



Acute Appendicitis in a Patient with Appendiceal Duplication: A Case Report

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Abstract

Acute appendicitis is a common pathology in emergency surgery. Some anatomic anomalies can complicate its correct diagnosis and management. Appendiceal duplication is a rare congenital abnormality. Its incidence rates 0.004% among patients operated for acute appendicitis or other surgical pathology. Preoperative diagnosis of appendiceal duplication is extremely difficult and it can be mistaken for other intra-abdominal disorders in some cases. In this paper we report a case of appendiceal duplication in a patient who underwent appendectomy on two separate admissions.

Keywords: Acute appendicitis; Appendiceal duplication; CRP; CT

Case Presentation

A 39-year old male presented at the Emergency Room after seven days of nausea, fever (38, 4°C) and a constant abdominal pain localized in the right iliac fossa. Patient's past medical history was not significant for any pathology. A physical examination revealed right lower quadrant pain with guarding and rebound tenderness. Laboratory results showed an elevated white blood cell count ($27 \times 10^3/\mu\text{L}$) with a left shift and an increased C-reactive protein level (267.6 mg/L). The abdominal computed tomography scan (CT-scan) detected an increased and thickened appendix and a voluminous inflammatory mass surrounded by dilated bowel loops. The patient underwent an open appendectomy through sub-umbilical laparotomy. Intraoperative findings included a significant localized peritonitis with purulent fluid caused by an inflammation of appendix extending to the cecal wall. Histological study revealed an acute suppurative appendicitis with a granulocyte inflammation of the surrounding tissues. Postoperative course was uneventful and the patient was discharged 7 days after surgery. The patient was readmitted to the Emergency Room four months later for fever (37,8°C), tachycardia and a sudden sharp pain localized in the lower quadrants and in the left flank of the abdomen. His abdomen was distended with generalized tenderness and rebound tenderness in the lower quadrants. Inflammatory markers were elevated (WBC $16.10 \times 10^3/\mu\text{L}$; CRP 102.6 mg/L). An abdomen CT-scan showed distended bowel loops with air fluid levels subsequent to a bowel loop obstruction suggesting for a volvulus of the small bowel. It was also documented an increased density of mesenteric tissue and the presence of a thickened cecum wall. The patient was taken to surgery. A moderate free fluid collection was found; the two ileum loops were twisted around each other due to peritoneal adhesions. Once the adhesions were divided, the volvulus was untwisted and the bowel, slightly discoloured, started to return to its normal colour and shine. The following exploration of the abdominal cavity revealed a partially retrocecal appendix showing signs of inflammation. So appendectomy was performed. Pathological examination confirmed an acute appendicitis. Post-operative period was uneventful and the patient was discharged on the sixth postoperative day.

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Discussion

The first case of duplication of the appendix was reported by Picoli in 1892 [1-3]. The possible anatomical and embryological variants of appendiceal duplication were described in Cave - Wallbridge classification, which was modified by Biermann [4-6] in 1993 as follows:

Type A: Single caecum with one appendix exhibiting partial duplication.

Type B: Single caecum with two obviously separate appendices.

B1: The two appendices arise on either side of the ileocaecal valve in a "bird-like" manner.

B2: In addition to a normal appendix arising from the caecum at the usual site, there is also

a second, usually rudimentary, appendix arising from caecum along the lines of the taenia at a varying distance from the first.

B3: The second appendix is located along the taenia of the hepatic flexure of the colon.

B4: The location of the second appendix is along the taenia of the splenic flexure of colon.

Type C: Double caecum, each bearing its own appendix and associated with multiple duplication anomalies of the intestinal tract as well as the urinary tract.

More recently other authors have reported the cases of "horseshoe appendix" which is characterized by two openings into a common caecum [7,8] and the extremely rare cases of "triple appendix" [9,10]. Although embryology of the normal appendix has been defined, pathogenesis of its duplication remains unclear. Cecum and appendix develop from the wall of the primitive midgut as conical dilatation at the end of the 5th week of gestation. The proximal part of the gut rapidly grows and generates cecum, while its distal part remains narrow to form the appendix. According to this embryological description appendicular duplication can be isolated or associated with cecal duplication [11,12]. Several theories were proposed to explain the pathogenesis of appendicular duplication. These include the split notochord theory, the median septum formation, failure of the normal regression of embryonic diverticula and partially twinning procedure [13]. Cave supposed that duplication of the appendix can be a result of a transient embryological structure or it can be a consequence of a more general affection of the primitive midgut [4]. Preoperative diagnosis of double appendix is extremely difficult. Routine radiological studies like ultrasonography and CT-scan cannot distinguish this intestinal abnormality [14]. Duplication of the appendix is generally identified incidentally during surgery for acute appendicitis of one of the two appendices or for other surgical pathology [2,8]. The second appendix can be overlooked in case of an important inflammatory pericecal process, a generalized peritonitis, an insufficient exploration or any variation in appendix position. In our case the patient underwent emergency appendectomy due to an acute peritonitis; the duplicated appendix was found on a second laparotomy performed a few months later for a suspected volvulus. An undetected second appendix may be asymptomatic, but if inflamed, it can lead to different clinical errors, delay in diagnosis and medico-legal consequences [15]. The diagnosis of acute appendicitis in patients with a previous appendectomy may be misunderstood with cecal diverticulum, colon cancer, volvulus, intussusception, small bowel obstruction or inflammatory bowel disease [16]. The diagnosis of acute appendicitis should be suspected in all cases of lower abdominal pain even if the patient had undergone appendectomy. In patients with double appendix, although only one of them is found inflamed, all of them should be removed to avoid

diagnostic confusion thereafter [2]. The treatment of choice for acute appendicitis is a laparoscopic appendectomy. However, in cases of doubtful diagnosis or acute abdomen with peritonitis, explorative laparotomy may be required. Although appendicular duplication occurs very rarely, it could present like an acute appendicitis on two separate cases in the same patient and it can lead to diagnostic difficulties with their potential complications. The accurate revision of the caecum during appendectomy should be advisory in order to identify any possible duplication of the appendix and to avoid clinical and medico-legal consequences.

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