Abstract

A 63-year-old man on long-term hemodialysis therapy was hospitalized due to right lower abdominal pain. CT scan demonstrated a multiple concentric structure in the ileocecal region. Colonoscopy showed a polyp-like tumor arising from the expected location of the appendix, with a dimple at the top. Barium enema study revealed a submucosal tumor-like filling defect in the cecum with non-filling of the appendix. A diagnosis of intussusception of the appendix (IA) was made. During the follow-up, IA reduced spontaneously. The present case report is the first description of IA in a patient on hemodialysis therapy. Furthermore, spontaneous reduction of IA is indeed rare.

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Key words: intussusception, appendix, CT, colonoscopy, spontaneous reduction, hemodialysis

Introduction

Patients with end-stage renal disease (ESRD) often suffer complications with various disorders in the gastrointestinal system (1). Intussusception of the appendix (IA) is a very rare disease, with a 0.01% of incidence among 71,000 specimens of the appendix (2). Only about 200 cases have been reported in the literature (3, 4). Various pathologic lesions have been thought to cause IA (5), whereas some reports state that IA may occur in an appendix even without an underlying abnormality (6, 7).

Levine et al (8) and Bachman and Clemett (9) suggested that IA could occur as a transient phenomenon based on the findings of barium enema studies. There have been, however, few reports that describe spontaneous reduction of this disorder in adults (10, 11). Here, we report the first case of IA in a chronic hemodialysis patient. Moreover, IA reduced spontaneously during follow up.

Case Report

A 63-year-old man started chronic hemodialysis therapy due to ESRD resulting from chronic glomerulonephritis in 1975. In 1991 he was diagnosed as having chronic hepatitis type C. He began complaining of pain around the navel and in the left lower quadrant of the abdomen late in January 2001. Subsequently he complained of pain in the right lower quadrant of the abdomen. The patient was admitted to Saiseikai Yahata Hospital on January 31 with a possible diagnosis of acute appendicitis. On admission he was an emaciated man with blood pressure of 158/98 mmHg and heart rate of 72 beats/min. He assumed a forward bending posture due to abdominal pain. Physical examination of the abdomen revealed tenderness in the right lower quadrant of the abdomen and rebound tenderness. Muscle guarding was absent. Abdominal masses were not confirmed and hepatosplenomegaly was not present. He had a Cimino-Brescia type arteriovenous fistula in the right forearm. The findings of the remainder of the examination were not remarkable.

Laboratory data on admission disclosed a slightly elevated C-reactive protein (0.9 mg/dl, normal range; 0–0.4) without leukocytosis. Hemoglobin was 10.7 g/dl, hematocrit 31.3%, and serum albumin 3.8 g/dl. The value for gamma-GTP was elevated (138 IU/l, normal range; 0–60). The values of urea nitrogen and creatinine were compatible with chronic renal failure on hemodialysis therapy. Serum potassium was within the normal range. The value for carcinoembryonic antigen (CEA) slightly exceeded the upper limit for normal subjects (5.2 ng/ml, normal range; less than 5), but it appeared to be normal when considering the different cut-off
value for hemodialysis patients (12, 13). CA19-9 was less than 2 U/ml. Abdominal computed tomography (Fig. 1) showed a concentric structure which consisted of high and low density tissue in the ileocecal region, suggesting edema of the bowel wall.

Because the patient’s complaints and findings of the physical examination were associated with neither leukocytosis nor significantly elevated CRP, a colonoscopy was done to permit a final diagnosis. As demonstrated in Fig. 2, a polyp-like cylindrical tumor arose from the expected location of the appendix. There was a dimple at the top and the mucosa around the dimple was reddish in color. Microscopic examination of the mucosal tissue around the dimple yielded normal colonic mucosa. Barium enema examination showed a submucosal tumor-like filling defect in the cecum with non-filling of the appendix (Fig. 3). The patient was diagnosed as having IA from the colonoscopic findings.

His complaint subsided shortly after admission in association with disappearance of the tenderness in the right lower abdomen. The value for C-reactive protein returned to its normal range.

Since surgeons could not exclude the possibility of a pathologic lesion which led to IA, laparotomy was considered. The patient, however, declined to undergo surgery because of his concern about perioperative complications and postoperative recovery. He agreed to undergo surgery if the symptom recurs. He was discharged on the ninth day after admission.

The patient did not suffer complications of recurrence of abdominal pain during the follow-up. Abdominal computed tomography at three and at six months later did not reveal the concentric structure in the ileocecal region. Moreover, fiberoscopic examination of the cecum at four months after discharge (Fig. 4) confirmed spontaneous reduction of IA.

**Discussion**

Patients with ESRD often suffer from various gastrointestinal...
Appenidiceal Intussusception

Figure 4. Second colonoscopy confirmed the disappearance of IA.

Most of these occurred in children under 10 years old, most commonly in males (5) (male : female ratio 4–5 : 1). However a review of papers since 1984 revealed cases occurring predominantly in adults with almost equal gender distribution (3).

The pathophysiology of IA can be divided into two groups: anatomic and pathologic (5). Anatomic conditions are characterized by 1) the fetal type of cecum with the appendix originating from its tip; 2) a wide appendicular lumen with the proximal lumen of greater diameter than its distal part; 3) a mesoappendix that is thin, free from fat, and with a narrow base; 4) a mobile appendicular wall capable of active peristalsis; and 5) an appendix that is free, unfixed by congenital peritoneal folds or adhesions. Pathological conditions leading to active peristalsis include falcations, foreign bodies, parasites, appendiceal neoplasms (polyps, adenocarcinoma, mucoceles, carcinoid), lymphoid follicles, and endometrial implants, respectively.

We must note that IA could occur without apparent pathologic lesions in the appendix (4, 21) as in the present case. Clinical presentation of IA is vague and various, ranging from asymptomatic (incidental finding at laparotomy), vague abdominal pain, right lower quadrant mass, and rectal bleeding to typical clinical symptoms reported in intussusception. Some patients, including the present case, might present with symptoms indistinguishable from acute appendicitis. Laboratory data were not helpful for determining the cause (22).

McSwain (23) has classified IA into five different anatomic types (Fig. 5): type 1, tip of the appendix intussuscepted into its proximal portion; type 2, middle part of the appendix intussuscepted into its proximal portion; type 3, the base of the appendix intussuscepted into the cecum; type 4, the proximal portion of the appendix forms the intussusceptum and is received into the distal portion; type 5, the complete inversion of the appendix intussuscepted into the cecum, with or without ileocecal or cecocolonic intussusception. The present case corresponds to type 3, the most common type, according to the colonoscopic findings.

Most patients with IA underwent laparotomy followed by appendectomy, cecotomy, or hemicolecotomy, because exploratory laparotomy was necessary in the majority of cases and because 52–63% of cases of adult intussusception were tumor-related (24). IA is now diagnosed more often by the radiologists or endoscopists preoperatively because of its distinctive features as shown in the Table 1 (8, 21, 25–29).

Although Levine et al (8) and Bachman and Clemett (9) suggested that IA could occur as a transient phenomenon based on their findings of barium enema study, spontaneous reduction of adult IA is indeed rare. Only two reports have described a colonoscopically-proved reduction of IA. Kuriyama et al (10) reported spontaneous reduction of IA in an asymptomatic 82-year-old man whose IA was identified during the exploration of occult fecal blood. Tomonaga et al (11) described an asymptomatic 74-year-old female who underwent colonoscopy for a medical check up. The first
colonoscopy revealed a so-called “inside-out” appendix, corresponding to type 5 IA (Fig. 5). When a second colonoscopy was done (9 months later), the IA had disappeared.

The present case report is the third description of adult IA in which spontaneous reduction during follow-up was confirmed colonoscopically. There are several differences between the preceding cases and the present case. Two patients were asymptomatic and the findings of the physical examination were normal, whereas our patient complained of pain in the right lower abdomen associated with tenderness. The value for C-reactive protein was slightly elevated in our case, in contrast to the value remaining within the normal range in the preceding cases.

In the present case, the time of spontaneous reduction of IA was uncertain. We consider it unlikely that IA was reduced during the admission and that air inflation during the colonoscopy contributed to the reduction of IA, since barium enema examination three days before discharge (4 days after colonoscopy) showed a submucosal tumor-like filling defect in the cecum with non-filling of the appendix (Fig. 3) indicating the persistence of the disorder.

The mechanism leading to IA in the present case was unclear. IA is a rare entity (2) and there has been no other report of IA in patients on hemodialysis. In addition only four cases of intestinal intussusception in patients with ESRD have been reported (18–20) suggesting that patients with ESRD are not more liable to develop intussusception. Therefore the association of ESRD and IA might be coincidental. Agha (24) stated that in those patients with no definable lead point, intussusception may be related to submucosal bowel edema, fibrous adhesions, or dysrhythmic contractions. Carr et al (18) suggested that uremic platelet dysfunction may result in intramural hematoma formation and subsequent intussusception.

In summary, we report a rare case of IA in a patient on chronic hemodialysis therapy. Moreover, IA reduced spontaneously during follow-up. Computed tomography, colonoscopy, and barium enema were helpful for the diagnosis of IA.

Table 1. Characteristic Findings of IA

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<tr>
<th>Ultrasound sonography</th>
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<td>target-like appearance (25)</td>
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<td>multiple concentric ring sign (26)</td>
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<th>Barium enema</th>
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<td>coiled-spring sign (8)</td>
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<td>cecal filling defect with non-filling of the appendix (27)</td>
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<th>Computed tomography</th>
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<td>well-demarcated cylindrical mass of soft tissue density (28)</td>
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<th>Colonoscopy</th>
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<td>mushroom-like polyloid tumor with a dimple at the top (21)</td>
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<td>foreskin and glans appearance (29)</td>
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References
9) Bachman AL, Clemett AR. Roentgen aspects of primary appendiceal
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