

A puzzling case presenting as a polypoid mass in the cecum in an adult patient: appendiceal intussusception

Erişkin bir hastada çekumda polipoid kitle olarak prezente olan şaşırtıcı bir olgu: apendiks intussusepsiyonu

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Abstract

Appendiceal intussusception is a rare condition in an adult patient and may not be initially considered in the differential diagnosis clinically or radiologically. It occurs due to an organic cause or idiopathically. We report a 74-year-old patient who had a polypoid lesion in the cecum on colonoscopy with no radiologic imaging and was ultimately diagnosed grossly and histopathologically as appendiceal intussusception. In conclusion, physicians are advised to consider appendiceal intussusception as an alternative diagnosis in all patients presenting with a suspected cecal mass.

Key words: Appendix vermiformis, intussusception, cecum, inverted appendix.

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Öz

Apendiks intussusepsiyonu yetişkin bir hastada nadir görülen bir durumdur ve başlangıçta klinik veya radyolojik olarak ayırıcı tanıda düşünülmemeyebilir. Organik bir nedene bağlı olarak veya idiyoatik olarak ortaya çıkar. Bu yazıda, kolonoskopide çekumda polipoid lezyon saptanan ve radyolojik görüntüleme yapılmayan, sonuçta gross ve histopatolojik olarak apendiks intussusepsiyonu tanısı konulan 74 yaşında bir hasta sunulmuştur. Sonuç olarak, hekimlere çekumda kitle şüphesi ile başvuran tüm hastalarda apendiks intussusepsiyonunu alternatif bir tanı olarak düşünmeleri önerilmektedir.

Anahtar kelimeler: Apendiks vermiformis, çekum, intussusepsiyon, apendiksin inversiyonu.

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Introduction

Appendiceal intussusception (AI) is a rare entity that can be difficult to diagnose preoperatively both clinically and radiologically. This entity is defined by the term 'intussusception' and is also referred to as 'appendiceal inversion' in the literature [1]. AI is more common in adults than in children. However, only 5% of all cases of intestinal intussusception occur in adults, the rest are diagnosed in children. Almost all pediatric cases are idiopathic, whereas adult cases usually present with an organic cause [2].

Here, we report a 74-year-old patient who was initially suspected to have a polypoid mass

in the cecum on colonoscopic examination and subsequently diagnosed with AI with clinicopathologic correlation during gross and histopathologic evaluation after wedge resection.

Case

A 74-year-old male patient was admitted to an external hospital with abdominal pain and underwent colonoscopy. Colonoscopy revealed a 10 mm diameter polypoid lesion at the appendix orifice at the base of the cecum, which was evaluated in favor of submucosal lipoma. As the patient also complained of inability to urinate, transurethral resection of the prostate

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was performed and the pathology diagnosis was nodular adenomyomatous hyperplasia. The patient was referred to our institute for the lesion described in colonoscopy. Routine blood tests including complete blood count, liver enzymes, electrolytes and CRP were within

normal limits. Re-colonoscopy was performed in our gastroenterology clinic and a 25-30 mm mass lesion with an orifice-like appearance at the base of the cecum, covered with normal mucosa was described (Figure 1, 2).

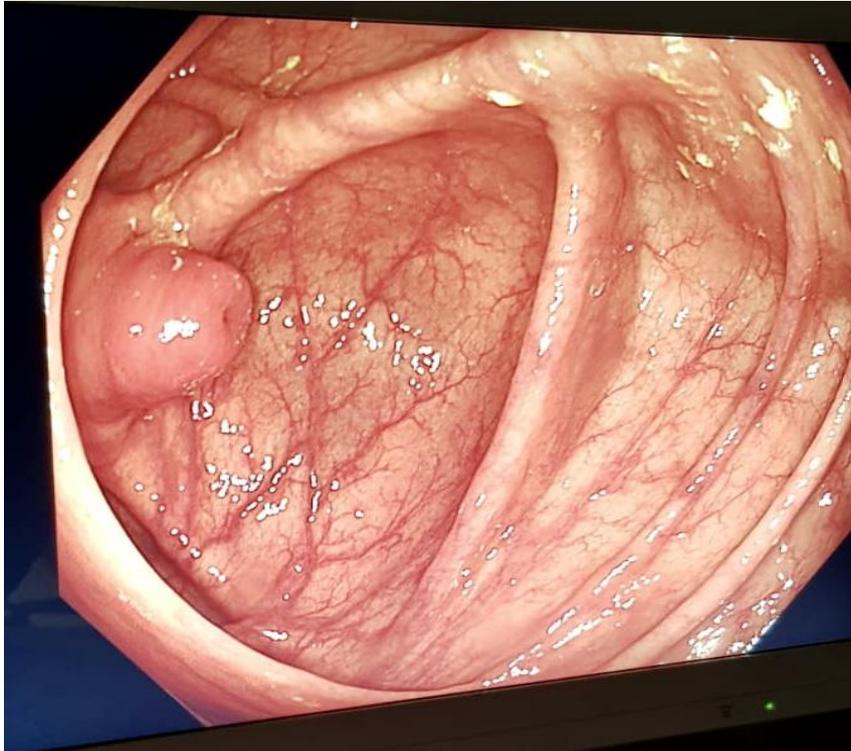


Figure 1. Colonoscopic view of the polypoid lesion located in the cecum with orifice-like appearance in the center

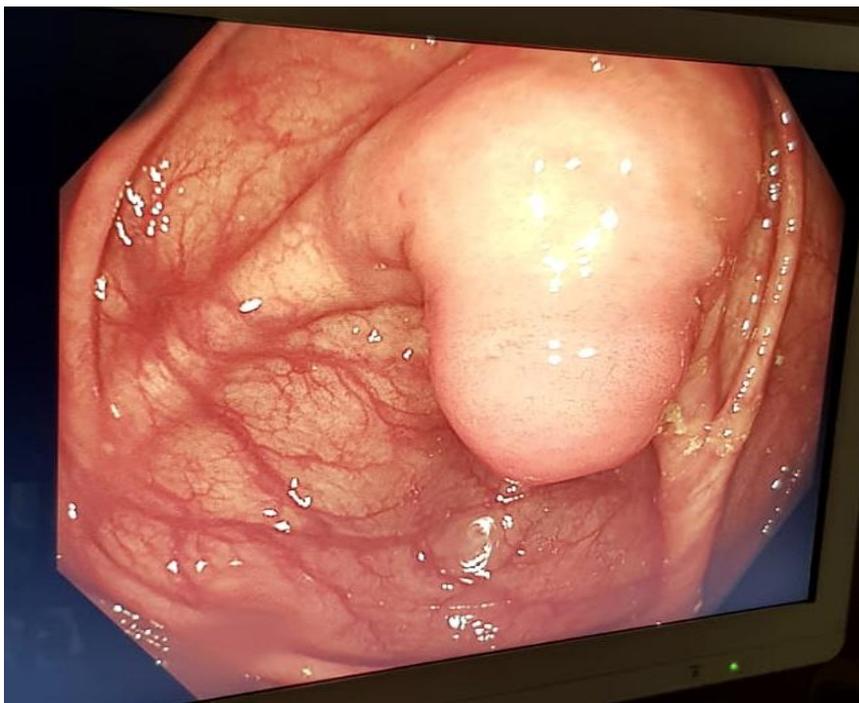


Figure 2. Colonoscopic view of the polypoid lesion from a different angle

A wedge resection including the appendix and partial cecum was performed and the specimen was photographed post-operatively (Figure 3). Gross examination gave the impression of intussusception of the appendix into the cecum, but the possibility of a tumoral mass could not be excluded. The entire specimen was sampled for histopathologic examination. Microscopically,

a calcified nodule in the subserosal adipose tissue was noted (Figure 4) and a dome-like appearance covered with mucosa consistent with appendix protruding into the cecum was observed (Figure 5, 6). In the light of these findings and clinicopathologic correlation, the case was diagnosed as AI into the cecum. No post-operative complications were noted and the patient was recommended for annual colonoscopic follow-up.



Figure 3. Post-operative photograph of cecal resection shows a polypoid lesion with an orifice-like area in the center, covered with shiny mucosa

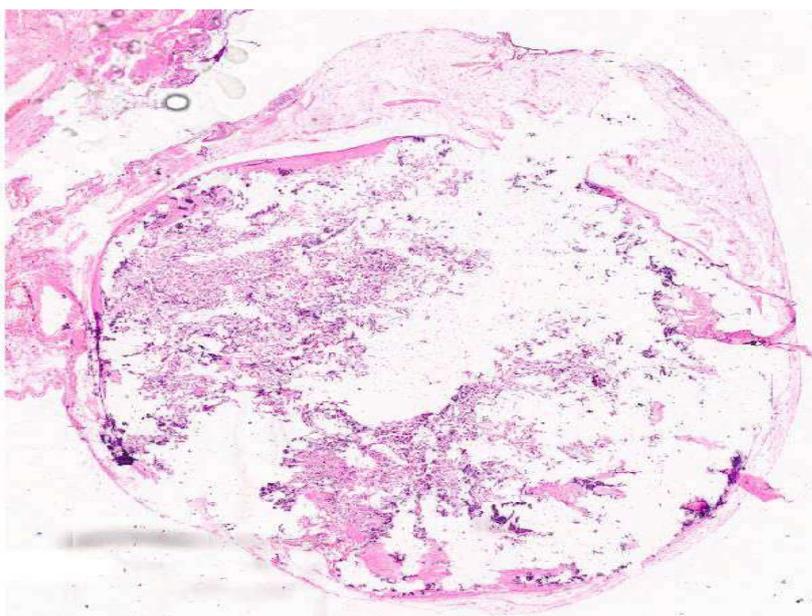


Figure 4. Histopathologic image of hyalinized nodule with foci of dystrophic calcification in subserosal adipose tissue, Hematoxylin&Eosin stain, x5 magnification



Figure 5. Dome-like appearance covered with mucosa, consistent with the diagnosis of appendiceal intussusception Hematoxylin&Eosin stain, x3 magnification



Figure 6. Closer observation of appendiceal mucosa with lymphoid aggregates Hematoxylin&Eosin stain, x8 magnification

Discussion

The pathogenesis of AI is not fully understood, but parasites, endometriosis, fecalitis, polyps, lymphoid follicular hyperplasia, neuroendocrine tumors, angiodysplasia, adenocarcinoma, mucinous neoplasia, abnormal mobility of the cecum or appendix, mobile mesoappendix and large appendiceal lumen seem to be the main causes of the disease [1, 3]. While endometriosis is the most common cause in adults, inflammation is the leading cause in children [4]. Cases of AI occurring without any of the above-mentioned causes have also been reported in the literature [3]. In our case, the calcified nodule in the subserosal adipose tissue may have caused motility disorder and thus intussusception.

AI may be categorized in 5 different anatomical groups according to where the intussusception starts, from the tip of the appendix to complete invagination of the appendix into the cecum, respectively (Type 1-5) [5]. Our case may be categorized as 'Type 3' because the intussusception starts at the junction of the appendix and the cecum.

Preoperative diagnosis of AI is difficult due to non-specific clinical features. Clinical symptoms of AI are classified in 4 different groups. In the first group, acute appendicitis-like features are seen. Patients in the second group have intermittent pain, vomiting, diarrhea and rectal bleeding. The third group has recurrent abdominal pain, vomiting and rectal bleeding (due to recurrent intussusception and self-reduction). In the fourth group, patients are asymptomatic [6]. Our patient was incidentally diagnosed with a cecal mass during colonoscopy for abdominal pain and belongs to the second group mentioned above in terms of clinical presentation.

Diagnosis of AI can be made by methods such as colonoscopy, barium radiography, ultrasonography and tomography. On computed tomography, target-like appearance or concentric ring sign on axial sections are findings that support the diagnosis. Failure to observe the appendix with a filling defect in the cecum on double contrast barium radiography may suggest the diagnosis of AI. Our patient did not have any radiologic imaging.

AI may appear as a polypoid lesion in the cecum on colonoscopy. A case of AI followed up

for more than one decade with a diagnosis of cecal polyp has been reported [3]. In our case, colonoscopy was performed and the lesion gave the impression of a polypoid mass.

On microscopic examination, the dome-like appearance covered with mucosa, associated lymphoid aggregates and ganglion cells in the muscular layer may suggest the diagnosis of AI [1, 7]. In our case, dome-like pattern covered with mucosa, lymphoid aggregates and muscularis propria compatible with appendix were also noted in accordance with the literature.

Treatment of AI may be achieved by reduction of the appendix. However, considering the recurrence rates after reduction, appendectomy is recommended. According to Chaar et al. [5], partial cecal resection, which will be applied with appendectomy instead of simple appendectomy, may be a more ideal treatment. In the same study, Chaar et al. [5] reviewed the literature data on AI and found that appendectomy was performed in 42% of adults and 71% of children. They also noted that ileocecectomy was performed in 27%, right hemicolectomy in 21% and subtotal colectomy in 1% of adults [5]. With the early recognition of intussusception, a large resection of the colon can be prevented. In particular, patients with risk factors such as endometriosis, mucocoele, and polyposis should be evaluated in terms of intussusception, and the treatment should be decided according to the patient, etiology and intraoperative findings [4]. Patient age may also influence the choice of treatment. For example, in pediatric patients, a minimally invasive method such as reduction is more favored as the likelihood of neoplasia is very low, whereas in adults, surgical options are more often preferred [8]. In our case, appendectomy and partial cecal resection were performed due to the possibility of a polypoid mass, and thus the incidentally diagnosed subserosal calcified nodule was also removed.

In conclusion, AI is a rare clinical condition that may be encountered especially by general surgeons, pediatric surgeons and gastroenterologists, but its preoperative diagnosis is difficult and its treatment is likely to be performed with colon resections with suspicion of malignancy. We think it is important to be aware of this entity and to consider it in the differential diagnosis.

Conflict of interest: No conflict of interest was declared by the authors.

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Informed consent: Information about the diagnosis and the procedure to be performed was provided to the patient or relatives of the patients, and a confirmation document was signed.

Authors' contributions to the article

N.C.D. and E.C., have constructed the main idea and hypothesis of the study. N.C.D., E.C., H.K., M.O. and U.O. developed the theory and arranged/edited the material and method section. E.C. and H.K., have done the evaluation of the data in the results section. Discussion section of the article written by N.C.D., E.C., H.K., M.O. and U.O. reviewed, corrected and approved. In addition, all authors discussed the entire study and approved the final version.