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Case Report

Acute appendicitis in twin appendices: case report

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ABSTRACT

A double appendix is a rare congenital malformation with a frequency ranging from 0.004 to 0.009% and only a small percentage of instances presenting as appendicitis. Although the cause of appendiceal duplication is unknown, it poses a difficult clinical picture in patients with right lower quadrant pain. In this case report, we presented two cases of operative treatment of acute appendicitis in a doubled vermiform appendix in 4 and 17 years old male patients. Both cases did not have any pertinent prior medical condition and presented with shifting right lower quadrant (RLQ) abdominal pain associated with nausea, vomiting and anorexia. Clinical exam, laboratory investigations as well as imaging findings were consistent with features of acute appendicitis. In both patients, the diagnosis of the duplicated appendices was unsuspected until two tubular structures arising from the cecal wall were discovered intraoperatively and confirmed on histopathological examination after appendectomy of the two appendices was performed. Hence, in all situations where acute appendicitis is suspected clinically and radiologically, surgeons must maintain a high level of suspicion for the potential of duplicated appendices to avoid missing the duplication and resulting post-operative difficulties and medicolegal concerns.

Keywords: Duplicate, Appendicitis, Appendectomy, Case report

INTRODUCTION

The appendix (or vermiform appendix) is a blind tube that connects to the cecum and appendicitis characterizes inflammation of the appendix. A dull, poorly localized, visceral pain usually starts in the center of the abdomen, and the pain localizes to the right iliac fossa as the inflammation progresses. Appendicitis can also cause anorexia, nausea and vomiting and low-grade fever. Appendicitis necessitates the removal of the inflamed appendix by a laparotomy or laparoscopic appendectomy.

Several congenital abnormalities can occur in the appendix and these anomalies include partial or complete duplication of the appendix, agenesis and branching appendix.[1-3] The reported incidence of duplication on those operated for acute appendicitis is only 1 in 25,000 appendectomy specimens.[4,5] The first case with a duplicate appendix was reported by Piccoli in 1892 and about 100 cases have been reported ever since, most of which were identified incidentally.[6] A double appendix in children necessitates a more thorough examination since it is frequently a symptom of the more complex developmental gastrointestinal tract (GIT), genitourinary or vertebral problems.[7,8] Furthermore, the duplicated appendices may become inflamed simultaneously or separately, mimicking other conditions and making clinical identification challenging.[3]

Here we presented operative treatment of two different cases of acute appendicitis in a doubled vermiform appendix, which caused acute abdomen, without any associated pathology.

CASE REPORT

Case 1

A 17 years old male patient presented with characteristic migratory abdominal pain of 2 days duration associated with nausea and one episode of vomiting, anorexia and low-grade fever of similar duration. He had no pertinent previous medical history. All his vitals were in the normal limits and had RLQ direct and rebound tenderness and positive Rovsing's sign on the abdominal examination. The white blood cells count (WBC) was 13,200 with a left shift of 86% and abdominal ultrasound showed markedly dilated appendix with a diameter of 10.2 mm and minimal peri appendiceal collection.



Figure 1: Appendectomy stump and an inflamed duplicated appendix identified during second laparotomy in a 17 years old male adolescent.

The patient was taken up for a laparotomy through McBurney's incision. Intraoperatively, there was about 10 ml of thin offensive pus in RLQ, 100 ml of reactive fluid in the pelvis and an acutely inflamed retrocecal appendix with perforation at midshaft and fecalith within the lumen. Additionally, there was a grossly non-inflamed healthy-looking duplicated appendix positioned at pre ileal location with a distinct base and mesentery entering to cecum ~1.5 cm apart from the inflamed appendix. Lavage and appendectomy were done for the inflamed appendix and the duplicated healthy-looking appendix was left intact.

On the second postoperative day, the patient still complained of pain at RLQ of the abdomen and had deep tenderness unexplainable by the surgical wound on examination. Complete blood cell count revealed a WBC of 8500 with 67% neutrophil and repeat abdominal ultrasound showed a dilated, diameter measuring 0.91 cm, noncompressible appendix perforated at its proximal shaft, but no fecalith with peri appendiceal complex fluid collection with echo debris measuring 7.8×2.6 cm.

With the impression of acute complicated appendicitis of duplicate appendix and postoperative collection, the abdomen was explored through an infraumbilical midline incision. The appendiceal appendectomy stump was healthy-looking and intact but the second pre-ileal appendix was perforated at the midshaft with an abscess cavity in the RLQ. The abscess cavity was dismantled, pus sucked out and an appendectomy of the second appendix was done and sent for histopathology. The specimen sent for histopathology was reported as showing features of acute appendicitis. The patient was put on intravenous antibiotics (ceftriaxone and metronidazole) and discharged on the 4th post relaparotomy day with per os antibiotics and analgesics. On follow up visits, the patient was doing fine.

Case 2

A 4 years old male child presented with RLQ abdominal pain of 5 days duration along with nausea, vomiting of ingested matter, anorexia and low-grade fever of similar duration. He was given unspecified syrup medications for four days before his presentation. This healthy child had a pulse rate of 116 bpm and the physical examination confirmed the presence of the hallmarks of acute appendicitis with direct and rebound tenderness in the RLQ of the abdomen.



Figure 2: Duplicated appendix identified during initial laparotomy in a 4 years old male child.

He had an elevated WBC of 15300 with neutrophilia of 75%. He was scanned with abdominal ultrasound and it revealed a single dilated and hyperemic appendix measuring about 0.8 cm with about 15 ml of peri appendiceal collection having echo debris. The abdomen was approached via McBurney's incision and the intraoperative findings included about 20 ml of thick

offensive pus in a cavity formed by the cecum, terminal ileum, omentum and anterolateral abdominal wall. There was a sub-cecal appendix with a healthy base and perforation at the tip and another acutely inflamed retrocecal appendix about 1 cm apart. Both the appendices had a mesoappendix containing an appendicular artery at their free margin.

Then the cavity was dismantled gently, the abscess sucked out and standard appendectomy was done for both appendixes. The specimen was sent for histopathology and reported as showing features of acute appendicitis. The patient was put on intravenous antibiotics (ceftriaxone and metronidazole) and discharged on the 4th postoperative day with per os antibiotics and analgesics. On follow up visits on the 7th and 30th postoperative days, the patient was doing fine.

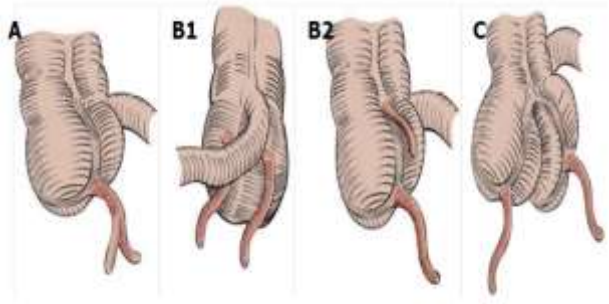


Figure 3: Classification modified by Cave-Wallbridge including the type A, subtypes B1, B2 and type C.[9]

DISCUSSION

The duplicate appendix was a rare congenital anomaly with an incidence of 0.004-0.009%.[5] The presence of a second appendix was often discovered incidentally after abdominal surgery, postmortem examinations or barium enema investigations.[10] Although duplicated appendices wee unusual, they had therapeutic and legal implications. Cave proposed the first classification for appendiceal duplication in 1936, which was later refined by Wallbridge et al.[11] Wallbridge et al divided appendiceal duplication into three forms in 1963: type A, type B and type C.[12]

According to the Cave-Wallbridge classification, type A duplications were defined as a single caecum that gave rise to various degrees of partial duplication on a normally localized appendix.[4,11,13] Two distinct appendages on either side of the ileocecal valve made up type B1 (bird-like or avian type). Type B2 contained one retrocecal appendix emerging from taenia coli convergence and a smaller second appendix along the anterior taenia at a variable distance from the first. Type C was made up of two separate ceca each with its appendix (Figure 3). Duplication in a horseshoe pattern considered as type D and triple appendices had also been reported.[1,2,14,15] Both of our patients presented in this paper could be classified as having type B1 as the appendices were arising from either side of the cecum (Figure 1 and 2). Most of the cases of double appendix studied by Wallbridge were of the B type accounting for 60% of cases.[7]

Most double appendices were asymptomatic but may present with symptoms of appendicitis even 5 months to 7 years after an appendectomy.[10] Similarly, in our first patient, the second appendix was perforated and complicated with an abscess on the second postoperative day of the initial appendectomy.[16] Both appendices might also be inflamed or perforated at presentation as in our second patient.[9,17] However, the incidence of appendicitis was difficult to quantify in patients with a duplex appendix for a variety of reasons, including the rarity of the condition and the fact that many instances may go unnoticed.[4]

In adults, it was frequently discovered incidentally during a laparotomy done for other disease conditions.[7] In children, however, concomitant malformations or duplications of the large intestine or the genitourinary system may be present, especially in types B1 and C which may serve as alarm signs for the successful identification of the duplication.[8,18] Among the various types of duplication, type B and especially subtypes where the second appendix lied retrocecally were of the highest risk to remain unnoticed and led to perforation and generalized peritonitis.[19] In our first case, however, the pre-ileal duplicated appendix ruptured and resulted in localized pelvic peritonitis.

The clinical features were indistinguishable for a single or duplicated appendix. Although preoperative diagnosis had been made with the aid of radiological studies such as a barium enema, abdominal ultrasound and CT scan, the majority of cases have been diagnosed at the surgery or on pathological examination.[20] The reported sensitivity and specificity of both for the diagnosis and especially the detection of the appendix was of less importance because these modalities were usually not included in the routine workup of otherwise healthy patients with right lower quadrant pain.[18] Both of our patients had preoperative abdominal ultrasounds, each performed by different consultant radiologists, but none pointed out the presence of a duplicated appendix supporting this evidence.

The difficulty of accurate preoperative diagnosis and the gravity of the associated complications of delayed diagnosis and management warranted a high index of suspicion for the duplication of appendix in all patients with lower abdominal pain, even if the patient reported a previous appendectomy.[19,21,22] The cecum should be visually inspected routinely to ensure that there were no appendiceal anomalies and laparoscopic or open appendectomy of both should be done to avoid future diagnostic confusion even if only one was inflamed.[4,7] In our first case, an inspection of the cecum and excision

during the first appendectomy could have prevented a second surgery for postoperative collection with perforation of the second appendix and the subsequent prolonged hospitalization and IV antibiotics. The diagnosis of a duplicate appendix warranted further investigation for possible underlying intestinal, genitourinary and vertebral malformations, especially if a type C abnormality was observed.[9,21]

CONCLUSION

We presented two different cases of appendiceal duplication which is an exceedingly rare condition with no known cause. As a result of the inadequate understanding of this odd but significant aberration, the presence of a duplicate appendix may go overlooked during surgery, necessitating additional surgical intervention for complications as in our first patient and perhaps could culminate in a lawsuit. Hence, the duplicated appendix should be kept in mind in patients presenting with right lower quadrant abdominal pain.

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