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# Complete Invagination of Vermiform Appendix with Adenocarcinoma: Case Report[1]

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Appendiceal intussusception is a very rare pathological condition, an incidence, as revealed by appendectomy specimens, of only 0.01 percent. There are various types among which complete invagination of the appendix is very rare. We encountered a case of intussusception of the appendix with complete invagination induced by appendiceal adenocarcinoma. A preoperative diagnosis of appendiceal adenocarcinoma and intussusception was not possible, but a final pathological report confirmed these conditions and retrospective analysis of a barium enema showed a finger-like filling defect of the cecum, a relatively specific finding in such cases. We describe a case involving a 39-year old man who one month earlier had noted the onset of pain in the right lower abdomen.

**Index words :** Appendix, neoplasms
Appendix, CT
Colon, intussusception

Primary adenocarcinoma of the appendix is a rare neoplasm (1 - 8) and intussusception of the appendix is also rare (1). The most common presentation of appendiceal adenocarcinoma is that of acute appendicitis (2). It has seldom been diagnosed preoperatively, however, and is frequently an incidental finding at the time of surgery for unrelated conditions (3). We report a case of adenocarcinoma in a 39-year-old man whose initial complaint was right lower abdominal pain, and which involved intussusception of the appendix with the leading point.

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## Case Report

A 39-year-old man was admitted to our hospital complaining of right lower abdominal pain, first noted one month earlier. He had a five-month history of epigastric pain and during that time his body weight had decreased by 8 kg. Physical examination revealed equivocal tenderness in the right lower quadrant without palpable mass. The white blood cell count was within normal range and the hemoglobin level was 9.5g/dl, and other laboratory findings were non-specific. Initial differential diagnoses were appendicitis, diverticulitis, or malignancy originating in the bowel.

Barium enema showed a fungating filling defect in the medial wall of the cecum and this was thought to be the result of extrinsic compression by a periappendiceal abscess or a submucosal mass such as lymphoma (Fig.1A). Retrospective review of the findings of barium enema revealed a movable finger-like filling defect at the top of the cecal mass (Figs. 1A, B). Abdominal CT demonstrat-

ed a 4.5 cm sized whirling soft tissue mass with central fat density in the cecal lumen (Fig. 1C). The 4 cm sized enhancing tubular structure seen at the top of the mass, which was missed at that time (Fig. 1D), was shown by pathologic exmaination to be a completely invaginated appendix. Initial CT diagnosis was cecocolic intussusception without mention of the appendix, the possible leading point being a lymphoma, a lipoma or a hidden malignancy. Three days after the CT scanning, colonoscopic examination revealed a large multidirectional polypoid mass near the ileocecal area, and the colonoscopic diagnosis was colon cancer of cecal origin.

On the basis of this diagnosis, a right hemicolectomy was performed but within the operational field, no definite abnormal mass was visible in the cecal area. On palpation, a movable, elliptrical hard mass was found in

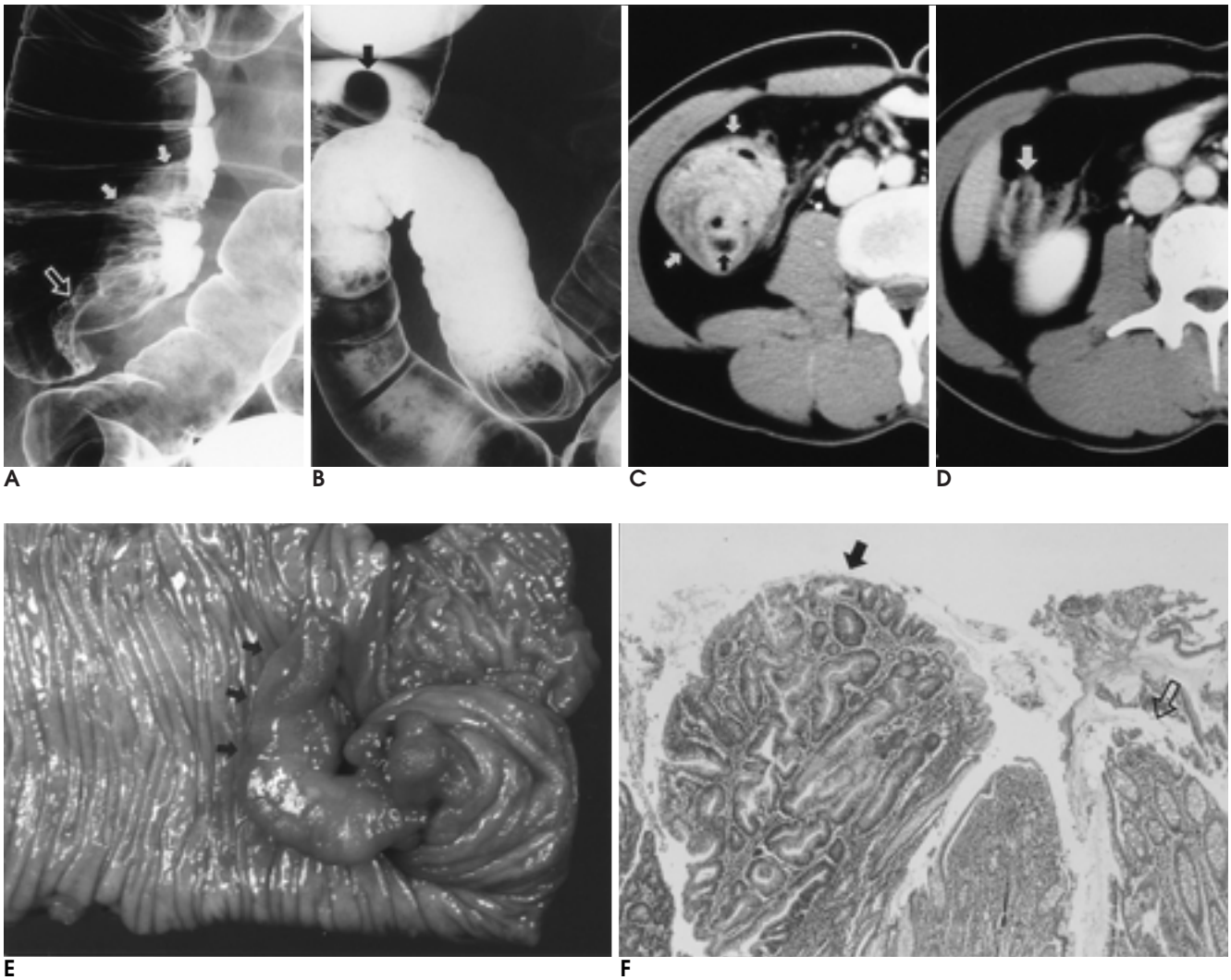


**Fig. 1.** 39-year old man with cecocolic intussusception with complete invagination and intussusception of the appendix with adenocarcinoma.

**A.** Barium enema shows fungating cecal mass (open white arrow) with finger-like tubular structure on the top of it (white arrows). The tubular structure abuts the medial wall of right colon, the dependent portion on left decubitus film. Initial diagnosis was a periappendiceal abscess or submucosal tumor of cecal origin.

**B.** Barium enema shows finger-like filling defect (arrow) floating on the barium filled cecal lumen.

**C.** Enhanced CT shows whirling soft tissue mass (white arrow) with central fat attenuation (arrow) in the cecal lumen.

**D.** Enhanced CT at the upper level of C shows enhancing tubular structure (white arrow) in the lumen of ascending colon, which turned out to be an inverted appendix on specimen correlation.

**E.** Resected specimen of cecum and ascending colon by right hemicolectomy shows an inverted appendix (arrow) with irregular mucosal surface surrounded by wrinkled intussuscepted cecal mucosal folds.

**F.** Microscopic examination reveals well-formed irregular glandular structures of adenocarcinoma arising from an appendix (arrow). The right side of the lesion shows normal surrounding cecal mucosa (open arrow) (HE stain, ×40).

the center of the cecum and a gross pathologic specimen revealed a totally inverted appendix with intussusception of this and a part of the cecum (Fig. 1E). Microscopic examination demonstrated the presence of a well-differentiated adenocarcinoma of the whole appendix infiltrating the serosa and spreading through the entire mucosal layer (Fig. 1F).

## Discussion

Intussusception of the appendix is a rarely reported pathologic condition, in Collins ' series of 71,000 appendectomy cases, only a 0.01 per cent incidence was reported (4). Fink and associates suggested that the etiological factors involved in intussusception of the appendix were partly anatomical and partly pathological. The anatomical variations cited include a fetal type cecum, a wide appendicular lumen, and a thin and mobile appendix. The pathological entities reported were worms, fecaliths, mucocele, papillomas, polyps, endometrial implants, and tumors (5).

Primary appendiceal malignancies account for 0.5% of all intestinal tumors. The four types of appendiceal neoplasm are carcinoid tumor (85%), mucinous adenocarcinoma (8%), colonic adenocarcinoma (4%), and adenocarcinoid tumor (2%) (6). Adenocarcinoma of the appendix is uncommon with an incidence ranging from 0.01% to 0.2% as revealed by appendectomy specimens (3).

Because the radiological appearance is nonspecific and is simulated by a variety of other lesions in this area, the preoperative diagnosis of adenocarcinoma of appendix is difficult. An inverted appendiceal stump, mucocele, carcinoma, adenomatous polyps of the cecum, appendiceal carcinoids, intussusception of the appendix, lipoma and the inflammatory changes secondary to appendicitis are conditions with which it may be confused (7).

The appendix originates from the cecum in such a way that its relationship to the ileocecal valve is constant (1, 7). It is found only between the cecal apex and this valve, and is always on the same side of the valve (1, 7). This rule may be helpful for distinguishing tumors of appendiceal origin from primary cecal neoplasms. Although an appendiceal carcinoma may have the appearance of an extrinsic mass, if the angle formed between the mass and the adjacent cecal wall is acute, an intramural or even an intraluminal tumor of the cecum is suggested (7). The preoperative diagnosis of intussusception of the appendix is also difficult and only a few cases can be diagnosed by barium enema. In fact, many cases have been diagnosed as filling defects or polypoid tumors of the cecum, as in our case (7). Appendiceal intussusception, when complete, may produce a linear or finger-like intraluminal filling defect with nonfilling of the appendix, the pathognomic of appendiceal intussusception, as in our case (9). A completely inverted or " inside-out " appendix is, howevere, extremely unusual (10). In instances of partial appendiceal intussusception without visualization of appendiceal lumen, differentiation from the cecal mass is also difficult. A coiled-spring defect in the cecum with nonfilling of the appendix has also been reported to be a characteristic finding of the appendiceal intussusception (9).

If the filling defect is on the opposite side of the ileocecal valve, the origin of the defect is most probably not appendiceal. If the defect remains constant and is homolateral with the valve, definite differentiation between irreducible partial appendiceal intussusception, masses of appendiceal origin, and other cecal lesions is not possible (1).

Clinically, almost 70 per cent of patients with appendiceal carcinoma have presented with signs suggesting acute appendicitis, and even during surgery, the diagnosis was correct in less than half of these cases (2). Most of the remaining patients presented with an abdominal mass which might be confused with other cecal lesions (2). Microscopically, almost all cases of appendiceal adenocarcinoma penetrated to the submucosal level, as in our case. The appendix is, therefore, subject to perforation and the peritoneum is seeded by the tumor. Right hemicolectomy is advocated for all appendiceal adenocarcinomas except those that are obviously Dukes ' A tumors and can be resected by simple appendectomy with histologically uninvolved margins (8).

In conclusion, when barium enema is performed, especially in elderly patients in whom appendicitis is suspected and a filling defect on the same side as the ileocecal valve is revealed, a rare but a important diagnosis of appendiceal carcinoma should be considered. If a linear or finger-like filling defect is demonstrated by barium enema, or CT images revealed an intraluminal tubular filling defect, the possibility of a completely invaginated appendix should also be considered.

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# 선암종에 의한 충수의 완전한 중적증[1]

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서영란 · 최철순 · 김호철 · 배상훈 · 김덕환[2]

충수의 장중적증은 충수 적출물 중에 0.01%의 발생 빈도를 갖는 매우 드문 질환이며, 충수의 선암종은 충수 적출물 중에서 0.01 - 0.2%의 발생빈도를 갖는 아주 드문 위장관의 악성종양이다. 저자들은 충수의 선암종에 의하여 충수의 장중적증과 동시에 맹장 - 맹장의 장중적증을 갖는 증례를 경험하였다. 수술전에 충수의 선암종 및 충수 및 맹장 - 맹장의 장중적증의 진단은 못하였으나, 수술 후에 충수의 선암종으로 인한 충수의 장중적증 및 맹장 - 맹장의 장중적증으로 확진이 되었고, 후향적으로 분석한 대장조영술에서도 비교적 특징적인 소견인 손가락 모양의 충만결손 소견이 보였다. 저자들은 한달 동안의 우하복부의 통증을 호소한 39세 남자환자에서 발생한 희귀한 증례를 보고한다.