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Ileocolic Intussusception Accompanied with Inflamed Appendix: 2 Case Reports 염증성 충수돌기를 동반한 회결장 장중첩증: 2예 보고

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Intussusception and acute appendicitis are common emergency conditions in children. They should be promptly differentiated in pediatric patients presenting with suggestive symptoms. However, both diseases may occur simultaneously. Herein, we present two cases of intussusception of the appendix accompanied with appendicitis.

Index terms Intussusception; Appendicitis; Ultrasonography; Tomography, X-Ray Computed

INTRODUCTION

Intussusception and acute appendicitis are common conditions with similar clinical manifestations, such as abdominal pain, vomiting, and/or irritability. Both require urgent attention and careful examination in children because the more the diagnosis is delayed, the greater the likelihood that severe complications, including peritonitis and possible death, will occur (1, 2). It is important to promptly differentiate these two conditions in pediatric patients presenting suggestive symptoms. In ileocolic intussusception, the appendix tends to be part of the intussusception (3). In rare cases, however, pathologic appendix and intussusception can coexist (3, 4). In this report, we present two cases of ileocolic intussusception with the pathologic appendix.

CASE REPORT

CASE 1

A 35-month-old girl visited the emergency department with abdominal pain. She had a history of recurrent intussusception, treated by pneumatic reduction 6 months ago. She complained abdominal pain and vomiting, which began 2 days ago. The pain was



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colicky and cyclic. In the emergency department, her vital signs were stable (body temperature, 37.2°C; pulse rate, 98 beats/min; respiratory rate, 20 breaths/min). On physical examination, there was tenderness in the right lower quadrant of abdomen and auscultation indicating a hypoactive bowel. There was no rebound tenderness or palpable mass in the abdomen.

Abdominal radiography and laboratory tests were performed. Abdominal radiography showed oval-shaped haziness in the right upper quadrant and localized bowel gas in the left upper quadrant without distension. Laboratory investigations revealed as followings: hemo-globin, 12.6 g/dL; hematocrit, 37.6%; platelet count, 439000/mm³; white blood cell count, 12360/mm³; neutrophils, 63.3%; monocytes, 4.9%; and lymphocytes, 28.1%, and elevated high-sensitivity C-reactive protein level (6.4 mg/L; normal range, 0–1 mg/L). We suspected intussusception and performed abdominal ultrasonography (US), which showed a multicentric, bowel-in-bowel mass appearance in the right lower quadrant, consistent with intussusception (Fig. 1A).

Pneumatic reduction was performed without complications. After 6 hours, the patient complained cyclic abdominal pain and US was repeated. The second ultrasonographic examination revealed recurrent intussusception and pneumatic reduction was successfully performed. After 12 hours, the patient complained abdominal pain again and on US recurrent intussusception was depicted. However, the third pneumatic reduction was failed. Afterward, contrast-enhanced abdominal and pelvic CT was performed, which showed ileocolic intussusception and wall thickening of the appendix, which was partially trapped within the intussusception (Fig. 1B).

In emergency surgery, the appendix was found to be inflamed and partially trapped within the intussusception. Thus, open appendectomy and manual reduction of the intussusception were performed. The surgically resected appendix appeared edematous and enlarged. Histologic examination showed multifocal inflammatory infiltration (Fig. 1C). There was no fecalith within the inflamed appendix on gross examination. Five days after surgery, the patient was discharged without specific complications.

CASE 2

A 35-month-old girl visited emergency department with vomiting and cyclic abdominal pain started one day ago. She was on medication due to acute nasopharyngitis diagnosed in a local pediatric hospital with symptoms of cough and rhinorrhea three days ago. In the emergency department, her vital signs were stable (body temperature, 36.8°C; pulse rate, 130 beats/min; respiratory rate, 30 breaths/min). On physical examination, we noted tenderness and abdominal guarding in the right lower quadrant. We found no rebound tenderness or palpable mass in the abdomen.

Abdominal radiography and laboratory tests were performed. Abdominal radiography showed paucity of bowel gas. Laboratory tests revealed as followings: hemoglobin, 13.4 g/dL; hematocrit, 38.8%; platelet count, 245000/mm³; white blood cell count, 9290/mm³; neutrophils, 77.9%; monocytes, 2.6%; and lymphocytes, 16.4%, and normal C-reactive protein level.

Initially, acute appendicitis was suspected and contrast enhanced abdominal CT was performed. On CT, bowel in bowel appearance in the right upper to lower abdomen was seen, suggesting ileocolic intussusception. In addition, enlarged appendix was noted with promi-

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Fig. 1. A 35-month-old girl presenting with recurrent intussusception with acute appendicitis.

A. Initial ultrasonographic images. The transverse image shows a multicentric, hypo- and hyperechoic, bowel-in-bowel mass appearance. The longitudinal image showing the multilayered bowel, echogenic mesentery fat, and two lymph nodes (arrows) within the intussusception.

B. Axial and coronal contrast-enhanced CT images show the appendix (arrows) partially trapped within the intussusception (asterisks). Prominent appendiceal wall enhancement and periappendiceal fluid collection are observed.





C. Microscopically, there is transmural neutrophil infiltration at the appendix (hematoxylin and eosin stain, × 200).

Fig. 1. A 35-month-old girl presenting with recurrent intussusception

with acute appendicitis.

Fig. 2. A 35-month-old girl presenting with ileocolic intussusception with inflamed appendicitis. Axial and coronal contrast-enhanced CT images show the appendix (arrows) partially trapped within the intussusception (asterisks). Enlargement of the appendix with prominent appendiceal wall enhancement and periappendiceal fat stranding are also noted.





DISCUSSION

Intussusception and acute appendicitis are two common and important conditions requiring adequate investigation at the proper time. They manifest with common symptoms, such as abdominal pain, vomiting, and/or irritability. Depending on the child's age at the time of diagnosis, differential diagnosis can be made; a child affected by intussusception tends to be younger than a child affected by acute appendicitis (4). Furthermore, there are some characteristic symptoms helpful for differentiation, such as currant jelly-like stool and cyclic irritability in intussusception and right lower quadrant pain in acute appendicitis. In many cases, if one condition is suspected, then the other may be overlooked (4).

Intussusception occurs when one segment of bowel and its associated mesentery enter into an adjacent segment, and it is one of the most common causes of intestinal obstruction in children under 3 years of age (3, 4). Generally, the cause of intussusception is idiopathic and most cases (75–90%) in children are ileocolic or ileocecal (3, 4). It is rare in children younger than 4 years of age to have a lead point as the cause of intussusception (4). When a lead point is present, the most common causes are Meckel's diverticulum and Henoch-Schönlein purpura (3-5). The various other causes include lymphoma, duplication, hemangioma, and intestinal polyps (5-7).

In our cases, patients were younger than 4 years of age (both are 35 months), thus we initially thought cases were idiopathic. In our first case, intussusception recurred twice and on the third recurrence, pneumatic reduction failed. After the failure of pneumatic reduction, we performed CT to determine any complications and to find a possible lead point that was not identified on previous US. On CT, we found the inflamed appendix partially entrapped within the intussusception. Our second case also showed intussusception with inflamed appendix on initial CT. Intussusception with appendicitis, such as in our cases, is rare in both pediatric and adult patients and has been described in only a few case reports in the English literature (3, 4, 8).

Appendicitis is a common and major surgical emergency in childhood. Appendicitis is caused by obstruction of the lumen by fecalith or lymphoid hyperplasia (8, 9). When intussusception and appendicitis coexist, as in our cases, it is confusing whether the inflamed appendix is a lead point of the intussusception or the intussusception caused strangulation and inflammation of the appendix. In our cases inflamed appendix was not invaginated but partially entrapped into intussusception on CT. We presumed luminal obstruction by intussusception might be the cause of luminal obstruction of appendix. For the first case, multiple trial of pneumatic reduction was failed and operative reduction was done. During operative treatment for intussusception, appendectomy was also performed. However, for the second case, pneumatic reduction succeeded and symptoms were resolved. After reduction, the lu-

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minal obstruction was removed and appendiceal inflammation might be improved. After three years of follow up periods, the second patient did not complain any symptoms suggesting acute appendicitis.

Although rare, the appendix can be a lead point for intussusception due to anatomical and pathologic factors. Anatomical factors include a fully mobile appendix, narrow meso-appendix, and poorly fixed or high caecum, whereas pathologic conditions include appendicular inflammation, calcified fecalith, polyp, tubulovillous adenoma, muccoele, mucinous cystadenocarcinoma, carcinoid tumor, and mucosa-associated lymphoma (3).

As the rarity of coexistence of intussusception and appendicitis, there is no established management yet. Treatment may vary depending on the patient's condition, and further research is needed. It is necessary to keep in mind that inflamed appendix and ileocolic intussusception can coexist and various conditions involving appendix can be a lead point as well.

Author Contributions

Conceptualization, H.S.M.; data curation, L.H.J., W.Y.J., H.M.E.; investigation, H.S.M., L.H.J., W.Y.J., L.K.H., H.M.E.; methodology, H.S.M.; project administration, H.S.M.; supervision, H.S.M.; writing—original draft, L.H.J., W.Y.J.; and writing—review & editing, W.J.Y., L.K.H.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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염증성 충수돌기를 동반한 회결장 장중첩증: 2예 보고

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소아 환자에서 장중첩증과 급성 충수돌기염은 흔한 응급 질환이다. 의심되는 증상이 있을 때 두 질환을 감별하는 것은 중요하다. 하지만, 드물게 두 질환이 한꺼번에 발생하는 경우도 있 다. 이에, 우리는 병적인 충수돌기를 동반한 장중첩증을 보였던 두 증례를 소개한다.

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