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# Adult Intussusception Caused by an Appendiceal Mucocele and Reduced by Colonoscopy

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Appendiceal intussusception is a very rare disease that is found in only 0.01% of patients who have undergone an appendectomy. Clinical symptoms vary but include acute appendicitis symptoms such as right lower quadrant abdominal pain or repetitive right lower quadrant crampy pain. Some patients are asymptomatic. Operative treatment is necessary to reduce an appendiceal intussusception in adults, but there is a debate about how to perform the reduction. Successful colonoscopic reductions have been recently reported for some cases. We report a case of appendiceal intussusception that was diagnosed, reduced by colonoscopy, and histologically confirmed as a mucinous cystadenoma after the operation.

**Key Words:** Appendiceal intussusception; Colonoscopy; Reduction; Mucinous cystadenoma

## INTRODUCTION

Appendiceal intussusception with a leading point in appendix is a very rare disease occurring only in 0.01% of appendectomy patients.<sup>1</sup> Clinical symptoms vary from acute appendicitis symptoms such as right lower quadrant pain to repetitive right lower quadrant crampy pain to intermittent lower gastrointestinal bleeding.<sup>2,3</sup> Some patients are asymptomatic. Adults and children require different treatment for their appendiceal intussusception. Children could be treated with barium enema or air reduction, while it is agreed that adult patients need operative treatment.<sup>4</sup> There is still some debate about how to perform the reduction, but there are also reports that colonoscopic reduction could be considered for patients with good condition.<sup>5-7</sup> We report a case of appendiceal intussusception that was diagnosed and reduced by colonoscopy, and histologically confirmed as a mucinous cystadenoma after the operation, together with a literature review.

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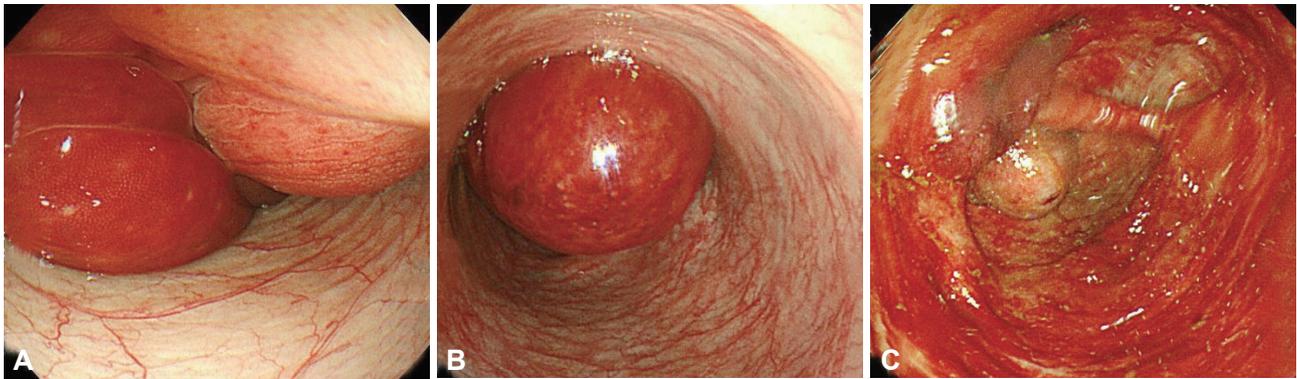
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## CASE REPORT

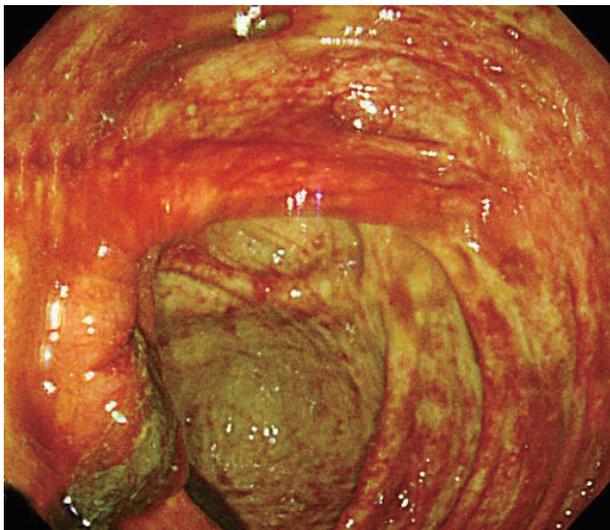
A 69-year-old male visited the hospital for diarrhea and abdominal discomfort lasting about 2 weeks. He did not have a medical history of hypertension or diabetes mellitus, but had a past medical history of tuberculosis diagnosis and complete recovery 1 year before the visit. He did not have any history of operation in the abdomen nor a symptom during defecation. He did not smoke or drink alcohol either. Diarrhea and abdominal discomfort started 2 weeks before the visit, without other specific symptoms. Vital signs at admission were measured as blood pressure 120/70 mm Hg, pulse rate 68/min, respiration rate 20/min, and body temperature 36.5°C. Not pale conjunctiva and anicteric sclera was found at physical examination. There was no splenomegaly or palpable mass from abdominal palpation, either. There was a mild tenderness at the left lower quadrant with indeterminate rebound tenderness. CBC result showed WBC 3,220/mm<sup>3</sup>, hemoglobin 13.0 g/dL, hematocrit 38.0%, platelet 263,000/mm<sup>3</sup>, PT 12.7 seconds (INR 1.09), and aPTT 29.8 seconds. The blood chemistry was analyzed as total bilirubin 0.75 mg/dL, total protein 7.5 g/dL, albumin 4.1 g/dL, AST 25 IU/L, ALT 19 IU/L, ALP 66 IU/L,  $\gamma$ -GTP 42 IU/L, triglyceride 72 mg/dL, cholesterol 149 mg/dL, BUN 28 mg/dL, and creatinine 0.9 mg/dL. Electrolyte was measured as Na 135 mmol/dL, K 3.9 mmol/dL, and Cl 101 mmol/dL. Urinalysis was normal but the stool occult blood test was weakly positive. Colo-



**Fig. 1.** Colonoscopic findings. (A) It shows huge coil-spring reddish polypoid mass which is occupying most of the lumen of the sigmoid colon. (B) The mass is migrated proximally during the air infusion. (C) It shows edematous and erythematous mucosal changes at the ileocecal lesion, and protruding mass is found at the appendiceal orifice.

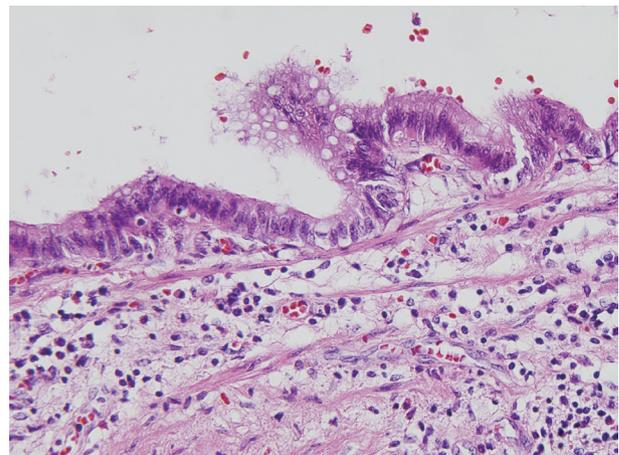


**Fig. 2.** Abdominal computed tomography finding. It shows wall enhancing cystic lesion at the appendiceal base (arrow).



**Fig. 3.** Colonoscopic finding. It shows slightly improved mucosal lesion.

noscopy was performed for the suspected infectious colitis, where a huge mass was found protruding to the middle of lumen of the sigmoid colon toward the anus. This mass was migrated toward the proximal by the air infusion and was clear-



**Fig. 4.** Microscopic finding. It shows appendiceal cyst is lined by columnar mucinogenic cells and inflamed wall (H&E stain, x400).

ed through the colonoscopy. Ileocecal lesion was found to have an edematous and erythematous mucosal change, and a cystic submucosal mass was found at the appendiceal orifice from histological examination after the colonoscopy (Fig. 1). An abdomen-pelvis computed tomography was performed, then, for it was suspected of an intussusception reduced by colonoscopy, and a wall enhancing cystic lesion was found at the appendiceal base (Fig. 2). When a follow-up colonoscopy was performed on day 4, ileocecal mucosal lesion appeared to have improved (Fig. 3). A laparoscopic appendectomy was performed on day 9, and a histologic examination shows appendiceal cyst is lined by columnar mucinogenic cells and inflamed wall (Fig. 4). The patient was discharged after the operation without particular complications.

## DISCUSSION

Appendiceal intussusception is a very rare disease that is appearing often as acute appendicitis, making it very difficult to diagnose before operation. Recent development of radiologic

examination and colonoscopy increased the possibility of diagnosis before operation, though.<sup>8</sup>

Appendiceal intussusception is caused by irregular appendiceal peristalsis developed by local irritation, and is more likely to occur in mobile mesoappendix wide appendicular lumen, and thin and mobile appendix.<sup>2</sup> Leading contributing factors include endometriosis, mucocele, villus adenoma, carcinoma, and adenocarcinoma; other less often factors include papilloma, hamartoma, mucosa-associated lymphoma, juvenile polyp, Crohn's disease, and melanosis coli.<sup>9</sup>

Gross<sup>10</sup> defined chronic intussusception as the disease lasting more than 5 days to 2 weeks. Adult patients tend to show chronic or subacute clinical symptoms, generally of long-term intermittent partial intestinal obstruction. Adult intussusception, especially appendiceal intussusception, varies from acute appendicitis symptoms such as right lower quadrant pain to repetitive right lower quadrant crampy pain to intermittent lower gastrointestinal bleeding. Some patients are even asymptomatic.<sup>2,3</sup> The most common symptom is the regular, intermittent abdominal pain. Our patient was hard to be suspected as intussusception only from physical examination, since he did not show any particular symptoms except for the abdominal discomfort and diarrhea that had started 2 weeks ago.

There is no finding for definite diagnosis of appendiceal intussusception, but several radiologic examinations might be useful. 'Coiled spring' sign of cecum or filling defect of appendix on double-contrast barium enema could be helpful for the diagnosis.<sup>11</sup> On abdominal ultrasonography, cecum might appear as concentric loop pattern or the appendix invaginated into the cecum.<sup>12</sup> Cecum appearing as a bulls-eye shape, sausage shape, or kidney shape are the characteristic findings of appendiceal intussusception on abdomen-pelvis computed tomography.<sup>13</sup> Our patient did not show such findings on abdomen-pelvis computed tomography, but a wall enhancing cystic lesion was found at the appendiceal base as the leading point of intussusception. Radiologic examination is therefore considered as a useful tool not only for the definite diagnosis of appendiceal intussusception but also for determining the presence and type of leading point and the possibility of malignancy.

Adults and children require different treatment for their appendiceal intussusception. Children could be treated with barium enema or air reduction, while it is agreed that adult patients need operative treatment.<sup>4</sup> There is still some debate about how to perform the reduction, but there are also reports of intussusception cases reduced by colonoscopy,<sup>5,6</sup> suggesting that colonoscopic reduction by non-operative air infusion could be a possibility. Such colonoscopic reduction accompanies the risk of dissemination of malignant lesion, peritoneal seeding, or venous embolism by excessive manipulation and the risk of bowel perforation accompanied by edema and ische-

mic change.<sup>14,15</sup> There are also reports, however, that pre-operation colonoscopy might be useful for it could find a leading point of intussusception and the air infused during the procedure could often induce the reduction of intussusception.<sup>5-7,16</sup> It is considered, therefore, that pre-operational reduction could be attempted for intussusception patients without severe bowel ischemia or malignancy.

Our case was first suspected as infectious colitis at admission; following the colonoscopy, however, he was diagnosed as intussusception, which was reduced by the injected air during the advancing of the scope following the mass. The ileocecal mucosal lesion was confirmed to have improved on the clinical symptom and follow-up colonoscopy after the reduction. The possibility of malignancy was found unlikely on abdomen-pelvis computed tomography, and laparoscopic appendectomy, not laparotomy, was performed later to improve the prognosis and shorten the hospitalization period. When an appendiceal intussusception does not accompany the possibility of other malignancies, colonoscopic reduction followed by an operation could better improve the inflammation than laparotomy only. This strategy is also deemed to minimize the extent of operation of the leading point, thereby enabling the patient to recover more rapidly.

We found that colonoscopy could play an important role in the diagnosis and treatment of appendiceal intussusception. More case reports on adult appendiceal intussusception reduced by colonoscopy and further studies on criteria for reduction are required.

#### Conflicts of Interest

The authors have no financial conflicts of interest.

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