Tubulovillous Adenomas in a Continent Ileostomy After Proctocolectomy for Familial Polyposis

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Tubulovillous lesions of the ileal mucosa are an uncommon finding. Until recent descriptions of duodenal polyps (1), these premalignant (2) lesions of familial polyposis were believed to be confined to the colonic mucosa. In 1979, Hamilton and associates (3) identified nine patients with ileal adenomas after colectomy for adenomatous polyposis. At our institution, 20 patients with colonic polyposis as their primary indication for total proctocolectomy have had a continent ileostomy created. Recently, we encountered our first patient in whom polyps developed in the continent ileostomy after total proctocolectomy.

CASE REPORT

The patient was seen at the Mayo Clinic at the age of 22 years with a strong family history of polyposis, including documented Gardner's syndrome in his paternal grandfather, father, two uncles, three of four siblings, and two cousins. Although the patient lacked soft-tissue or bony abnormalities, it is likely he had a form of Gardner's syndrome. A proctoscopic examination to 24 cm showed several hundred small polyps. In 1956, he underwent total abdominal colectomy with ileorectostomy. Pathologic examination revealed "innumerable adenomatous polyps" (0.2–2.0 cm), and a grade 1 adenocarcinoma *in situ* was found in the tip of the largest polyp examined. The terminal portion of the ileum was normal.

Proctoscopy in 1966 showed several hundred polyps, which were subsequently fulgurated on numerous occasions. In 1972, a biopsy specimen from one of the polyps showed an adenomatous polyp with villous features, and the patient underwent proctectomy, with creation of a continent ileostomy. Pathologic examination revealed that the rectum contained "multiple, small, sessile tubulovillous adenomas." The resected portion of the ileum showed inflammation, with a sessile adenomatous polyp 5 mm in diameter.

In 1978, endoscopic examination, because of pouch leakage, revealed numerous polyps, mainly in the area of the nipple. One year later, multiple polyps, including a 2-cm polypoid lesion in the nipple, were seen and biopsied. A tubulovillous adenoma was identified. A roentgenogram of the upper gastrointestinal tract and the small bowel revealed that no additional polyps were present. Because of the progression in polyp size and persistent incontinence, excision of the Kock pouch was advised. Accordingly, the continent ileostomy was excised, and a standard Brooke ileostomy was created.

The surgical specimen consisted of a 23-cm length of small intestine, including the stoma and pouch. A sessile tubulovillous adenoma (3.6 × 2.2 × 1.8 cm) was present in the pouch, extending to within 2.5 cm of the stoma (Figure 1). Innumerable smaller mucosal adenomas, ranging in size from 0.9 cm to those perceptible only by microscopy, also were present in the distal ileal component of the pouch (Figure 2). Approximately 30 adenomas large enough to be grossly identified were present. The mucosa between the sites of adenomatous proliferation showed histologic features of normal ileum. There was no evidence of colonic metaplasia; specifically, a normal ratio of goblet to absorptive epithelial cells was present, and clusters of Paneth cells and crypt progenitor cells were identified in the usual distribution (Figure 2). Microscopic as well as gross examination of the proximal portion of the pouch revealed no evidence of mucosal metaplasia or adenomatous change. All polyps appeared to be located in the portion of the pouch created from the most distal portion of the ileum.

DISCUSSION

This case is the first report of adenomas occurring in a continent ileostomy. Hamilton and others (3) have noted similar changes in patients with a Brooke ileostomy after proctocolectomy for famil-

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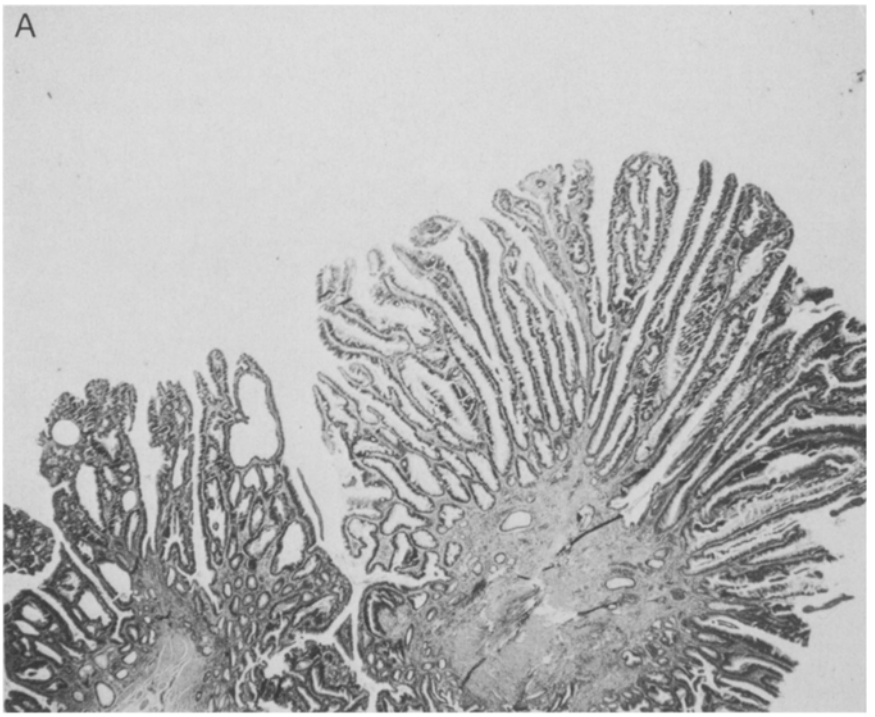

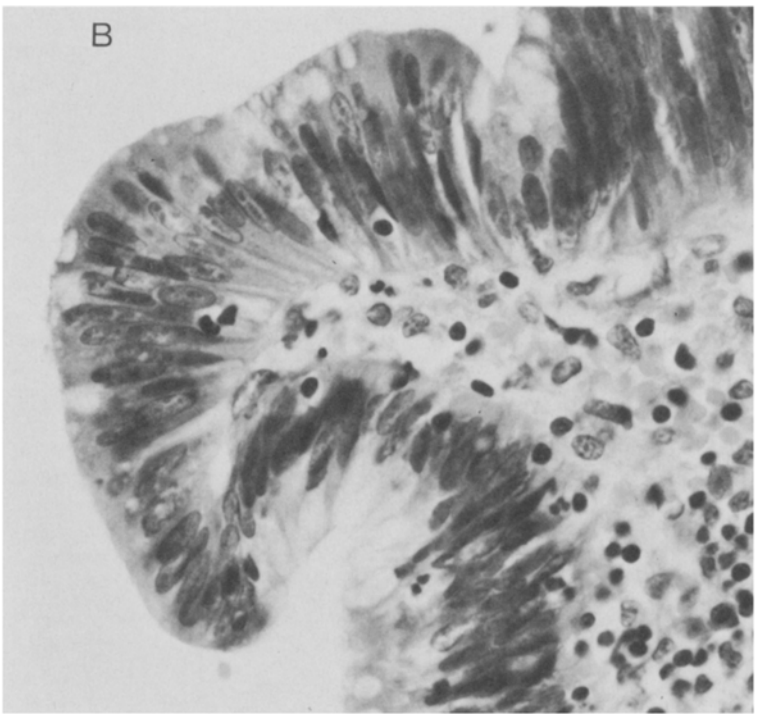

Fig 1. (A) Portion of largest adenoma that was adjacent to stoma, demonstrating tubulovillous epithelial growth pattern. (Hematoxylin and eosin, ×16.) (B) High magnification of largest tubulovillous adenoma demonstrating atypical hyperchromatic cellular composition of adenomatous mucosa. (Hematoxylin and eosin, ×640.)

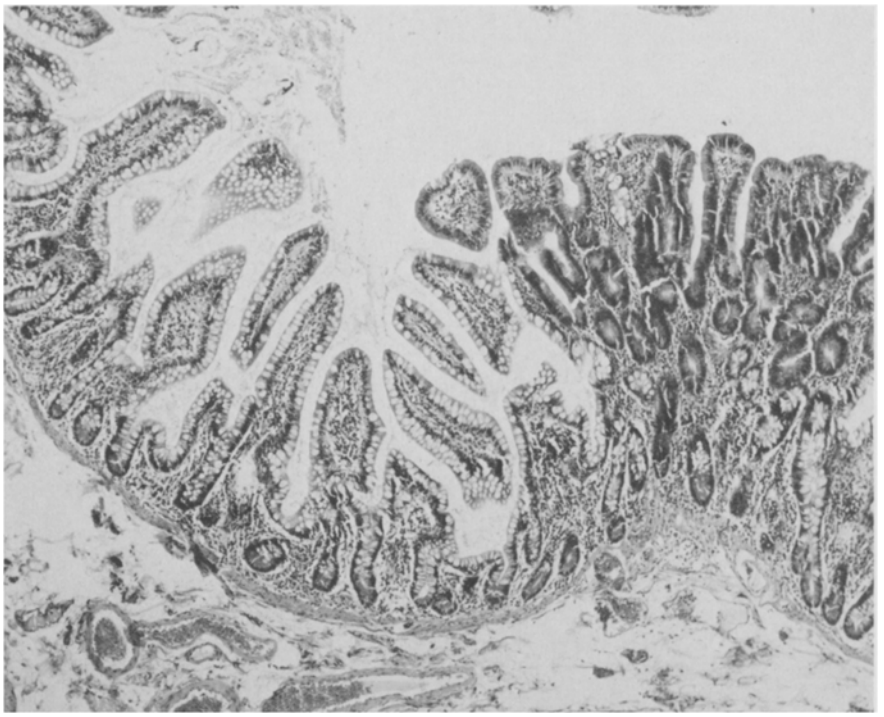

Fig 2. Microscopic focus of adenomatous proliferation in distal ileal component of pouch, approximately 1 mm in diameter (right half of field). Uninvolved mucosa (left half of field) shows normal ileal mucosal histology, with preservation of villous crypt architecture and mixture of goblet cells and absorptive cells. (Hematoxylin and eosin, ×64.)

ial polyposis. Gadacz and others (4) have shown that the ileal mucosa of a reservoir undergoes colonification with time. Such metaplasia, however, was not seen in this specimen and therefore was not responsible for polyp formation. Similarly, the bacterial contents of the pouch come to assume an intermediate level between that of normal ileum and that of normal colon. It has been a concern that these morphologic and microbial changes may be causally related to a premalignant degeneration.

This case demonstrates several features. First, as noted by Hamilton and others (3), the development of tubulovillous adenomas may occur in the ileum of patients with hereditary polyposis. Interestingly, the distribution of adenomas within the pouch seemed to correspond to that part of the pouch created from the terminal portion of the ileum and unrelated to suture lines. The more proximal portions of the pouch did not develop polyps, despite being exposed to the same intestinal milieu. This may be fortuitous, or it may reflect the fact that these polyps tend to develop in the more distal portions of the ileum for some unknown reasons.

Also of concern was the rapid increase in size of these adenomas. The presence of tubulovillous lesions and the premalignant implications of these lesions were of great concern and prompted us to recommend removal of the pouch. It remains unclear if the proliferation of the polyp was a product of the Gardner's syndrome or the pouch itself.

Significant conclusions cannot be drawn on the basis of this one observation, and recommendations cannot be made, other than that patients with familial polyposis who have a continent ileostomy should be followed carefully for production of adenomas in their pouch. If adenomas develop, excision of the pouch would seem reasonable.

SUMMARY

This is a case report of the first description of tubulovillous adenomas in a continent ileostomy following proctocolectomy for Gardner's syndrome.

REFERENCES

1. Yao T, Iida M, Ohsato K, Watanabe H, Omae T: Duodenal lesions in familial polyposis of the colon. Gastroenterology 73:1086–1092, 1977

2. Schnur PL, David E, Brown PW Jr, Beahrs OH, ReMine WH, Harrison EG Jr: Adenocarcinoma of the duodenum and the Gardner syndrome. J Am Med Assoc 223:1229–1232, 1973
3. Hamilton SR, Bussey HJR, Mendelsohn G, Diamond MP, Pavlides G, Hutcheon D, Harbison M, Shermeta D, Morson BC, Yardley JH: Ileal adenomas after colectomy in nine patients with adenomatous polyposis coli/Gardner's syndrome. Gastroenterology 77:1252–1257, 1979
4. Gadacz TR, Kelly KA, Phillips SF: The continent ileal pouch: Absorptive and motor features. Gastroenterology 72:1287–1291, 1977