CASE REPORT

The connection between acute otitis media and the acute abdomen

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SUMMARY

A female aged 9 years with a recent episode of acute otitis media (AOM) presented to her primary care physician with complaints of severe abdominal pain with right lower quadrant rebound tenderness, suggestive of an acute surgical abdomen. Neurological examination was normal on presentation. She was transferred to the local children's hospital for workup of appendicitis, during which she began exhibiting ataxia and slurred speech. Further evaluation revealed mastoiditis, venous sinus thrombosis and subdural empyema. Appendicitis was ruled out. We describe the first documented case of neurological complications of AOM presenting as an acute surgical abdomen without initial neurological findings.

BACKGROUND

Our case highlights many important lessons that primary care physicians will encounter. First, we present an example of when prematurely focusing on one diagnosis may distract the clinician from the true diagnosis and lead to diagnostic delay. Moreover, we illustrate an example of the interplay between distant body systems; connecting abdominal pain to a neurological infection. We believe that this case provides a fund of knowledge for other primary care physicians.

Complications of acute otitis media (AOM) are varied and can result in significant harm. Of these, most notable are intracranial disease such as venous sinus thrombosis (VST) and cerebral abscess, and extracranial disease such as mastoiditis. VSTs among children most commonly present as isolated, gradual-onset, localised headaches.¹ Although less likely, 'thunderclap' and migraine-type headaches may occur.²⁻⁵ Focal neurological findings, encephalopathy and seizures are possible findings as well. Intracranial abscesses are associated with the classic triad of headache, fever and focal neurological findings; however, less than half of cases present with this triad.⁶ Common findings for intracranial abscesses also include lethargy, nausea and vomiting.⁷ Mastoiditis is characterised by postauricular tenderness, erythema, fluctuance, auricular displacement or protrusion and ear pain.^{7,8}

In the case of our patient, she presented with a primary concern of acute onset abdominal pain in the setting of an episode of AOM. Her presentation has never been reported in the literature as the presenting complaint for VST, mastoiditis or subdural empyema.

CASE PRESENTATION

Our patient is a female aged 9 years with no significant past medical history who presented to her primary care provider on 21 January 2016 with the complaint of 5 days of fever, bilateral ear pain, anorexia, non-bloody and non-bilious emesis and 2 days of generalised abdominal pain. Two days prior to presentation, she was seen at an outside emergency department for AOM. Because of a penicillin allergy, she was given azithromycin 500 mg for 1 day, followed by 250 mg for 4 more days. However, after the first dose, she began to vomit, and did not complete her antibiotic therapy. The remaining review of systems were negative. On arriving to the primary care paediatrician, initial physical examination revealed that she was tired and ill-appearing with purulent drainage from her left ear and acute tenderness in the right lower quadrant of her abdomen. She exhibited rebound and guarding. Her urinalysis in the outpatient clinic was negative, and due to concern for acute abdomen and serious bacterial infection she was sent to the emergency department of the local Children's Hospital.

INVESTIGATIONS

Initial labs at the emergency department showed a white blood cell count of $23 \times 10^3/\mu\text{L}$ and a C reactive protein of 28.6 mg/L. Chemistry was within normal limits. Her left ear was again noted to have purulent drainage, and her abdomen continued to be very tender to palpation with guarding. She was initially admitted to the surgical service given concern for appendicitis. On her first day of admission, she received a limited abdominal ultrasound, which was negative for appendicitis. During the ultrasound, the patient was noted to have slurred speech and right-sided weakness. Neurology was then consulted, and felt that her symptoms were more consistent with a seizure disorder, therefore, they recommended an electroencephalogram (EEG) which showed focal left paracentral slowing. After receiving the results of the EEG, neurology recommended a brain MRI and magnetic resonance venography (MRV). These studies were done the same day, and they revealed meningeal enhancement of the frontal and parietal lobes consistent with meningitis, as well as left venous sinus thrombosis and left mastoiditis. Haematology was then consulted. They recommended a hypercoagulable workup, which was negative. Due in part to her persistent right-sided weakness, a subsequent brain MRI was obtained, 6 days after the first,

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which showed a subdural empyema. The patient's venous sinus thrombosis, subdural empyema, meningitis and cerebritis were attributed to a complicated otitis media and mastoiditis. Blood culture drawn at the time of admission grew coagulase-negative staphylococcus at 24 hours, which was thought to likely be a contaminant. A lumbar puncture revealed cerebral spinal fluid (CSF) with a white blood cell count of 153 cells/ μ L, with 59% neutrophils and 41% monocytes. The CSF protein level was 71 mg/dL and glucose 67 mg/dL. CSF culture had no growth. CSF was positive for *Streptococcus pneumoniae* by PCR (negative PCR for *Neisseria meningitidis* and *Haemophilus influenzae* type b). Subdural empyemic fluid culture had no growth, and culture of her ear drainage grew scant coagulase-negative staphylococcus.

Studies completed during this hospitalisation:

Hospital day 1—ultrasound abdomen limited

Hospital day 1—EEG

Hospital day 1—MRI and MRV brain with contrast

Hospital day 3—CT orbit, ear, temporal bone with and without contrast

Hospital day 3—CT neck with contrast

DIFFERENTIAL DIAGNOSIS

- Mesenteric adenitis (working diagnosis).
- Lemierre's syndrome with otogenic variant.
- Mesenteric ischaemia and cavernous sinus thrombosis secondary to hypercoagulable state induced by serum inflammation.
- Primary otitis media with spread to adjacent tissues causing central nervous system infection with secondary bacteraemia/fungaemia, leading to haematogenous spread to the abdomen.
- Abdominal pain secondary to recurrent vomiting in the setting of elevated intracranial pressure.

TREATMENT

The patient was started on vancomycin and ceftriaxone empirically on admission. Infectious disease consultants recommended adding metronidazole on day 2 of hospitalisation given the unclear aetiology of her infection and her persistent fevers. She was treated with intravenous ceftriaxone and metronidazole for 2 weeks inpatient, followed by an additional 4 weeks outpatient (via a peripherally-inserted central catheter (PICC) line). On discovery of the venous sinus thrombosis, the patient was started on a heparin drip and later in her hospital course transitioned to enoxaparin and heparin XA (low molecular weight heparin). She also underwent tympanostomy tube placement and left mastoidectomy on day 2 of hospitalisation, followed by burr hole irrigation of the subdural empyema 5 days later. The patient was followed by neurology, haematology, neurosurgery, otolaryngology, infectious disease and physical therapy during the course of her stay.

Procedures completed during this hospitalisation:

Hospital day 2—bilateral myringotomy tubes, and mastoidectomy with wash out

Hospital day 4—single lumen PICC line placement in left brachial vein

Hospital day 7—burr hole irrigation of subdural empyema

OUTCOME AND FOLLOW-UP

The patient's abdominal pain resolved within the first 2 days of her hospital stay. Patient was discharged from the hospital after 22 days. Follow-up brain MRI 3 weeks after hospital discharge

showed resolution of the subdural empyema and the venous sinus thrombosis, with resolving cerebritis, and evolving post-surgical changes of the left mastoid process. Her antibiotics were discontinued at that time, and her PICC line was removed the following day. By her follow-up appointment with haematology 7 weeks after discharge, patient had 'returned to her baseline normal activity without any evidence of ongoing infection' or sequelae.

On recent follow-up, now 11 months after initial presentation, the patient exhibits a full recovery. She has graduated from physical therapy, and is back to practicing competitive gymnastics. Our patient is no longer on any medications, and has been discharged from the haematology, infectious disease, neurosurgical and neurology specialists. She does continue to see the otolaryngologist and audiologist for follow-up care while her tympanostomy tube remains in place.

DISCUSSION

We present the case of a female aged 9 years with multiple sequelae of AOM demonstrating an atypical clinical presentation. VSTs are considered exceptionally rare, with the incidence reported at 0.67/100 000 in children.⁹ Studies also show that VSTs are more common in neonates and infants, rather than children or adolescents.⁹ Furthermore, while mastoiditis is more common than VSTs, the incidence of mastoiditis is exceedingly rare in a child aged 9 years. Various studies have demonstrated that mastoiditis occurs most often in children <2 years of age, with one report of a median age of 48 months.^{10–12} Moreover, as mentioned above, the clinical presentation for these particular sequelae largely present with neurological findings. Our patient did not initially exhibit any of the aforementioned neurological findings.

Demographically, statistically and clinically, our patient exists as an anomaly to the current literature. Our case highlights the essential nature of acknowledging the connection between distinct organ systems. Additionally, focusing on the possibility of an acute emergency can be a distraction from investigating its aetiology, and may lead to diagnostic delay.

In an attempt to connect otitis media with abdominal pain, we propose the diagnosis of mesenteric adenitis. Mesenteric adenitis is associated with group A streptococcal (*Streptococcus pyogenes*) pharyngitis, which is also present in cases of AOM.^{13 14} Mesenteric adenitis has been shown to present very similarly to appendicitis.¹⁴ A potential cause of our patient's severe abdominal pain with rebound and guarding may have been her *S. pneumoniae* otitis media contributing to the development of mesenteric adenitis. It is possible that the mesenteric lymph node enlargement was missed on the limited abdominal ultrasound, as the ultrasound focused primarily on the appendix. Along with this theory, we also postulate that a primary otitis media may have occurred, with spread into contiguous tissues causing mastoiditis, subdural empyema and venous sinus thrombosis. This infection may have then become haematologically disseminated, with resultant seeding of the infection in the abdomen, which prompted the need for intravenous antibiotics. Interestingly, her ear fluid culture grew coagulase-negative staphylococcus, an organism also found in her blood culture, which correlates this theory. However, this was considered to be a contaminant, and not a true infection. This theory is less likely considering the bacteria that was isolated in her CSF was *S. pneumoniae*, and that her blood cultures were presumably negative. Furthermore, we considered the link between elevated intracranial pressure and repetitive vomiting to be a potential

explanation for her abdominal pain. However, abdominal pain due to vomiting would not typically present as severe right lower quadrant pain with rebound tenderness and guarding.

Another potential explanation to connect these symptoms include the otogenic variant of Lemierre's syndrome. Lemierre's syndrome is a suppurative thrombophlebitis involving the internal jugular veins, most often caused by *Fusobacterium necrophorum*. Thrombosis of the jugular veins occur, followed by septic emboli to the lungs.¹⁵ A case reported by Masterson *et al* revealed an otogenic variant of Lemierre's syndrome, in which a young girl suffered bilateral mastoiditis with subsequent internal jugular vein thrombosis.¹⁶ While this explanation is plausible, it is weakened by the fact that she did not present with pulmonary symptoms, which is a classic finding in Lemierre's syndrome. Chest X-ray was not performed during the course of this illness.

Moreover, considering her elevated CRP suggesting serum inflammation, she may have been in a hypercoagulable state. This hypercoagulable state could have presented with two separate infarctions, one being her documented venous sinus thrombosis, and the other being a thrombus in a mesenteric vessel. This theory is supported by her improvement while on anticoagulants and decreased abdominal pain with bowel rest (a hallmark aspect of managing mesenteric ischaemia). However, considering her otitis media, mastoiditis and subdural empyema, it is more likely that the thrombus in her cerebral sinus was due to contiguous spread of infection with vascular insult, rather than primary vascular thrombosis.

Unfortunately, due to the high morbidity associated with appendicitis, it is likely that the clinician will narrowly focus on this diagnosis, at the expense of a broad diagnostic workup. In this scenario, focusing on the diagnosis of appendicitis resulted in a distraction away from the patient's ear infection and its potential sequelae. Had neurological symptoms not developed, she may have suffered from a higher morbidity, due to delayed diagnosis. Luckily she had been admitted to a Children's Hospital, under the care of multiple specialists, where she was able to have an EEG and then subsequent brain MRI with MRV with little time delay. It is interesting to note that this patient had an EEG performed prior to any imaging of her brain. At the time, the patient had been admitted to the surgical service for a likely appendicitis, and the recommendation to obtain an EEG by the consulting neurology team had been followed without consideration of prior emergent brain imaging. Also interesting to note is that no further workup (beyond the initial limited ultrasound) or discussion regarding the cause of her acute abdominal pain is apparent in review of her medical records. There appears to have been a somewhat myopic focus on the symptom that was most severe at a given time (initially abdominal pain, later the neurological symptoms), without much effort to search for connections or provide more holistic care.

In conclusion, our unique case provides important teaching points for any physician treating patients in the ambulatory clinic. We found in this case, that prematurely focusing on one diagnosis led to diagnostic delay. Furthermore, we highlighted that distant body systems may be unsuspectingly related, requiring a 'whole-body' approach for accurate diagnosis. An important lesson to be taken away from this case is that one unifying diagnosis is more likely than two separate, unrelated

diseases. Fortunately, our patient is healing appropriately and remains well overall.

Learning points

- ▶ Prematurely locking onto a diagnosis may distract a clinician from the true diagnosis.
- ▶ Distant body systems may be unsuspectingly related, therefore, clinicians should always exercise a 'whole-body' approach.
- ▶ For a set of signs and symptoms, one unifying diagnosis is more likely than two separate aetiologies.
- ▶ There is a potential connection between abdominal pain and neurological infections that is not well-defined by our current literature. Mesenteric adenitis may be one possible explanation.

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