Intussusception of vermiform appendix with microscopic melanosis coli: A case report

Mikroskopik melanozis koli bulunan appendiks vermiformisin intussussepsiyonu: Bir olgu sunumu

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Intussusception of the appendix is a rare occurrence. Due to the similarity of its symptoms with appendicitis, preoperative diagnosis of this condition is extremely difficult. In this report, we present appendiceal intussusception with histological melanosis coli that occurred in a patient on long-term anthranoid laxative use for chronic constipation. Melanosis coli in the appendiceal tissue, as an indicator of chronic laxative intake, may be a clue implying that the appendical exposure to hyperperistalsis for a long time in our case led to the intussusception. We conclude that colonoscopy may help in preoperative diagnosis of appendiceal intussusception in patients with suspicious appendicitis, particularly in those using laxative medication.

Key words: Appendix, intussusception, melanosis coli, anthraquinone, laxative

Appendiks intussussepsiyonu ender görülen bir durumdur. Apandisit semptomlarına benzerlik gösterdiği için, bu durumun preoperatif tanısı son derece zordur. Burada, kronik konstipasyon nedeniyle uzun süredir antrakinon türü laksatif kullanan bir hastada melanozis kolinin histolojik bulgularını gösteren bir appendiks intussussepsiyonu gelişimi sunulmaktadır. Hastamızda kronik laksatif alımının bir göstergesi olarak appendiks dokusunda melanozis kolinin varlığı, appendiksin intussussepsiyon geliştirmek üzere uzun bir zaman süreci boyunca hiperperistaltizme maruz kaldığına ilişkin bir ipucu olabilir. Özellikle uzun süreli laksatif kullanan ve şüpheli apandisti bulguları olan bir hastada kolonoskopi yapılması bu ender patolojinin preoperatif tanısının konulmasına yardımcı olabilir.

Anahtar kelimeler: Appendiks, intussussepsiyon, melanozis koli, antrakinon, laksatif

INTRODUCTION

Intussusception of the appendix into the cecum is an uncommon condition which is rarely diagnosed preoperatively. Specific anatomic and/or pathological conditions that cause irritation or increased peristalsis should be present for this event to occur. Because of the rarity of the disease and the similarity of its presentation with that of appendicitis, preoperative diagnosis is extremely difficult (1-3). Here, we report an unusual case of appendiceal invagination with histological characteristics of melanosis coli, both of which were thought to originate from chronic anthranoid laxative use for chronic constipation.

CASE REPORT

A 48-year-old woman underwent evaluation for a two-month history of intermittent abdominal pain. She denied weight loss, hematochezia, fever, or change in urinary function. She had been using an anthranoid laxative for about four years because of chronic constipation. Physical examination revealed abdominal tenderness at right lower quadrant without rebound tenderness. Laboratory studies, including a complete blood count, urinalysis and serum chemistry, were unremarkable. Ultrasound showed a slightly tender, non-compressible hypoechoic mass 2 x 3.5 cm in diameter in the right lower abdomen, surrounded by a hollow

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234 AKBAYIR et al.

bowel-like structure. Colonoscopy disclosed a polypoid lesion protruding from the appendiceal orifice with absence of macroscopic discoloration in appendiceal and colonic mucosa (Figure 1A-B). A close-up endoscopic observation revealed that this lesion with smooth surface and waxy consistency was an invaginated appendix circumscribed by mucosal folds, protruding into the cecal lumen like a cone shape. Overlying mucosa was moderately edematous but smooth. When pushed by a biopsy forceps from tip of appendix into its base, invagination could only be partially reduced. Histopathological evaluation of biopsies taken from the

A

Figure 1. A-B) Endoscopic appearance of intussuscepted appendix protruding into the cecal lumen

appendix showed the presence of brown pigment deposits in histiocytes in the lamina propria, which is consistent with microscopic melanosis coli (Figure 2).

The patient, who had no complaint except for episodes of right lower abdominal colicky pain, did not accept surgical management although it was explained to her in detail that the appendix must be removed.

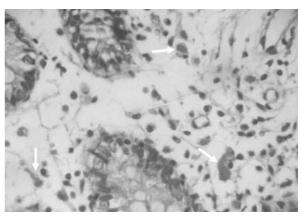


Figure 2. Histiocytes containing brown pigment deposits in the lamina propria of appendiceal mucosa (*arrows*) (Hematoxylin and eosin, original magnification x 200)

DISCUSSION

Review of the literature concerning appendiceal invagination revealed that the symptoms of this lesion include right lower quadrant pain, and occasionally nausea and vomiting, and that it was encountered in 0.01% of surgically removed appendices (1). Pathological factors such as fecalith, parasites, endometriosis, carcinoid, carcinoma, adenoma, mucocele and follicular lymphoid hyperplasia are accountable for intussusception of the appendix (4, 5).

Because of similar symptoms, most patients are misdiagnosed as having appendicitis and undergo surgical exploration without prior endoscopic evaluation. Therefore, diagnosis of this rare lesion by endoscopic appearance is another distinctive aspect in the presented case. The other aspect to emphasize is co-existence of melanosis coli and inside-out appendix in our patient. Melanosis coli is a brownish discoloration of the colonic mucosa caused by the accumulation of pigment in macrophages of the lamina propria. The association between melanosis coli and chronic use of anthraguino-

ne laxatives is well described and is further supported by the development of characteristic pigmentation in laboratory animals after administration of anthraquinones (6). The condition is widely regarded as benign and reversible, and disappearance of the pigment generally occurs within one year of stopping laxatives (7). On the other hand, the appendix is reported to be a common location for melanosis coli (8). Melanosis coli in the appendical tissue, as an indicator of chronic anthranoid laxative use, may be a clue implying that appendical exposure to hyperperistalsis for a long time in our patient led to the intussusception.

To our knowledge, there is no data concerning the association of melanosis coli and appendiceal invagination in the literature. Beyond a simple association, there may be a cause and effect relationship between these two conditions. Even though other pathological factors could not be excluded due to unavailability of surgical specimen, it may be

speculated that increased colonic motility related to chronic laxative use could have led to or contributed to the formation of inside-out appendix in our case. Apart from pathological factors mentioned above, anatomical factors such as fetal type cecum, mobile appendix, wide appendicular lumen and a directed hyperperistalsis against intraluminal obstruction are thought to be pathogenetic mechanisms. The endoscopic appearance of the invaginated appendix with regular and smooth mucosal surface in our patient may suggest that increased peristaltic activity may itself be the only responsible factor for invagination rather than appendicular luminal obstruction.

We conclude that right lower abdominal pain and/or mass in patients who are known to be chronic laxative users may indicate appendiceal invagination. If the diagnosis of acute appendicitis is equivocal, colonoscopy can be of benefit in the differential diagnosis.

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