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Appendiceal Intussusception: A rare case of tubulovillous variant

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Abstract

Intussusception of the bowel is telescoping of a proximal segment of the gastrointestinal tract within the lumen of the adjacent segment. Occurrence in adults is rare, accounting for only 5% of all cases of intussusceptions. Cases in which the appendix invaginates into the cecum are even rarer. A villous adenoma as a cause of intussusception is even more uncommon, in comparison to other benign appendiceal lesions.

We report a case of 56-year-old male with of 5-day history of progressive abdominal pain. CT of the abdomen showed a 15 cm colocolonic intussusception involving the ascending and transverse colon. Confirmatory colonoscopy revealed a tubular mass partially obstructing the cecum, secondary to the intussusception. Pathology report of the surgical specimen from a right hemicolectomy demonstrated that the appendix was completely intussuscepted within the cecum. It measured 7.5 cm in length by 2.5 cm in diameter, with a prominent folding containing a tubulovillous adenoma, without invasion or adjacent lymph node metastases. This rare case presentation of tubulovillous appendiceal intussusception is an important illustration of the diagnostic clinical work up, clinical reasoning and management of said pathology.

Keywords

intussusception; appendiceal intussusception; tubulovillous adenoma

Introduction

Intussusception of the bowel is telescoping of a proximal segment of the gastrointestinal tract within the lumen of the adjacent segment. Occurrence in adults is rare, accounting for only 5% of all cases of intussusceptions, with only 1%-5% presenting as bowel obstruction [1].

Appendiceal intussusception is extremely unusual and is only found in 0.01 per cent of patients who undergo appendectomy [2]. Despite the first case report of appendiceal inversion described by McKidd in 1858 [3], the first chronological scientific review evaluating the literature was not published until over a century later, when in 1963 Collins reviewing over 71,000 cases of appendectomy, reported the rare inci-

dence of appendiceal intussusception [4].

Several case reports exist in regard to appendiceal intussusception, but few specifically with the finding of tubulovillous adenoma [5,6]. A tubulovillous adenoma is a form of polyp that has malignant potential, and is considered to have a higher risk of malignant transformation than simple tubular adenomas [7]. Most cases of villous adenomas are in the rectum and sigmoid [8]. When located in the appendix, they usually present as simple appendicitis [6]; Stoppa et al (2009) reported this lesion in 0.06% of appendectomy specimens [9]. Cases of intussusception are extremely rare.

Case Report

This is the case of a 56-year-old male with notable past medical history of significant daily chronic alcohol abuse and tobacco use. He presented to the ED with a 5-day history of progressive abdominal pain, nausea, vomiting, and diarrhea in association with recent 10lb weight loss. He denied any prior abdominal pain episodes in the past and presented due to persistent, but not necessarily worsening pain. He did not have a PCP nor routine medical care. Of note, patient denied any previous history of EGD or colonoscopy.

At the time of evaluation, the patient stated that the pain was located in the mid abdomen, non-radiating, with no exacerbating or alleviating factors; he also mentioned it was associated with vomiting, non-bilious, non-bloody diarrhea, liquid, moderate amount, blackish-gray in color.

Physical exam showed generalized tenderness, non-focal in the abdomen with absence of guarding or any other peritoneal signs. An abdominal CT was performed in the ED, which showed a 15 cm colocolonic intussusception involving ascending and transverse colon with marked wall thickening.

The patient was admitted to the hospital and the next day underwent a barium enema with air contrast. No colonic mass or stricture was appreciated and there were no signs of extravasation of contrast. The ileocecal intussusception extending up to the mid transverse colon was subsequently reduced up to the cecum. Despite this, it was noted by the GI team, that during the procedure neither air nor contrast reflux into the small bowel could be achieved despite several maneuvers.

In spite of initial relief of abdominal pain with the barium enema, the pain recurred and did not subside even with pain management service involved. At this juncture, the only tumor marker elevated was CEA at 6.4 (normal 0-5) with CA-19-9 19.8 (normal less than 30), CA-125 2.5 (normal is less than 30).

A colonoscopy was then performed and a partially obstructing large mass (measuring 9 cm in length x 30mm diameter) was found in the cecum. No bleeding was present. This was biopsied with a hot forceps for histology. Pathologic diagnosis from the ascending colon biopsy was reported as a tubular adenoma. Based on this result, a plan for laparoscopic right colectomy with ileocolic anastomosis was scheduled for the next day. Once the resected right colon was delivered from the abdomen onto the bench table, we opened the cecum, where the appendix, which was noticeably big, with a remarkably nodular aspect, appeared to be invaginated (Figure 1). This was sent to pathology for further examination; the report showed a tubulovillous adenoma with high grade dysplasia, that failed to display any evidence of invasion; all margins of

resection were free of disease, as were all lymph nodes sampled.

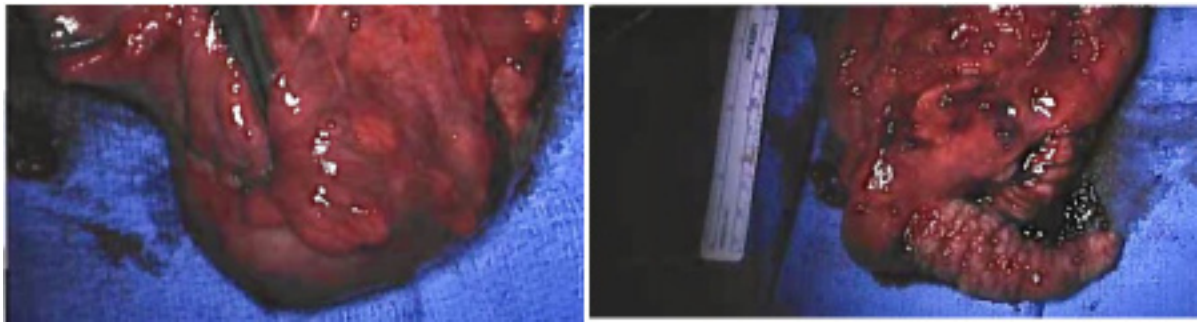

Figure 1: Resected right colon with invaginated appendix.

Discussion

Despite the rare entity of appendiceal intussusception first reported by McKidd et al., 1858 [3], it took Collins a full century later, when reviewing over 71,000 cases of appendectomy, to note that the greatest difficulty with any intussusception of the appendix, is diagnosing the condition preoperatively [4]. He states most diagnoses are made intraoperatively, with only 32% of cases determined prior to surgical intervention (Collins, 1963).

As in this case, suspicion of a possible mass lesion causing intussusception is founded upon with imaging modalities, such as barium enema and CT scan, usually followed up with colonoscopy. In the radiological community there has been a steady move toward more diagnostic clues when non operative methods are likely to fail. In a retrospective review of the last 12 years of irreducible intussusception; Marion et al (2006) reported a new radiographic sign that they coined appendix sign (radiographic visualization of the appendix without reflux of air or contrast into the small intestine), which was shown to have an association with failure of nonoperative management, and may be a further indication of necessity of surgical intervention [7]. During successful radiographic reduction, the small bowel is almost always visualized before the appendix; this was not the case in our patient, as air or contrast reflux into the small bowel could not be achieved despite several maneuvers.

In fact, the pitfall of colonoscopy is that a partially or completely invaginated appendix may be mistaken for a polyp during colonoscopy [8]. These authors recommend a progressive sequential work up starting with CT abdomen, followed by barium enema with sequential colonoscopy prior to any surgical intervention.

Despite concordance in the literature of the existence of intussusception within the appendix, there still remains questions to the adequacy or margins needed for resection. There are no clear guidelines for the management of this disease [10]. During the laparoscopic approach, a right hemicolectomy is usually warranted when there is concern for malignancy, but even less is known in the case of tubulovillous adenoma where all margins of resections are disease free, such as in this case. This raises the question, that perhaps a two stage approach should be considered for appendiceal intussusception to reduce the morbidity associated with a right hemicolectomy.

In such situations, a simple appendectomy could be performed while awaiting pathology profile of clear margins of resection. A standalone intussuscepted appendix can undergo unnecessary partial resection of the ileum, cecum or hemicolectomy for misdiagnosis or misinterpretation of malignancy [11,12]. Despite this, literature exists stating that strong malignancy suspicion based on the preoperative evaluation or during surgery merits consideration for a right hemicolectomy [13,14].

It is worth noting that appendiceal adenomas have a strong predisposition to the development of invasive adenocarcinomas, and as such, surgeons may be more reluctant to limit the margins of resection. One option is an intraoperative frozen section, which from our surgical experience is often times unavailable, untimely, and is less used clinical practice.

It is a general consensus that once the margin invasion is found, a right hemicolectomy is warranted [6]. There is no clear consensus in the literature dictating whether laparoscopic is of equal merit to an open approach. One advantage of laparoscopic intervention for appendix intussusception is the fact that it allows observation within the entire abdominal cavity and minimizes physical contact with possible malignancies [15,16] and potential spread.

Our patient underwent laparoscopic right hemicolectomy without any complications. Indeed, as the technique of laparoscopic appendectomy becomes more refined, so too in tandem should the development of clearer guidelines for managing appendectomy intussusception lesions laparoscopically with better clarification of margins needed.

Conclusion

In conclusion, there are two main take home messages from this case report. Firstly, diagnosis of appendiceal intussusception will likely need a multifactorial approach, including abdominal CT, barium enema and colonoscopy, as there is no standalone form of evaluation withstanding operative approach that can confirm the diagnosis. Secondly, it is clear that further research is warranted to confirm margins needed for a benign appendiceal intussusception, such as a tubulovillous adenoma in this case, which is extremely rare in the appendix and as a form of intussusception

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