

# A very rare association: acute apendagitis and appendicular agenesis, case report

## Abstract

**Background:** Apendagitis is an entity with an incidence about 8.8: 1,000,000 habitants. It represents difficult diagnosis pathology, completed many times only after surgical exploration. The association of Apendagitis with vermiform appendix agenesis has never been reported before in literature.

**Case:** A 4 years-old female present at emergency room with abdominal pain of 2days of evolution. At physical exam with tachycardia, tachypnea, temperature of 38°C, dehydrated, abdominal pain localized in right iliac fossa, Mc Burney sign, other appendicular signs absents, without bowel sounds. Laboratories report leucocytes 14800 with neutrophils 78%. Without US or CT scan image in the hospital during night, acute appendicitis diagnosis was carried out and an appendectomy programmed. During surgery and after Cattel maneuver the vermiform appendix was not found and a Meckel diverticulum discarded. Mc Burney incision was extended and a necrotic epiploic appendix found in the ascendant colon, it was removed. Diagnosis of Apendagitis and appendicular agenesis was registered and after twodays of analgesia treatment, patient was discharged without complications.

**Conclusion:** Apendagitis diagnosis is a rare entity that would be consider in cases of acute abdominal pain because would be misdiagnosed as pathologies like diverticulitis, appendicitis or Meckel diverticulum. The preoperative diagnosis can only be achieved accurately by CT-scan and discarding other abdominal pain causes, avoiding unnecessary surgeries and allowing medical management for this entity.

**Keywords:** epiploic apendagitis, apendagitis, vermiform appendix agenesis

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## Introduction

First described in 1956 by Lynn, Apendagitis was described as a benign and self limited disease secondary to a colon epiploic appendix torsion, with ischemia or venous thrombosis. It is a rare entity that could be clinically confound with diverticulitis, appendicitis or a Meckel diverticulum, with diagnosis confirmation in the majority of cases only after surgical exploration.

Vermiform appendix agenesis was first described by Morgagni in 1718 and is a rare entity too, with an incidence of 1:100,000 laparotomies for suspected appendicitis. In the present we report the case of a 4 year-old girl which presented an acute abdomen, diagnosed as acute appendicitis by clinical and laboratory findings, but that at surgical exploration resulted in Apendagitis and vermiform appendix absence, an association never reported before.

## Case report

A 4-year-old is presented in the emergency department complaining of abdominal pain. She had no pathological background. The abdominal pain begins 48hours before in the umbilical area and increased progressively, with migration to right iliac fossa, without association with nausea or threw up, but with fever of 38.4 °C last 6hours. At physical exam with 95 beats per minute, 27 breaths per minute, 38.2°C. Irritable, referring abdominal pain, dehydrated

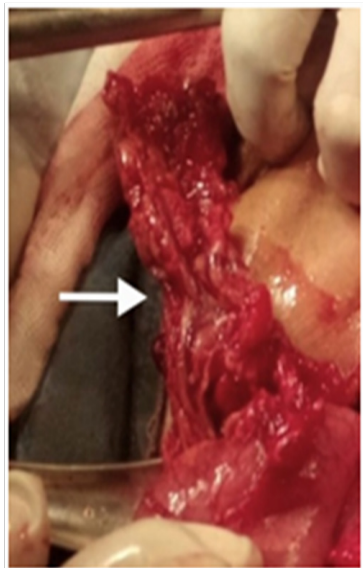
teguments, tachycardia, tachypnea with suitable bilateral ventilation, distended abdomen without diminished bowel sounds, pain localized in right iliac fossa, Mc Burney sign without other appendicular signs absent. Laboratories report Hemoglobin 13.6mg/dL, Leucocytes 14800/mm<sup>3</sup> with Neutrophils 78%, Platelets 122000, Glucose 115mg/dL, Cr 1mg/dL, and urinalysis without pathologic findings. This case presented in the night, when in this second level hospital we did not have auxiliary imaging studies like ultrasound or CT scan. With clinical data and laboratories the acute appendicitis diagnosis was established and surgery programmed. During surgery cecum was localized and explored to identify vermiform appendix but it was not identified, for this reason a Cattel maneuver was developed and posterior (Figure 1) and anterior (Figure 2) cecum surfaces explored. Appendix was not and agenesis was the conclusion. Ileum and jejunum were explored searching a Meckel diverticulum without success. When ascendant colon was explored we found an epiploic appendix crooked, with thickening, inflammatory aspect and ischemia patches, and this was removed. The final diagnosis was Apendagitis and appendicular agenesis, received analgesic therapy only and was discharged after twodays without complications. At 30days follow up she remained asymptomatic. The histopathology study reports inflammatory cell infiltration with predominance of eosinophilic cells, ischemia and necrosis secondary to vascular thrombosis, with final diagnosis of Apendagitis.



**Figure 1** Ileocecal junction, posterior cecal view without vermiform appendix.



**Figure 2** Ileocecal junction, anterior cecal view without vermiform appendix.



**Figure 3** Epiploic appendix (White arrow) with ischemia, inflammatory signs and thickening.

## Discussion

Apendagitis is a rare cause of abdominal pain considered as a differential diagnosis from appendicitis.<sup>1</sup> It is a benign pathology with an estimated incidence of 8.8: 1000000 patients and in some series 2% of hospitalized patients with appendicitis or diverticulitis diagnosis.<sup>2,3</sup>

Lynn introduce de term Apendagitis in 1956 as a benign and self-limited entity secondary to vascular pedicle torsion or venous drainage

thrombosis, with a presentation between 4<sup>o</sup> to 5<sup>o</sup> decade of life, and only a few case reports of this pathology in child.<sup>4,5</sup>

Epiploic appendixes are between 50 to 100 fat tissue structures, pedicle, aligned in two separate rows, arranged in front of and behind the free colon tapeworm. It extends from the cecum to the recto-sigmoid union and are covered with peritoneum. The epiploic appendixes are between 0.5 to 5cm. long and 1-2cm thickness. They are more numerous and long in the sigmoid colon. They have one or two arterioles and a central vein.<sup>6-9</sup> Their anatomy and vascular pedicle predispose the torsion, with ischemia and inflammation, known as Apendagitis.<sup>3</sup>

It is called primary Apendagitis when the origin is the torsion or thrombosis of the epiploic appendix with ischemia and inflammation, generally presented in the sigmoid colon or cecum. Secondary Apendagitis is associated with inflammation of another organs like in diverticulitis, appendicitis, colecistitis, etc.<sup>3,10</sup>

Clinical signs include intense abdominal pain, localized in the same point, frequently associated with some movement like postprandial exercise. Is frequently localized in left and right iliac fossa, between 10 to 30% present a palpable mass, and sometimes is associated with fever. Nausea, throw up or diarrhea are not associated frequently but in a few cases a high leucocyte count could be present.<sup>9,11</sup> Symptoms would solve after 7 to 10days only with analgesics.<sup>12</sup>

Diagnosis is very difficult to be achieved clinically but some imaging studies could help us like ultrasound, doppler ultrasound, CT scan or MRI, with the last two being the ones with more sensibility.<sup>10-13</sup> Appendicular agenesis was first described by Morgagni in 1718 and is rarely found, in fact the incidence is about 1: 100,000 laparotomies for suspected appendicitis.<sup>14,15</sup>

Collins classify appendicular malformations in V types as follows:

- i. Type I, absence of appendix and cecum
- ii. Type II, rudimentary cecum and absence of appendix
- iii. Type III, normal cecum without appendix
- iv. Type IV, normal cecum and rudimentary appendix

Type V, giant cecum without appendix

The most frequent malformation is the type II, and in the present case there was a malformation type III.<sup>16,17</sup>

Although the technologic advances in imaging studies, the ileocecal region and appendix still being difficult to assess areas and the pre-operative diagnosis of appendicular agenesis is extremely difficult, and frequently completed during surgery.<sup>18</sup>

The correct diagnosis of Apendagitis confirmed by CT scan would allow us to apply an ambulatory management with analgesics. Surgical intervention is only recommended if there exist persistence of symptoms and clinical signs of suspected Apendagitis after medical management.<sup>19,20</sup>

## Conclusion

In patients with localized abdominal pain without another symptom, the diagnosis of apendagitis must be considered. Recognizing the previous mentioned clinical characteristics and with the help of imaging studies, the accurate diagnosis could be achieved and avoid an unnecessary surgery, with medical management with analgesics as the standard of care and excellent outcomes.

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## Conflict of interest

The author declares no conflict of interest.

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