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Laparoscopic Management of Enterocutaneous Fistula as a First Presentation in Pediatric Crohn's Disease

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ABSTRACT

Background: Enterocutaneous fistula is an uncommon presentation of Crohn's disease in pediatrics and managing this complication is challenging for surgeons due to multiple factors, including the patient's age and nutritional, septic status. The timing and approach of surgical intervention are also critical in these cases.

Case Report: In our case, a 14-year-old female patient presented with umbilical feculent discharge. She was initially complaining of chronic abdominal pain, weight loss for 1 year, she underwent drainage for umbilical abscess before but the wound not healed and continued discharge. A contrasted CT scan abdomen and pelvis revealed the following findings:

- The cecum and ileocecal junction are characterized by mural thickening, a narrow lumen and surrounding fat stranding.
- An umbilical fistula connected to bowel loops with an anterior wall defect and related fatty soft tissue. These findings raised our suspicion of Crohn's disease.

After optimization the patient condition, she underwent a laparoscopic exploration and ileocecal resection and anastomosis. The pathology results confirmed our diagnosis of Crohn's disease. With the support of a multidisciplinary team for managing sepsis, correcting malnutrition and initiating steroids and biological therapy pre- and post-operatively, we successfully discharged the patient home symptom-free and gaining weight.

Conclusion: In conclusion, despite adequate conservative treatment, one-third of pediatric patients with Crohn's disease develop complications such as fistula, stricture and obstruction, which may require surgical intervention at any stage of the disease. Enterocutaneous fistula in such pediatric age group it is presented with aggressive unique mass penetrating anterior abdominal wall forming ilioumbilical fistula is not found in the literatures.

Managing an enterocutaneous fistula in a pediatric patient with Crohn's disease requires a high index of suspicion and multidisciplinary care, integrating surgical expertise for appropriate timing and approach to surgical intervention.

Keywords: Crohn's disease, Enterocutaneous fistula, Umbilical fistula

1. Introduction

Crohn's Disease (CD) is a chronic relapsing inflammatory disease that mainly affects the gastro- intestinal tract. It is thought to develop as a result of the abnormal immune reaction triggered by several environmental factors in genetically susceptible individuals¹. The incidence of CD is rapidly increasing worldwide and up to 25% of patients are diagnosed during childhood or adolescence². Pediatric onset CD tends to have a more complicated behavior (stricture or penetration) than elderly onset CD at diagnosis³.

Fistulae may develop from the intestine since CD affects the intestine transmurally. In addition to developing between intestinal areas, fistulas can also arise between the intestine and nearby organs like the skin, urologic organ or gynecologic organ. While they can occur anywhere in the intestine, intra-abdominal fistulae most commonly occur in the ileocecal region, often in conjunction with abscesses⁴. The enteroenteric fistulas might not need surgery if they don't exhibit any symptoms. Surgery is necessary, though, if they are symptomatic. Resection is the best course of action for an inflammatory colon, although primary closure can also be used to treat non-inflammatory bowel or secondary impacted adjacent organs like the bladder or vagina⁵. Surgery in CD is not aiming to cure the disease but rather to relieve symptoms and complications. Generally suggested surgery indications are listed in (Table 1)⁶.

Table 1: Operative Indications in Crohn's Disease.

List of Surgery Indications
Complex perianal fistula or abscess
Intestinal stricture or obstruction
Intra-abdominal abscess
Fistula (bowel to bowel, bowel to skin, bowel to adjacent organ)
Bowel perforation
Massive intestinal bleeding
Growth retardation
Neoplastic changes
Fulminant disease which is not responds to the medical treatment

2. Case Presentation

The 14-year-old female patient had multiple visits to a pediatric clinic for 1 year complaining of epigastric and lower abdominal pain, loss of appetite, dysphagia and weight loss. After undergoing an upper GI endoscopy and biopsy, the histopathology revealed a diagnosis of eosinophilic esophagitis. Treatment started and mild improvement was noticed. However, she was still underweight.

later on, the patient developed an umbilical abscess and underwent incision and drainage in other facility, received antibiotics and a daily dressing for discharge. A two months later, she visited the pediatric surgery clinic with a complaint of umbilical discharge, which they described as bowel content associated with maceration skin of abdominal wall around the wound (Figure 1). The patient underwent an abdominal CT scan with IV and oral contrast, which revealed an umbilical enterocutaneous fistula tract (Figure 2). Her laboratory investigations, including hemoglobin level, renal and liver function tests, electrolyte and inflammatory markers, were within the normal range. We implemented multidisciplinary management of the patient's condition, which included the use

of antibiotics, steroids, mesalazine and nutritional supplements. After optimization, the patient was booked for laparoscopic exploration of the abdomen.



Figure 1: The umbilicus of the patient showing the opening of the fistula.

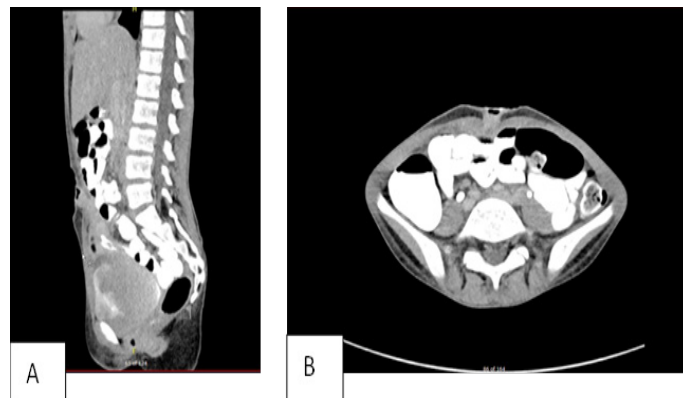


Figure 2: A) Sagittal section of CT scan abdomen showing umbilical enterocutaneous fistula. B) Transverse section of the abdomen with air reaching the umbilicus.

The intra-operative findings showed that the ileocecal was adherent to the umbilical fistula, forming a phlegmon covered by the omentum. We observed short mesentery and fat creeping, both of which are indicative of Crohn's disease. We performed an ileocolic resection until we reached the limit of healthy tissue with end-to-side anastomosis. Histopathology for the resected segment showed severely active chronic inflammation with non-caseating granulomas compatible with Crohn's disease, negative for dysplasia or malignancy (Figures 3 and 4).

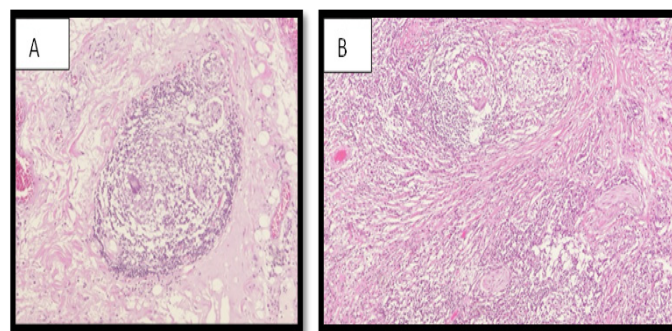


Figure 3: A) Microscopic cut showing the lymph node containing non-caseating granuloma. B) Microscopic picture of transmural inflammation

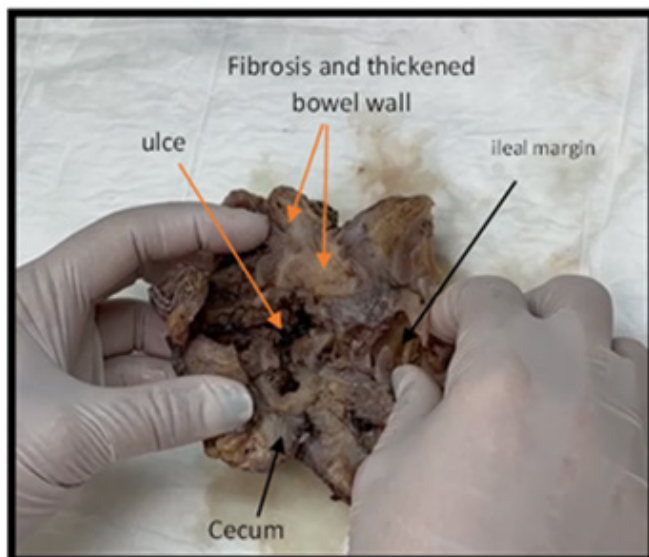


Figure 4: Macroscopic cut of the resected liocecal part.

During hospitalization for three weeks, the patient was started on medical treatment for Crohn's then gradually weaned off parenteral nutrition prior to discharge. She returned to the clinic with weight gain and no symptoms. The operation wound was healed. She was following with gastroenterology continued biological treatment Ustekinumab (Stelara).

3. Discussion

The incidence of Crohn's disease in the pediatric population in Saudi Arabia is relatively lower than suggested in the Western literature. A multicenter national study reported that the incidence of pediatric-onset Crohn's disease (CD) was approximately 0.27 per 100,000 children aged 0 to 14 years between 2003 to 2012⁷.

Crohn disease has a wide spectrum of clinical presentations and rarely can present with complications such as a bowel stricture or fistula⁸. The exploration of spontaneous umbilical fistula cases in patients with Crohn's disease reveals a compelling narrative that spans decades. With only three significant studies identified, the scarcity of literature highlights the rarity of this condition, further underscored by the fact that the most recent research dates back to 1989^{9,10}. Among these cases, the youngest documented patient was a 13-year-old female, published in 1971¹¹. This limited yet poignant body of work not only sheds light on an unusual complication of Crohn's but also invites further inquiry into the long-term implications and management strategies for such rare occurrences. The need for updated research is clear, as understanding these unique presentations could enhance care for affected individuals and contribute to a more comprehensive grasp of Crohn's disease as a whole.

Diagnostic laparoscopy was used to determine what surgical treatment was eventually required with conventional laparotomy. the aim off this study to prove that the laparoscopic approach in pediatric Crohn's complications is diagnostic and therapeutic and better cosmetic results with shorter return to normal activity and bowel function, being incidence of major complications unaffected by choosing approach¹².

In conclusion to a study done on 80 Crohn's patients that underwent ileocecal resection during childhood showed that majority of patients were satisfied or very satisfied with their ileocecal resection (81%). In agreement with thire cohort, a previous study reports that 80% of patients were satisfied with surgery and would choose to undergo the procedure again if necessary¹³.

4. Conclusion

This case highlights the challenges and successful management of an enterocutaneous fistula in a pediatric patient with Crohn's disease. It reinforces the importance of multidisciplinary care, integrating surgical expertise, detailed diagnostic evaluation and meticulous postoperative management to achieve favorable outcomes in complex pediatric cases.

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