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Title: Malignant transformation of an ileostomy stoma scar – an unusual presentation

Short title: Malignant stoma scar

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This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: [10.1111/ans.16171](https://doi.org/10.1111/ans.16171)

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Number of figures: 3

Text word count: 748

Author Manuscript

Case report

A 45-year-old female presented to an external hospital with six weeks of left abdominal wall swelling and pain. She had a history of gastroschisis and had undergone multiple surgeries by the age of six months to achieve abdominal closure, including ileostomy formation and reversal. She was otherwise well and denied any history of recent trauma. Examination of the abdominal wall at the time revealed normal appearing skin and mature scars.

Figure 1

A computed tomography (CT) scan showed two rim-enhancing collections located within the abdominal wall, situated deep to the scar from her previous stoma. The patient underwent radiologically guided percutaneous drainage of the collections with insertion of a pigtail drain. However, the collections recurred within a week of drain tube removal and open surgical debridement was subsequently performed. Two purulent, communicating cavities were found and debrided. Culture of their contents returned no growth but histological examination showed fibrofatty tissue and skeletal muscle, including the wall of a cavity lined by organising fibrin and granulation tissue. There was a mixed acute and chronic inflammatory infiltrate with fibrosis, dystrophic calcification and focal atypical epithelial proliferation, raising the possibility of adenocarcinoma.

Further imaging with magnetic resonance exhibited a 36x36x27mm subcutaneous tumour involving the left external oblique, internal oblique and transversalis but not communicating

with the abdominal cavity. In a search for occult primaries, upper and lower gastrointestinal endoscopy, cystoscopy and pelvic ultrasound were performed and were all normal. Tumour markers were also within normal limits.

Figure 2

After referral to a quaternary cancer centre and discussion at a multidisciplinary meeting, the patient underwent wide resection of the tumour with full thickness resection of the abdominal wall. This included peritoneum which, given prior surgery, was adherent to the abdominal wall. Clear margins were confirmed on frozen sections. The defect was reconstructed with a composite polyester/collagen mesh and a fasciocutaneous rotational flap. Formal histopathology from this excision altered the diagnosis to a poorly differentiated squamous cell carcinoma (SCC).

Figure 3

Discussion

There were two unusual aspects of this cases that warrant report. The first revolves around the origin of SCC deep within the abdominal wall. We hypothesise that this case represents a Marjolin's ulcer that has arisen within chronic scarring. To our knowledge, this is the first report of such a case in an old stoma site.

Marjolin's ulcer refers to formation of malignancy in a chronic scar, classically arising from burns, previous skin grafts or chronic osteomyelitic sinuses¹. SCCs are the most common subtype although basal cell carcinomas, melanomas, sarcomas, histiocytomas, schwannomas and mesenchymal tumours have all been reported². The pathogenesis of Marjolin's ulcers is multifactorial and likely includes environmental and genetic factors. The recurrent irritation and ulceration of an unstable scar stimulates cell proliferation and spontaneous mutations³ whilst also inciting a chronic inflammatory process; this "smouldering" inflammation is now recognised as a key factor in the tumour microenvironment and promotes neoplastic initiation, proliferation and metastasis⁴⁻⁶. This association can be seen in other cancers such as gastric cancer with *H. pylori* infection, colon cancer with inflammatory bowel disease and cervical cancer with some strains of HPV⁷. The theory of an "immunologically privileged" site has also been postulated, where the obliteration of local lymphatics through injury enables developing malignancies to circumvent immune-surveillance mechanisms⁸. Mutations in p53 and Fas with the potential to disrupt apoptotic and homeostatic processes have also been identified^{9,10}.

The second unusual aspect of this case was the presentation with a subcutaneous collection. Most reported Marjolin's ulcers arise as chronic nonhealing ulcers in areas of previous trauma³. Due to the unusual presentation, the patient underwent drainage and debridement and had a delayed diagnosis. This case highlights that malignant transformation can occur in any chronic scar and a high index of suspicion is required when investigating any abnormal scar transformations.

From a reconstructive perspective, vascular supply to the anterior abdominal wall following gastroschisis has not been described and in this scenario was made even more challenging by significant scarring from multiple previous surgeries. A pre-operative CT angiogram identified the presence of a deep inferior epigastric artery (DIEA) on the ipsilateral resection side but not on the contralateral lower abdominal wall, demonstrating an aberrant blood supply to the anterior abdominal wall. As such, the rotational flap used to close the defect was based inferomedially on the existing DIEA and its perforators, ignoring the existing midline laparotomy scar.

Conclusion

- Marjolin's ulcers are uncommon but a high index of suspicion should always be held when scars present with indolent transformations, even in the absence of cutaneous ulceration.

Disclosure statement

The authors declare that there is no conflict of interest regarding the publication of this article.

Figure legends

Figure 1: Scarred abdomen with discharging sinus in left upper quadrant

Figure 2: MRI image demonstrating intramuscular component of left abdominal wall malignancy

Figure 3: Intra-operative defect demonstrating full thickness abdominal wall defect and closure with composite polyester/collagen mesh and a fasciocutaneous rotational flap

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ANS_16171_Figure 2.PNG

ANS_16171_Figure 3.PNG

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Title:

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Date:

2020-07-16

Citation:

Lu, D., Zinn, R., Mitchell, C., Ryan, A. & Gyorki, D. (2020). Malignant transformation of an ileostomy stoma scar: an unusual presentation. ANZ JOURNAL OF SURGERY, 90 (9), pp.1770-1772. <https://doi.org/10.1111/ans.16171>.

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