

CLINICAL VIGNETTE

The Case of Intractable Hiccups and Backaches

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The primary care physician is often the first to encounter atypical presentations of common illnesses. Due to the complex and yet nonspecific nature of gut sensory innervation and the commonality of referred pain, it can be helpful to adopt an anatomic rather than a strict pattern recognition approach to the diagnosis of GI complaints. We present a man with two atypical concurrent presentations of relatively common conditions (pancreatitis and colitis), and review literature on presentation and epidemiology relevant to primary care practice.

A 51 year old male smoker of South Asian ancestry with no relevant past medical history had an episode of diverticulitis six years ago. Colonoscopy revealed transverse, descending, and sigmoid diverticula, one hyperplastic polyp in the transverse colon, and one tubular adenoma in the sigmoid colon without high-grade dysplasia. The patient remained asymptomatic until six years later when he developed abdominal cramps and back pain, with CT demonstrating “exuberant wall thickening of proximal and mid sigmoid colon, most likely the sequela of diverticulitis, with small hypodense foci within the colonic wall likely small intramural abscesses”. His symptoms resolved after a short course of metronidazole and ciprofloxacin.

Two months later, the patient presented with two days of nausea followed by refractory hiccups so severe they prevented sleeping. Upon further questioning, the patient also reported fevers greater than 100°F the day prior, mild shortness of breath, and mild bilateral back aches over the past two days. He denied abdominal or epigastric pain, cough, upper respiratory symptoms, diarrhea, recent sick contacts, foreign travel, rashes, or suspicious food intake. Physical exam showed a heart rate of 120, blood pressure of 125/90, and a temperature of 96°F. Lungs were clear to auscultation bilaterally, his abdominal exam was soft, nontender, and non-distended in all quadrants. Back soreness was not reproducible on exam. He had no costovertebral angle tenderness, and the remainder of his exam was unremarkable. Labs were remarkable for a white blood cell $14.08 \times 10^3/uL$, platelets $127 \times 10^3/uL$, sodium 133 mmol/L, chloride 91, potassium 3.7, creatinine of 0.8mg/dL, aspartate amino transferase 101 U/L, alanine amino transferase 90 U/L, alkaline phosphatase 176 U/L, and total bilirubin 2.2 mg/dL. Chest x-ray was within normal limits, and a muscle relaxant (cyclobenzaprine) failed to provide relief for the hiccups. Abdominal CT scan the next day revealed: “1. Interval development of heterogeneous enhancement of the pancreatic tail with surrounding stranding, compatible with pancreatitis. 2.

Colonic diverticulosis with near interval resolution of the previously visualized peri-sigmoid stranding with persistent sigmoid colonic wall thickening, compatible with resolving acute diverticulitis, with evidence of chronic diverticular disease with no abscess.”

With recurrence of his nausea and vomiting, the patient was referred to the emergency room, where labs were noteworthy for lipase 159 U/L, creatinine 1.4mg/dL (baseline 0.8mg/dL), and blood ethanol level of 276mg/dL. He was given parenteral hydration, demonstrated good oral intake, and discharged home with a presumptive diagnosis of alcoholic pancreatitis. As the patient’s condition continued to improve with resolution of his symptoms over the next several days, no antibiotics were prescribed. The patient was referred to gastroenterology, but deferred follow up.

He presented three times with recurrent hiccups, left sided abdominal pain, increasing abdominal distention, and decreased bowel movements but without any fever, nausea, or vomiting. He was treated with regimens of ciprofloxacin and metronidazole of various duration with temporary relief. He finally presented to gastroenterology several months later with residual intermittent abdominal pain radiating to the back, 20 pounds of weight loss, but no nausea, vomiting or bowel changes.

CT abdomen and pelvis revealed: “*Circumferential wall thickening of the splenic flexure and entire descending colon extending into the proximal sigmoid with surrounding peri-colonic stranding. A few colonic diverticula are noted, however the length of the inflamed colon suggests more diffuse process. Mild prominence of the terminal ileum without significant surrounding fat stranding to suggest acute inflammation.*”

Upon repeat 1 month later: “*Persistent long segment colitis. Of note, the distal descending colon has significant persistent circumferential thickening and luminal narrowing with intussusception.*”

MRCP showed: “*1. Pancreatic tail hyperenhancing lesion, 1.0 cm, similar to spleen on all sequences, most likely an intrapancreatic splenule. 2. Bowel wall thickening and enhancement, extending from the sigmoid colon to the splenic flexure, likely representing colitis, more extensive and persistent than expected for diverticulitis. Given possible*

terminal ileal wall thickening and enhancement, findings may represent inflammatory bowel disease such as Crohn's colitis.
3. *Minimal intrahepatic bile duct irregularity, possibly artifactual, although could also represent early changes associated with inflammatory bowel disease if bowel abnormality is proven to represent IBD.*"

Two subsequent colonoscopies with diagnostic and pediatric scopes failed to pass a segment of the sigmoid colon due to a narrow, friable stricture. The rectal region was unremarkable. One biopsy taken showed an inflammatory polyp without atypical features. The patient was prescribed a six week course of ciprofloxacin and metronidazole, with plans for repeat imaging and subsequent colectomy.

Discussion and Conclusion

Our patient had an atypical presentation of presumed alcoholic pancreatitis masquerading as hiccups in the absence of epigastric pain. However, an alternate explanation is acute and chronic pancreatitis related to inflammatory bowel disease (IBD).¹ Some cases of pancreatitis are believed to be secondary to IBD related Primary Sclerosing Cholangitis or Cholelithiasis, while others have a less clear pathophysiology.

Our patient's second atypical presentation was of colitis masquerading as back pain in the presence of a normal initial abdominal exam. While the final pathological diagnosis is still pending, his four courses of recurrent and now persistent colitis over 9 months as well as his remarkable imaging findings in the absence of compromised immunity or other infectious risk factors make IBD strongly suspicious. It is concerning for Crohn's Disease due to absence of rectal disease in the presence of significant colonic disease, possible involvement of the biliary tree and terminal ileum on MRI, absence of gross bleeding, and his smoking history.

Crohn's Disease may have an especially long prodromal period, up to 3-7 years on average, with symptoms such as bloating, diarrhea, abdominal pain, heart burn, weight loss, and fatigue.^{2,3} In the meantime, patients are often diagnosed with other conditions such as Colitis from enteric pathogens or Diverticulitis, both of which can be common in the bi-modal age distribution (around 20 and 50 years old) that is seen in IBD patients. It is important for the primary care provider to have a healthy index of suspicion even for patients with somewhat atypical symptoms such as back pain (in this case), or the aforementioned heartburn, weight loss, and fatigue. Recurrent symptoms of diverticulitis even with a previously indicative colonoscopy may warrant further work up, especially during inflammatory episodes.

The third atypical aspect of this patient's presentation was his South Asian descent⁴, in contrast to the commonly seen association with European heritage. The epidemiology and presentation of Crohn's disease in Asian patients may not follow the same patterns of Caucasian cohorts. Major differences include a loss of the bimodal age distribution, a more equal gender distribution, and less perianal disease.⁵ Unlike cardiovascular disease, there are no validated risk

calculators to assist in diagnosis of Crohn's. More research is needed to identify the most helpful initial clinical features of Crohn's Disease so that we may shorten the delay to diagnosis and treatment of this debilitating condition.

REFERENCES

1. **Ramos LR, Sachar DB, DiMaio CJ, Colombel JF, Torres J.** Inflammatory Bowel Disease and Pancreatitis: A Review. *J Crohns Colitis.* 2016 Jan;10(1):95-104. doi:10.1093/ecco-jcc/jjv153. Epub 2015 Sep 7. Review. PubMed PMID: 26351384.
2. **Pimentel M, Chang M, Chow EJ, Tabibzadeh S, Kirit-Kiriak V, Targan SR, Lin HC.** Identification of a prodromal period in Crohn's disease but not ulcerative colitis. *Am J Gastroenterol.* 2000 Dec;95(12):3458-62. PubMed PMID: 11151877.
3. **Mekhjian HS, Switz DM, Melnyk CS, Rankin GB, Brooks RK.** Clinical features and natural history of Crohn's disease. *Gastroenterology.* 1979 Oct;77(4 Pt 2):898-906. PubMed PMID: 381094.
4. **Nguyen GC, Chong CA, Chong RY.** National estimates of the burden of inflammatory bowel disease among racial and ethnic groups in the United States. *J Crohns Colitis.* 2014 Apr;8(4):288-95. doi: 10.1016/j.crohns.2013.09.001. Epub 2013 Sep 24. PubMed PMID: 24074875.
5. **Kalaria R, Desai D, Abraham P, Joshi A, Gupta T, Shah S.** Temporal Change in Phenotypic Behaviour in Patients with Crohn's Disease: Do Indian Patients Behave Differently from Western and Other Asian Patients? *J Crohns Colitis.* 2016 Mar;10(3):255-61. doi: 10.1093/ecco-jcc/jjv202. Epub 2015 Oct 29. PubMed PMID: 26519461; PubMed Central PMCID: PMC4957468.

Submitted June 28, 2017