

Unusual Presentation of Shigellosis: Acute Perforated Appendicitis and Peritonitis

Şigelozisin Nadir Bir Prezantasyonu: Akut Perfore Apendisit ve Peritonit

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Abstract

Shigella spp. is one of the most common agents that cause bacterial diarrhea and dysentery in developing countries. Clinical presentation of shigellosis may vary over a wide spectrum from mild diarrhea to severe dysentery. We report the case of 5.5-year-old previously healthy boy, who presented to our clinic with abdominal pain, vomiting, and constipation. On examination, we noticed abdominal tenderness with guarding at the right lower quadrant. With the diagnosis of acute appendicitis, open appendectomy was performed. Exploration of the abdominal cavity revealed perforated appendicitis and generalized peritonitis. *Shigella sonnei* was isolated from the peritoneal fluid culture. The patient completely recovered without any complications. Surgical complications, including appendicitis, could have developed during shigellosis. There are few reported cases of perforated appendicitis associated with *Shigella*. Prompt surgical intervention can be beneficial to prevent morbidity and mortality if it is performed early in the course of the disease. (*J Pediatr Inf* 2015; 9: 45-8)

Keywords: *Shigella* spp., acute appendicitis, peritonitis, surgical complication

Özet

Shigella spp. gelişmekte olan ülkelerde bakteriyel ishal ve dizanterinin en sık etkenidir. Şigelozisin klinik bulguları hafif ishalden ciddi dizanteriye kadar değişen geniş bir klinik yelpaze göstermektedir. Beş buçuk yaşında daha önce sağlıklı olan erkek hasta karın ağrısı, kusma ve kabızlık ile başvurmuştu. Fizik muayenede sağ alt kadranda hassasiyet saptandı. Hastaya akut apendisit tanısı ile açık apendektomi işlemi yapıldı. Abdominal kavitenin cerrahi olarak açılması ile perfore apendisit ve yaygın peritonit tespit edildi. Peritoneal sıvı kültüründe *Shigella sonnei* üredi. Hasta komplikasyon gelişmeksizin tam olarak iyileşti. Şigelozisin seyri sırasında apendisitte dahil olmak üzere cerrahi komplikasyonlar gelişebilmektedir. Literatürde *Shigella* ilişkili perfore apendisit olgusu oldukça az sayıda bildirilmiştir. Erken cerrahi girişim morbidite ve mortaliteyi önlemede yararlıdır.

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Anahtar kelimeler: *Shigella* spp., akut apendisit, peritonit, cerrahi komplikasyon

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Introduction

Shigella spp., a group of Gram-negative, small, non-motile, non-spore forming, and rod-shaped bacteria are the causative agents of shigellosis (or bacillary dysentery). *Shigella* spp. are classified into four species: group A (*Shigella dysenteriae*), group B (*Shigella flexneri*), group C (*Shigella boydii*), and group D (*S. sonnei*). Among the *Shigella* spp., *S. flexneri* is the most

common species in developing countries, whereas *S. sonnei* is most common in industrial countries. In Turkey, before 1987, the most commonly isolated *Shigella* species was *S. flexneri*; however, following this year, *S. sonnei* has been more commonly encountered. *Shigella* spp. is one of the most common agents causing bacterial diarrhea and dysentery in developing countries (1). The clinical presentations of shigellosis range from asymptomatic infection to severe

dysentery. Shigellosis is associated with several intestinal and extra-intestinal complications. There are few reported cases of perforated appendicitis complicated with peritonitis due to *Shigella* spp. (2, 3). We report a case of perforated appendicitis and severe peritonitis that is related to *S. sonnei* infection.

Case Report

A 5.5-year-old boy, who was reported to be previously healthy, was referred to our hospital due to complaints of abdominal pain, vomiting, and constipation for 3 days. His body temperature was 38.5°C; blood pressure, 100/50 mmHg; and heart rate, 124/min. Physical examination revealed abdominal tenderness with guarding at the right lower quadrant. Rebound tenderness was also noticed. The rest of clinical examination was unremarkable. Laboratory studies revealed white cell count was $20.3 \times 10^3/\mu\text{L}$, hemoglobin, 11.8 g/dL; platelets, $436 \times 10^3/\mu\text{L}$; and C-reactive protein, 114 mg/L. Serum electrolyte levels and renal and liver function tests were within the respective normal ranges. Urinalysis was normal. Abdominal ultrasonography revealed a tubular structure, compatible with plastron appendicitis, with a diameter of 30×20 mm. Open appendectomy was performed after the diagnosis of acute appendicitis was established. Exploration of the abdominal cavity revealed a large amount of purulent fluid in the abdominal and pelvic cavity. The appendix was perforated and surrounded by omentum. It was then removed and the purulent collection of fluid was drained and copiously irrigated with normal saline. Intravenous antibiotic therapy with ampicillin, ampicillin, and clindamycin were initiated. The patient resumed full oral intake in 48 hr; this was followed by a rapid recovery. On the second postoperative day, *S. sonnei* was isolated from the peritoneal fluid culture. Antibiotic sensitivity testing revealed sensitivity to ampicillin, amoxicillin-clavulanate, ampicillin-sulbactam, cefazolin, cefuroxime-axetil, gentamicin, amikacin, tobramycin, ciprofloxacin, imipenem, piperacillin-tazobactam and resistance to cefalotin and trimethoprim-sulfamethoxazole. Antibiotic therapy was not changed and the blood culture result was negative. Stool was not cultured because the patient had not had diarrhea. Antibiotic therapy was discontinued, and he was discharged from the hospital on sixth day after the operation. Histological investigation of the appendix revealed acute perforated appendicitis and peritonitis.

Discussion

Worldwide, shigellosis causes an estimated 160 million cases of infection and >1 million deaths annually. Most deaths occur in children <5 years of age (4).

Symptoms of shigellosis include high fever, generalized toxicity, anorexia, nausea, crampy abdominal pain, and diarrhea. It is known that one of Hippocrates' aphorisms pointed out the severity of a certain type of dysentery in the pediatric population, which might be a guide towards a potential diagnosis of shigellosis (5). Most patients recover without complications within seven days; however, intestinal and extra-intestinal complications could occur during the course of the infection (1, 6).

Toxic megacolon, colonic perforation, appendicitis with or without perforation, intra-abdominal abscesses, and intestinal obstruction were the reported surgical complications that are related to *Shigella* infection (2, 7). In one series of 173 autopsied cases of shigellosis, perforation was found post-mortem in three cases. Perforation has been most commonly reported in neonates or severely malnourished children and may occur with either *S. dysenteriae* type 1 or *S. flexneri* infection (7). All the reported cases of *Shigella* peritonitis have been secondary to appendicitis, colonic obstruction, or transmural colitis leading to colonic perforation (2, 3). In our patient, *Shigella* peritonitis developed secondary to perforated appendicitis.

Cultures of inflamed or gangrenous appendices typically yield 10-14 different organisms, which generally reflect colonic microflora (8). *Escherichia coli*, *Bacteroides fragilis*, *Klebsiella* spp., and *Proteus* spp. were the most common pathogens isolated from these cultured appendices. *Pseudomonas* species have been reported to be a major pathogen in gangrenous or perforated appendicitis. *Yersinia enterocolitica* and *Y. pseudotuberculosis*, *Campylobacter*, and nontyphoidal *Salmonella* might be the organisms causing acute appendicitis (9). In 1961, White et al. (10) examined appendectomy materials of 160 pediatric cases of acute appendicitis: *S. sonnei* was detected 12 (7.5%) of them who had diarrhea before or after operation. In 1974, Leigh et al. (11) examined 153 appendices of adult and pediatric patients, *Shigella* spp. were not in any of them. In literature, there are few reports of gangrenous appendicitis and localized peritonitis, associated with *Shigella* spp. gastroenteritis in pediatric and adult patients (2, 3, 12-16). Interestingly, our patient did not have diarrhea. For this reason, the diagnosis of shigellosis was made after the result of the peritoneal fluid culture was obtained. *Shigella* spp. are known to invade the mucosa of the colon. The non-motile bacteria travel from one colonic epithelial cell to another through the cytoplasm, by a unique mechanism called F-actin polymerization. *Shigella* spp. spread laterally to infect and kill the adjacent epithelial cells; in addition, the bacteria spread vertically and reach the lamina propria of the colonic mucosa (17, 18). It is hypothesized that by the same mechanism the bacteria can enter the blood stream and/or travel across the colonic wall to reach the outer perito-

neal surface of the colon (17). *Shigella* spp. may cause perforated appendicitis using this mechanism without causing diarrhea or colitis.

Since 2000, only one *Shigella* spp.-related appendicitis case has been reported, in literature (19). *Shigella* spp. are among the specific infectious agents causing acute appendicitis, however, they are only rarely identified as causative agents. Furthermore, there is a reduction in the incidence of *Shigella* spp. globally. It's reported that the incidence has been slowly diminishing for the past three decades, from 6.4 per 100,000 per year between 1968 and 1988 to 5.6 per 100,000 per year between 1989 and 2002; an all-time low of 3.5 per 100,000 was reported in 2006 (20).

Shigella spp. were generally cultured from diarrheal stool in most reported cases with appendicitis with/without peritonitis. Therefore, the authors could not determine whether *Shigella* spp. were the cause of appendicitis/peritonitis or if it was coincidental (3, 12, 15). *S. sonnei* was isolated from the peritoneal exudates of the present case, who did not have diarrhea or other symptoms of shigellosis. In addition, only *S. sonnei* was isolated, without any concomitant microorganism. Due to the sole isolation of *S. sonnei*, in the absence of any other concomitant microorganisms, we suggested that the causal relationship between *S. sonnei* infection and appendicitis is clear; the clinical presentation of colitis can mimic acute appendicitis. On the other hand, bacillary dysentery-like disease may be one of the presentation signs of acute appendicitis. In most reported cases of *Shigella* appendicitis, an indefinite diagnosis of *Shigella* colitis delayed surgical management. It is a well known fact that antibiotic treatment alone cannot always prevent perforation, particularly in malnourished children, who are presumed to have thin intestinal walls (1, 6, 15, 16). Surgical complications caused by *Shigella* spp. may be fatal. Over past 40 years, the authors have reviewed the surgical complications of shigellosis in children. Since 1961, fifty-six infants and children with *Shigella* appendicitis and its complications have been included in this review. Among these patients, 13 were reported to have died (2). Ten of them had intestinal obstruction that was not operated on; two newborn infants with intestinal perforation had not been operated on either. Only one of these patients had been operated on for colonic perforation (2).

In conclusion, *Shigella* spp. may cause perforated appendicitis and peritonitis, even in the absence of diarrhea. Although the development of appendicitis in the course of shigellosis is rare, pediatricians and pediatric surgeons should be alerted to the risk of surgical complications of shigellosis, because of the significant morbidity and mortality associated with a delayed diagnosis.

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