Ileostomy polyps, adenomas, and adenocarcinomas

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Abstract
Ileostomy polyps are uncommon and poorly described. The aim of this study was to undertake a retrospective clinicopathological review of ileostomy polyps. Seven patients with 60 polyps arising on ileostomies performed for ulcerative colitis were studied. The histopathological evaluation of archival ileostomy biopsy specimens, polypectomy or excision specimens, and clinical review of patient records was undertaken. Fifty of 60 polyps were inflammatory cap polyps and six further polyps were composed of granulation tissue only. They occurred anywhere on the stoma at any time after ileostomy construction and were strongly associated with overt stomal prolapse. Four neoplastic polyps were identified in two patients 27–36 years after ileostomy construction; all occurred at the muco-cutaneous junction. One patient presented with a 2 cm polypoid invasive adenocarcinoma while in the second a 1.7 cm polypoid mucinous adenocarcinoma and a 0.7 cm ileal tubular adenoma with high grade dysplasia occurred at the site of excision of a cap polyp showing focal low grade adenomatous dysplasia six years previously. Neoplastic and non-neoplastic polyps could not be differentiated clinically. It was found that most ileostomy polyps are inflammatory cap polyps associated with stomal prolapse. Less common are polypoid adenomas or adenocarcinomas arising at the muco-cutaneous anastomosis >20 years after ileostomy construction. To prevent ileostomy carcinoma it is recommended that a biopsy of all polyps at the muco-cutaneous anastomosis and of any non-prolapse associated polyps elsewhere on the stoma occurring >15 years after ileostomy construction is done.

Methods
Clinical details were collated on all ileostomates with stomal polypoid lesions presenting to the Department of Surgery at the University Hospital of Wales during 1984–1994. Pathological examination was carried out consecutively by two pathologists (RA and GTW) on routinely prepared 4 μm haematoxylin and eosin stained formalin fixed paraffin wax embedded sections. Elastic and van Gieson’s stains for connective tissue and high iron diamine/Alcian blue staining for the differentiation of sulphated from non-sulphated mucus glycoproteins was carried out on selected cases, the last to assess ‘colonic’ metaplasia of the background mucosa of the ileostomy.

Case reports
Case one
In 1955, a 33 year old man underwent proctocolectomy and ileostomy for ulcerative colitis. Thirty years later, in 1985, a 2 cm polyp

Keywords: ileostomy, inflammatory polyp, ulcerative colitis, prolapse, adenoma, adenocarcinoma.

Protocolectomy with eversion ileostomy is an established surgical treatment for patients with chronic idiopathic inflammatory bowel disease in whom medical treatment has been unsuccessful, and in both ulcerative colitis and familial adenomatous polyposis (FAP) as a prophylactic procedure in cancer prevention. One ileostomal complications are noted in approximately 80% of patients and include skin excoriation, stoma retraction, prolapse, stenosis, abscess and fistula formation, hernia formation, and ileitis. In most cases these are minor and well tolerated. More recently adenocarcinoma arising at ileostomies has been reported as an uncommon complication, usually occurring 20 or more years after stoma construction in patients with either ulcerative colitis or FAP and some of these have presented as ileostomy polyps. Descriptions in published works of other polypoid lesions occurring at ileostomies are very scanty but occasional reports do mention the existence of inflammatory polyps. We present the clinicopathological features of seven patients with ileostomy polyps presenting to a gastrointestinal surgical unit over a period of 11 years.

Figure 1: Two inflammatory ‘cap’ polyps composed of acute on chronically inflamed ileostomy mucosa. The ‘cap’ of inflammatory granulation tissue is particularly well developed in the right hand polyp. Case 1. Haematoxylin and eosin × 20.
that had developed over a period of eight months was removed from the mucocutaneous anastomosis. Histological evaluation was reported as showing an inflammatory polyp. In 1991 an ileal refashioning procedure was performed after stomal prolapse. Macroscopically 6 cm of small bowel was excised and eight polyps were present ranging in size from 2 to 7 mm. In addition two larger polypoid lesions were noted at the mucocutaneous anastomosis site, the larger measuring 17×8 mm and the smaller 7×4 mm. On the small polyps five had microscopic features identical to inflammatory 'cap' polyps of the large intestine (Figs 1 and 2, histological features detailed later) while the other three were composed of vascular granulation tissue only. Background ileal mucosa showed changes interpreted as being caused by mucosal prolapse (Fig 3). Both the larger polyps at the mucocutaneous anastomosis were inflamed neoplastic polyps showing varying degrees of adenomatous dysplasia (Figs 4 and 5). An invasive well differentiated mucinous adenocarcinoma was present in the base of the larger polyp (Fig 5). No lymph nodes were identified in the small amount of mesentery attached to the specimen. On follow up the patient remained well. In 1992 and 1993 a further two and 11 inflammatory cap type polyps respectively were removed from the ileostomy site. No dysplastic change was noted in the epithelium of these polyps. Histopathological review of the original polyp at the mucocutaneous anastomosis showed many features of an inflammatory cap polyp, but in addition there were foci of glandular architectural and cytological atypia that were considered to represent low grade dysplasia (Fig 6).

**Case two**

In 1960, a 28 year old man underwent a total colectomy and ileostomy for severe acute ulcerative colitis. The patient remained well until 1974 when he presented with stomal prolapse. This was treated conservatively at the patient's request until 1994 when increasing pain, prolapse, mucous discharge, and the appearance of multiple stomal polyps prompted a stomal revision. Fourteen cm of ileum bearing 20 pink sessile polypoid lesions was excised. The polyps ranged in size from 3–8 mm and were scattered over an 11 cm length of ileum, sparing only the 3 cm adjacent to the mucocutaneous anastomosis. Histologically, all 20 polyps showed features of inflammatory cap polyps. There was no evidence of dysplasia or malignancy. The background ileostomy mucosa showed mild non-specific chronic inflammation and fibromuscular thickening of the lamina propria in keeping with mucosal prolapse.

**Case three**

A 25 year old woman underwent a total colectomy with ileostomy in 1984 for severe active ulcerative colitis. The following year, prolapse of her ileostomy was noted and a small polyp identified at the stomal margin. Histologically this represented a 3 mm inflammatory cap polyp. A second reconstructive procedure was performed in 1991. No further stomal complications or polyp formation are detailed in the clinical records.
vascular granulation tissue. Stomal prolapse with the development of more polyps occurred in the same year (Fig 7) necessitating an ileal resiting procedure with resection of 8 cm of stomal ileum. Eight ulcerative polyps were noted, ranging in size from 3 to 20 mm. The appearances in all cases were of inflammatory cap polyps. There was no evidence of neoplasia.

Case five
In 1975 a 50 old man with ulcerative colitis underwent a subtotal colectomy and proctectomy with colostomy formation. The following year a paracolostomy hernia and prolapse developed. In 1984 a completion colectomy with ileostomy was performed. Over the following year two 5 mm polyps were removed from the ileostomy and biopsy showed in both cases vascular granulation tissue. In May 1992 a further 10 mm polyp was excised from the ileostomy site. Histologically this was an inflammatory cap polyp.

Case six
A 38 year old man underwent a panproctocolectomy and ileostomy for ulcerative colitis in 1961. The patient remained well until 1986 when he developed myelodysplasia. In 1988 a 2 cm exophytic lesion developed at the ileostomy site. Attempted excision biopsy was performed and showed a well differentiated ileal adenocarcinoma occurring at the mucocutaneous anastomosis and invading the underlying muscularis propria and the adjacent skin and subcutaneous tissue (Fig 8). The tumour had been incompletely excised and further excision of the tumour and resiting of the stoma was undertaken. Again local tumour excision was incomplete and in February 1991 recurrent adenocarcinoma was confirmed at the original ileostomy site. Radical excision of the right abdominal wall was now performed but the patient died on the sixth postoperative day after an extensive intra-abdominal haemorrhage. Necropsy examination showed no residual tumour.

Case seven
In November 1989 a 77 year old woman presented with ileostomy bleeding and peristomal excoriation 19 years after proctocolectomy for ulcerative colitis. Local stoma care resolved the initial problems but further excoriation of the stoma with an 8 mm bleeding polyp developed in September 1992. Histological examination of the excised lesion showed an inflammatory cap polyp with no evidence of dysplasia or malignancy.

Clinical and pathological features (Table)
The study group comprised five men and two women. In all cases the original surgery was performed for ulcerative colitis and the ileostomies were of Brooke type. A total of 60 polyps were identified, ranging in size from 3
Ileostomy polyps, adenomas, and adenocarcinomas

Clinical pathological features of seven patients with ileostomy polyps

<table>
<thead>
<tr>
<th>Case</th>
<th>Age*</th>
<th>Sex</th>
<th>Presenting symptoms</th>
<th>Oncset</th>
<th>Histological type</th>
<th>Maximum size (mm)</th>
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<tr>
<td>1</td>
<td>63/M</td>
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<td>30</td>
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<tr>
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<td>Ileostomy polyp</td>
<td>22</td>
<td>Cap</td>
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*At diagnosis of ileostomy polyp, fyears after ileostomy construction; **see text.

evidence of a preinvasive lesion, either flat dysplasia or a raised adenoma, was seen in this case. The tumour was deeply invasive into the wall of the ileum and the adjacent skin and subcutaneous tissue and complete local excision was only achieved by radical excision of the abdominal wall, from which the patient did not recover. The second (case 1) was an invasive mucinous adenocarcinoma that was clearly arising in an inflamed tubular adenoma (Fig 5), again at the mucocutaneous anastomosis. A second, distinct polypoid adenomatous lesion was also present nearby: this showed high grade dysplasia but no unequivocal invasive malignancy (Fig 4). A polyp removed six years earlier from this site, while having the overall features of an inflammatory cap polyp, also showed cytological and architectural features of low grade glandular dysplasia particularly at the mucocutaneous anastomosis (Fig 6), although this was only recognised on retrospective review. No evidence was obtained from high iron diamine/Alcian blue staining to suggest that the neoplastic polyps had arisen with colonic metaplasia of the ileostomy mucosa.

Neoplastic polyps appeared in the two patients at 27 and 36 years after ileostomy construction. Non-neoplastic cap polyps appeared at any time, ranging from 1–38 years after the original surgery.

Discussion

The findings of this study, taken in the light of previous publications, show that polypoid lesions occurring on ileostomies are of two broad types: inflammatory cap polyps that are related to stomal prolapse, which are frequently multiple and which may occur at any site on the ileostomy, and less frequent neoplastic polyps—adenomas or adenocarcinomas—that occur at or close to the mucocutaneous anastomosis more than 15 years after ileostomy construction. The original surgery in all our cases was for ulcerative colitis, but patients with FAP may be equally affected. While the clinical appearances of the two types of polyp were similar, microscopic examination allowed a clear distinction to be made in all but one polyp (case 1), which showed a focus of adenomatous dysplasia in an otherwise typical inflammatory cap polyp arising at the mucocutaneous anastomosis.

Originally described in the rectosigmoid colon, inflammatory cap polyps have also been identified at the site of haemorrhoids, prolapsing colostomies, diverticular mucosal folds, in the anorectal region, and in association with the 'solitary rectal ulcer' syndrome. There are compelling reasons to believe that they are manifestations of mucosal prolapse. Inflammatory cap polyps of the small intestinal mucosa have not been described previously as such, although we are aware of reports of an 'inflammatory myoepithelial polyps' of intussuscepting ileum that had similar morphological features and of polypoid mucosal prolapse in a pelvic ileal reservoir. The histological features to 20 mm. Multiple polyps were found in four of seven cases, in all of which there was overt stomal prolapse. Two polyps were associated with stomal bleeding and skin excoriation.

Grossly all of the polyps appeared as pink lobulated sessile lesions. Of the 60 polyps identified, 50 had histological features similar to inflammatory cap polyps of the large intestine. They were composed of actively inflamed ileal mucosa with villous loss, crypt elongation, and striking capillary vascular proliferation in the superficial lamina propria, often forming a 'cap' of inflammatory granulation tissue (Figs 1 and 2). Admixed fine fascicles of smooth muscle extending between the crypts from the muscularis mucosae were noted throughout the polyps and fibrohyaline thickening of the lamina propria was seen in six. Surface erosion with or without an overlying cap of inflammatory granulation tissue was identified in 36 of 50 polyps. Background ileal mucosa adjacent to the polyps frequently showed changes similar to that seen in colorectal mucosal prolapse (Fig 3) with blunting or loss of villi and hyperplasia, dilatation or tortuosity of crypts. The surface epithelium was often attenuated, eroded or displayed focally a serrated pattern similar to that seen in colorectal metastatic (hyperplastic) polyps. There was no evidence of sulphomucin production to suggest colonic metaplasia. Biopsy specimens of six of 60 polyps were composed of vascular granulation tissue with variable degrees of acute inflammation only, and it is probable that these also represented the superficial portions of inflammatory cap polyps. Cap polyps occurred anywhere on ileostomy mucosa (Fig 7) with no particular predilection for the vicinity of the mucocutaneous anastomosis. They were strongly associated with overt stomal prolapse, which was reported clinically in five of six patients with cap polyps. They were also commonly multiple, in that four patients with prolapse had three, eight, 10, and 20 cap or granulation tissue polyps respectively. Two of four neoplastic ileostomy polyps were invasive adenocarcinomas. One (case 6) was a well differentiated tubular adenocarcinoma (Fig 8) occurring at the mucocutaneous junction in a patient with stomal retraction. No
in the background ileal mucosa of our cases were similar to those of rectal mucosal prolapse and we believe that the 50 cap polyps we describe on prolapsing ileostomies bearing an eversion fashioned stoma have a similar aetiology. Six polyps were composed entirely of granulation tissue. All arose in ileostomies featuring prolapse and as granulation tissue is frequently one of the hallmarks of the superficial zones of cap polyps, 10, 11 we consider it probable that these polyps are essentially similar in nature to the 50 more typical examples.

Adenocarcinoma arising at ileostomy sites is uncommon. We have been able to find only 20 cases in the English medical literature since 1967 8 and in this series two further cases are added. In many of the reported cases, as in those described here, the tumour has presented as an exophytic mass adjacent to the mucocutaneous junction. Histological examination usually shows a well differentiated mucinous adenocarcinoma, often with a favourable prognosis after adequate local excision and stomal resiting. 11 The aetiology of the tumour is unclear but the long latent period between ileostomy construction and the appearance of neoplasia suggests that longstanding regenerative epithelial hyperproliferation caused by chronic irritation at the mucocutaneous anastomosis, perhaps from trauma around the flange or chemical agents used as stoma adhesives, may be important. Changed bacterial flora 16 within and around the ileostomy may also play a part. A predisposition to small intestinal neoplasia may be contributory in patients with underlying FAP but not in those with ulcerative colitis, although examples of adenocarcinoma of the terminal ileum attributed to chronic 'backwash' ileitis are described in colitis patients. 17, 18 In some reported cases of ileostomy carcinomas so called 'colonic' metaplasia of the ileal epithelium has been noted adjacent to the invasive lesion 5 suggesting a prior metaplastic step in the carcinogenic process: we were unable to show this in our two cases. More significantly, a small number of cases in published reports have described preinvasive epithelial dysplasia adjacent to the carcinoma 5 and in one of our cases (case 1) invasive mucinous adenocarcinoma was clearly arising in a pre-existing adenoma, showing that at least some ileostomy carcinomas may be prevented by early recognition and extirpation of polypoid adenomas. Moreover a 20 mm cap polyp showing focal low grade adenomatous dysplasia at the mucocutaneous junction had been removed from the site of this tumour six years previously. While it is possible that this tumour arose within the cap polyp, dysplasia has not been reported previously in colorectal cap polyps. It is perhaps more probable, therefore, that this polyp had arisen on the basis of prolapsing adenomatous tissue.

It is of interest that in all of our patients with ileostomy polyps the original surgery was for ulcerative colitis despite the fact that the cohort of ileostomates reviewed also contained many patients with Crohn's disease. However, the number is much fewer and the length of follow up shorter than for those with ulcerative colitis.

These factors may well explain the absence of neoplastic lesions in Crohn's disease patients and no conclusion can be drawn regarding whether adenomatous lesions might affect ileostomies in Crohn's disease. The absence of Crohn's disease patients from the larger number of ileostomates with cap polyps may be more significant, reflecting the clinical experience that ileostomy prolapse is less common in Crohn's disease where ileostomy retraction and complications of recurrent Crohn's disease are a greater problem.

The latency period between ileostomy construction and the appearance of stomal adenocarcinoma in most previously published cases has been approximately 25 years 6 and the two cases presented here, with periods of 27 and 36 years respectively, are similar. Ileostomy cap polyps by contrast, being related to local mucosal prolapse, may appear at any time. It has been suggested that ileostomy adenocarcinoma may be seen increasingly frequently in the future, as the ileostomy population grows, and as ileostomy increases in the population. 5 As a strategy for the prevention of ileostomy carcinoma we therefore recommend biopsy of all polyps appearing at the mucocutaneous anastomosis, and of any polyps elsewhere on the stoma that are not associated with prolapse, in patients who have ileostomies of more than 15 years duration.