

Bowel obstruction due to knotting as an atypical presentation of acute appendicitis: case presentation and clinical comparison with 13 international cases

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Abstract

Context

Acute appendicitis is one of the main indications for an emergent abdominal surgical intervention. The classic presentation includes abdominal pain, nausea and vomiting. However, it has a variety of atypical presentations, especially in late adulthood.

Subject and methods

We report a case of an elderly patient with a positive past abdominal surgical history whose initial symptoms and imaging suggested bowel obstruction with significant clinical deterioration. The surgical management consisted of an exploratory laparotomy, finding an intestinal obstruction at the ileocecal valve due to a necrotic appendix wrapped around it; named Appendiceal Tie Syndrome. We reviewed the literature to look for similar cases in MEDLINE database. Of the eligible papers, we selected only case reports. We performed a clinical comparison of our case with the previous reports.

Results

In the reviewed articles, 13 reported cases were similar to ours. Including our patient, seven cases were older than 60 years of age and 10 were male. The most common duration of symptoms was 3 to 4 days prior surgery. Among the 14 cases of acute appendicitis with knotting, five reported bowel resection. In the majority of cases that involved elderly patients, a bowel resection was necessary due to small bowel necrosis. In contrast, in our case, there was no evidence of necrosis, nor need of bowel resection, despite the age of the patient and the prolonged evolution of 63 hours from the onset of symptoms and surgical resolution. The possible explanation for the preservation of the intestine is that the strength of the ileocecal valve helped to safeguard its vascularity preventing necrosis.

Conclusion

This case let us emphasize the importance of early detection and surgical resolution of an atypical acute appendicitis particularly in patients with signs and symptoms of bowel obstruction. Furthermore, it is critical not to underestimate the possible complications of this apparently simple diagnosis because it can cause high morbidity and mortality. In addition, we considered that our findings, supported by other published case reports, suggest an association between appendix knotting and a history of past abdominal surgeries.

Resumen

Palabras clave: Apendicitis aguda, Obstrucción intestinal, Síndrome de anudamiento, Anudamiento apendicular, Presentación atípica.

Obstrucción intestinal debido a anudamiento como presentación atípica de la apendicitis aguda: presentación de un caso y comparación clínica con 13 casos internacionales

Contexto

La apendicitis aguda es una de las principales indicaciones para una intervención quirúrgica abdominal de emergencia. La presentación clásica incluye dolor abdominal, náusea y vómito. Sin embargo, existe una gran variedad de presentaciones atípicas, especialmente en los pacientes adultos mayores.

Sujeto y métodos

Se presenta el caso de una paciente adulta mayor con antecedentes de cirugías abdominales previas, cuyos síntomas y exámenes de imagen iniciales sugirieron un proceso de obstrucción intestinal con deterioro clínico significativo. El manejo quirúrgico consistió en una laparotomía exploratoria. Entre los hallazgos se encontró una obstrucción intestinal en la válvula ileo-cecal debido a que el apéndice en estado necrótico lo estaba envolviendo, lo que se denomina Síndrome por Anudamiento Apendicular. Se efectuó una búsqueda en MEDLINE y se realizó una comparación clínica con los reportes de casos internacionales.

Resultados

Se identificaron otros 13 casos similares reportados internacionalmente. Incluyendo nuestro paciente, siete casos fueron mayores de 60 años de edad y 10 eran hombres. La duración más común de los síntomas fue de 3 a 4 días antes de la cirugía. Entre los 14 casos de anudamiento apendicular, cinco reportaron una resección intestinal. En la mayoría de los casos correspondientes a pacientes adultos mayores, la resección intestinal fue necesaria debido a necrosis del intestino delgado. En contraste, en nuestro caso no hubo evidencia de necrosis, ni necesidad de resección intestinal, a pesar de la edad del paciente y la evolución prolongada de 63 horas desde el inicio de los síntomas y la resolución quirúrgica. La posible explicación para la preservación del intestino es que la fuerza de la válvula ileocecal ayudó a salvaguardar la vascularidad y prevenir la necrosis.

Conclusión

Este caso permite enfatizar la importancia de una detección y resolución quirúrgica temprana en una apendicitis aguda con presentación atípica, particularmente en pacientes con signos y síntomas de obstrucción intestinal. Es muy importante no subestimar las posibles complicaciones que este diagnóstico aparentemente simple, ya que puede causar alta morbilidad y mortalidad. Además, nuestros hallazgos y los casos publicados, sugieren una posible asociación entre el anudamiento apendicular y los antecedentes de cirugías abdominales previas.

Introduction

Acute appendicitis is a common condition that results from the inflammation of the veriform appendix which requires a rapid diagnosis and treatment. In the United States, the reported crude annual incidence of acute appendicitis is 11 cases per 10000 people [1]. The initial point of the diagnosis and management decision focuses on the clinical presentation, including the typical pain in the right iliac fossa in 96% of cases, nausea in 80% and vomiting in 73% [2]. Despite this, the classic presentation occurs in only 31% of patients [2]. There have been reports of acute appendicitis with atypical presentations, especially in older adults, including bowel obstruction [2]. This clinical scenario is less common and can be explained as the result of 4 different mechanisms: by adynamic ileus, adhesion formations, direct obstruction of the intestine by the inflamed appendix and intestinal obstruction due to mesenteric ischemia [3]. In all four mechanisms the predominant clinical presentation is bowel obstruction, which masks the classic presentation of appendicitis and lengthens the time of diagnosis and treatment. This could cause complications and fatal consequences for the patient [4]. Moreover, the diagnosis may be delayed even more in the elderly patient because the clinical presentation is different from the general population. In fact, the classic pain of appendicitis is only present in 35% of the aging population and there is a higher risk of complications (2.5 times) and mortality (12 times) [5, 6]. Altogether we recommend considering the diagnosis of acute appendicitis in a typical scenario of bowel obstruction. We report a case of acute appendicitis in an 82 year-old patient with an intestinal obstruction by strangulation of the ileocecal valve and her briefly comparison with other 13 international cases.

Subject and methods

Case presentation

An 82 year-old female patient came to the emergency department with abdominal pain and bloating. The pain started 26 hours prior to hospital admission and initially began as diffuse, progressive, burning abdominal pain with 6 out of 10 intensity. The pain subsequently localized to the epigastric region and finally migrated to the right lower quadrant. It was accompanied by nausea, four episodes of bilious vomiting and difficulty passing flatus. Additionally, three days prior to the onset of abdominal pain, she suffered from constipation. She normally has forceful bowel movements every 2 days. Past medical history included hypothyroidism diagnosed 13 years ago and treated with levothyroxine 50 µg daily as well as chronic constipation for 13 years treated with lactulose 15 ml every other day. Four years ago, she had a hysterectomy for an unspecified benign uterine tumor. Around this time, she also had a left inguinal hernia repair.

Physical examination at admission showed normal vital signs, dry oral mucosa, distended abdomen and decreased bowel sounds. She had diffuse tenderness predominantly with deep palpation in the right lower

quadrant, positive Blumberg, McBurney, and Dunphy signs, and negative Rovsing and Murphy signs. No masses were palpated. On rectal examination, she had an adequate sphincter tone, the rectal vault was empty with absence of macroscopic blood. The remainder of the examination was noncontributory. A nasogastric tube was placed initially without immediate presence of liquid.



Figure 1 Abdominal X-ray. Pneumatization and dilation of small bowel loops with characteristic arrangement of obstructive abdomen. Absence of gas in the rectal vault.

Laboratory tests showed 7590 leukocytes/mm³ with 89.1% neutrophils. On admission, an abdominal X-ray (**figure 1**) demonstrated pneumatization and dilated small bowel loops characteristic of abdominal obstruction. The CT scan also showed dilated bowel loops and air-fluid levels (**figure 2-A** and **2-B**). The patient subsequently was admitted with the diagnosis of bowel obstruction. Nine hours after admission she developed leucopenia 860/mm³ with 45.3% neutrophils and a C-reactive protein (CRP) of 190.80 mg/L. Thirty-three hours after admission, the clinical picture worsened, with greater abdominal distention accompanied by 1460/mm³ leukocytes, 55.5% neutrophils, a CRP of 216.7 mg/L, and a procalcitonin (PCT) of 4.42 ng/mL.

An exploratory laparotomy was performed 63

hours from the onset of symptoms. During the procedure, approximately 800 ml of inflammatory peritoneal fluid was identified as well as a distended jejunum and ileum. An intestinal obstruction at the ileocecal valve was found due to a necrotic posterior appendix wrapped around it- this is known as Appendiceal Tie Syndrome (**figure 3**). The other intestinal loops and the mesentery were normal and there was no evidence of intestinal necrosis. Histopathological macroscopic results reported a cecal appendix measuring 7x1 cm, covered with an opaque serosa and fibrinoid-purulent material. On microscopy, a cecal appendix was observed with areas of necrosis and abundant polymorphonuclear inflammatory infiltrate that involved the entire wall thickness (**figure 4**). The diagnosis of necrotizing acute appendicitis was confirmed.

Search strategy

We reviewed the literature to look for similar cases in MEDLINE database using the terms "acute appendicitis" AND "bowel obstruction", yielding 839 results, "atypical appendicitis presentation" yielding 119 results, "appendiceal tie syndrome" yielding 2 results. Of the eligible papers, we selected only case reports.

Clinical comparison with other cases

Data were extracted from selected articles using pre-defined parameters about the case reported: sex, age, and duration of symptoms; type of surgery performed, findings at operation, need of intestinal resection, and past-history of abdominal surgery. We performed a clinical comparison of our case in these variables with the previous reports.

Results

In the reviewed articles, 13 reported cases were similar to ours [3, 7-16]. The first one published in 1973 [7] and the last in 2015 [15]; **table 1**. Nine articles reported a single patient [3, 8-15], and two articles reported simultaneously two different patients [7, 16]. Including our patient, seven cases were older than 60 years of age and 10 were male. Duration of symptoms ranged from 1 to 7 days prior surgery, but the most frequent duration was between 3 and 4 days in seven cases: **table 1**.

Among the 14 cases of acute appendicitis with knotting published in the literature, five required bowel resection [7-10, 13], seven preserved the bowel integrity [3, 7, 11, 12, 14] and in two cases no description was offered [16].

In the majority of cases that involved elderly patients, a bowel resection was necessary due to small bowel necrosis [8-10]. In contrast, in our case, there was no evidence of necrosis despite the age of the patient and the time (63 hours) elapsed between the onset of symptoms and surgical resolution.

Rajan et al [15], reported a case of a 65 y/o male elderly patient in which there was no need for resection. Similarly, in our case, intestinal resection was not required, regardless of the extensive time of evolution. The possible explanation for the preservation of the intestine in this case is that the strength of the ileocecal valve helped to safeguard its vascularity preventing necrosis.



Figure 2-A - Abdominal Computed Tomography, coronal view. Dilation of small bowel loops due to obstruction

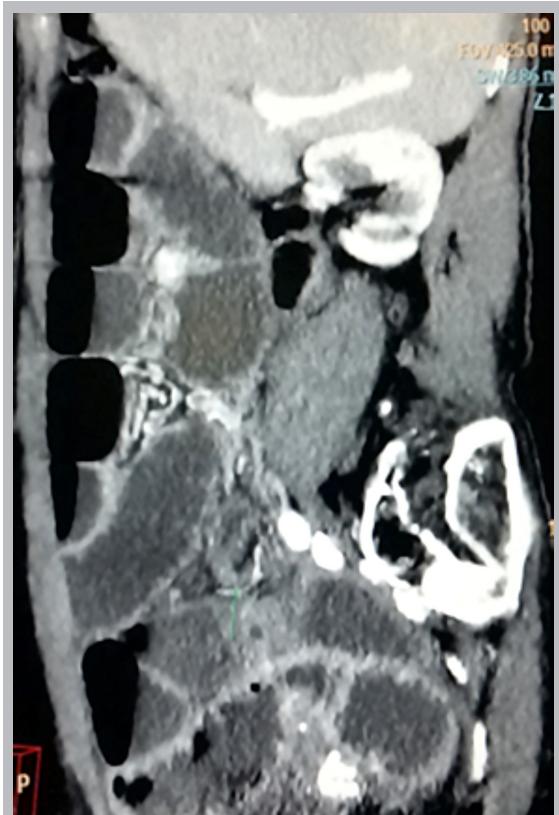


Figure 2-B - Abdominal Computed Tomography, sagittal view. Dilation of small bowel loops and air-fluid levels



Figure 3 - Exploratory laparotomy. Appendiceal knotting around the ileo-cecal valve causing bowel obstruction.

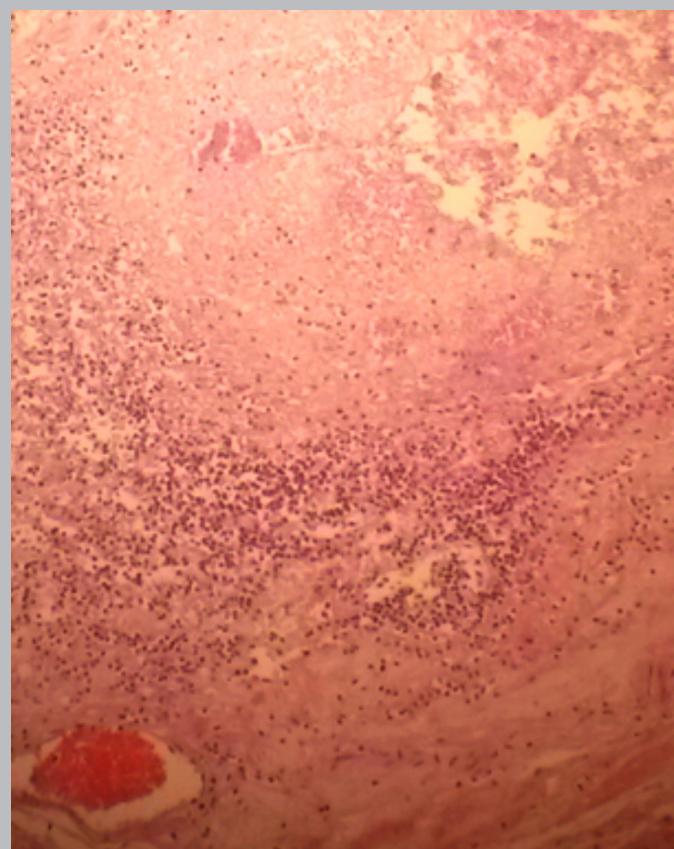


Figure 4 - Microscopy of the appendix. Polymorpho-nuclear inflammatory infiltrate involving the entire thickness of the appendiceal wall.

Discussion

Acute appendicitis is a condition that needs to be diagnosed in a timely manner in order to avoid complications such as an inflammatory mass, an abscess, a ruptured appendix, and generalized peritonitis, among others^[2]. Taking into account that it can present atypically at different ages, health professionals should recognize the clinical variants of this common disease. Bhandari et al.^[3] classified the presentation of different cases of acute appendicitis with strangulation into two types: the

first with predominant features of appendicitis and the second with classic features of bowel obstruction. Our case corresponds to the second type. In particular, acute appendicitis was found during an exploratory laparotomy of a patient with apparent acute intestinal obstruction and clinical deterioration. Thus, one should consider that acute appendicitis can be masked by obstructive symptoms, as in the case presented, and must be part of the differential diagnosis in a patient with abdominal pain, especially in the elderly.

The singularity of the case presented lies in the arrangement and location of the appendix around the ileocecal valve. This unusual anatomic location caused complete obstruction, delaying the clinical diagnosis for 63 hours. According to the literature reviewed, the location of appendiceal knotting is mainly in the distal ileum and none in the ileocecal valve as described in this case report^[10, 11, 14, 15].

The mechanism of the appendiceal knotting is that the tip of this organ is adhered to its body forming an annular structure for the passage of bowel loops. Bose et al.^[7] proposes that it is difficult to explain the adherence of the appendix to the intestine but the most likely mechanism is that the inflamed appendix adheres to the hyperactive bowel loops in the acute phase. It is less likely that this particular location of the initial uninflamed appendix resulted in the knotting of the ileocecal valve^[7].

One hypothesis proposed is that the existing anatomical location of the appendix around the ileocecal valve possibly occurred by the previous abdominal surgeries that the patient went through. For this reason, we must consider that a positive abdominal surgical history relates to the presentation of abdominal obstruction in acute appendicitis, particularly in patients with Appendiceal tie syndrome. In the cases reported (**Table 1**), most authors do not mention the history of abdominal surgery. Of all the elderly patients, only 3 cases described past surgical history. No previous surgeries were reported in one case and hysterectomies were reported in the remaining two cases. Hence, there might be an influence of past surgeries in tie syndrome especially in older adults. However, more investigation on this topic is required to establish a real association.

It is important that health professionals consider acute appendicitis with an atypical presentation as the etiology for intestinal obstruction. With this in mind, it can be treated as soon as possible to avoid complications. One suggestion is to use the parameters in the blood count, CRP and PCT, as indicators for surgical need in patients with intestinal obstruction. More than 10000 leukocytes/mm³ has a positive likelihood ratio for appendicitis of 2.47 (95%CI: 2.05-2.95, p<0.001) and if this value is greater than 12000/mm³, the positive likelihood ratio is 2.75 (95%CI: 1.99-3.80, p<0.05)^[17]. Moreover, if the C-reactive protein is greater than 10 mg/L, it has a positive likelihood ratio for appendicitis of 1.97 (95%CI: 1.58-2.45, p<0.001) and if it is greater than 20 mg/L the positive likelihood ratio is 2.39 (95%CI: 1.67-3.41, p<0.05)^[17]. Additionally, PCT raising more than 0.57 ng/

Table 1: Cases of mechanical bowel obstruction due to knotting in 13 patients with acute appendicitis reported in literature compared with the case reported.

Case	Autor, year [Reference]	Sex, age (years)	Duration of symptoms (days)	Findings at operation	Surgery	Intestinal resection	Past history of abdominal surgery
1	Bose, 1973 [7]	M, 50	1	Gangrene of the distal ileum. Inflamed and retro-ileal appendix. Its distal half was wrapped around a loop of ileum, producing a strangulating obstruction.	Retrograde appendectomy + bowel resection	Five feet of gangrenous ileum	Not reported
2	Bose, 1973 [7]	M, 35	1	Small intestine distended and bluish. Volvulus of the ileum. Appendix encircling the loop of ileum, causing mechanical obstruction and strangulation of the gut. Inflammation of the tip of the appendix and adherent to the small intestine. Meckel's diverticulum in the terminal part of the ileum.	Retrograde appendectomy	The volvulus was undone and the Meckel's diverticulum removed.	Not reported
3	Assenza, 2005 [8]	F, 78	1	Bloody peritoneal fluid. Inflamed appendix wrapping around the last loop of ileum, producing volvulus of bowel	Ileocecal resection + Reconstruction of bowel	Caecum and about 70 centimeters of ileum	Not reported
4	Menon, 2007 [9]	M, 81	1	Inflamed appendix wrapped around the mid ileal loop forming an appendico-ileal knot, resulting in strangulation of small bowel.	Laparotomy + appendectomy + bowel resection	20 cm of ischaemic bowel	Not reported
5	Bhandari, 2009 [3]	M, 24	7	Hemorrhagic fluid. Dilated jejunal loops. Strangulation of the ileal segment by a band of inflamed appendix and omentum producing a window underneath, through which the intestine had protruded.	Appendectomy	No resection was needed.	Not reported
6	Harrison, 2009 [16]	F, 62	4	Inflamed appendix. Small bowel obstruction.	Appendectomy + adhesiolysis	Not reported	None
7	Harrison, 2009 [16]	M, 83	4	Inflamed appendix causing small bowel obstruction	Laparotomy + adhesiolysis + appendectomy.	Not reported	Not reported
8	Lukong, 2009 [11]	M, 10	6	Inflamed appendix with the tip knotted at the terminal ileum and an inflammatory mass surrounding the tip. Herniation of part of the terminal ileum. Dilated appendix and stomach. Collapsed distal ileum and colon. Ischaemic portion on the terminal ileum.	Laparotomy + appendectomy	No resection was needed.	None

Table 1. Continuation

Case	Autor, year [Reference]	Sex, age (years)	Duration of symptoms (days)	Findings at operation	Surgery	Intestinal resection	Past history of abdominal surgery
9	O'Donnell, 2009 [10]	F, 86	3	Distention of proximal small bowel with an ischaemic segment. The appendix had surrounded a loop of the terminal ileum, causing obstruction. Buried tip of the appendix resulting in a tour-niquet type-effect on the small bowel. Chronic inflammation of the appendix.	Laparotomy + appendicectomy + bowel resection	40 cm segment of small bowel	Histerectomy
10	Chatterjee, 2013[13]	M, 26	3	Hemorrhagic fluid. Dilatation of the jejunum and ileum. The tip of the inflamed appendix was adherent to the ileum forming a ring like structure with herniation of the ileum. Gangrenous ileal segment.	Laparotomy + bowel resection	Seg-mental ileocolic resection	None
11	Inoue, 2013 [12]	M, 3	3	Dilatation of a loop of ileum due to obstruction by the adherent and encircling vermiciform appendix. At the middle of the appendix was adhered to the me-senterium and formed the hernia orifice. Necrotic distal appendix due to torsion.	Laparotomy + appendicectomy	No resec-tion was needed.	Not reported
12	Awale, 2015 [14]	M, 20	4	Distended loops of small bowel. Constricting ring around the terminal ileum created by a phlegmonous appendicitis.	Laparotomy + appendicectomy	No resec-tion was needed	None
13	Ranjan, 2015 [15]	M, 65	4	Small bowel obstruction caused by knotting due to encircling of the small bowel by appendix. Gangrenous inflammation of periappen-dicular tissue.	Laparotomy + appendicectomy	No resec-tion was needed	None
14	Torres, 2015 [Case re-reported]	F, 82	1	Inflammatory peritoneal fluid. Distended jejunal and ileum loops. Intestinal obstruction at the ileocecal valve due to necrotic appendix attached to posterior region.	Laparotomy + appendicectomy	No resec-tion was needed	Histerectomy

Sex: M: Male; F: Female.

ml is a marker of intestinal ischemia in patients with intestinal obstruction, and therefore could help in the decision for surgery^[18]. Consequently, a high clinical suspicion and the use of laboratory tests could lead to a prompt diagnosis of bowel obstruction due to acute appendicitis.

Conclusion

The diagnosis of acute appendicitis is challenging due to its diverse presentations, especially in older adults. It is therefore important to know the atypical presentations of this common disease and consider the diagnosis of acute appendicitis if the clinical symptoms suggest

intestinal obstruction. Intestinal obstruction in some cases of acute appendicitis occurs due to knotting of the appendix around the bowel loops causing ischemia and necrosis, which in turn significantly increases morbidity and mortality. A high clinical suspicion accompanied by laboratory tests and relevant imaging studies can lead to an early diagnosis and resolution without surgical complications.

Informed consent statement

The study participant provided informed written consent.

Conflict-of-interest

No conflicts declared

Financial disclosure

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Institutional review board statement

The study was reviewed and approved by Institutional Review Board of the Universidad San Francisco de Quito

Author contributions

Torres F generated the idea of the manuscript, made the documentation of the case and the study design for clinical comparison with other cases, reviewed the draft and corrected the final version of the article. Rodán GA and Cobo G made the review of the literature and wrote the draft. Dorn H reviewed and corrected the draft and approved the final version of the article. Torres F was the leader of the surgical team.

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