

Appendiceal intussusception induced by primary serrated adenocarcinoma: a case report

Intussuscepção do apêndice vermiforme ocasionado por adenocarcinoma serrilhado primário: relato de caso e revisão de literatura

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ABSTRACT

Intussusception is defined as the intussusception of an intestinal segment, either the small or large intestine, into the lumen of the adjacent segment. Any segment of the gastrointestinal tract may be involved in this process; however, the most common segments to be affected are the jejunum and ileum, in addition to the colonic segments. In this case report, we report and discuss a case of intussusception of the cecal appendix secondary to serrated adenocarcinoma, which occurred in a male patient at a hospital in the city of Juiz de Fora, Minas Gerais - Brazil. Once the diagnosis was made, right hemicolectomy with primary anastomosis was performed. Histopathological analysis revealed low-grade serrated adenocarcinoma — moderately differentiated (pT 3, pN 0). Primary neoplasms of the vermiform appendix are a rare condition, possibly associated with appendiceal intussusception, an event also considered rare in the literature, which requires a careful diagnostic approach. Treatment is surgical and will depend on the classification of the intussusception, but in suspected cases of associated malignancy, hemicolectomy with lymphadenectomy is well indicated.

Keywords: Intussusception; Appendix; Adenocarcinoma.

RESUMO

A intussuscepção é definida como a intussuscepção de um segmento intestinal, seja o intestino delgado ou grosso, no lúmen do segmento adjacente. Qualquer segmento do trato gastrointestinal pode estar envolvido nesse processo; no entanto, os segmentos mais comuns a serem afetados são o jejuno e o íleo, além dos segmentos colônicos. Neste relato de caso, relatamos e discutimos um caso de intussuscepção do apêndice cecal secundária a adenocarcinoma serrilhado, ocorrido em um paciente do sexo masculino em um hospital de ensino da cidade de Juiz de Fora, Minas Gerais, Brasil. Uma vez feito o diagnóstico, foi realizada hemicolectomia direita com anastomose primária. A análise histopatológica revelou adenocarcinoma serrilhado de baixo grau —

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moderadamente diferenciado (pT 3, pN 0). As neoplasias primárias do apêndice vermiforme são uma condição rara. Essa condição pode estar associada à intussuscepção apendicular, evento também considerado raro na literatura, que requer uma abordagem diagnóstica cuidadosa. O tratamento é cirúrgico e dependerá da classificação da intussuscepção, mas em casos suspeitos de malignidade associada, a hemicolectomia com linfadenectomia é bem indicada.

Palavras-chave: Intussuscepção; Apêndice cecal; Adenocarcinoma.

INTRODUCTION

Intussusception is defined as the invagination of an intestinal segment (intussusceptum), either the small or large intestine, into the lumen of the adjacent segment (intussusciens), like a “telescope”¹.

Any segment of the gastrointestinal tract (GIT) can be involved in this process. However, the most common segments to be affected are the loops of the jejunum and ileum, in addition to colonic segments².

Intussusception of the cecal appendix is considered a rare event, with approximately 0.01% of cases reported in the medical literature³. In 1858, the first historical case was reported by M’Kidd (1858)⁴: a 7-year-old boy who was affected by intense and intermittent attacks of abdominal pain, with no change in bowel habits - no signs of blood or mucus in the stool, and no evidence of palpable masses on abdominal physical examination. Pain crises were managed with morphine and repeated inhalations of chloroform. The child died of exhaustion. During the autopsy, complete invagination of the vermiform appendix into the cecum was evidenced⁵.

In this case report, we report and discuss another case of appendiceal intussusception secondary to serrated adenocarcinoma, which occurred in a hospital in the city of Juiz de Fora, Minas Gerais, Brazil.

CASE REPORT

Patient FMC, male, 49 years old, without comorbidities, with a report of diffuse abdominal pain evolving for three months, admitted to our service with severe abdominal pain and melena for one day, with no other complaints. The patient was hemodynamically stable, afebrile. On physical examination, there was no pain on palpation and no peritoneal irritation. Laboratory investigation showed a leukocyte count of 6.67×10^3 and hemoglobin of 11.8 mg/dl; positive fecal occult blood test. Computed tomography (CT) of the abdomen was performed, which showed signs of ileocecolic intestinal intussusception with thickening of the wall of the intussusception segment (Figure 1).

The patient underwent exploratory laparotomy, which revealed invagination of the entire vermiform appendix into

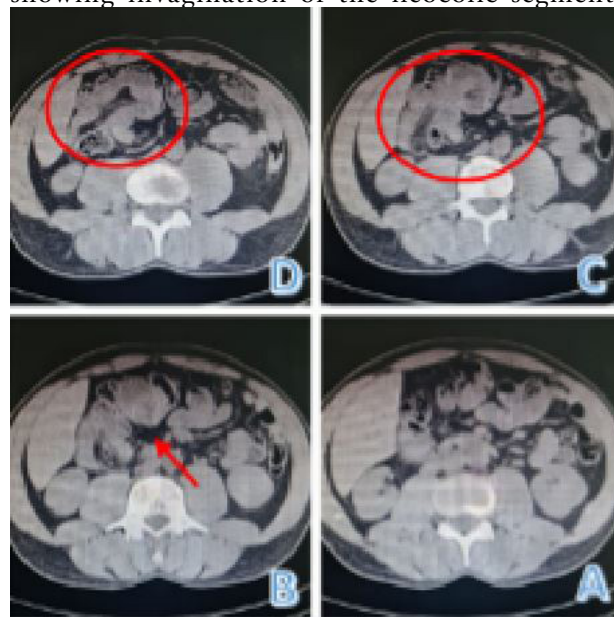
the cecum (Figure 2), with no other intraoperative findings. Right hemicolectomy was performed (Figure 3) with primary end-to-end ileocolonic anastomosis. The surgical specimen was sent for anatomopathological analysis.

The open surgical specimen revealed an ulcerative-vegetative, verrucous, granular lesion in the appendiceal topography.

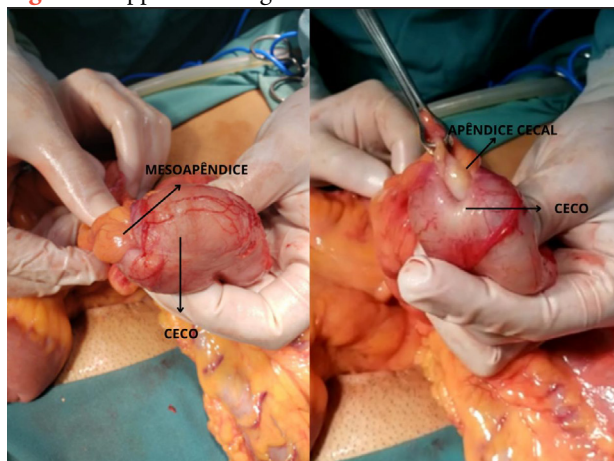
The histopathological examination of the specimen revealed low-grade serrated adenocarcinoma — moderately differentiated, measuring 4.3×3.5 cm. Invading up to the subserosa (focally), there was an absence of angiolymphatic and perineural invasion, and an absence of metastasis in 13 regional lymph nodes studied. Pathological staging was pT 3, pN 0.

During the immediate postoperative period, the patient presented adynamic ileus, without major complications. The recovery in the immediate postoperative period was rapid and progressed well, and the patient was discharged from hospital on the fourth postoperative day.

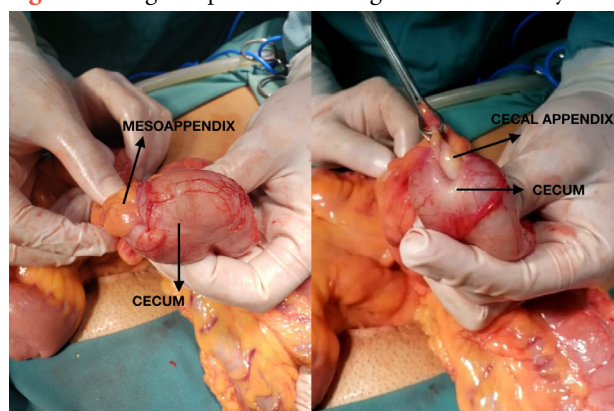
Figure 1. Abdominal computed tomography images showing invagination of the ileocolic segment.



Source: Personal archive.

Figure 2. Appendix invaginated into cecum.

Source: Personal archive.

Figure 3. Surgical specimen after right hemicolectomy.

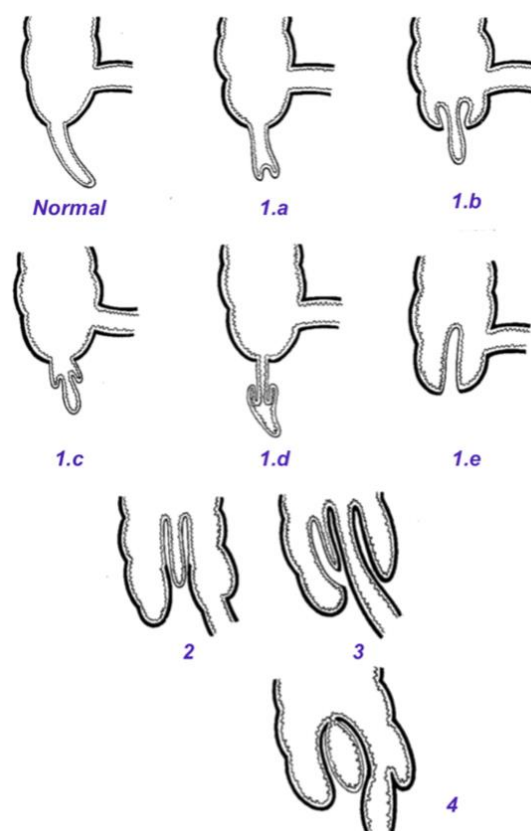
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DISCUSSION

Invagination of the vermiform appendix is a very rare event. Because it is a difficult condition to diagnose, many cases are identified intraoperatively — as is the case of our patient — or even postoperatively^{4,6}. Some factors may be involved in the pathophysiology of this condition. Anatomical factors, such as a wide appendiceal lumen, especially the proximal lumen; a sparse mesoappendix, free of fat and with a narrower base; a healthy, mobile appendiceal wall capable of active peristalsis; a free appendix, not fixed by congenital or inflammatory adhesions. Factors that alter appendiceal motility and physiological peristalsis, such as the presence of fecaliths, worms and foreign bodies in the appendiceal lumen; conditions that affect the appendiceal wall, such as mesenteric adenitis, papilloma, polyps and endometrioma⁵.

Clinical signs and symptoms in adults are usually chronic, with most patients presenting nonspecific symptoms suggestive of intestinal obstruction. Abdominal pain is the most common, followed by nausea and vomiting. Palpable masses are rarely identified^{7,8}.

According to the Forshall (1953)⁵ classification (Figure 4), we have 1.a - Invagination of the tip of the appendix

Figure 4. Classification of appendiceal intussusception.

Source: Forshall (1953)⁵.

(the intussusceptum) into the proximal appendix (the intussusciens); 1.b - Invagination starting at the junction of the appendix with the cecum, the appendix being the intussusceptum and the cecum the intussusciens; 1.c - Invagination starting along the length of the appendix; 1.d - Retrograde intussusception from the proximal appendix to the distal appendix; 1.e - Complete invagination of the appendix into the cecum (our case); 2 - Compound intussusception (compound or secondary intussusception of the cecocolic type induced by an invaginated appendix as the apex with appendiceal intussusception); 3 - Any type of appendiceal intussusception complicated by ileocolic intussusception; 4 - Invagination of appendiceal mucocele into the cecum⁵.

The treatment of this condition is basically surgical. The surgical procedure is based on this classification, in which types 1.a, 1.c and 1.d, appendectomy is sufficient since the lesion does not include the base of the appendix. On the other hand, in types 1.b, 1.e and 2, the ligated base of the appendix can lead to continued intussusception after isolated appendectomy, so partial colectomy (cecum) should be considered in principle. However, treatment may vary based on histopathological results. In type 3, it is necessary to identify the origin of the ileocecal intussusception and remove the trigger point. In type 4, if during surgery and/or preoperative evaluation there is a high suspicion of malignancy, oncological right hemicolectomy should be

Source: Forshall (1953)⁵.

considered. On the other hand, if the malignant potential appears to be low or doubtful, it can be treated similarly to types 1.b, 1.e, 2 and 3^{9,10}.

CONCLUSION

While colorectal neoplasms have a high incidence, primary vermiform appendix neoplasms are a rare condition. This condition may be associated with appendiceal intussusception, an event also considered rare in the literature, which requires a careful diagnostic approach. Treatment is surgical and will depend on the classification of the intussusception, but in suspected cases of associated malignancy, hemicolectomy with lymphadenectomy is well indicated.

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AUTHORS' CONTRIBUTIONS:

We describe contributions to the papers using the taxonomy (CRediT) provided above:

Conceptualization, Investigation, Methodology, Visualization & Writing – review & editing: MB Weiss; MFV Martins. *Project administration, Supervision & Writing – original draft:* MB Weiss; GA Rosa; MVF Martins. *Validation & Software:* GA Rosa. *Resources & Funding acquisition:* MVF Martins. *Data curation & Formal Analysis:* MVF Martins.

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