Squamous cell carcinoma at an ileostomy site—Fifty-four years following colectomy for ulcerative colitis: A case report and literature review

Farshid Ejtehadi *, Metin Nizamoglu, Rangasamy Sivakumar

Mid Yorkshire Hospitals NHS Trust, Colorectal Unit, United Kingdom

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A B S T R A C T

INTRODUCTION: Carcinoma arising at an ileostomy site is an extremely rare occurrence. The rate of malignancy arising at an ileostomy site is reported as being 2–4 of every 1000 cases. The development of squamous cell carcinoma at the mucocutaneous junction of an ileostomy is extremely rare. PRESENTATION OF CASE: We present a case of a 76-year-old male who developed squamous cell carcinoma at an ileostomy site fifty-four years after total colectomy as management for ulcerative colitis. DISCUSSION: Our literature review has identified only four similar cases previously published in English literature. All cases of squamous cell carcinoma developing in ileostomy have occurred after a minimum of twenty-six years following ileostomy. This suggests that the etiology may be due to chronic factors. CONCLUSION: Patients with chronic stomal inflammation, bleeding or persistent induration and/or mass formation should be followed up closely and investigated for recurrence or development of a new malignancy. There should be a low threshold to obtain an early definitive tissue diagnosis by taking biopsies to prevent local or systemic invasion.

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1. Introduction

The rate of malignancy arising at an ileostomy site is reported as being 2–4 of every 1000 cases,1 making it very rare. Adenocarcinoma of the ileostomy site is well described in the literature, particularly in patients with inflammatory bowel disease or familial adenomatous polyposis.

The development of squamous cell carcinoma (SCC) at an ileostomy site is exceptionally rare. A literature search for cases of SCC of the ileostomy site identified four SCC occurring at end ileostomy following total colectomy and one at the mucocutaneous junction of an ileal conduit following cystoprostatectomy.

We present a case of SCC developed at the ileostomy site after total colectomy and ileostomy formation.

2. Case report

A 76-year-old Caucasian man who had undergone a total colectomy and an end ileostomy fifty-four years ago for ulcerative colitis was referred to us following development of an ulcerative lesion arising from the ileostomy, extending to the abdominal wall tissue and producing pain, discomfort and bleeding for several months.

The patient had parastomal mass for several months, which was dealt with by various community stoma teams as an asymptomatic parastomal hernia. The mass also was causing pain and intermittent bleeding. The symptoms were thought to relate to para-stomal hernia and stoma-related granulation, both of which are very common in patients with long-term stoma. Consequently, he was referred to surgical clinic for evaluation and assessment of his parastomal symptoms.

His other past medical history were appendectomy, gout, hypertension and chronic obstructive pulmonary disease. He was a retired mineworker, and a non-smoker with moderate alcohol intake. His exercise tolerance was limited to twenty-five yards; hence he mobilizes using a wheelchair.

Biopsies taken from this lesion, at the time of original referral, revealed skin tissue which was widely infiltrated by a well-differentiated squamous cell carcinoma extending down from the epidermis. No peri-neural or lympho-vascular invasion was identified (Fig. 1). He also had a CT scan of his chest, abdomen and pelvis that did not show any metastasis. Biopsy and CT staging were discussed at the local multidisciplinary team (MDT) meeting (Fig. 2).

With regards to considering wide local excision and re-siting the stoma, because the cancer was already at an advanced stage and the patient’s co-morbidities included severe COPD and 25 yards exercise tolerance, he was considered unfit for major reconstructive surgery. Due to his co-morbidities and physical state, the meeting resulted in a decision to proceed with radiotherapy to prevent occlusion of his ileostomy. He underwent radiotherapy to his abdominal wall (30 gray in 10 fractions). The patient had a very
Table 1

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Fig. 1. A large ulcerating squamous cell carcinoma of the ileostomy site. The photograph was taken a year after diagnosis following 2 cycles of palliative chemotherapy.

Fig. 2. Photomicrographs of skin tissue which is widely infiltrated by a welldifferentiated squamous cell carcinoma extending down from the epidermis; hematoxylin–eosin stain 250×.

good clinical response to the radiotherapy, but due to recurrence of the tumor he had to have the second course of radiotherapy within six months (5/20 in 5 fractions). Despite these measures the tumor continued to progress. Hence he was discharged into the community with palliative care team support.

3. Discussion

Squamous cell carcinoma developing at the ileo-cutaneous junction following ileostomy is a rare complication. Our literature review has identified four cases previously published. The first reported case was of a primary squamous cell carcinoma occurring on a skin-grafted ileostomy stoma twenty-six years following a procto-colectomy for Crohn’s. The second case described a para-stomal squamous cell carcinoma involving an ileostomy forty-four years after a procto-colectomy for ulcerative colitis. The third case involves squamous cell carcinoma developing around an ileostomy forty-one years after a procto-colectomy and formation of an end ileostomy for a patient with ulcerative colitis. The fourth case again described squamous cell carcinoma developing in an ileostomy fifty-one years following procto-colectomy for severe ulcerative colitis. All four patients were treated with radical excision and re-siting of their stoma without major complication.

There has also been a case of primary squamous cell carcinoma at the mucocutaneous junction of an ileal conduit twenty-seven years following a radical cystoprostatectomy and ileal conduit formation. This was managed by soft tissue excision and reconstruction of the abdominal wall.

Table 1 summarizes the demographic data, underlying diagnosis, initial operation and time scale following surgery of patients presenting with SCC at an ileostomy site.

All four cases of squamous cell carcinoma developing at ileostomy have occurred within a minimum of twenty-six years following ileostomy formation, suggesting a chronic pathogenic process after many years of repetitive physiological stress. The exact mechanism is unknown however this may be due to recurrent irritation. Ulcerating squamous cell carcinoma arising in an area of chronic irritation is commonly known as Marjolin’s ulcer. The various aetiologic mechanisms include release of local toxins after injury, induction of dormant neoplastic cells, and activation of injury-induced pre-neoplastic cells by a co-carcinogen. Chronic stomal irritation secondary to prolonged exposure to toxins, such as stool and/or urine, or recurrent infections can cause a localized response resulting in metaplasia. Metaplasia is not cancerous; however, it is a pre-malignant condition that does not of itself require therapy but requires monitoring for progression to cancer with removal of risk factors of cancer formation, if possible.

The cases stress the need for appropriate education and training in stoma care to prevent chronic irritation and for early identification of problems. Furthermore, bulging at the stoma site should not be assumed to be para-stomal hernia without appropriate clinical examination or further investigation. Also, patients who have chronic stomal inflammation, bleeding or persistent induration and/or mass formation should be investigated for recurrence or development of a new malignancy. There should be a low threshold to obtain an early definitive tissue diagnosis to prevent local or systemic invasion. Therefore, obtaining a biopsy of any abnormal tissue at the ileostomy or ileo-cutaneous junction is recommended, particularly in cases with changes in a long-term stoma, to facilitate early diagnosis of this rare complication.

Conflict of interest statement

No conflict of interest.

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Ethical approval

A copy of the written consent is available on request.

Author contributions

F. Ejtehadi and M. Nizamoglu were involved in the case study and review of the literature, R. Sivakumar was the responsible consultant and was involved in revision of manuscripts.

References