Is Early Surgical Treatment of Crohn's Disease Harmful or Beneficial in Children?

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ABSTRACT

Objectives: Crohn's disease is an increasingly common health problem in children. If untreated or treated inappropriately, it eventually results in complications. This report presents a case of suspected Crohn's disease with inflammation of the pelvic region with a review of the literature.

Case: An 11-year-old female was suspected of having Crohn's disease, but laboratory tests and biopsy by endoscopy failed to confirm the diagnosis. She had intra-abdominal inflammation, an enteroenteric fistula, and an abscess that did not resolve despite antibiotic treatment for 1.5 months. She was referred to us for diagnosis. We performed surgery with a provisional diagnosis of Crohn's disease; the pathology was consistent with Crohn's disease.

Conclusion: Surgery is indicated for the therapy and diagnosis of Crohn's disease under certain conditions. The treatment of Crohn's disease requires a multidisciplinary team.

Key words: children, crohn's disease, surgery

ÇOCUKLARDAKİ CROHN HASTALIĞINDA ERKEN CERRAHİ TEDAVİ YARARLI MI YOKSA ZARALI MI?

ÖZET

Amaç: Crohn hastalığı, çocuklarda giderek yaygınlaşan bir sağlık sorunudur. Bu hastalar uygun tedavi veya tedavi edilmediği takdirde komplikasyonlara sebep olabilmektedir. Bu çalışmada Crohn hastalığında şüphelenilen ve pelvik bölgede inflamasyon ile başvuran olgu literatür ile değerlendirilmiştir.

Olgu: Crohn hastalığı şüphesi olan 11 yaşında kız hasta fakat yapılan laboratuvar testlerinde ve endoskopi ile alınan biyopsi ile tanı konulamamıştır. Bir buçuk ay antibiyotik kullanmasına rağmen karın içi inflamasyonu, abse ve enteroenterik fistülü devam etmiştir. Tanı için bize refere edilen hasta, Crohn hastalığı ön tanısıyla ameliyata alındı. Hastanın ameliyattaki makroskobik bulgular ve patoloji Crohn hastalığı ile uyumluydu.

Sonuç: Cerrahi bazı koşullarda Crohn hastalığının tedavisi ve teşhisi için endikedir. Crohn hastalığının tedavisi multidsipliner bir ekip gerektirir.

Anahtar sözcükler: çocuk, crohn hastalığı, cerrahi

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Crohn's disease (CD) is an idiopathic, granulomatous, life-long, chronic relapsing inflammatory bowel disease (IBD) (1-4). CD can affect any segment of the digestive tract from the mouth to the anus, and inflammation may extend throughout the intestinal wall (1) CD is an important pediatric condition because 20–30% of patients with CD present during childhood (1,5). The worldwide incidence of pediatric CD is 2.5–11.4 per 100,000, (6) and is increasing worldwide (1,5). The rates are higher in Europe and North America than in the rest of the world (2).

Diagnosing CD can be challenging. Various biomarkers can aid in the diagnosis (2). The cumulative incidence of fistulas in patients with CD is 17–50%, (7) although enteroenteric fistulas are rarely the initial presentation of CD in children. These are related to chronic transmural inflammatory disease with segmental lesions (8).

The primary aim of treatment is symptom control, with prolonged remission and consequent improvement in the long-term (2). CD has a variable course, although the majority of patients develop complications requiring surgery (9,10). Surgery is not curative in CD, but surgical resection is performed for emergencies or failure to respond to medical treatment.

A delay in surgical intervention can lead to septic and infectious complications in the setting of intestinal perforation, enteroenteric fistula, and abscess. Therefore, we present a patient who was not diagnosed at endoscopy before requiring surgical intervention for a septic enteroenteric fistula and review the literature.

Case

An 11-year-old female with suspected inflammatory bowel disease was referred to us by a pediatric gastroenterologist, after failing to make a diagnosis.

One year earlier, she had lost 5-kg weight in a 6-month period. A pediatrician recommended a diet and alimentary support. She subsequently gained weight, but then lost it again. She developed abdominal pain and was referred to a pediatric nephrologist and then a gastroenterologist for weight loss, abdominal pain, and anemia. The personal and familial histories were non-contributory. Ultrasonography (USG) at that time showed grade 2 hydronephrosis. Laboratory tests showed anemia, leukocytosis with toxic granulation, an elevated C-reactive protein level (CRP; 70 mg/L), and erythrocyte sedimentation rate (ESR; 104 mm/h). Urinalysis showed leucocytes and pyuria. Computed tomography (CT) and magnetic resonance imaging (MRI) showed small calcifications in the right suprarenal area, grade 2 hydronephrosis of the right kidney, thickening of the walls of the distal segment and terminal ileum, high vascularity in the pericolonic area, thickening of the sigmoid colon mesentery lymphadenopathy, and a suspected abscess measuring 32×25 mm in the presacral region. Upper endoscopy was normal. A colonic stricture prevented passage of a colonoscope (Figure 1). There was a necrotic region before the stricture. A biopsy was not diagnostic. Antibiotic therapy

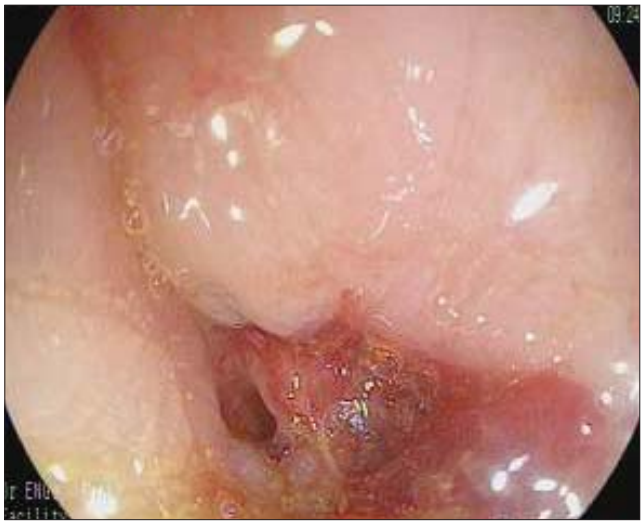

Figure 1. A colonic stricture prevented passage of a colonoscope

Figure 2. Colonography shows luminal narrowing and a fistula

with meropenem and vancomycin was started. The CRP remained around 50 mg/L and the presacral mass and fistula remained. Colonography showed a fistula and stricture (Figure 2).

The inflammation did not resolve despite antibiotic treatment for 1.5 months. As no diagnosis had been made, treatment for IBD was not given. At this time, the patient was referred to us by the gastroenterologist.

On physical examination, the abdomen was relaxed and no tenderness or rebound was detected.

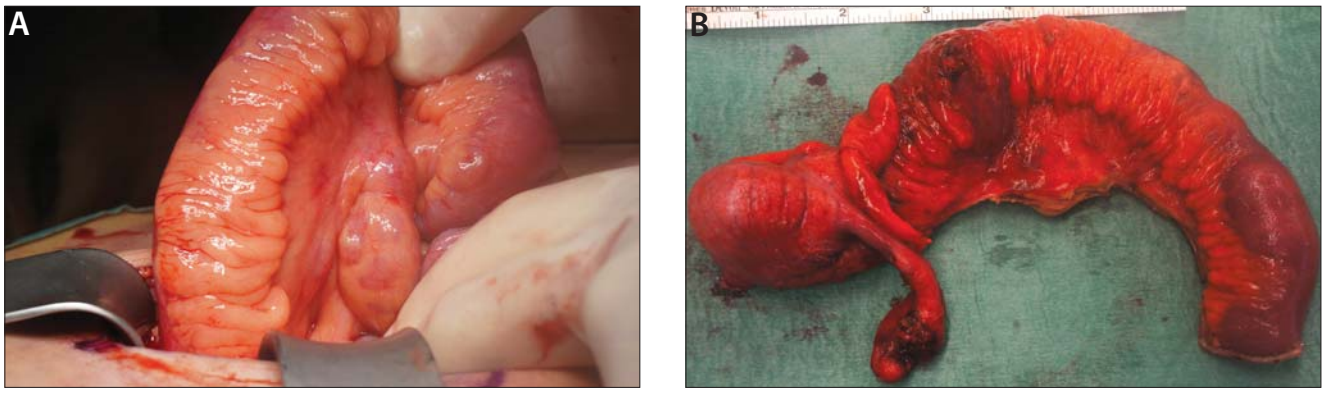

Figure 3. Thickening of the mesenteric margin and fat wrapping on the ileal wall **(A)**. There was a self-limiting fistula and adhesion in the descending colon **(B)**.

The patient underwent surgery with a provisional diagnosis of enteroenteric fistula and abscess related to IBD. At laparotomy, the terminal ileum was inflamed and edematous, with mesenteric lymphadenopathy and adhesions with the sacrum. The involved ileal segment showed thickening of the mesenteric margin and fat wrapping on the ileal wall. There was a self-limiting fistula and adhesion in the descending colon (Figure 3 A-B). She underwent resection of the terminal ileum and cecum, repair of the fistula of the colon, and an ileostomy. The patient was discharged on postoperative day 3 without any problems. The pathology was consistent with Crohn's disease. She underwent medical treatment for CD. Two months later, the ileostomy was closed. She is being followed-up on by a gastroenterologist and is receiving treatment for CD.

Discussion

Crohn's disease was first described about 100 years ago (4,11). However, the etiology and cause are unknown and there is no cure. CD is strongly influenced by environmental changes (5). However, little is known of the interactions among various environmental factors, the gut microbiota, and the host mucosal immune system (4) Our patient had no family history of CD and she was from a low socioeconomic level.

The symptoms depend on the location and extent of disease involvement (1). The terminal ileum is the most common location in children, and children younger than 5 years have a higher rate of colonic involvement (1). CD may be diagnosed at any age, most frequently in the second decade of life. Children diagnosed with CD before the age of 6 years had lower rates of surgery than those diagnosed at >10 years of age (12). Children can have atypical symptoms and infection increases the mortality rate in patients with CD (3). Our patient initially needed surgery for CD involving both the ileum and colon.

The diagnosis of CD can be challenging in some cases, (13) and the most important initial step in the diagnostic pathway is to consider the possibility of CD (14). Abdominal imaging such as USG, MRI, and CT enterography are useful for making a diagnosis, evaluating disease activity, and identifying complications (7). Numerous serological markers of CD have been identified, including anti-*OmpC* IgG, anti-IL-12, anti-*Saccharomyces cerevisiae* antibodies (ASCA), *perinuclear anti-neutrophil cytoplasmic antibodies* (p-ANCA), ESR, and CRP. Genetic markers and endoscopy are also useful. Usually, CD is diagnosed from the combined clinical, endoscopic, laboratory, radiological, pathological, and operative findings (2). Nevertheless, our patient could not be diagnosed until surgery.

It can be difficult to differentiate CD from acute infectious enterocolitis, ulcerative colitis (UC), intestinal tuberculosis, or intestinal Behçet's disease. If the treatment for CD is delayed due to a late diagnosis, the rate of complications can increase (2). As our patient could not be diagnosed, she was initially admitted with a complication of CD.

A family history is a strong risk factor for developing IBD (2). Our patient lacked a family history of IBD. The complication most often associated with CD in childhood is poor weight gain (1). Our patient initially sought medical advice for weight loss, but was not diagnosed until surgical intervention was required.

The treatment goals in children are to achieve clinical, endoscopic, and biological remission of active disease, to promote growth and adequate nutrition, to improve the physical and social quality of life, and to decrease the need for surgical intervention (1,4). However, the majority of patients are treated conservatively, which increases the number requiring elective or urgent surgery (1,5). In childhood, 15–20% of the patients with CD require surgical

intervention within 3 years of diagnosis because of severe illness (1). We believe that surgical intervention can lead to clinical improvement, decreased medication requirements, and improved septic condition. Targeting early CD and achieving deep remission might be the best method of altering the disease course and increasing the quality of life of the patient (5).

Medical therapy is usually effective in terms of inducing the remission of symptoms, but is sometimes ineffective and has many risks, including bone loss, myelosuppression, and lymphoma (1). Surgical resection is almost inevitable in the course of Crohn's disease and the rate of surgery increases with time (10). Most patients eventually develop complications related to strictures or perforation (16).Complications, rather than medically intractable disease, are now the main reason for surgical treatment (4). The most common indications for surgery in CD are the appearance of intestinal complications and the failure of medical treatment (17). Intestinal resection at diagnosis is associated with a reduced need for surgery during the follow-up period (17).

We hypothesized that patients with suspected CD with a fistula and high levels of markers of infection would benefit from surgery resulting in a diagnosis and reducing the number of medications. Our patient had an enteroenteric fistula. Delayed surgical intervention might increase the rates of septic and infectious complications. Patients who underwent surgery before developing complications—such as perforation, fistula, or abscess—had lower complication rates (1). A delay in surgery might result in increased septic and infective complications. A previous study hypothesized that patients undergoing surgical intervention had lower complication and recurrence rates compared with those treated medically or with percutaneous drainage (1). The resection of inflamed bowel in children with CD is not a cure, but resection of affected isolated segments is considered an integral part of the therapeutic armamentarium (1). The most common indications for surgical interventions are stricture, abscess, enteroenteric fistula, obstruction, and perforation. Our patient had an enteroenteric fistula.

In conclusion, in selected cases of CD, early surgical intervention might be useful to avoid the later loss of long bowel segments. Early operative intervention in a patient with a complication of CD might be the best method of altering the disease course and improving the quality of life of the patient.

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