Incarcerated solitary cecal diverticulum: A case report of the primary documented occurrence and literature review.

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ABSTRACT:

The solitary cecal diverticulum is an unusual cause of right lower quadrant pain that should be considered in the differential diagnosis of right lower quadrant pain. Approximately one thousand cases have been described in medical literature since its initial report in 1912. We present the case of an 18 year old male with the first documented report of a strangulated cecal diverticulum and a review of the current literature on the management of this condition.
Solitary cecal diverticula are unusual causes of RLQ pain most commonly found incidentally during appendectomy. With an estimated incidence of one in three hundred preoperatively diagnosed appendectomies, a cecal diverticulum will be found. Due to the rarity of the disease, it is unlikely to be seen by most surgical residents during their training. Historical clues include prolonged length of abdominal discomfort or prior subacute incidents of similar pain. Resection with primary closure is indicated when found during laparoscopy. A trial of antibiotics is warranted when only inflammation is found on computerized tomography, with expectant resection.

CASE PRESENTATION:

An 18 year old black male presented with sharp, constant right lower quadrant pain. He had been well until four days prior when he developed periumbilical pain that had initially been dull, intermittent, and without migration. He had multiple episodes of this in the past and attributed it to constipation. On the day of presentation, he awoke with sudden worsening of this abdominal pain which had migrated to his right lower quadrant causing anorexia with nausea, but without vomiting or urinary symptoms. He had one liquid, bloodless bowel movement which lessened the pain for a short time prior to presentation in the ED.

This young man had a medical history significant for Kawasaki's disease treated with IgG at four months of age, a surgically corrected patent ductus arteriosus at age 2, vesicular urethral reflux, and mild intermittent asthma for which he had not received treatment in over one year and had no history of intubations due to asthma.

On arrival to the ED, he was pyrexic 38·2 C, pulse 94, BP 106/74 and mildly tachypneic at 20 breaths per minute but without any other signs of respiratory distress. The patient was obese, with Hippocratic facies, and in to pain. He located the epicenter of pain 2 cm to the right and 2cm inferior to his umbilicus. His abdominal exam revealed no surgical scars, tenderness to percussion in the RLQ with guarding, rebound tenderness, psoas sign, and Dunphry's sign. Obturator, Rovsing, and Aaron signs were negative. His laboratory values revealed a leukocytosis of 10,100 with an 82·8% left shift. Serum chemistry was within

normal limits. Prior to surgical consult, an ultrasound was performed showing a 1·2 cm tubular structure which was non-compressible with a thickened wall and a small amount of free fluid in the pelvis.

The patient was determined to have an Alvarado score of 9 and he was taken to the operating room for a laparoscopic appendectomy after intravenous fluid resuscitation. Intraoperatively, a normal appearing appendix was identified and an incidental appendectomy performed. On further inspection of the cecum, a one centimeter violet mass was discovered. This mass had herniated from the lateral aspect of the cecum through the mesentery. This was reduced, however the tissue remained necrotic and friable, and it was excised and the defect suture ligated. Pathological examination revealed fragments of fibro-adipose tissue with marked acute and chronic inflammation (see inserts below). Separate pieces of colonic wall with marked inflammation and necrosis but no colonic mucosa was identified. The patient was discharged home postoperative day eleven after a difficult hospital course.

DISCUSSION:

Solitary cecal diverticulitis was initially described in 1912 by Potier and colleagues, approximately 1000 cases have been recorded since first reported.[6,15] It is an uncommon cause of RLQ pain that is found incidentally at laparoscopy or laparotomy in approximately one in three hundred patients.[11] This occurs more commonly in Asians.[6] In western countries 85% of all diverticula occur in the descending and sigmoid colon. In Asian countries, the incidence of right sided diverticular disease can be 71% of those affected[9]. While in European and American studies 1 to 2 percent of right colonic specimens may contain cecal diverticula, Asian studies have revealed that the incidence can be as high as 43 to 50%.[15]

The accepted theory of embryological development is that a cecal outpouching develops within the sixth week, and by the seventh week, in the vast majority of cases, the protuberance has atrophied. Should the transient appendix not atrophy, a cecal diverticulum is formed.[3,12] This theory is supported by the work of Kelly and Hurdon in 1905 where 50 embryos were found to contain a temporary outgrowth from the cecum which would atrophy by the seventh week. Gladstone in 1924 added to the discussion that although the formation of the diverticulum simulates the true appendix, it should be considered a separate entity as it atrophies and the appendix veriformis differentiates later.[3] This theory is supported by evidence such as the three year old girl found to have an inflamed solitary cecal diverticulum in 1931 by Odqvist.[12]

The typical presentation of a patient with solitary cecal diverticulitis consists of RLQ pain, fever and occasionally peritoneal signs.[19] Cecal diverticular disease presents similarly to appendicitis with subtle dissimilarities. The course tends to be longer and more insidious, the pain begins and remains in the right lower quadrant and the patient will be nontoxic in appearance. Nausea and vomiting are uncommon symptoms.[9] Only 9% of solitary cecal diverticula are correctly diagnosed preoperatively.[10, 11,12,19] 80% of cecal diverticula are located within the area one to two centimeters from the ileocecal valve.[19] 60% are located on the anterior aspect of the cecum and when severely inflamed can perforate and cause a generalized peritonitis. Those that are on the posterior aspect will not produce peritonitis and may mimic a perforating carcinoma.[12] Dearden described a case series of three patients found to have a solitary cecal diverticulum. The common pattern that emerged was a prolonged history, days to weeks, of abdominal discomfort eventually leading to a sudden severity in the right lower quadrant, mimicking acute appendicitis.[7] Kachroo also describes a patient with vague abdominal pain for 3 days in the right upper quadrant with clinical findings necessitating operative intervention. After entering the abdomen, a hemicolectomy

was performed due to concern of a malignancy and pathologic examination determined that cecal diverticulitis was the culprit of this pain.[10]

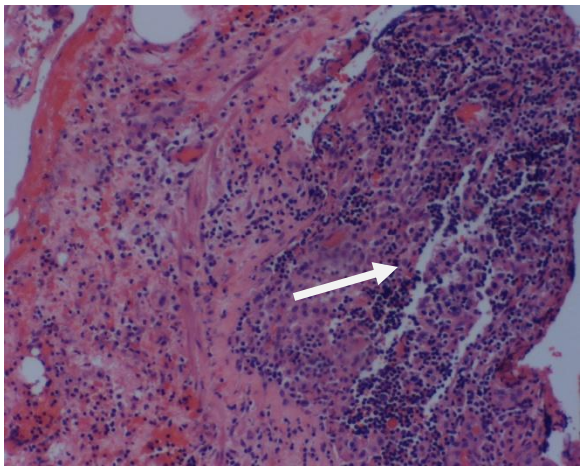
Preoperative diagnosis of right sided diverticulitis is difficult without imaging. Computerized Tomography is the most reliable diagnostic tool with 98% sensitivity.[11] Ultrasonography has also been shown to be highly sensitive and specific, 91·3 and 99·5% respectively. A circular hypoechoic or anechoic area along the colon with segmentally thickened colonic wall is highly suspicious for a diverticulum.[4,19] Differential diagnosis of acute right lower quadrant pain should include appendicitis, Crohn's disease, tubo-ovarian etiology in women, perforated cholecystic or gastric disease, and tumor of the ascending colon, ileocecal valve or appendix. This clinical entity may mimic not only appendicitis but localized carcinoma, ischemic colitis or perforation.[10] Like other forms of diverticular disease, bleeding may occur and other rare conditions such as angiodysplasia, Osler-Weber-Rendu syndrome, Sturge-Weber, and Bonnet-Dechaume-Blanc syndrome should be ruled out with appropriate testing.[17] In an audit of 1492 suspected appendectomies at a Hong Kong facility, 17 neoplastic etiologies were discovered and 42 inflammatory conditions identified (2 granulomatous appendicitis and 40 diverticular disease).[13] It should be remembered that the Asian population has higher risk of right sided diverticular disease.

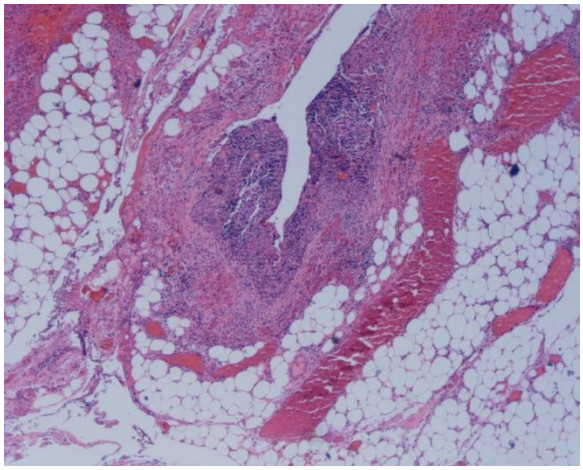
The surgical management remains controversial. Hemicolectomy is indicated in all cases where carcinoma is suspected. Chui *et al*, reported on using cecoscopy to rule out malignancy when the diagnosis is uncertain[5]. Since its inception in 2002, no other reports have been found to confirm their findings. In young, seemingly healthy patients, diverticulectomy with appendectomy is standard of care. Depending upon the level of training of the surgeon, a laparoscopic diverticulectomy has also been shown to be feasible and safe by several small studies and case reports.[1,18] Others have proposed that in all cases of suspected appendicitis, laparoscopic treatment should be employed. Per a Cochrane review by Sauerland *et al*, women benefit most from laparoscopic therapy as their anatomy has offers a wider range of diagnostic possibilities.[16]

In the situation of an inflamed solitary diverticulum, several studies have reported equivocal efficacy of antibiotics compared to surgical resection. The literature is ripe with reports with treatments ranging from sole antibiotic therapy for uncomplicated cases to cecectomy to right hemicolectomy.[8,14] Canver and Freier state that 14% of cecal diverticula will become acutely inflamed but they do not offer evidence on how they came to this conclusion.[2] This leads to the question: if an incidental solitary cecal diverticulum is found, should it be excised? The other issue is that the majority of reports are for the acutely inflamed diverticulum. Therapy largely depends upon the physical exam and imaging of the patient to determine the extent of the disease. Much of the evidence for the treatment of cecal diverticula has been inferred from the treatment of ascending colonic diverticulitis. Due to the rarity of the disease and presentation similar to appendicitis, there are no randomized prospective trials to determine optimal outcomes. Dearden mentions in her paper that diverticulectomy may be sufficient or in certain cases drainage and antibiotic therapy may be necessary. If the perforation is too large, however, a Mickulicz procedure with excision of the diverticulum and secondary ileocolostomy may be necessary. Despite the five fold increase in morbidity in emergent situations, a hemicolectomy may also be necessary[7]. These conclusions are based upon work with diverticula and diverticulitis throughout the colon. Contrarily, Kachroo mentions that the solitary cecal diverticulum is unlikely to resolve with antibiotics and conservative management and typically ultimately requires resection, and a delay of surgical intervention may result in increased morbidity and mortality.[10]

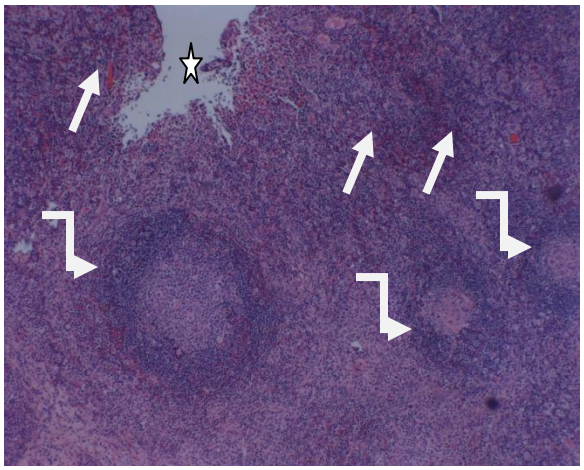
In conclusion, therapy depends upon proper diagnosis and the underlying circumstances of the patient, the availability of imaging to assess the ascending colon and correct determination of the disease etiology. If a normal appearing appendix is discovered during

the procedure, it is imperative to search for an alternative source of disease. Most choose to resect and primarily close the colonic defect, but based on the level of inflammation, a cecectomy or hemicolectomy is an alternative. Our experience has determined that these rare masses may become incarcerated in extremely rare cases like any other portion of bowel, and that resection is curative. Imaging is necessary to evaluate and plan the potential resection but most cases will not be diagnosed until the operation begins. Although this is an unusual case of pain, it should be suspected with a prolonged course with an insidious onset in a patient with symptoms similar to appendicitis.

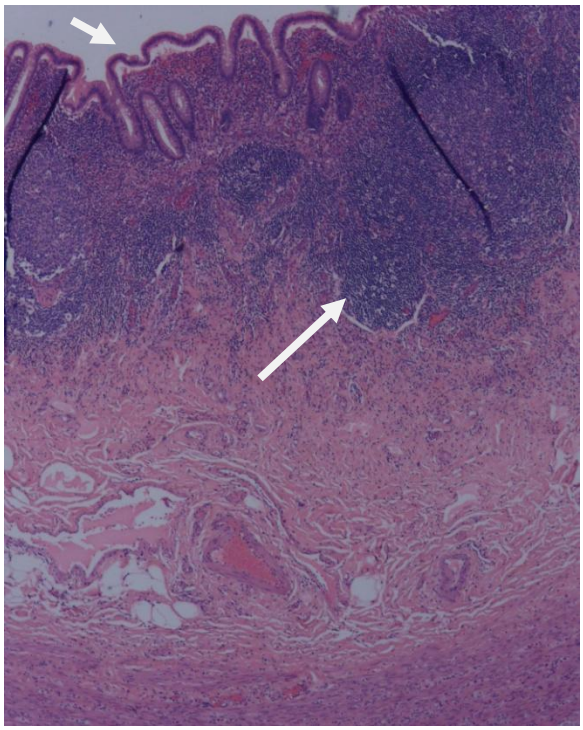




Images obtained from necrotic solitary cecal diverticulum (necrosis noted by arrow) that had herniated through mesentery, note the lack of mucosa, the significant amount of neutrophils and that all layers of the bowel wall are presented, except mucosa. Note in the top image the residual smooth muscle tissue with adipose tissue on left side and inflammatory on right, representative of a diverticulum. The image below represents the above image seen at lesser magnification power.



The above image represents colonic wall with acute (multiple arrows) and chronic (curved arrows) inflammatory changes and lymphoid follicles located within the wall. Mucosal layer is not identified. (Location where mucosa should be identified by star)



Above – veriformis appendix wall, normal in appearance with lymphoid follicles (long arrow) located in the submucosa (mucosal layer intact, short marrow)

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