Complete Appendiceal Intussusception Induced by Primary Appendiceal Adenocarcinoma in Tubular Adenoma: a Case Report

Minoru Takahashi1, Toshio Sawada1, Takahiro Fukuda1, Taiki Furugori1 and Hiroyuki Kuwano2

1Department of Surgery, Gunma Cancer Center Hospital, Ohta, Gunma and 2First Department of Surgery, Gunma University School of Medicine, Maebashi, Japan

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A case of complete intussusception induced by appendiceal carcinoma is reported. The patient was a 49-year-old man complaining of rectal bleeding. Barium enema and colonoscopy revealed a cecal polyp; it was interpreted as an inverted appendix with a tumor. Computed tomography showed an invaginated appendix into the cecal cavity. During surgery, the appendix was found to be inverted completely into the cecum; ileocecal resection with regional lymph node dissection was performed. Microscopic examination revealed well-differentiated adenocarcinoma in tubular adenoma. Diagnosis of intussusception with carcinoma of the appendix is often difficult because appendiceal carcinoma with intussusception of the appendix is a rare condition. Although this condition can be diagnosed by radiographic imaging or colonoscopy, computed tomography has also been useful. The clinical manifestation of appendiceal intussusception with primary appendiceal tumor resembles a large cecal polyp, but its treatment differs greatly. Failure to recognize this condition may result in unexpected complications such as consequent peritonitis in case of endoscopic removal.

Key words: appendiceal intussusception – appendiceal carcinoma – colonoscopy

INTRODUCTION

Intussusception of the appendix vermiformis in adults is a rare condition caused by anatomical and pathological factors such as tumors and is rarely diagnosed before surgery. Although most appendiceal tumors are benign, tubular adenoma is an unusual lesion. Furthermore, carcinoma of the appendix is a distinctly rare phenomenon. The combination of both a carcinoma and intussusception has been regarded as extremely rare (1). Here, we report a case with intussusception of the appendix induced by primary appendiceal adenocarcinoma and discuss the clinical features, classification, preoperative diagnosis and therapy of this condition together with a review of the literature.

CASE REPORT

A 49-year-old man with rectal bleeding was admitted to our hospital for further investigation. Double contrast barium enema examination showed a large polypoid tumor in the medial part of the cecum below the ileocecal valve (Fig. 1a); colonoscopy revealed a cecal polyp, interpreted as an inverted appendix with a tumor (Fig. 1b). Computed tomography also showed an invaginated appendix into the cecal cavity (Fig. 2). Although pathological examination of biopsy specimens from the tumor revealed tubular adenoma with severe atypia, an in situ diagnosis of appendiceal carcinoma was made clinically because of the tumor shape and size. During surgery, the appendix and mesoappendix were found to be inverted completely into the cecum. We performed ileocecal resection with regional lymph node dissection because possible malignancy had to be considered. The resected specimen was found to con-
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tain a tumor arising in the appendix. The tumor was $3.5 \times 2.5$ cm in size in the cecal cavity; the appendix had invaginated completely into the cecum at its base (Fig. 3a). Microscopic examination revealed well-differentiated adenocarcinoma in tubular adenoma and no metastasis in the 13 dissected lymph nodes (Fig. 3b). Pathologically this appendiceal carcinoma was diagnosed as stage 0 (pTis, pN0, pM0).

DISCUSSION

Intussusception of the appendix is an uncommon pathological condition. The incidence of invagination of the appendix is 0.01% in a large autopsy series (2). The etiology of appendiceal intussusception has been proposed by Fink et al. to be divided into anatomical and pathological causes (3). Anatomical variations include a fetal-type cecum, a wide appendicular lumen and a thin, mobile appendix. Reported pathological conditions include worms, endometrial implants and tumors. Various classifications of appendiceal intussusception have been attempted. Forshall divided cases into a primary type and a secondary or compound type (4). Langsam et al. classified the disorder into four types according to the relationship of the intussusceptum and intussusciptens (5). In our case, the appendiceal tumor as a leading point seemed to have induced the complete appendiceal intussusception.

Preoperative diagnosis of intussusceptions of the appendix is often difficult because it is a rare clinical entity; only a few cases can be diagnosed by barium enema and colonoscopy. Many cases have been diagnosed as filling defects or polypoid tumors of the cecum. Careful endoscopic examination, identifying the appendiceal orifice, should be required in the case of cecal polyp. When differential diagnosis is difficult, computed tomography or abdominal ultrasound is more useful. A definite finding of intussusception of the appendix in CT is the invaginated appendix in the cecal cavity.

Tumors of the appendix are uncommon; also, most tumors are benign. Adenoma of the appendix is also a rare condition. Only 50 cases of appendiceal adenoma were reported among 30 000 appendectomies (6). Moreover, primary adenocarcinoma is a very rare entity, which in most cases arises from a pre-existing adenoma (7). Ohno et al. first reported appendiceal intussusception induced by tubulovillous adenoma with carcinoma in situ similar to our case; the combination of both a carcinoma in adenoma and intussusception has been regarded as extremely rare (8).

Treatment of appendiceal intussusception is mainly surgical. The procedure varies from reduction of intussusception with appendectomy to right hemicolectomy. A right hemicolectomy or ileocecal resection with lymph node dissection should be performed when carcinoma is diagnosed preoperatively or during surgery. In the last decade, laparoscopic procedures have been applied to the treatment of almost all colonic diseases, including both benign and malignant lesions. As Schmidt et al. reported laparoscopic management of appendiceal intussusception, laparoscopic procedures will be more useful as a minimally invasive treatment (9).

The clinical manifestation of appendiceal intussusception with primary appendiceal tumor resembles a large cecal polyp with a wide stalk, but its treatment is completely different. Endoscopic removal should be performed carefully in cases of polypoid lesions in the cecum, taking into consideration the possibility of an invaginated appendix. Failure to recognize...
this condition may result in unexpected complications such as consequent peritonitis (10).

References