Royal Australasian College of Surgeons

Primary adenocarcinoma arising from an ileostomy site: a late complication of end ileostomies

A 55-year-old male was referred for a gradually increasing large parastomal hernia and difficulty managing his stoma. He had a background of familial adenomatous polyposis (FAP) requiring total colectomy and ileorectal anastomosis in 2010 with further completion proctectomy and end ileostomy due to poor compliance with rectal surveillance and high-risk rectal lesions in 2012. He was non-compliant with FAP surveillance. On examination, there was a large polypoidal lesion measuring 6 × 5 cm at the ileostomy site, separate from the mucocutaneous junction (Fig. 1).

A partially reducible parastomal hernia was also evident with no obstructive symptoms. A CT scan delineated the hernia and the extension of this lesion (Fig. 2). We subsequently proceeded with an elective resection of the lesion with parastomal hernia repair. Intraoperatively, a 5 mm margin of peristomal skin was taken and 5 cm of ileum resected proximal to the lesion. The uncertainty regarding the nature of the lesion led us to primary closure of the parastomal hernia with interrupted mattress nylon sutures, refashioning of stoma and subcutaneous drain (fig. 3). His post-operative stay was uneventful. Histopathology revealed a well differentiated adenocarcinoma arising from high grade dysplasia which measured 60 x 45 x 30 mm with ulceration but no evidence of vascular invasion, 8 mm from the nearest margin.

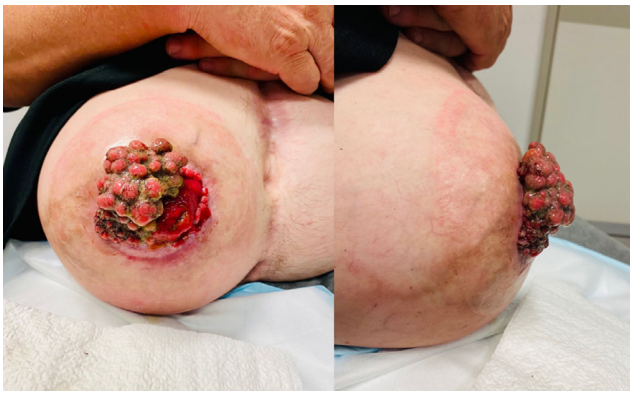

Fig. 1. Exophytic polypoid lesion arising from the end ileostomy, separate from the mucocutaneous junction and large parastomal hernia.

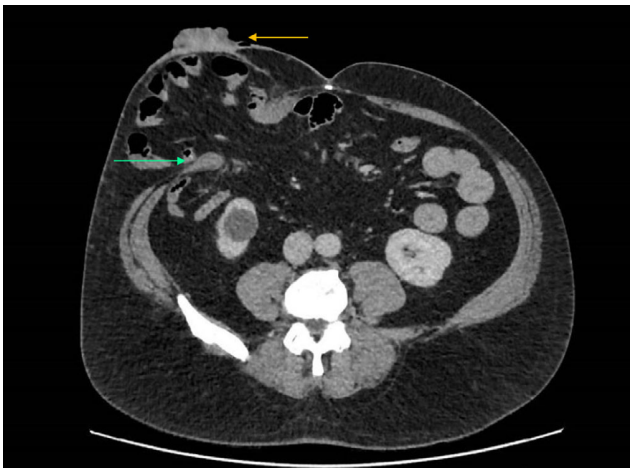

Fig. 2. Axial phase CT scan demonstrating superficial soft tissue lesion at end ileostomy site (yellow arrow) and a non-obstructed parastomal hernia with a wide neck (green arrow).

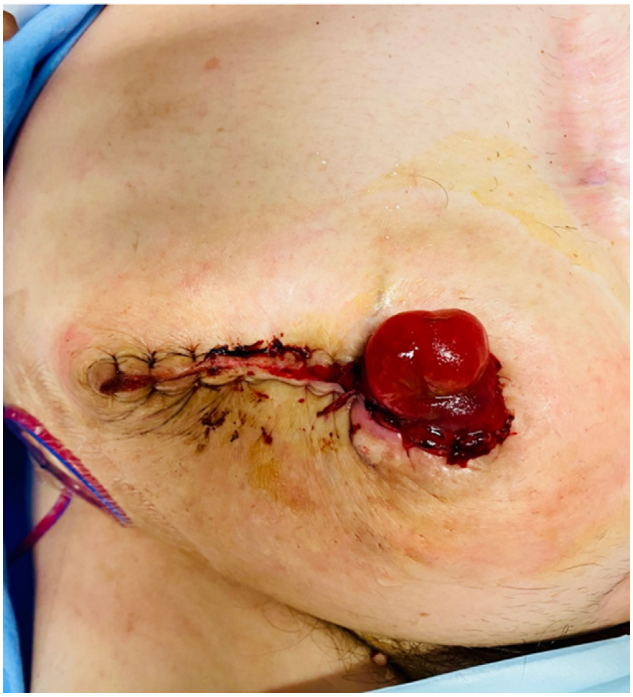

Fig. 3. Post en-block excision of lesion (including peristomal skin and 5 cm of distal small bowel), parastomal hernia repair and refashion of ileostomy.

Colorectal multidisciplinary meeting discussions recommended surgical surveillance.

We report a rare case of primary adenocarcinoma arising from an ileostomy site. In 2008, Quah and colleagues reported a total of 46 such cases described worldwide.[1] Though our case was related to FAP, the condition has also been reported in the context of ulcerative colitis. The first case of adenocarcinoma arising from an ileostomy site was reported in 1969 by Sigler and Jedd[2] in a patient who underwent pan-proctocolectomy for ulcerative colitis. The first case related to FAP was described in 1982 by Roth and Logio, occurring 7 years post formation of ileostomy.[3]

Metzger and colleagues analysed the time frame of developing adenocarcinoma and found an average of 27 years following placement of the ileostomy[4]; in our case, there were less than 10 years. The pathophysiology of occurrence appears to be secondary to chronic chemical and physical irritation of the ileal mucosa, resulting in ileal metaplasia or dysplasia, which progresses to adenocarcinoma if unchecked.[1,4]

Controversy arises on how best to surveil patients for this condition given its rarity, there are grim consequences if not treated early. Some studies have suggested yearly ileoscopy for patients who have had their stoma for more than 15 years[4]; however, as in our case, this condition can occur before this time frame. Therefore, we recommend yearly physical examination by a stoma nurse, biopsy of any suspicious lesions and if low grade dysplasia is detected, then yearly ileoscopy. In the presence of high-grade dysplasia and adenocarcinoma, we recommend discussion at colorectal MDT followed by en-block resection and refashion of stoma.

Author contributions

Mathew Ollapallil Jacob: Conceptualization; data curation; supervision; writing – original draft; writing – review and editing. **Kugendran Ponniah:** Data curation; writing – original draft; writing – review and editing. **Bhovineey Ramanathan:** Visualization; writing – original draft; writing – review and editing. **Jimmy Eteuati:** Conceptualization; supervision.

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