

Sarcomatoid squamous cell carcinoma with overlying Bowen's disease in a patient presenting with chronic toe ulceration in diabetes

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Key words

- Diabetic foot
- Non-healing ulcer of toe
- Osteomyelitis
- Squamous cell carcinoma of toe

Article points

1. It is important to send histology, as well as microbiology, if ulceration looks abnormal.
2. The patient history is as important as the clinical examination.

Authors

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Primary cutaneous sarcomatoid carcinoma, also known as spindle cell carcinoma, is an exceedingly rare and aggressive malignancy. Bowen's disease is a rare, slow growing skin disorder that presents as a scaly and red cutaneous patch. The authors report a case of sarcomatoid squamous cell carcinoma with overlying Bowen's disease presenting as a non-healing traumatic ulcer of toe in a patient with type 2 diabetes.

Primary cutaneous sarcomatoid carcinoma, also known as spindle cell carcinoma, is an exceedingly rare malignancy accounting for only 1% of squamous cell carcinomas (SCC) (Wernheden et al, 2022). It is a highly aggressive variant characterised by biphasic epithelial and mesenchymal components of uncertain pathogenesis and prognosis (Lee, 2020). Bowen's disease is a rare, slow-growing skin disorder that presents as a scaly, red cutaneous patch.

Here, we report a case of sarcomatoid squamous cell carcinoma with overlying Bowen's disease presenting as a non-healing traumatic ulcer of toe in a patient with type 2 diabetes.

Case presentation

A 75-year-old man with type 2 diabetes presented to community podiatry in July 2021 with a year-long history of a non-healing ulcer to his left third toe. He reported having dropped a chair on it and it had failed to heal since then.

He was initially treated with antibiotics and simple dressings as an outpatient. Serial clinical photographs were taken over this period (*Figure 1*). The first X-ray identifying loss of bone was taken in August 2021 and reported as showing bony destruction of terminal phalanx and middle phalanx of left third toe. He was seen every 2 weeks by podiatry to monitor progress. Serial X-rays were

performed (*Figure 2*). He had a 6-week course of antibiotics to treat the suspected osteomyelitis.

He was seen in the diabetic foot multidisciplinary clinic on August 6, 2021 and the plan was to change footwear to relieve recurrent trauma and facilitate a trial of healing.

He was discussed again at a virtual meeting on October 28, 2021, and the decision was for phalangeal amputation of toe due to chronic hypergranulation and increasing pain from the toe.

A repeat X-ray in November, prior to surgery, showed further erosive changes of the medial aspect of the head of the proximal phalanx.

On December 16, he attended the day-case unit for phalangeal amputation. On closer inspection pre-operatively the wound looked less like overgranulation and more like a melanoma. The surgery was performed as planned (*Figure 3*).

Deep tissue was sent for histology and microbiology. Microbiology samples grew *Proteus mirabilis*, *Staphylococcus aureus* and *Pseudomonas aeruginosa*. Skin margins demonstrated sarcomatoid squamous cell carcinoma, with overlying Bowen's disease (*Figure 4 and Box 1*). An aggressive and invasive skin cancer was diagnosed.

Following the primary histology result from the biopsy taken during amputation the patient was referred to dermatology on a 2-week referral pathway.



Figure 1. A: The case on July 29, 2021. B: August 19. C: September 2. D: September 16. E: September 30. F: October 14. G: October 28. H: November 12, 2021.

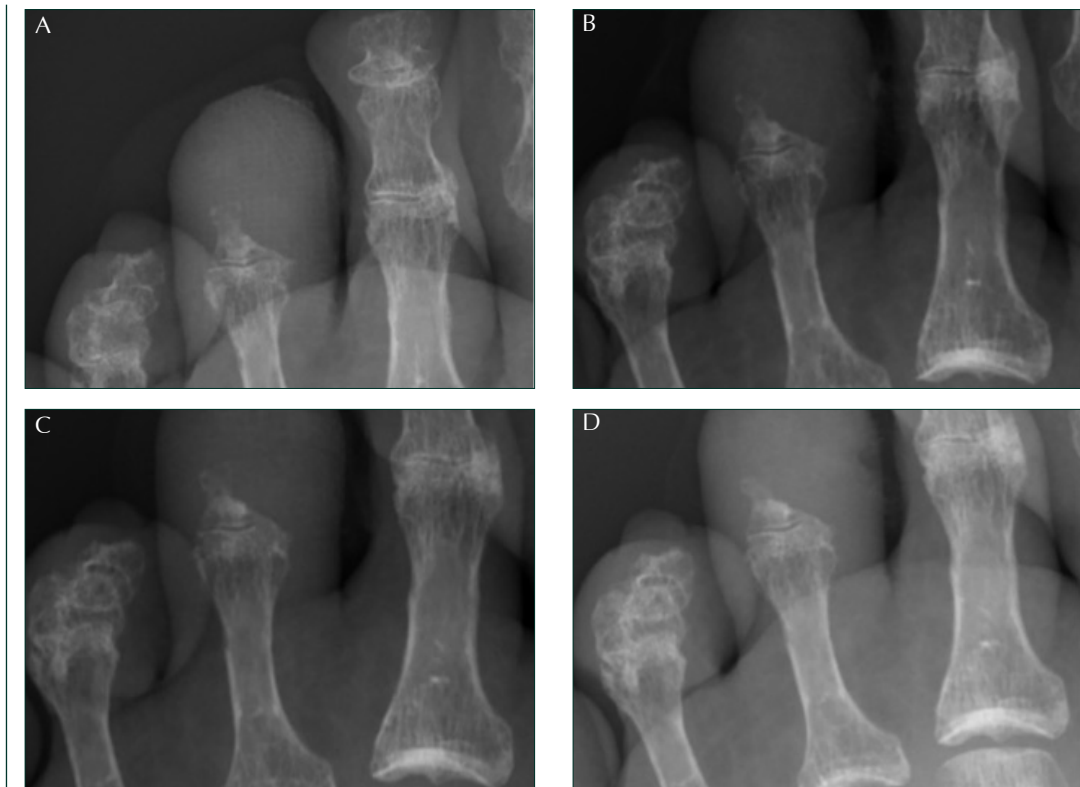


Figure 2. A: X-ray on August 6, 2021. B: August 27. C: September 27. D: October 8.



Figure 3. Post-amputation on December 20, 2021.

Melanoma multidisciplinary meeting staged the lesion as T4N0M0 and it was deemed “a very high-risk SCC”.

He was booked for a staging CT of the head, thorax, abdomen and pelvis to look for secondary spread, of which there was none.

He was reviewed in the dermatology follow up clinic on June 16, 2022. The wound had healed well and there was no ongoing evidence of SCC, Bowen's, lymphadenopathy or concerning skin lesions elsewhere. He will be reviewed quarterly going forward.

Conclusion

A 75-year-old man underwent a phalangeal amputation of his toe for a diabetic foot ulcer which appeared unusual at the time of surgery. Sarcomatoid squamous cell carcinoma with overlying Bowen's disease was the final diagnosis.

No metastatic spread found on CT. The patient continues on the high-risk follow-up schedule under dermatology.

Recommendations

Although it is more common that foot lesions in this cohort of patients are related to the complications of diabetes and microvascular disease, malignancy should not be overlooked. Histopathology is an important adjunct when treating diabetes-related foot lesions, especially if the clinical presentation is unusual. A low threshold for sending samples may avoid missing such conditions. ■

Lee ET (2020) A spindle cell squamous cell carcinoma on the cheek presenting with in-transit metastases and a satellite lesion. *Arch Craniofac Surg* 21(1): 58–63

Wernheden E, Trustrup H, Pedersen Pilt A (2020) Unusual presentation of cutaneous spindle cell squamous cell carcinoma: a case report. *Case Rep Dermatol* 12(1): 70–5

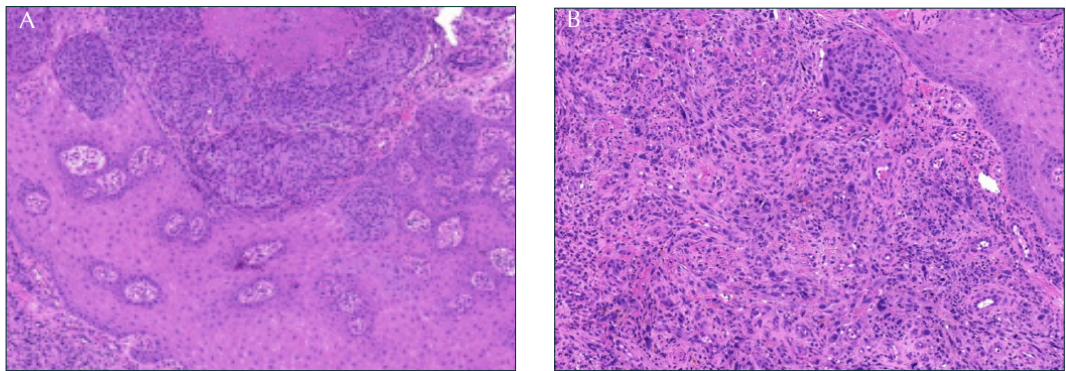


Figure 4: Histology. A: Intraepidermal nests of atypical cells in keeping with Bowen's disease. These were positive for cytokeratin immunohistochemistry and negative for melanoma markers. B: Dermal infiltrate of atypical spindle cells. These were also negative for melanoma markers, as well as a panel of cytokeratins. There was focal EMA expression. In view of the overlying Bowen's disease, we interpreted this as poorly differentiated sarcomatoid squamous cell carcinoma arising in Bowen's disease (see Box 1 for full report).

Box 1. Histology and pathology reports.	
Histology report	Pathology report
<p>TNM staging: pT1pN0pM0.</p> <p>Histology: Squamous cell carcinoma — 28899001.</p> <p>Grade of differentiation: Not appropriate or cannot be assessed.</p> <p>Pathology report text: skin, left third toe.</p> <p>Clinical history: Lesion left third toe. Overgranulation from osteomyelitis? Melanoma?</p> <p>Macroscopic: Cream and tan pieces of tissue measuring 10 4 5 4 2 mm in aggregate. All in one cassette for six levels. NRL. (SR)</p> <p>Microscopic: These are fragments of ulcerated skin showing infiltration by atypical epithelioid and spindled cells. There are numerous atypical mitotic figures (up to 10/mm²). There are nests of similar atypical cells in the junction. The appearances are of a malignant neoplasm and are likely to represent malignant melanoma. In the best orientated fragment, the tumour is at least 3 mm in thickness. However malignant cells are widely present at the deep aspect of this biopsy. Immunohistochemistry for confirmation is in progress, as well as BRAF mutation analysis.</p> <p>Diagnosis: skin, left third toe: malignant neoplasm, likely melanoma (at least 3 mm Breslow thickness, pT3b).</p>	<p>A wide panel of immunohistochemistry has been performed. Both the nested in-situ component and the dermal epithelioid and spindled component are negative for melanoma markers (S100, SOX10, melanA and HMB45). The junctional component is positive for cytokeratins AE1/3, MNF116, CK5/6 as well as p63, and is, therefore, assessed as Bowen's disease rather than melanoma-in-situ.</p> <p>The dermal component shows focal EMA expression, but is negative for all cytokeratins and p63.</p> <p>On balance, in view of the overlying Bowen's disease and the focal EMA expression, the appearances are interpreted as sarcomatoid/spindle cell squamous cell carcinoma. This case has been reviewed at the departmental skin pathology meeting.</p> <p>Final diagnosis: skin, left third toe: sarcomatoid squamous cell carcinoma, with overlying Bowen's disease</p>