



A Rare Cause of Right Lower Quadrant Abdominal Pain: Isolated Cecal Necrosis

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ABSTRACT

Aim: Isolated cecal necrosis (ICN), a variant of ischemic colitis, is a rarely seen clinical condition. This aim of this case series was to evaluate the diagnosis and management of ICN.

Method: Patients operated between December 2013-January 2020 with the presumptive diagnosis of acute abdomen and intra-operatively diagnosed as ICN were evaluated retrospectively.

Results: There were 17 patients of whom nine (52.9%) were male. The mean age of the patients was 55.8 (range: 22-85) years. All the patients had at least one co-morbid disease, the most frequent of which were coronary artery disease, hypertension, and chronic renal failure. Fourteen (82.35%) underwent right hemicolectomy, and ileotransversostomy, while two (11.8%) had right hemicolectomy and Mikulicz ileocolostomy, and one (5.9%) underwent partial colonic resection with Mikulicz ileocolostomy due to limited cecal necrosis, which was diagnosed earlier. Six (35.3%) patients died.

Conclusion: ICN must be kept in mind in the differential diagnosis of acute abdomen with right lower quadrant localization, especially in patients with co-morbid diseases. Due to delayed diagnosis and complications, such as perforation, ICN had a high rate of morbidity and mortality.

Keywords: Acute abdomen, acute appendicitis, cecal necrosis, mesenteric ischemia, tocilizumab

Introduction

Isolated cecal necrosis (ICN), a rarely seen clinical condition, is a form of non-occlusive mesenteric ischemia. ICN mostly occurs due to decreased blood flow in the right colon, which may occur as a result of hypovolemic shock, hypotension, hemodialysis, chronic heart disease, cardiac arrhythmia, atherosclerosis, and drug use.¹ Most patients diagnosed with ICN attend the emergency service with right lower quadrant abdominal pain, and the primary diagnosis is usually acute appendicitis.^{2,3} Although ICN and acute appendicitis have similar clinical symptoms and physical examination findings, the surgical procedure, incision type, and postoperative course are very different.² Thus, being aware of ICN and making the differentiative diagnosis from acute appendicitis is fundamental. This study aimed to evaluate the diagnosis and management of ICN with a case series of 17 cases.

Materials and Methods

This study was designed as a retrospective observational study. Seventeen patients diagnosed with ICN between December

2013-January 2020, were retrospectively analyzed. All patients were operated due to acute abdomen. Definitive diagnosis was made during the operation and with subsequent histopathological examination. Demographics features, clinical symptoms, laboratory and imaging data, co-morbidities, surgical procedure, and postoperative follow up data were extracted from the hospital records.

Approval from the University of Health Sciences Turkey, İzmir Tepecik Training and Research Hospital Institutional Research Ethics Board was obtained (approval number: 2021/04-17). Written informed consent was obtained from each patient who participated in this study.

Statistical Analysis

IBM SPSS Statistics 22 program was used in the statistical analyses when evaluating the findings of the study. Descriptive statistical methods (mean, standard deviation, and frequency) were used in the comparison of qualitative data.



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Results

In total there were 17 patients diagnosed with ICN during the study period, with a mean age of 55.8 (22-85) years. Nine (52.9%) of the patients were male with a mean age of 63.5 years, and eight were female with a mean age of 62.3 years. All the patients had presented to the emergency service with right lower quadrant abdominal pain, that persisted for at least one day, and nausea. During the physical examinations, all of the patients had tenderness and rebound tenderness at the right lower abdominal quadrant. Abdominal computed tomography (CT) imaging was performed on all patients. Ten out of seventeen (58.8%) had an Alvarado score between 7-8, and abdominal CT was performed to exclude the diagnosis of acute appendicitis. Seven (41.2%) of the abdominal CTs were performed due to a medical history of appendectomy. In the evaluation of the abdominal CT images, all of the patients had pericecal inflammation, cecal wall thickening and, in all patients who had not undergone previous appendectomy, appendix vermiformis was seen normally (Figure 1). Comorbidities present were as follows: four (23.5%) were on hemodialysis due to chronic renal failure; five (29.4%) had coronary artery disease; eight (47.1%) had hypertension; three (17.7%) had congestive heart failure; five (29.4%) had cardiac arrhythmia; four (23.5%) had diabetes mellitus; three (17.7%) had chronic obstructive pulmonary disorder; one (5.9%) patient was followed due to pancreatitis; one (5.9%) patient had lung cancer; one (5.9%) patient who had an iliac artery stent due to peripheral vascular disease; and one (5.9%), who was also the youngest patient, had aplastic anemia.

Fourteen (82.25%) patients underwent right hemicolectomy and ileotransversostomy, two (11.8%) had right

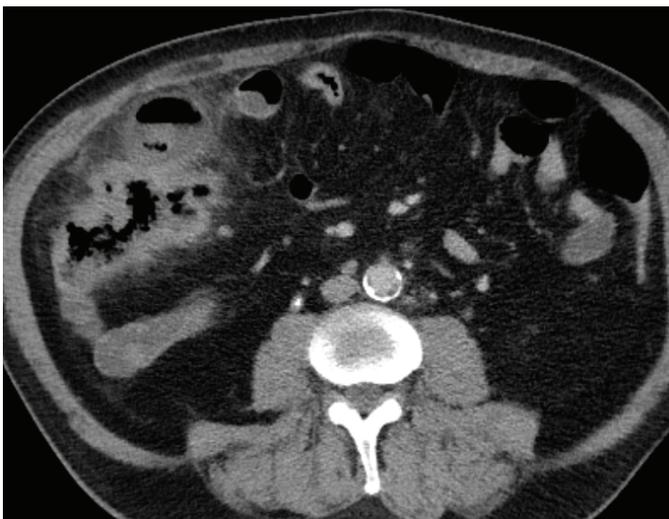


Figure 1. Computerized tomography shows circumferential bowel thickening, peri-cecal inflammation and heterogeneity

hemicolectomy with and Mikulicz ileocolostomy, and one (5.9%) underwent partial colon resection with and Mikulicz ileocolostomy due to limited cecal necrosis, which was diagnosed earlier. Histoathological evaluation confirmed all patients had ICN without malignancy (Figure 2). Pathology also reported transmural necrosis and serositis isolated in the cecum with no evidence of embolism, malignancy, or vasculitis (Figure 3). The mean operation time was 117 minutes (55-170 minutes). Six (35.3%) patients died during the postoperative follow-up period. The mean hospitalization duration was 11.3 days (1-28 days). The most common postoperative complication associated with the surgery was superficial surgical site infection, which occurred in three (17.7%), and evisceration was seen in two (11.8%) cases. The patients with surgical site infection were given antibiotic treatment, and the wound dressings were changed daily. Two patients with evisceration were re-operated. Two patients who underwent right hemicolectomy with an ileostomy had fecal peritonitis with clinical suspicion of sepsis, so inotropic drugs were initiated during the surgery and continued postoperatively in the intensive care unit. However, both died on the first postoperative day due to septic shock. Two patients, both with congestive heart

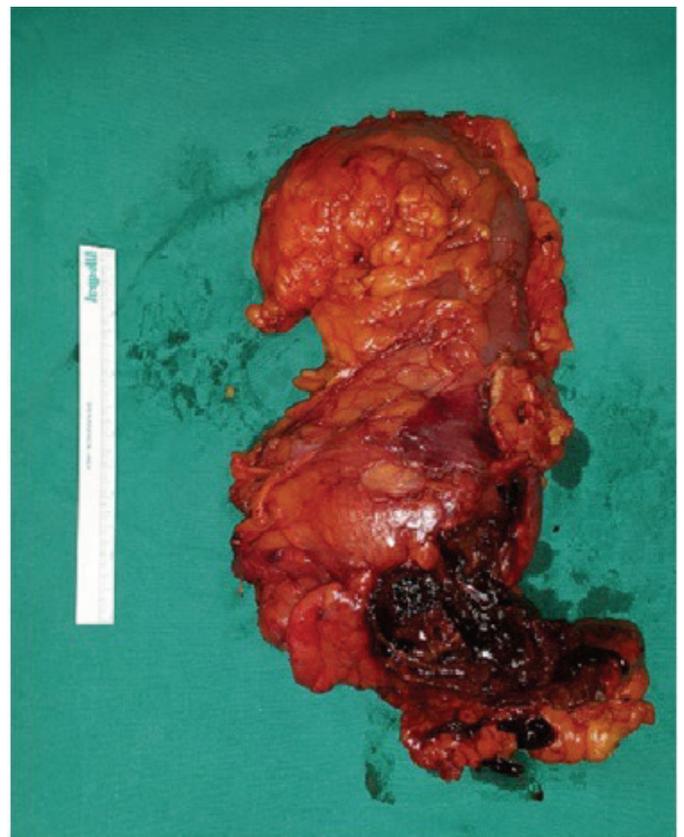


Figure 2. Macroscopic appearance of resected right hemicolectomy specimen which supported the diagnosis of isolated cecal necrosis

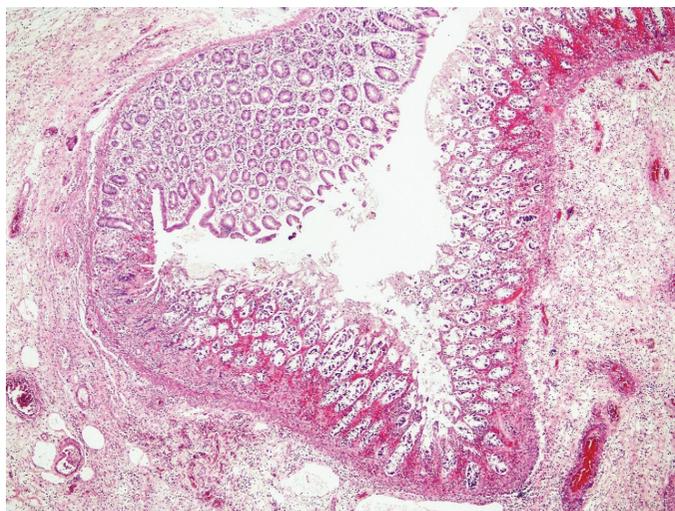


Figure 3. Sharp passage of normal colon mucosa and ischemic large intestine segment; active chronic inflammatory granulation tissue extending to the serosa, which microscopically showed a sharp transition with normal mucosa (hematoxylin and eosin, x40)

failure, died due to cardiopulmonary insufficiency on the postoperative fifth and seventh days. The patient who had a prior diagnosis of lung cancer and the patient who had obstructive sleep apnea syndrome and chronic obstructive pulmonary disorder died due to ventilator-associated pneumonia on the postoperative 16th and 23rd day. The duration of the hospital stays varied, since all patients had different and multiple comorbidities. Two patients had pneumonia, and the duration of hospitalization was 12-28 days.

Discussion

Ischemic colitis is a rarely seen clinical entity, which mostly affects elderly patients after the sixth decade, and is more common in female patients.¹ The disease is classified, according to its etiopathogenesis, into two groups depending on whether it occurs due to a vascular obstruction as the occlusive form, or due to poor blood flow caused by other underlying reasons as the non-occlusive form.^{2,3} However, some studies classified colonic ischemia into three groups. These include the occlusive and non-occlusive forms plus phlebosclerotic colitis, which occurs due to mesenteric vein thrombosis as a result of venous obstruction caused by fibrotic sclerosis and calcification of the walls of the mesenteric veins.⁴

Vascular circulation serving the cecum is provided from the anterior and posterior cecal arteries and arterial anastomosis between them.^{5,6} Ischemia of the cecum is rarely seen because of the rich collateral circulation that comes from the ileal branch and colic branch of the ileocolic artery.

ICN is a rarely seen, clinical form of ischemic colitis. Poor mesenteric perfusion, systemic hypotension, dialysis,

trauma, shock, chronic heart disease, cardiac surgery, drugs, hypercoagulability, portal hypertension, smoking history, diabetes mellitus, hypertension especially among young patients, dyslipidemia, systemic chemotherapy, oral contraceptives and cocaine abuse, and pancreatitis (as in one of our patients) have all been reported to be associated with the development of ICN.^{3,6,7,8} Frossard et al.⁹ reported two young female patients with ICN, which occurred as a cause of a high level of circulating estrogens due to pregnancy and oral contraceptive medication. In our study, two patients had a history of new-onset monoclonal antibody drug use, which might conceivably have been involved in the development of ICN. One patient had rheumatoid arthritis and was on Tocilizumab treatment, a recombinant humanized anti-interleukin-6 receptor monoclonal antibody.¹⁰ The other patient had aplastic anemia for which they were receiving Eculizumab, a fully-humanized immunoglobulin G monoclonal antibody to complement component C5.¹¹ Although Tocilizumab is used for rheumatoid arthritis, more recently it is being used in the treatment of Coronavirus disease-2019 (COVID-19) infected patients.^{12,13} ICN should be kept in mind in the etiology of right lower quadrant abdominal pain, which develops suddenly in COVID-19 patients under tocilizumab treatment.

ICN is a diagnostic challenge, as it is rare and thus not well-known, and is an atypical presentation of acute colonic ischemia. The presumptive diagnosis is usually based on the combination of clinical suspicion, physical examination, and radiological imaging methods. There is no specific serum marker for ICN.⁶ ICN generally presents with right lower quadrant abdominal pain and tenderness, fever, diarrhea or hematochezia, and leucocytosis. These manifestations may mimic acute appendicitis, cecal diverticulitis, stercoral perforation, or cecal carcinoma and therefore, early diagnosis is challenging.^{14,15,16} Guitart Giménez et al.⁶ reported that the most commonly seen findings in CT images were thickening of the cecal wall and ischemic cecal mural thickening and the ascending colon or pneumatosis of the cecal wall. In our study, cecal wall thickening and ascending colon were the most commonly seen findings. According to previous studies and case reports, most ICN cases were preoperatively diagnosed as acute appendicitis.^{3,16,17,18,19} As suggested by Kohga et al.⁷ this misleading preoperative diagnosis could be prevented with preoperative abdominal CT scan, a hypothesis with which we are in agreement. The use of colonoscopy in the diagnosis of ischemic colitis is still controversial. Although some authors recommend colonoscopy in the diagnosis of ischemic colitis, others suggest that colonoscopy may increase the risk of perforation

due to the increased colonic intraluminal pressure.^{3,14,20} Diagnostic laparoscopy is considered a useful option to make a definitive diagnosis and eliminate acute appendicitis from the differential. Also, laparoscopy can be helpful to implement an operational strategy which includes the incision type. Based on the results of diagnostic laparoscopy, the appropriate incision type can be planned. In our study, a superior and inferior midline incision was preferred in all patients. We performed diagnostic laparoscopy in two patients with a history of appendectomy, although their physical examinations were compatible with acute appendicitis. Perko et al.¹⁸ reported that a 73-year old patient with lower quadrant pain was diagnosed with acute appendicitis, but during the operation, ICN was detected, and partial resection was performed laparoscopically. Although these patients had multiple comorbid diseases, appropriate patients could be managed by laparoscopic partial resection if the surgeon had sufficient laparoscopic surgical experience.^{7,18} We recommend that the duration of the operation be kept as short as possible in this type of emergency operation because the patients tend to have many comorbidities. Studies have shown that the primary surgical treatment was resection of the necrotic bowel segment and anastomosis or ostomy, depending on the abdomen condition, via open technique or laparoscopically.^{3,7,8,18} In treatment, partial cecal resection or right hemicolectomy is the most commonly preferred method, according to the size of cecal necrosis and presence of peritonitis.^{3,7,8} In our study, right hemicolectomy with ileotransversostomy was the most commonly used method, followed by right hemicolectomy with an ileocolostomy. In our study, the surgical methods were chosen according to the patient's general condition and the preference of the staff surgeon. Fourteen patients were operated with right hemicolectomy and ileotransversostomy. In two patients, right hemicolectomy and Mikulicz ileocolostomy was preferred due to fecal peritonitis and edematous intestinal wall, which increases the risk of anastomotic leak. One patient was operated with partial colonic resection with Mikulicz ileocolostomy, since the cecal necrosis was limited and early diagnosis was achieved. Previous reports have suggested that some cases of ICN showed poor prognosis.³ In contrast, many patients with ICN showed an uneventful postoperative course. It has been reported that early diagnosis and urgent resection of the damaged intestine are essential to improve the postoperative outcomes of ICN.^{3,7} Although Çakar et al.³ reported that the prognosis was poor and the mortality rate was 83%, Gundes et al.¹⁷ reported a lower mortality rate of 38% which was similar to our study with a rate of 35.3%. We believe that the increased mortality rate reported by Çakar et al.³ was due to delayed diagnosis in the emergency

service. Patients with co-morbid disease attending the emergency department with right quadrant pain should be evaluated carefully with a detailed physical examination. Diagnostic laparoscopy, which allows the exploration of all of the intra-abdominal organs, should be performed without delay when there is a suspicion of cecal necrosis.

Study Limitations

The limitations of our study were its retrospective nature and inclusion of uncommon cases due to the small number of cases.

Conclusion

ICN should be kept in mind during the evaluation of right lower quadrant abdominal pain, especially in patients with co-morbid diseases. For diagnosis of ICN, clinical suspicion, and findings generally confirmed by imaging methods, especially CT, and diagnostic laparoscopic surgery may be helpful and also aid in planning surgery. When diagnosis is delayed, ICN may be life-threatening.

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Ethics

Ethics Committee Approval: Approval from the University of Health Sciences Turkey, İzmir Tepecik Training and Research Hospital Institutional Research Ethics Board was obtained (approval number: 2021/04-17).

Informed Consent: Written informed consent was obtained from each patient who participated in this study.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.D.A., M.Ü., E.K.A., T.K., B.Ç., Concept: S.D.A., M.Ü., Design: S.D.A., M.Ü., Data Collection or Processing: S.D.A., E.K.A., Analysis or Interpretation: S.D.A., T.K., B.Ç., Literature Search: S.D.A., Writing: S.D.A.

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