

Case Report

Shigellosis with Acute Appendicitis and Peritonitis: A Case Report

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Abstract: Shigellosis is a form of bacterial diarrhea caused by gram-negative bacteria *Shigella* species. It is common in developing countries and results from contaminated food, poor sanitation conditions, or direct person to person contact. *Shigella* can cause infection in all age groups. High-risk group include very young, elderly, and immunocompromised person. *Shigella* species is relatively resistant to acid in the stomach, and few organisms are required to cause the disease. Once ingested, it multiplies in the small intestine and enters the colon. In the colon, it produces shigella enterotoxins and serotype toxin 1, resulting in watery or bloody diarrhea. Clinical presentation of shigellosis may vary over a wide spectrum from mild diarrhea to severe dysentery. We report the case of 7 years old previously healthy boy, who presented to our hospital 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.

Keywords: *Shigella Sonnei*, Acute Appendicitis, Peritonitis, Surgical Complication

1. Introduction

Shigella is a genus of gram-negative bacilli that causes human gastrointestinal infections, sometimes with extraintestinal manifestations [1]. Four species (serogroups) are defined on the basis of serologic or biochemical reactions, namely, *Shigella dysenteriae*, serogroup A; *Shigella flexneri*, serogroup B; *Shigella boydii*, serogroup C; and *Shigella sonnei*, serogroup D [1, 2].

Species classification has important therapeutic implications because the species differ in both geographic distribution and antimicrobial susceptibility [1,

3-5]. In developed countries, *S. sonnei* is the most common species; reports from several locations show an increase in its relative prevalence in the last several years [2, 4, 6-8]. The serogroups are further classified into at least 37 serotypes and 13 subserotypes. [1, 2]

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.

2. Case Report

A 7-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 $16.26 \times 10^3/\mu\text{L}$, hemoglobin, 11.8 g/dL; platelets, $344 \times 10^3/\mu\text{L}$. 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 acute appendicitis.

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 amoxicillin-clavulanat, gentamicin, 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, and confirmed by Matrix Assisted Laser Desorption Ionization - Time of Flight (MALDI-TOF). Antibiotic sensitivity testing revealed sensitivity to amoxicillin-clavulanat, cefotaxim, aztreonam, gentamicin, amikacin, tobramycin, ciprofloxacin, imipenem, piperacillin-tazobactam. Antibiotic therapy was not changed.

Stool was not cultured because the patient had not 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.

3. Discussion

Shigellosis still remains an important public health problem in developing countries. It is usually a self-limiting disease remaining confined to the gastrointestinal tract [9-11]. The disease is transmitted by feco-oral route and has an incubation period of 12 hours to one week. The spectrum varies from mild to severe and fatal disease [11].

The distribution of *Shigella* species is dynamic over time and place [1]. No imported shigellosis cases caused by *S. dysenteriae* or *S. boydii* were found. The stable bacterial species and low input cases could potentially be explained by the lower population mobility in Taiyuan than Beijing, Shanghai and Guangzhou. Besides, rates of *S. sonnei*-infection have not significantly increased with economic growth in the past 10 years. However, in other developed regions in China, the predominant *Shigella* species from *S. flexneri* to *S. sonnei* shifted in Beijing, Shanghai, and

Guizhou.

Common symptoms may include mild abdominal discomfort to severe diffuse colicky abdominal pain (70% to 90%). Patient reports of small volume mucoid diarrhea (70% to 80%) that precedes bloody diarrhea (30% to 50%). Other symptoms include fever, nausea, vomiting, anorexia, lethargy, and tenesmus. Rare but severe symptoms include delirium, encephalopathy, anuria, seizures, meningismus, and coma.

Physical examination of patients suffering from shigellosis may indicate lethargic or toxic individuals. Vital signs may reveal fever, tachycardia, tachypnea, and hypotension. An abdominal examination may show a distended abdomen with hyperactive bowel sounds. Tenderness may be present especially in the lower abdomen due to the involvement of sigmoid colon and rectum. 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].

Peritonitis can develop as a result of inflammatory, infectious and ischemic injuries, and perforation of gastrointestinal and genitourinary systems. Among the Gram negative agents, species that are members of *Escherichia coli*, *Klebsiella sp.*, *Proteus sp.*, *Pseudomonas sp.* and *Enterobacter sp.* genus are the dominant species in the etiology. Among anaerobic bacteria, *Bacteroides fragilis* and *Clostridium species* are identified as causative agents. The causative agents in peritonitis that develop following appendix, colon, or rectum originated infections are generally Gram negative or anaerobic bacterias. In the present case, peritonitis developed secondary to *S. sonnei*.

S. sonnei was isolated in the peritoneal fluid as a result of appendix perforation in a patient presenting with constipation, fever and right lower quadrant pain.

In 1961, White et al. [13] 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. [14] 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, 15-19]. 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 [20, 21]. 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 peritoneal surface of the colon [20]. *Shigella* spp. May cause perforated appendicitis using this mechanism without causing diarrhea or colitis.

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, 15, 18]. *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.

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, 18, 19].

Surgical complications caused by *Shigella* spp. may be fatal. Over past 40 years, the authors have reviewed the surgical complications of shigellosis in children.

Review of the literature shows that there are several case reports describing association with appendicitis and Shigellosis. Even though many case reports date prior to 21st century, there are a few recent case reports. Huynh et al. (2015) reported a 34-year-old man presented with a 6-day history of diarrhea, abdominal cramps and fever and testing the initial blood culture correctly identified the organism as *S. sonnei*. Special attention should be paid to exclude signs of peritonism or peritonitis, which will indicate serious illnesses that might require surgical care. As reported recently in literature of Ghosh et al. (2011), there is a risk of epidemic due to nalidixic acid resistant *S. sonnei* in the near future. This might pose therapeutic challenges in treatment of such cases.

4. Conclusion

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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