A rare case of a 52-year-old man with multiple comorbidities including a history of ulcerative colitis and subsequent total proctocolectomy with end ileostomy was presented for the evaluation of persistent papules around his stoma. Biopsy confirmed squamous epithelium with areas transitioning to enteric mucosa consistent with intestinal metaplasia around his ostomy site.

Intestinal metaplasia is an uncommon benign cellular transformation that can occur in patients with an ostomy.1-4 The etiology remains elusive, but chronic irritation and trauma have been suggested as possible risk factors.5 We present an uncommon case of intestinal metaplasia without adenomatous or dysplastic changes arising along the periphery of an ostomy site in a patient with ulcerative colitis.

1 INTRODUCTION

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2 REPORT OF A CASE

A 52-year-old man with a history of deep venous thrombosis, iron-deficiency anemia, gastroesophageal reflux disease well controlled on proton pump inhibitors, and ulcerative colitis (diagnosed at age 13 and is status-post total proctocolectomy with end ileostomy at age 15) presented to our clinic with a chief complaint of bumps adjacent to his stoma that had been present for less than 6 months. He denied any changes in size or pigmentation. He denied symptoms of pruritus, tenderness, bleeding, or purulent drainage. On review of systems, he denied fevers, chills, oral ulcerations, weight loss, dysphagia, odynophagia, nausea, or vomiting. His ostomy output had been normal, without blood in the effluent in the past 5 years. Patient had no personal history of cancers, but admitted a family history of cutaneous malignancy, specifically basal cell carcinoma in sister, brother, uncle, and father. On his physical examination, his abdomen was soft, flat, nondistended, and nontender, with good bowel sounds in all 4 quadrants. No intraabdominal masses or hepatosplenomegaly were appreciated. However, there were firm mildly hyperkeratotic polypoid nonulcerated papules in the peristomal area of the right lateral abdomen (Figure 1). Differential diagnosis included granulation tissue, verruca vulgaris, and malignancy. A shave biopsy was obtained, and histologic examination revealed squamous epithelium with areas transitioning to enteric columnar mucosa (Figure 2, Figure 3). No evidence of malignant cells was identified on the tissue sections. The pathology finding

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was deemed consistent with ostomy site-associated intestinal metaplasia. No further treatment was necessary for the patient since there were no dysplastic features on histology. For management and follow-up, the results were discussed with the patient’s gastroenterology team to educate on the risk of malignant transformation and encourage close surveillance.

FIGURE 1 Hyperkeratotic polypoid papules on the lateral aspect of the ileostomy site in right lateral abdomen

FIGURE 2 Hematoxylin-eosin staining of biopsy specimen showing colonic glands merging with the squamous epithelium of the skin (magnification 40×)

FIGURE 3 Hematoxylin-eosin staining of biopsy specimen showing colonic glands merging with the squamous epithelium of the skin (magnification 100×)

3 | DISCUSSION

There are numerous indications for ostomy creation in complex acute or chronic gastrointestinal disorders, including traumatic injury, inflammatory bowel disease, and malignancy. Despite diligent stoma care, reports of stomal and peristomal complications such as stenosis, prolapse, retraction, parastomal hernia, parastomal dermatitis, and parastomal bleeding are commonly reported. There have been few reports of intestinal metaplasia without adenomatous or dysplastic changes adjacent to an ostomy site. Most of these cases occurred approximately 10 years after ostomy placement, similar to our patient. Predisposing factors that influence cutaneous to intestinal metaplasia remain largely unknown. It has been postulated that chronic inflammation, dynamic change in host bacterial population, or dissemination of intestinal tissue during surgery may play a role.

Although rare, some of the reported cases of intestinal metaplasia along ostomy sites have displayed concomitant adenoma or dysplasia in cases of peristomal adenocarcinoma and/or squamous cell carcinoma. As such, close clinical monitoring is imperative to identify malignancy early and improve patient morbidity/mortality rates. Clinical presentations reported in patients with ileostomy carcinomas include palpated mass, small bowel obstruction, peristomal bleeding, pain, stomal prolapse, or retraction. Evidence of these symptoms should warrant a biopsy or immediate referral to gastroenterology for imaging.

4 | CONCLUSION

Intestinal metaplasia can occur as a parastomal complication. Fortunately, the risk of malignancy is low, but this possibility
necessitates close clinical surveillance. We recommend diligent regular monitoring at a minimum of 10-15 years post-stomy creation as majority of the malignant transformations have occurred within this timeframe. Clinicians should maintain a low threshold for obtaining a biopsy of a parastomal lesion as early identification is imperative in decreasing the risk of malignant conversion.

CONFLICT OF INTEREST
The authors declare that there are no conflicts of interest regarding the publication of this paper.

AUTHOR CONTRIBUTIONS
NSU: is a primary case report author and reviewed the literature. MAS: performed histopathology evaluation. MWN: reviewed the case report and is a editor.

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