Acute abdomen secondary to complete tubular colonic duplication

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Abstract

We report the case of a 6-month-old infant who presented with a complete duplication of the large intestine, debutting clinically with acute abdomen and severe metabolic disorders. We discuss the pathogenesis and morphology of the lesions, diagnostic difficulties and peculiarities of surgical treatment.

KEY WORDS: Acute abdomen, ascites, colon, duplication

INTRODUCTION

Colonic duplications are congenital anomalies reportedly present in 0.2% of the pediatric population.[1] Such malformations have been described all along the gastrointestinal tract, although they are rarely found in the colon (only 5%-6%). Complete tubular forms are even more unusual.[2]

Since this condition is usually clinically asymptomatic, diagnosis prior to surgery is difficult and can only be confirmed during a laparotomy.[3] According to some authors, 80% of the cases are symptomatic before the age of two, presenting with acute abdomen secondary to perforation or massive bleeding.[4,5] Moreover, others report that diagnosis in adulthood is also very difficult, because of a very non-specific clinical presentation.[6]

We report the case of a complete colonic duplication presenting with ‘ascites and acute abdomen’. The duplication began at the terminal ileum, 4-5 centimeters from the ileo-caecal valve; which showed 2 caeca and 2 appendices, as well as two complete tubular colonic formations, ending in a blind pouch located at the rectal level. One normal anus was present. Given the low incidence and unique clinical presentation of this anomaly, we hope that publishing our case will enhance diagnostic precision and contribute to improved surgical treatment.

CASE REPORT

A 6-month-old patient was admitted into the Pediatric Intensive Care Unit with an initial diagnosis of urinary septic shock. His personal history reflected the presence of posterior urethral valves and left vesicoureteral reflux (VUR), accompanied by severe hydronephrosis and ipsilateral renal atrophy. Three months prior to the septic event the patient had undergone a cystoscopic resection of the valves.

At the moment of the admission he presented with intense hypotonia, high fever, painful tympanic abdomen on palpation and a severe general state. Hemodynamic data reflected signs of severe hemodynamic instability. Blood tests showed dehydration and severe metabolic acidosis.

Over the next 18 hours the patient required vasoactive drugs to maintain hemodynamic signs. Physical examination revealed signs of acute abdomen with a large accumulation of fluid between the bowels. The abdominal x-ray did not show any conclusive findings. The abdominal ultrasound showed a large dilated bowel with no typical obstruction pattern. A precise preoperative diagnosis could not be established so an urgent laparotomy was
performed.

The laparotomy revealed a peritoneal cavity filled with serous fluid and the terminal ileum duplicating at 4-5 cm distance from the ileo-caecal valve, ending in two caeca and two appendices [Figure 1], which were continued with a total tubular colonic duplication. Both colonic tracts had a common wall [Figure 2], intimately connected to each other. The anti-mesenteric edge was hugely dilated and showed clear ischemic signs in its transversal portion, ending in a blind pouch located at the rectal ampulla level, at 3-4 cm from the normal anus. We resected the total duplicated gut, excluding the rectum, by combining conventional surgery with the use of a linear mechanical stapler to resect the most distal portion due to its difficult access.

The histopathology of the lesion revealed hemorrhagic necrosis of the resected gut. This could be related to the massive dilation and decreased blood supply of the anti-mesenteric gut.

The urine and blood cultures were negative. The final diagnosis was acute abdomen and shock. Eighteen days later, following restoration of intestinal passage, the patient had recovered acceptably and was discharged from the hospital, with antibiotic prophylaxis for VUR. He was followed up in clinic for a year, with abdominal ultrasounds and also seen by the nephrologists and urologists. At the moment, he is asymptomatic.

DISCUSSION

Colonic duplications are the most unusual type of intestinal tract duplications, and total tubular forms are even rarer. Several authors describe the high association of this type of anomalies with genitourinary or lower vertebral column, such as prostatorectal fistula, anorectal or scrotal abnormalities and myelomeningocele.[7, 8] The pathogenesis of the entity is not clear, and many theories have been postulated about it. This wide variety of explanations, no fully accepted theory or demonstrated, suggesting that the origin of intestinal duplications may be multi-factorial and that the current molecular genetics research will probably give us an acceptable explanation regarding these anomalies in the near future.[9]

In the present case, the duplication was associated with posterior urethral valves, leading to a clinical picture of left hydronephrosis and renal atrophy. This association of lesions should be included in the spectrum of previously reported associations, some of them very similar in urologic nature.[6, 7]

The most interesting aspect of this case was its clinical outcome: ascites and acute abdomen with severe hemodynamic and metabolic instability that endangered the patient's life. This type of clinical debut is rare, usually discovered as a casual finding,[3] because of its asymptomatic course or its clinical appearance in adulthood. The lack of a unique clinical picture of the entity constitutes a serious diagnostic dilemma.[3] Either way, there is no uniform pattern of clinical onset, but always seems to appear in the early stages of life.[10]

From a morphologic perspective we agree with most authors that cystic duplication at any point along the colon's anatomy is the most frequent type of anomaly,[6, 9] followed by partial tubular duplications (next to the main intestinal tract). Very few cases of complete cecoapendicular duplications, such as the one presented here, exist in the literature.[6]

With regard to proposed surgical techniques, most of the authors prefer a complete excision of the duplicated tract,[8] although some authors support the creation of a common channel, using mechanical sutures for the common wall resection to avoid the accumulation of fecal material in the duplicated gut, as was described by Yucesan.[11] In our case however, this technique could not be used since the ischemic and inflammatory component of the duplicated gut clearly required the excision of the piece.

Footnotes

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REFERENCES


**Figures and Tables**

**Figure 1**

Cecoappendiceal duplication. The arrows indicate both vermiform appendices

**Figure 2**
Complete tubular colonic duplication. The arrows indicate the two colonic tubes. The outer fragment shows ischemic signs corresponding to the portion that was removed during surgery.