

Case Report

Meckel's diverticulum: misdiagnosis and delayed presentation with ureterohydronephrosis

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ABSTRACT

MD occurs in 2% of the population and is the most common congenital malformation of the gastrointestinal tract. Most patients are asymptomatic and complications are found in about 5%. This study reports an exceptional case where a longstanding Meckel's diverticulum (MD) was initially misdiagnosed as inflammatory bowel disease and further complicated with ureterohydronephrosis. A 14-year-old male presented with abdominal pain and fever, minor elevation of inflammatory markers, the ultrasound showed a liquid collection and suggested complicated acute appendicitis. Laparoscopy showed a thickened small bowel segment, low grade inflammation of the appendix and no collection; appendectomy was performed. Follow-up ultrasound showed bowel wall thickening and collections, which further suggested inflammatory bowel disease. One month after discharge, the patient returned with abdominal pain and subfebrile temperature. Imaging studies showed pelvic plastron, purulent fluid, collections and right ureterohydronephrosis. Treatment for complicated inflammatory bowel disease was ineffective, there was worsening of the ureterohydronephrosis, however, the collections were small and persisted roughly unchanged, which suggested a different etiology. Scintigraphy with technetium-99m pertechnetate was positive and surgery was proposed. A MD was found intimately adherent to the posterior abdominal wall, involving the ureter. En bloc enterectomy and primary anastomosis were completed successfully. Follow-up ultrasound 11-days after surgery was normal. After 6-months, the patient was asymptomatic. This case represents a diagnostic challenge since ureterohydronephrosis was never described as a complication of MD and it was an important element in the diagnosis.

Keywords: Meckel's diverticulum, Meckel's diverticulitis, Meckel's complications, Ureterohydronephrosis, Pediatric surgery

INTRODUCTION

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract and is caused by incomplete obliteration of the omphalomesenteric duct.¹ MD occurs in the terminal ileum within 100 cm from the ileocecal valve and has an average length of 3 cm.²⁻⁵ It occurs in approximately 2% of the population, however, the real prevalence is hard to

determine because most patients are asymptomatic.⁶ The diagnosis is either an incidental finding or subsequent to a complication of the MD.⁴ If the patients develop symptoms/complications, they usually present in the first 10 years of life with an average age of 2.5 years.⁷ The typical presentation is painless rectal bleeding in a child younger than 2 years, since MD accounts for approximately 50% of all lower gastrointestinal bleeding episodes at that age.² Bleeding results from mucosal

ulceration caused by acid secretion from ectopic tissue in the MD, most commonly gastric.¹⁻² Most studies report intestinal obstruction as the most common complication in the pediatric age group and it is caused by intussusception, omphalomesenteric bands or adhesions, which can also be a leading point for volvulus or internal hernia.¹⁻² Mass effect by ectopic tissue, especially if pancreatic, can also be a leading point for intussusception or volvulus.⁸ Inflammation, or diverticulitis, results from luminal obstruction at the base of the MD, which can further lead to bacterial overgrowth and infection and, ultimately, to perforation and peritonitis.¹ The aim of this study is to report an exceptional presentation of MD with ureterohydronephrosis, a complication that had not been previously described.

CASE REPORT

A previously healthy 14-year-old male presented with a 3-day abdominal pain and fever, mild tenderness on the right iliac fossa (RIF), normal white blood cell count with neutrophilia, minor elevation of C-reactive protein and an ultrasound (US) showed a liquid collection (28×20×20 mm) that was interpreted as an abscess, suggestive of complicated acute appendicitis (Figure 1a).

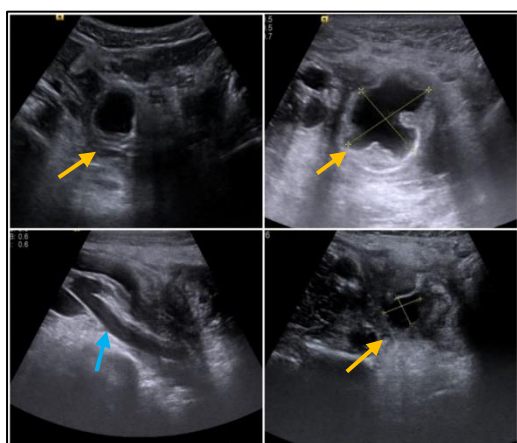


Figure 1: Serial US; (a) the first US, liquid collection of 28×20×20 mm (yellow arrow) interpreted as abscess and suggestive of complicated acute appendicitis; (b) POD 4 after laparoscopic appendectomy, showed three intrabdominal collections, one with persistent characteristics and size (28×23 mm) (yellow arrow); (c) and (d) 1 month after laparoscopic appendectomy, right ureterohydronephrosis (blue arrow), persistent liquid collection of 19×11 mm (yellow arrow) and purulent fluid (white asterisk).

Exploratory laparoscopy showed an unperforated appendix with low grade inflammation, no abscesses or collections, clear peritoneal fluid and thickened small bowel segment with marked inflammatory changes in the RIF/pelvic area; transumbilical laparoscopic-assisted appendectomy was completed without complications. Peritoneal fluid culture was negative, the postoperative period was uneventful, however, for clarification, the

patient repeated US on postoperative day (POD) 4, which showed bowel wall thickening and three intraabdominal collections (28×23 mm, 43×23 mm, 20×7 mm) (Figure 1b). There was suspicion of inflammatory bowel disease (IBD) and the patient was discharged on POD 10 with oral antibiotics and follow-up by pediatric gastroenterology.

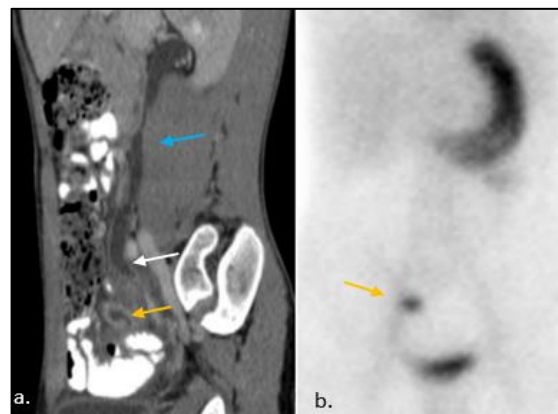


Figure 2: Further imaging; (a) computed tomography, sagittal plane, with ureterohydronephrosis (blue arrow), complete ureteric obstruction (white arrow) and liquid collection with 30×25×20 mm (yellow arrow); (b) scintigraphy with technetium-99m pertechnetate showing ectopic gastric mucosa on the RIF (yellow arrow).

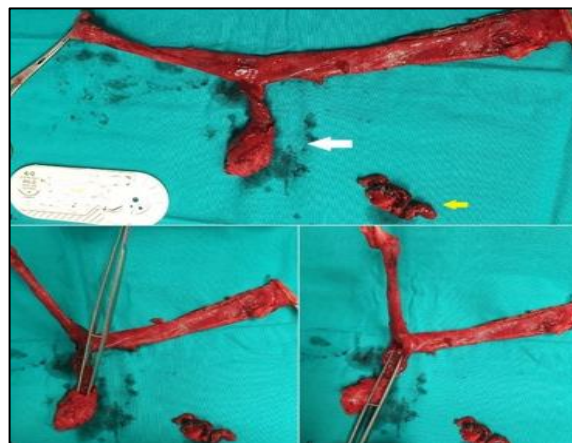


Figure 3: Enterectomy involving the MD; (a) MD (white arrow), MD's wall that was adherent to the posterior surface of the small bowel mesentery and posterior abdominal wall (yellow arrow); (b) MD distal lumen; (c) MD proximal lumen.

One month after discharge, the patient returned to the hospital with abdominal pain and subfebrile temperature. The pain was intermittent, but subsisted since discharge, and additionally, 1.5 kg of weight loss was verified. The abdominal examination revealed mild RIF tenderness. Laboratory investigations were normal. US showed an abdominal plastron in the RIF/pelvic region, hyperechoic fat, purulent fluid, a collection of 19×11 mm, and right

ureterohydronephrosis (ureter 8mm, renal pelvis 17 mm) (Figure 1c, d). The main diagnostic hypothesis was complicated IBD and the patient was put on nil per os, parenteral nutrition, intravenous antibiotics and anti-inflammatory medication. The patient had no pain nor fever, laboratory investigations were persistently normal, fecal calprotectin was normal, and the collections persisted roughly unchanged on serial US. Further imaging with computed tomography scan showed two collections, one with air-fluid level with 30×25×20 mm partially involving the ureter and conditioning ureterohydronephrosis by complete ureteric obstruction (Figure 2a), and an adjacent one with 25×20×20 mm.

The worsening of the ureterohydronephrosis caused by small sized and persistent collections mandated further investigation for a different etiology, namely a duplication cyst or a MD. On the 6th day after admission, scintigraphy with technetium-99m pertechnetate was positive for ectopic gastric mucosa (Figure 2b).

The patient was submitted to exploratory laparoscopy, where adhesions in the RIF/pelvic area were incompletely divided because of their complexity which warranted conversion to Rockey-Davis laparotomy. A 10 cm MD was found intimately adherent to the posterior surface of the small bowel mesentery and posterior abdominal wall, where the ureter crosses the bifurcation of the common iliac vessels; there were no abscesses or collections, however there were two free fecaliths. Enterectomy involving the MD (Figure 3) and manual end-to-end anastomosis were completed successfully, as well as division of intestinal adhesions, removal of fecaliths and drain placement.

Postoperative period was complicated by intermittent nausea and alimentary vomiting, but the patient maintained regular bowel movements. An upper contrast study using gastrografin on POD 6 showed gastroparesis and globally delayed contrast progression, without obstruction. Low-dose erythromycin resolved the complication and on POD 10 the patient was tolerating normal diet. US on POD 11 was normal and the patient was discharged on POD 13. Histological description was of a MD mostly lined with gastric mucosa, without dysplasia. At 6-month follow-up the patient is asymptomatic and the ultrasound remains normal.

DISCUSSION

MD is a rare condition and the clinical presentation was utterly unusual, which made the diagnosis challenging. Nonspecific presentations of MD are usually confused with more common diagnosis such as appendicitis or IBD, especially when outside the typical age of MD's presentation.⁹ In this case, MD was initially misdiagnosed as complicated acute appendicitis. Chen et al reported in a large single center retrospective study 6 patients misdiagnosed as appendicitis that were later reoperated for persistent symptoms of abdominal pain or melena.⁹ However, in this case, marked inflammatory changes in a

small bowel intestinal segment directed the diagnosis to complicated IBD, which was more compatible with the age and the weight loss. In literature, few cases are described where a MD was initially misdiagnosed as Crohn's disease because of bowel wall thickening found in imaging studies.¹⁰⁻¹² Retrospectively, we can understand that the initial presentation was probably of a contained perforation of a Meckel's diverticulitis and the thickened small bowel segment was possibly the adjacent ileum to the MD. The length and location of the MD resulted on vague abdominal signs and symptoms, without peritonitis or guarding, and minor elevation of inflammatory markers, plus the negative peritoneal fluid culture were inconsistent with perforated Meckel's diverticulitis.

Ureterohydronephrosis develops secondary to urinary tract obstruction which can be caused by intrinsic or extrinsic compression.¹³ The intrinsic causes are the most frequent, and involve problems within the kidneys and urinary tract.¹⁴ External compression can be caused by masses, usually retroperitoneal, but also pregnancy, malignancy, trauma, retroperitoneal fibrosis, among others.¹³ Also, it was described that an inflammatory mass or abscess as a complication of Chron Disease can lead to hydronephrosis by external compression.¹⁵ In this case, considering the size of the collections, it was unlikely that the ureterohydronephrosis was caused by mass effect or compression, especially when the obstruction was worsening and the collections persisted roughly unchanged. These findings warranted further clarification and final diagnosis was possible through scintigraphy with technetium-99m pertechnetate, which has a reported sensitivity of 85% and the specificity of 95%.³

With the surgical findings we concluded that the ureter was most likely involved by severe contiguous transmural inflammation of the MD and not compression. Also, there was at least one persistent liquid collection that was continuously interpreted as an abscess that was actually an inflamed segment of the MD.

CONCLUSION

This is an exceptional case where a longstanding MD was initially misdiagnosed as inflammatory bowel disease and further complicated with ureterohydronephrosis. As far as we know, ureterohydronephrosis was never described in literature as a complication of MD and it was an important element in the diagnosis. We conclude by reiterating the ever most importance of intestinal exploration for MD when the severity of appendicitis or ultrasound findings do not match the preoperative evaluation.

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