An Unusual Cause for Massive Inflation

Abstract:
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Abstract
Chagas disease is a rare condition but with an increasing incidence. Megacolon is a known sequela. Surgical management remains the only disease modifying treatment option with variable long-term success. We highlight an interesting case to emphasize attention to this rare condition as a differential diagnosis in any patient presenting with massive intestinal dilatation.

Introduction
Worldwide the most common cause of megacolon is due to an infective process (Trypanosoma cruzi). With increasingly global integration, infective causes of megacolon are more prevalent in developed countries. Surgery is the only disease modifying treatment option. Surgical management has had innumerable modifications over the years with the intention to improve outcomes.

Case Report
We report a sixty-nine year old gentleman that presented with a one-week history of abdominal distention, nausea and absolute constipation. A grossly distended abdomen was noted on initial plain radiography. Subsequent computerized tomography (CT) scan showed massive dilatation of both the colon and rectum without evidence of a focal transition point. Interestingly, on further review of the patient's history, he had spent a significant amount of time living in South America. The possibility of transmamamorial infection was considered. Cardiac investigations (echocardiogram) did not detect any abnormality. Additionally, barium swallow showed no involvement of the upper gastrointestinal tract. A conservative management approach was refractory. Due to long-standing colonic stenosis the prospect of elective bowel resection was discussed with the patient. Four months later, he was electively admitted for a total abdominal colectomy and ileorectal anastomosis. The colon and rectum were grossly distended at surgery with no transition point (Figure 1). Day-eight post-operatively the patient developed severe lower abdominal pain. CT scan observed no collection, but a significant volume of free air was detected. The patient was returned to theatre for exploration. A small area of necrosis along the suture line was properly repaired and a defunction ileostomy fashioned. Recovery was uneventful thereafter. Ileostomy was reversed three-months later, and at twelve-month review he had made a full recovery. Histopathology observed an eosinophilic infiltrate, transmural inflammation of the distal colon, and though T. cruzi was not identified, this was felt to be the causative factor. Additionally, immunohistochemistry with neuronal markers showed thinning of the nerve fibres with prominent residual ganglion cells in the outer plexus in keeping with T. cruzi infection.

Discussion
Carlos Chagas first described Chagas disease in 1909. The causative agent is the protozoan T. cruzi, which is transmitted by insects. It is endemic in parts of South America, but not confined to this region. Increasing migration patterns have made it a worldwide issue. More than 18-million people worldwide have T. cruzi, with an estimated 20,000–deaths annually. Acute T. cruzi though largely asymptomatic, can have flu-like manifestations with varying severity, typically within eight-weeks, less than 5% of patients die acutely. Approximately, 30% of patients develop a chronic infective state, with long asymptomatic phases without electrocardiographic or radiological abnormalities. Major morbidity and mortality relate to the development of cardiac and mega-syndromes. 4.5% develop a mega-syndrome (megacolon or megaesophagus) characterized by gross dilatation and thickening of their walls. Irreversible destruction and degeneration of the intra-mural intestinal nervous system with loss of coordinated motility results in progressive constipation, eventually resulting in faecal impaction and/or volvulus. Diagnosis is through direct demonstration of a parasite in human tissue/blood, which is only detectable in the acute phase. Alternatively, antibody testing and/or enzyme-linked immuno-sorbant assays are required. Treatment of Chagas disease is difficult with limited success. Anti-trypanosomal drugs (Nifurtimox/Benznidazole) have been utilized, but cytotoxic effects limit their generic application. Ultimately, drug therapy has been largely unsuccessful with little evidence to show that the progression of cardiac or digestive tract patholgy. Surgical management is a significant challenge, as the dilated organ distorts anatomy making dissection and ligation of vessels difficult. The two most commonly used procedures include the low anterior resection and the Duhamel-Haddad technique (2-stage technique). The latter remains popular in South America. Despite the technique, recurrence remains an appreciable concern with a lack of long-term follow-up data.

In conclusion, this case highlights a rare condition, and the importance of clinical suspicion when a patient presents with massive organ dilatation and a history of tropical travel.

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References