wide spectrum of opportunistic infections. We describe the case of a previously healthy teenager with a large splenic abscess secondary to *Fusobacterium nucleatum*, successfully managed with percutaneous drainage and intravenous antibiotics. Identification of the organism was achieved using anaerobic culture of the aspirated fluid and matrix-assisted laser desorption/ionization time of flight, later confirmed by 16S ribosomal RNA metagenomic sequencing of the fluid. *Fusobacteria* are typically associated with oropharyngeal infections but are very rarely implicated in splenic abscesses. Aerobic and anaerobic blood cultures should be drawn when an intra-abdominal infection is suspected in a paediatric patient, and empiric antimicrobial therapy should be administered with coverage for gram-positive, gram-negative, and anaerobic bacteria.

Keywords

Splenic abscess, splenomegaly, Fusobacterium, paediatric

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Introduction

Isolated splenic abscesses are a dangerous, but fortunately uncommon, condition in children requiring a high degree of clinical suspicion. A clinical presentation of fever, upper abdominal pain, and leukocytosis should raise the possibility of splenic abscess; risk factors include infections at a distant or contiguous site, splenic trauma, hemoglobinopathies, and an underlying state of immunocompromise.¹ Due to the wide variety of causative pathogens, broad empiric antimicrobial coverage and comprehensive evaluation for the source of infection are required. Historically, the treatment strategy for splenic abscesses included splenectomy and antimicrobial therapy; however, accumulated evidence has demonstrated that percutaneous aspiration with antimicrobial therapy has a high success rate.²⁻⁴ Whenever possible, spleen-preserving techniques should be used in order to maintain the immunologic function of the organ.^{1,3}

Fusobacterium species are fastidious anaerobic bacteria that are present in large populations in the oral cavity of both healthy and diseased individuals.⁵ Although commonly implicated as a periodontal pathogen, *Fusobacterium nucle-atum* has also been implicated in a wide range of human diseases.⁵ We present a case of a previously healthy teenage boy with an isolated splenic abscess secondary to *F. nucleatum*,

which was successfully treated with percutaneous aspiration and a 6-month course of antimicrobial therapy.

Case

A previously healthy, fully immunized 12-year-old male was transferred to our tertiary care paediatric centre from a community hospital with a 1-month history of abdominal discomfort, anorexia, intermittent fevers, and significant weight loss, accompanied by new onset left shoulder pain and dyspnea. He first sought medical attention 2 weeks into his illness

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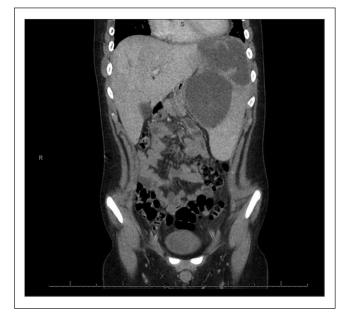


Figure 1. Abdominal CT scan with contrast.

due to fevers and emesis, at which time he received intravenous fluids for presumed gastroenteritis. Aerobic blood cultures obtained at the time were negative. Despite resolution of fever and emesis, his abdominal pain persisted prompting a second hospital visit.

On further history, he reported an all-terrain vehicle accident approximately 3 months prior, during which time he suffered blunt-force trauma to the left side of his torso with short-lived abdominal pain that resolved spontaneously. He did not recall any respiratory tract symptoms, ear or throat infections, and had no recent dental work. Exposures were only significant for domestic dogs. He did not have any history of foreign travel. He had not been prescribed antibiotics. No individuals in his household were unwell.

On physical examination, he was afebrile with heart rate, blood pressure, and respiratory rate within the normal range for his age. He was noted to have occasional desaturations to 90%-94% SpO₂ on pulse oximetry. He complained of severe left-sided abdominal pain, shoulder pain, and chest pain on deep inspiration, requiring morphine for adequate analgesia. He had decreased air entry to the left lung base. The left side of his abdomen was extremely tender with marked splenomegaly. The remainder of the physical exam was unremarkable.

Bloodwork demonstrated a haemoglobin of 117 g/L, white blood cell count of 13.4×10^9 cells/L (81% neutrophils), and C-reactive protein of 162 nmol/L. INR (international normalized ratio) was persistently elevated in the 1.2–1.3 range. Electrolytes, liver enzymes, and serum creatinine were normal. Albumin was slightly low (33 g/L; normal range: 35-52 g/L); however, the remainder of liver function tests were normal. Aerobic blood cultures were drawn prior to initiation of antibiotics and were negative; anaerobic blood cultures drawn after initiation of

antimicrobial therapy were also negative. HIV serology was non-reactive.

Abdominal computed tomography (CT) scan with contrast done at the community hospital immediately prior to transfer demonstrated a splenic length of 25.4 cm, with multiple low density focal areas throughout the spleen that appeared multi-loculated. The largest lesion measured 17 cm in diameter (Figure 1). There were no lesions in the liver and no evidence of intra-abdominal thrombosis. Chest X-ray was significant for basilar atelectasis and a small simple leftsided pleural effusion with no lung lesions. X-rays of the left shoulder done in the community were not suggestive of fractures or osteomyelitis. Echocardiogram was unremarkable. Empiric therapy with piperacillin-tazobactam was initiated to provide broad-spectrum coverage. He received diuretics due to dyspnea. The elevated INR was thought to be due to poor nutritional intake in the context of abdominal pain, and he subsequently received vitamin K prior to drainage of the collection.

Final diagnosis

The differential diagnosis at the time included splenic abscess, parasitic infection, and post-traumatic splenic rupture. Image-guided percutaneous drainage was performed with a total of 810 mL of malodorous purulent fluid drained, and a pigtail catheter left in situ. Initial Gram stain of the fluid revealed 1+ white blood cells and 1+ gram-negative bacilli. After 5 days of incubation, growth on the anaerobic culture plate was confirmed as *F. nucleatum* using matrix-assisted laser desorption/ionization time of flight (MALDI-TOF) mass spectrometry.

Clinical course

The diagnosis of *F. nucleatum* splenic abscess was confirmed by direct 16S ribosomal RNA (rRNA) amplification and sequencing of the fluid performed at the British Columbia Centre for Disease Control. The isolate was susceptible to penicillin and metronidazole. The patient's antimicrobial coverage was narrowed to ceftriaxone and metronidazole to provide ongoing polymicrobial and anaerobic coverage.

After lesion drainage, the patient reported immediate improvement in his symptoms; his abdominal discomfort diminished, his dyspnea resolved, and he regained his appetite. Repeat CT 6 days post-drainage indicated persistent, extensive multi-loculated collections, despite ongoing catheter drainage (Figure 2); consequently, a second guided drainage was performed. After 19 days, the pigtail catheter was removed and the patient was discharged home with a peripherally inserted central catheter line in situ to receive ongoing antibiotic therapy with ceftriaxone and metronidazole. An abdominal ultrasound was performed prior to discharge, which demonstrated persistent splenic collections measuring up to 7.1 cm in diameter (Figure 3). Approximately

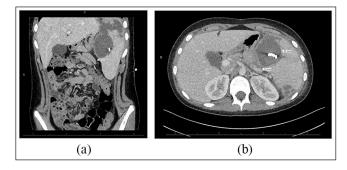


Figure 2. Repeat abdominal CT scan with contrast 6 days post abscess drainage: (a) coronal section and (b) transverse section.



Figure 3. Abdominal ultrasound pre-discharge. Residual splenic collections are seen, including one measuring 7.1 cm in the sagittal plane at the medial upper pole.

2 months post-discharge, the patient was switched to amoxicillin-clavulanate oral therapy; at that time, the largest splenic collection seen on ultrasound had reduced to $2.8 \text{ cm} \times 2.5 \text{ cm} \times 2.8 \text{ cm}$. Five months post-discharge and after approximately 6 months total antimicrobial therapy, the splenic collections had stabilized and showed only residual scarring and calcifications (Figure 4). Consequently, antimicrobial therapy was discontinued. Repeat ultrasonography 5 months later did not demonstrate any re-accumulation of fluids and showed resolution of splenomegaly.

Discussion

Isolated splenic abscesses are a dangerous, but fortunately uncommon condition in children requiring a high degree of



Figure 4. Abdominal ultrasound approximately 5 months postdischarge. Findings were suggestive of resolved splenic abscesses with residual scarring, calcifications, and splenomegaly.

clinical suspicion.¹⁻³ The triad of fever, upper quadrant abdominal pain, and leukocytosis is not specific but should raise suspicion for a splenic abscess.^{2,3,6} In addition to these signs and symptoms, patients may complain of left shoulder pain due to involvement of diaphragmatic pleura, and shortness of breath in the presence of splenomegaly and pleural effusion. Abdominal CT scans have >90% sensitivity and specificity for detecting splenic abscesses and are considered the imaging modality of choice.^{2,3,6} Ultrasound imaging in children has been shown to have 100% sensitivity in two recent case series and has been useful in guiding percutaneous aspiration, as was the case with our patient.^{2,3} Interestingly, our patient was afebrile with only mild leukocytosis at the time of presentation, despite not having received antibiotics. Due to the wide variety of causative pathogens implicated in these infections, broad empiric antimicrobial coverage and comprehensive evaluation for the potential source of infection are required in all individuals presenting with splenic collections.

Predisposing conditions for paediatric splenic abscess include bacterial infection at a distant or contiguous site, splenic trauma, hemoglobinopathies, and an underlying immunocompromised state.^{2,3,6} Patients should be evaluated for a potential source of infection, including an ear–nose– throat focus, endocarditis, or disseminated intra-abdominal infection sites. Rates of positive blood cultures are difficult to estimate due to the small number of cases and variety of pathogens implicated. Many centres do not routinely perform anaerobic blood cultures in children; while the overall incidence of true anaerobic bacteremia in children is low, these blood cultures should be drawn when an intra-abdominal infection is suspected.⁷ Our patient's review of systems was non-contributory, except for a recent episode of gastroenteritis. We hypothesized that his splenic abscess may have developed as a complication of a gastrointestinal infection, possibly seeding a healing hematoma that he acquired during an all-terrain vehicle injury.

Enteric gram-negative bacteria, including Klebsiella pneumoniae, Escherichia coli, and Salmonella enterica, are the most commonly reported organisms causing isolated splenic abscesses in adults and children.^{2,6,8} Typhoid fever, caused by Salmonella enterica serovar Typhi, is a frequent etiology of paediatric splenic abscess in endemic regions.^{2,3} Gram-positive bacteria implicated in splenic abscesses include viridans group streptococci, Staphylococcus aureus, and Enterococcus spp.; these bacteria have been more commonly reported in association with infective endocarditis.^{6,9} Our patient's echocardiogram was unremarkable. Anaerobic organisms have been associated with polymicrobial splenic abscesses,⁶ although only a single organism was isolated in our case. Our patient had been receiving broad-spectrum antibiotics for several days prior to surgery, however. Fungal infection should be considered in immunocompromised patients.^{2,3,6} Our patient did not have risk factors for Bartonella henselae, Brucella spp., Mycobacterium tuberculosis, or Burkholderia pseudomallei, all of which are known to cause visceral abscesses. Cystic echinococcosis is caused by incidental ingestion of Echinococcus granulosus eggs. Again, our patient did not have risk factors for infection with this tapeworm.

Fusobacteria are anaerobic gram-negative bacilli found in oral, gastrointestinal, and female genital flora. Fusobacterium necrophorum and F. nucleatum are implicated in the majority of invasive human infections.¹⁰ Splenic abscesses due to Fusobacterium spp. have been described in case reports as complications of Lemierre's syndrome,¹¹ infective endocarditis,¹² and splenic hematoma secondary to trauma with co-existing respiratory tract illness.13 Pylephlebitis secondary to Fusobacterium spp., also known as gastrointestinal or abdominal variant of Lemierre's syndrome, is a rare clinical entity described in case reports in the literature. The portal vein septic thrombophlebitis seen in this syndrome is believed to be secondary to contiguous spread or spread from regions drained by the portal system.¹⁴ Review of the literature revealed one case report of splenic abscesses associated with hepatic abscesses and the gastrointestinal variant of Lemierre's syndrome.14,15 Fusobacterium spp. are susceptible to a broad spectrum of antibiotics, with small numbers of isolates demonstrating penicillin and erythromycin resistance. Recommended empiric treatment includes a third-generation cephalosporin and metronidazole to account for potential beta-lactamase production and polymicrobial infection.¹⁶

Source control (i.e. drainage) is usually indicated in collections larger than 3–5 cm in diameter.^{3,8} Spleen-preserving interventions with image guided placement of a percutaneous drain are becoming increasingly common with good outcomes in children;^{2,3} however, fluid re-accumulation necessitating repeat drainage may occur, as evident in our case. Empiric treatment of splenic abscesses usually includes broad grampositive, gram–negative, and anaerobic coverage.⁶ This can be achieved through the combination of a third-generation cephalosporin and metronidazole or piperacillin-tazobactam monotherapy. Once the causative organism(s) is identified, the antimicrobial regimen can be narrowed accordingly. Recommended duration of treatment depends on the organisms identified, source control, and other organ systems involved. Durations of 3–8 weeks have frequently been cited in the literature.

Conclusion

Isolated splenic abscesses can be caused by a wide variety of potential pathogens. Initial empiric management should include broad-spectrum antimicrobial therapy targeting gram-positive, gram-negative, and anaerobic organisms. Aerobic and anaerobic blood cultures should be drawn when an intra-abdominal infection is suspected in a paediatric patient. Source control with abscess drainage is generally indicated when collections are greater than 3–5 cm in diameter, and fluid should be sent for culture. *Fusobacterium* species are typically associated with oropharyngeal infections but can rarely be implicated in splenic abscesses and are often associated with polymicrobial infections.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

Written informed consent was obtained from the patient and their guardian (parent) for their anonymized information to be published in this article. The guardian (parent) was the legally authorized representative of the minor subject

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