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Case Report OMICS International

Fulminant Pseudomembranous Colitis and Abdominal Compartment Syndrome

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Abstract

Patients with cystic fibrosis have higher rates of *Clostridium difficile* colonization than the general population. However, complicated forms of presentation are rare. We present a case of pseudomembranous fulminant pancolitis with abdominal compartment syndrome in a 9-year-old boy with cystic fibrosis.

Conclusion: The incidence of *Clostridium difficile* infection is higher in patients with cystic fibrosis. Although it is rare, fulminant colitis can be associated with abdominal compartment syndrome. This is the only reported case of fulminant *Clostridium difficile* colitis associated with abdominal compartment syndrome in children who survived without total colectomy.

Keywords: Abdominal compartment syndrome; *Clostridium difficile*; Cystic fibrosis; Pseudomembranous colitis

Abbreviations

ACS: Abdominal Compartment Syndrome; *C. difficile*: *Clostridium difficile*; CDI: Infection with *Clostridium difficile*; CT: Computed Tomography; CF: Cystic Fibrosis

Introduction

The clinical spectrum of infections with *Clostridium difficile* (*C. difficile*) ranges from asymptomatic carriers, mild forms with self-limiting diarrhea and unspecific abdominal pain, to complicated forms such as fulminant colitis, toxic megacolon or intestinal perforation, which can even cause death [1]. Patients with Cystic Fibrosis (CF) have higher rates of *C. difficile* colonization than the general population. However, complicated forms of presentation are rare [2]. Infection with *Clostridium difficile* (CDI) is a known risk factor for Abdominal Compartment Syndrome (ACS) [3], which is defined as intraabdominal pressure over 20 cm H_2O associated to secondary organ dysfunction. We present a case of pseudomembranous fulminant pancolitis with ACS in a 9-year-old boy with CF.

Clinical Case

The patient is a 9-year-old boy who was admitted as an emergency with fever and odynophagia accompanied by hyporexia and slight abdominal pain of 2 months of evolution.

In his personal history, he had been diagnosed with CF with the genotype G542X/G542X at birth when he presented with meconium ileus. He showed a mainly digestive involvement with steatorrhea, refractory malabsorption and hepatic steatosis. In the respiratory level, he showed bilateral diffuse bronchiectasis with repeated processes of colonization by *P. aeruginosa* and *S. aureus*. The last acute respiratory episode took place 15 days before admission and it was treated with oral amoxicillin and clavulanic acid. He also presented with distal polyarthritis.

Hemogram	Biochemistry	Coagulation	Blood gases
Leukocytos 17270/µL	Amylasa: 261 U/L	PT 51%	pH 7.3
Neutrophilos 12380/µL	GOT 38U/l, GPT 59U/l	APTT 31 s	pCO2 40
PCR 10.38 mg/dl	Bilirubin 0.68 mg/dL	DD 3 µg/ml	pO2 23
PCT 91 ng/ml	GGT 71U/l	Fibrinogen 315 mg/dl	bicarbonate 18.4
	LDH 437U/l		BE -6.7
	Natremia: 122 mmol/L		lactate 3.8

Table 1: Analysis values.

The patient has a treatment of pancreatic enzymes, fat-soluble vitamins, omeprazole, ursodeoxycholic acid, taurine, medium-chain triglycerides, hyperproteic and hypercaloric diet and respiratory physiotherapy.

In the physical examination, the patient revealed HR 153 bpm, AP 91/58 mmHg (p5) and transcutaneous oxygen saturation 95%. His general condition was slightly affected. The nutritional status was deficient. Mucus was sticky. He showed polypnea with 40 breaths per minute, with no more distress signs. In the cardiorespiratory auscultation, he showed fine crackles mainly at the bases of lungs. The abdomen was distended without masses and a hepatomegaly of 4 cm was found. All other parameters were normal.

The analysis revealed leukocytosis with neutrophilia and increased levels of acute-phase reactants, hyponatremia, alteration of the hepatic profile, coagulopathy, and metabolic acidosis (Table 1).

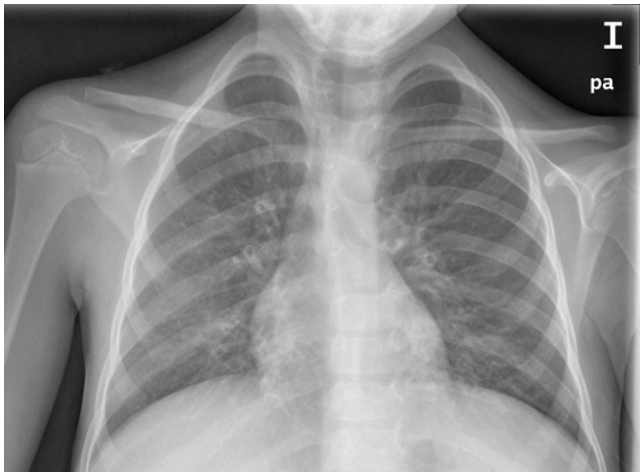

Figure 1: bronchiectasis.

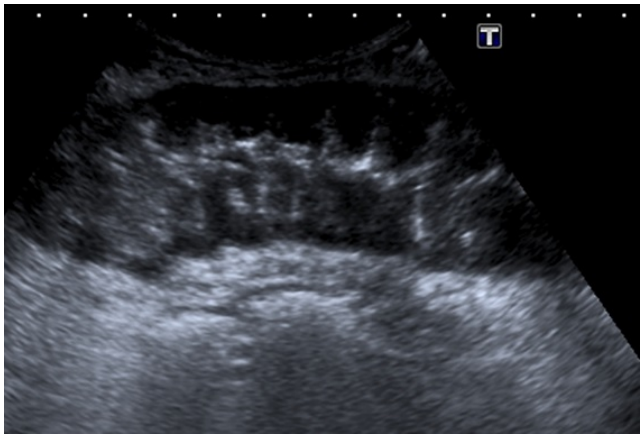

Figure 2: Abdominal ultrasound showed a marked thickening, edema and inflammation of the loops of the small and large intestines, with finger-like impressions.

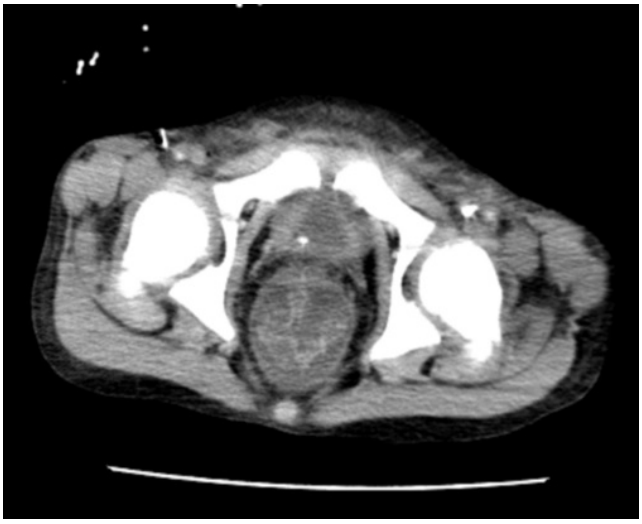

Figure 3: Thickening, edema and inflammation of the loops of the small and large intestines, with finger-like impressions.

The thoracic X-ray (Figure 1) revealed bronchiectasis and the abdominal ultrasound (Figure 2) showed a marked thickening, edema and inflammation of the loops of the small and large intestines, with finger-like impressions. The findings were confirmed with an abdominal CT scan (Figure 3).

The patient clinical condition worsened and evolved towards multiple organ failure with abdominal distension, fecaloid vomiting, diarrhea, oliguria, decreased level of consciousness, hypotension, tachycardia (AT 68/34 mmHg, HR 140 bpm), and ejection fraction which decreased to 50% in the echocardiography. Normal saline solution was used as a volume expander in doses up to 110 ml/kg over the first hours. The patient later required a perfusion of dopamine (20 µg/kg/min), noradrenaline (1 µg/kg/min), adrenaline (1 µg/kg/min), hydrocortisone (50 mg/m^2 for 48 hours) and terlipressin (0.025 mg/kg/dose). He also required endotracheal intubation and continuous veno-venous hemofiltration.

The symptoms and images were compatible with pseudomembranous pancolitis, and antibiotic therapy was started with oral metronidazole 30 mg/kg, intravenous amikacin 20 mg/kg, and intravenous ceftazidime 150 mg/kg. A test for *C. difficile* toxin was requested, and the results were positive. The patient showed partial response and the treatment was switched to rectal vancomycin enemas, oral vancomycin (60 mg/kg/day) and intravenous meropenem (15 mg/kg/day), with maintained intravenous amikacin 20 mg/kg/day.

The patient presented with abdominal compartment syndrome and intraabdominal pressure over 25 cm H_2O, with hemodynamic and respiratory compromise. In view of these facts, a decompressive laparotomy was performed and a Bogota bag was placed.

After surgery, the patient showed profuse rectal bleeding which was treated with cooled serum enemas and red blood cell transfusion. This led to hemodynamic improvement, a reduction of inotropic agents and surgical closure after 15 days.

Discussion

CF is an autosomal recessive genetic disorder caused by mutations in the gene that encodes the protein which regulates the management of ions in the membranes (CFTR), with an incidence in our environment of 1 per 3000 living newborns [4]. Around 1600 mutations have been identified, and the most common one is the 508del, which accounts for 75% of the cases. The G542X mutation, which could be observed in our case, appears in 1-2% of the global population, although an increased incidence has been observed in the Spanish population, in which it reaches 7.7% [5].

Patients with CF show higher rates of *C. difficile* colonization than the general population; around 32-50%, compared with 2% in the general adult population [6]. There are certain risk factors that predispose patients to colonization, such as immunosuppression, inflammatory bowel disease and a history of abdominal surgery. However, the main risk factor is the frequent exposure to antibiotics, used for the control, eradication and prophylaxis of respiratory diseases, as in our case [7].

The diagnosis of infection with *Clostridium difficile* needs to be suspected in patients with CF, even in the absence of diarrhea [6]. When the suspicion is considered, different tests for the detection of the toxin in the feces through immune-enzyme analysis can be performed, although false negatives are reported in up to 32% of the cases [7]. Imaging tests (radiography, ultrasound or abdominal CT scan) reveal a thickening of the colon wall with typical finger-like projections, due to mucous edema. This finding is highly suggestive, although it is not diagnostic [6]. Our patient showed an ultrasound which suggested the possibility of pseudomembranous colitis, and an early antibiotic treatment was prescribed with oral metronidazole and intravenous ceftazidime and amikacin. Afterwards, the CT scan images and the positive result for fecal *C. difficile* confirmed these suspicions.

The incubation period of CDI is 1-10 days, although the infection may appear up to 8 weeks after the use of antibiotics [6]. Generally, cases of colitis develop with diarrhea with blood, pain and abdominal distension. In 1-3% of the cases, it can be accompanied by general symptoms such as a fever, nausea and malaise [8]. Complicated forms such as fulminant colitis, toxic megacolon or intestinal perforation, which may cause death, are less common [6].

Our patient presented with fulminant colitis complicated with ACS, which has a fatal prognosis, because the mortality rates for fulminant colitis are 30-90% [9], and for ACS they are up to 60% [10].

ACS is defined as intraabdominal pressure over 20 cm H_2O associated to secondary organ dysfunction (in brain, lungs, heart, kidney and spleen) [11]. The higher the degree of intraabdominal hypertension, the more severe the systemic dysfunction will be. It is important to carry out an early diagnosis for an early surgical decompression [10]. CDI is a known risk factor for ACS.

The treatment of choice in most cases of colitis with *Clostridium difficile* in children is oral metronidazole, although it is usually associated to oral vancomycin in severe forms, because recent studies in adults have shown superior results of oral vancomycin in these cases [2].There are other new treatments, such as fidaxomicin and intestinal microbiota transplantation, with promising results, especially in recurrent forms of presentation [12]. The duration of treatment is 10-14 days [2]. An improvement of symptoms usually appears in the first 48-72 hours. A surgical intervention is recommended in absence of improvement or in cases of peritonitis, worsening of CT images or multiple organ failure with shock refractory to vasoconstrictors [13]. According to the latest systematic reviews, the treatment of choice in severe forms of infection with *C. difficile* is early total colectomy with ileostomy. However, this approach shows high mortality rates of around 30-80% of cases [14-16]. There are other, less invasive surgical alternatives, but they are only recommended in selected patients in the early stages of the disease [16]. Our patient survived after decompressive surgery and the placement of a Bogota bag with deferred surgical closure, without the need for a colon resection. Schiowith et al. [17] describe a case of an adult with toxic megacolon associated to ACS, secondary to *C. difficile* infection, who also survived without surgical resection [17]. Shaikh et al. describe another case in adults with colitis caused by *C. difficile* associated to ACS which did not require colectomy. In the pediatric age, we have not found in the literature any case of fulminant colitis associated to ACS which showed a favorable evolution without the need of intestinal resection.

Conclusions

Patients with cystic fibrosis show higher rates of *C. difficile* colonization than the general population. Due to increased risk of *C. difficile* infection in CF patients, it is important to suspect pseudomembranous colitis even without diarrhea, like in our case. Although *C. difficile* infection represents a risk factor which has already been described for ACS, the association of fulminant colitis with ACS is rare, and even more so in the pediatric age.

Despite the torpid evolution with fulminant colitis, compartment syndrome and very high surgery risk, this study highlights the successful resolution of this case. To our knowledge, this is the only reported case of fulminant colitis caused by *C. difficile* associated to abdominal compartment syndrome in the pediatric age group who has survived without the need of a total colon resection.

Compliance with Ethical Standards

Funding: This study was not funded.

This article does not contain any studies with animals performed by any of the authors.

Informed consent was obtained from all individual participants included in the study.

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