

Appendicitis in double cecal appendix: Case report

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Author contributions: Alves JR conducted the patient care in the emergency, surgery and photography service during the intraoperative period (Figure 1) and the postoperative medical care, was in charge of general supervision of students, writing, translation, final review and article submission; Maranhão IGO, Oliveira PVV performed the literature review on the anatomical variations of the cecal appendix, and are co-authors of the manuscript. All the authors read and approved the final manuscript.

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anatomopathological examination of the surgical samples showed acute inflammation in the two cecal appendices. So, performing a routine retroperitoneal release and a complete cecum evaluation during such surgical procedures is recommended and suggested due to the possibility of not identifying a second cecal appendix.

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Key words: Appendix; Anatomic variation; Appendicitis; Appendectomy; General surgery

Core tip: Double cecal appendix is a rare (about 100 cases reported worldwide) anatomical variation often incidentally diagnosed in the face of inflammation in the organ. The current paper presents the first case reported in South America. The case is extremely important for the study of this possible anatomical variation since the lack of a diagnosis in a second cecal appendix can cause further complications for the patient and the physician. Moreover, it is associated with the presence of other anatomical variations, such as intestinal, genitourinary and bone. Such variations will be investigated in cases of the aforementioned diagnosis.

Abstract

Double cecal appendix is a rare anatomical variation. Approximately 100 cases have been reported worldwide. It is usually diagnosed incidentally during emergency appendectomies due to inflammatory processes in the cecal appendix. Case presentation: male, white, 36 years old, obese, presenting with pain in the lower abdomen for 24 h followed by nausea, vomiting and mild fever. He was subjected to additional tests, with the leukogram showing leukocytosis and abdominal ultrasonography depicting cecal appendix with thickened wall, locally associated with small quantities of liquid and intestinal loop obstruction. He underwent laparotomy, revealing acute appendicitis. Another intestinal loop obstruction was identified next to the ileum, leading to recognizing another cecal appendix after local dissection. Double appendectomy and segmental ileectomy were performed although not needed. Results of the

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INTRODUCTION

Double cecal appendix is a rare anatomical variation, found in 0.004%^[1] to 0.009%^[2] of performed appendectomies. Approximately 100 cases of double cecal appendix^[3-5] have been described worldwide so far, with no case reports in South America^[2,3,6-37].

CASE REPORT

A male, white, 36 years old, slightly obese [body mass



Figure 1 A photograph taken during a laparotomy procedure depicting an inflamed double cecal appendix. Minor (black arrow) and major (green arrow) inflamed cecal appendix. Surgeon's hand is on the left side of the picture, holding the proximal segment of the ileum (arrow with white edges).

index (BMI) = 31.1 kg/m²], presented with abdominal pain in the lower abdomen for 24 h, followed by nausea, vomiting and mild fever (axillary temperature = 37.9 °C). He was subjected to blood tests that only showed leukocytosis without left shift. In addition, abdominal ultrasonography depicted cecal appendix with thickened wall, locally associated with small quantities of intra-abdominal fluid and local obstruction of intestinal loops.

He underwent laparotomy with a McBurney's incision. The presence of an inflamed cecal appendix in its usual position after lysis of adhesions and cecum release was identified. Another intestinal loop obstruction was identified near the ileum. After the release of dense adhesions, it was possible to recognize the presence of a second cecal appendix, also with an inflammatory aspect (Figure 1), with its origin along the taenia coli.

A double appendectomy and segmental ileectomy in the part of the devascularized intestinal loop, resulting from ileum dissection, was performed in order to provide the release and excision of the second cecal appendix. Both appendices showed no sign of perforation despite the inflammatory aspect, *i.e.*, the occurrence of increased dimensions, thickened and erythematous wall, associated with fibrin and local tissue fragility.

The anatomopathological examination of the surgical samples corroborated the diagnosis of inflammation in both cecal appendices and resected segment of small intestine (ileum), with subserosal congestion and acute fibrinous serositis with eosinophils.

The patient had no postoperative complications and was discharged on the third day after surgery.

DISCUSSION

Since 1892 after the first case of double cecal appendix^[27] was reported, less than 100 cases have been reported worldwide^[3]. It demonstrates the rarity of such variations and why the current reported case is the first one to be described in South America^[2,3,6-37].

Over time, some authors have presented classifica-

tions to categorize anatomical variations of cecal appendix. The first classification was developed in 1936 by Cave^[28]. His classification was modified in 1962 by Wallbridge^[29]. Since then, a number of authors have made some changes to it, leading to the modified classification by Cave-Walbridge, which is now the most widely used^[17,30].

The classification modified by Cave-Walbridge categorizes double cecal appendix into three types: A, B and C. Type A is characterized by the presence of two cecal appendices with a common origin in a single cecum. In type B, two appendices emerge from different cecal origins from a single cecum. This type is also subdivided into B1 and B2. In subtype B1, the two appendices emerge from a single cecum, one from each side of the ileocecal valve, symmetrically. On the other hand, in subtype B2, one of the appendices is in its usual position and the second one is located alongside the taenia coli. Finally, type C is characterized by the existence of two caeca, each with a cecal appendix (Figure 2).

The present reported case describes the occurrence of a patient with double cecal appendix type B. There are reports of other rarer forms presenting with anatomic variations of the cecal appendix, such as the horseshoe appendix^[31] and the triple appendix^[32].

The existence of an cecal appendix duplication is asymptomatic and its diagnosis only comes during investigations on inflammation processes^[3,17,33,34]. This is what happened in our patient's case. According to clinical data, he had no complaints related to his cecal appendix duplication until the occurrence of acute appendicitis.

Despite the rarity of anatomical variations in the cecal appendix, the awareness of them is of great importance to surgeons. An inadequate surgical evaluation of the cecum due to unawareness of such variations can leave a second or third cecal appendix^[17,30] unidentified. This may lead to further reoperations, diagnostic difficulties and medicolegal problems regarding malpractice because of the possibility of new inflammation in the remaining appendices^[17,30].

For instance, this happened in a child whose cecal appendix duplication was not identified in the first appendectomy. Five months later, another laparotomy was needed in order to remove a second appendix which had also become inflamed^[35]. Such a situation is most commonly found in patients with double cecal appendix type B^[30]. It is worth mentioning that there is an increase in the postoperative morbidity and mortality^[17,30] in patients in whom anatomical variations of the cecal appendix are not identified.

Finally, the importance of being aware of the association between double or triple cecal appendix and other anatomical variations, intestinal, genitourinary and osseous, should be highlighted^[36,37]. These are most often associated with duplications of the cecal appendix types B1 and C^[3]. Thus, when two or three cecal appendices are identified, investigating these other anatomical variations is recommended^[3].

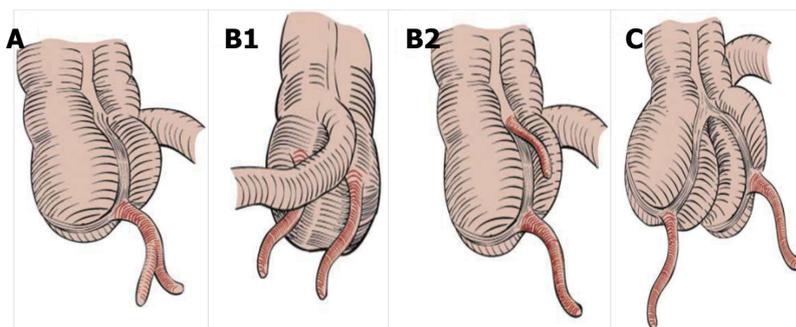


Figure 2 Classification modified by Cave-Wallbridge^[30], including type A, subtype B1, subtype B2 and type C.

As a final conclusion, although double or triple cecal appendices are rare, surgeons must be aware of them and identify cecal appendix anatomical variations. Such a procedure is recommended when doctors surgically approach a patient with acute appendicitis. They should perform a complete cecum evaluation after the retroperitoneal release in order to avoid further complications. Surgeons should remember that in the face of such changes, they will need to investigate the presence of intestinal, genitourinary or bone anatomic variations.

ACKNOWLEDGEMENTS

We thank the patient for allowing the disclosure of his medical report and intraoperative photographic records.

COMMENTS

Case characteristics

Male, white, 36 years old, slightly obese, presenting with acute appendicitis.

Clinical diagnosis

Abdominal pain in the lower abdomen for 24 h, followed by nausea, vomiting and mild fever (axillary temperature = 37.9 °C).

Differential diagnosis

Causes of acute inflammatory abdomen.

Laboratory diagnosis

Leukocytosis without left shift.

Imaging diagnosis

Abdominal ultrasonography depicting cecal appendix with thickened wall, locally associated with small quantities of intra-abdominal fluid and intestinal loop local obstruction.

Pathological diagnosis

Inflammation in both cecal appendices.

Treatment

Laparotomy with a McBurney's incision, followed by the performance of a double appendectomy and segmental ileectomy.

Related reports

Double cecal appendix is a rare (about 100 cases reported worldwide) anatomic variation most often incidentally diagnosed in face of inflammation of that organ.

Term explanation

The classification modified by Cave-Wallbridge categorizes double cecal appendix.

Experiences and lessons

The surgeon must be aware and identify cecal appendix anatomical variations. The procedure is recommended when surgeons surgically approach a patient with acute appendicitis. It is worth performing a complete cecum evaluation after the retroperitoneal release.

Peer review

This case report is well designed and presents a wide range of information

about the subject, spreading the right messages and broadly contributing to the literature.

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