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## Case Report

# Linguine technique for excision of lentigo maligna and poorly defined non-melanotic skin cancer – A case series

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## ABSTRACT

Accurately identifying the clinical margins of lesions such as lentigo maligna, lentigo maligna melanoma and other non-melanotic skin cancers can often be a clinical challenge. Irregular, poorly-defined peripheral margins accompanied by the presence of subclinical disease only detectable histologically can mean rates of incomplete excision are increased. We seek to highlight the use of the linguine technique for excision of lesions with poorly-defined peripheral margins. We describe in detail the step-by-step process for undertaking the technique, highlighting its advantages and disadvantages with a review of the related literature. We present three cases where the senior author has employed the linguine technique for the excision of lentigo maligna, lentigo maligna melanoma and extramammary Paget's disease.

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## Introduction

Curative excision of any skin cancer is dependent on the operating surgeon taking adequate histologically confirmed surgical margins to ensure complete removal of disease. Evidence-based guidelines exist to direct clinicians on the appropriate peripheral and deep margins to take, however in some clinical situations adequate margin control may be hampered by problems identifying peripheral margins.<sup>1–3</sup> Within the head and neck, this is particularly the case for the excision of lentigo maligna (LM) where surgery must take account of proximity to important facial features. As a subtle, slow growing pigmented change that occurs predominantly within sun-exposed areas of the body, LM is characterised by a spectrum of actinic change ranging from simple junctional melanocytic hyperplasia to melanoma-in-situ.<sup>4,5</sup> Affected patients have an estimated 5% lifetime risk of developing invasive disease (lentigo maligna melanoma, LMM)<sup>6</sup> meaning treatment, either surgical excision (conventional surgery or Mohs micrographic surgery (MMS)) or ablative radiotherapy<sup>4</sup> is necessary.

LM is a challenge for plastic surgeons. Oncologically surgical excision is difficult because clinical margins are frequently ill-defined and atypical melanocytes may extend beyond visible peripheral margins. Consequently, recurrence rates are high with studies reporting rates between 8 and 20%.<sup>7</sup> The large surface areas of lesions and predilection for the face mean that post-excision reconstruction can be challenging with surgeons tasked with reconstructing sizeable soft-tissue defects that can cross multiple cosmetic subunits. The linguine technique, also known as the spaghetti or perimeter technique is a staged margin-controlled strategy for ensuring complete excision of lesions appropriate for sensitive surgical sites. In this case-series of three patients, we detail our experience using the linguine technique for the excision of LM, LMM and non-melanotic skin cancer.

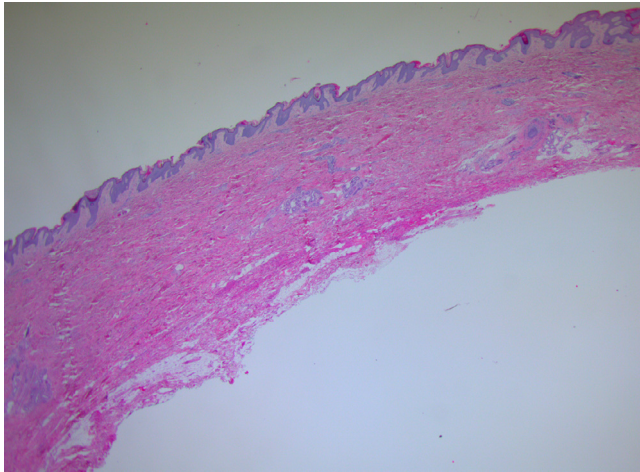
## Methods

### *First-stage procedure*

During the first-stage procedure, the peripheral margin of the lesion is marked using bright-theatre lights and dermoscopic magnification. At 4 mm from the clinical margin, a 2 mm wide strip of tissue is marked circumferentially around the lesion, divided into labeled linguina segments and photographed. The complete circumferential strip is then excised with the defect closed primarily using a continuous non-dissolvable monofilament suture. The central lesion is left *in situ* and undisturbed. The excised circumferential strip of tissue is divided into multiple equally sized linguine segments (approximately 3 cm in length). Each linguina is then marked to indicate orientation, sutured to a numerically labeled foam board in a straight and taut position, placed in formalin and submitted for histological examination. Ensuring each linguina is straight and taut is an important step to prevent distortion during fixation. Distortions can hamper histological interpretation. Clear labeling including a hand-drawn map of the linguine on the operation note and pathology request form, pre-operative and intra-operative photographs as well as close communication with designated histopathology colleagues are all undertaken to ensure accurate histological interpretation of the specimens. In the laboratory each strip is inked at one longitudinal end for orientation and fixed in a paraffin block with straightness maintained. The technique allows the entirety of each segment to be assessed with a clean, uninterrupted and orientated view of the epidermis along the long axis of each specimen (see [Figure 1](#)). If peripheral margin involvement is identified, it is possible for surgeon and histopathologist to accurately identify the involved location using the corresponding clinical diagram. Multiple sections of each segment are cut until all segments are confirmed as being negative.

### *Second-stage procedure*

The second stage procedure is undertaken as soon as the histology result from the first-procedure is available. If all peripheral strips are verified free of disease, the full lesion is excised with an appropriate deep margin and a further peripheral margin is marked beyond previous circumferential strip biopsy with the exact peripheral margin determined by the type of skin cancer present. The second-stage deep margin is checked by traditional bread-loafed sliced histological analysis. In the case that



**Figure 1.** High power histological image demonstrating a linguine segment orientated in the long-axis facilitating uninterrupted assessment of the epidermis.

one or more linguine segments from the first-stage procedure demonstrates tumour involvement, a further wider margin is taken and a third-stage procedure is performed. The site of involvement can be precisely identified from the meticulous intra-operative mapping and photos. Upon confirmed histological clearance, appropriate reconstruction is then carried out, usually with a full-thickness skin graft or local flap.

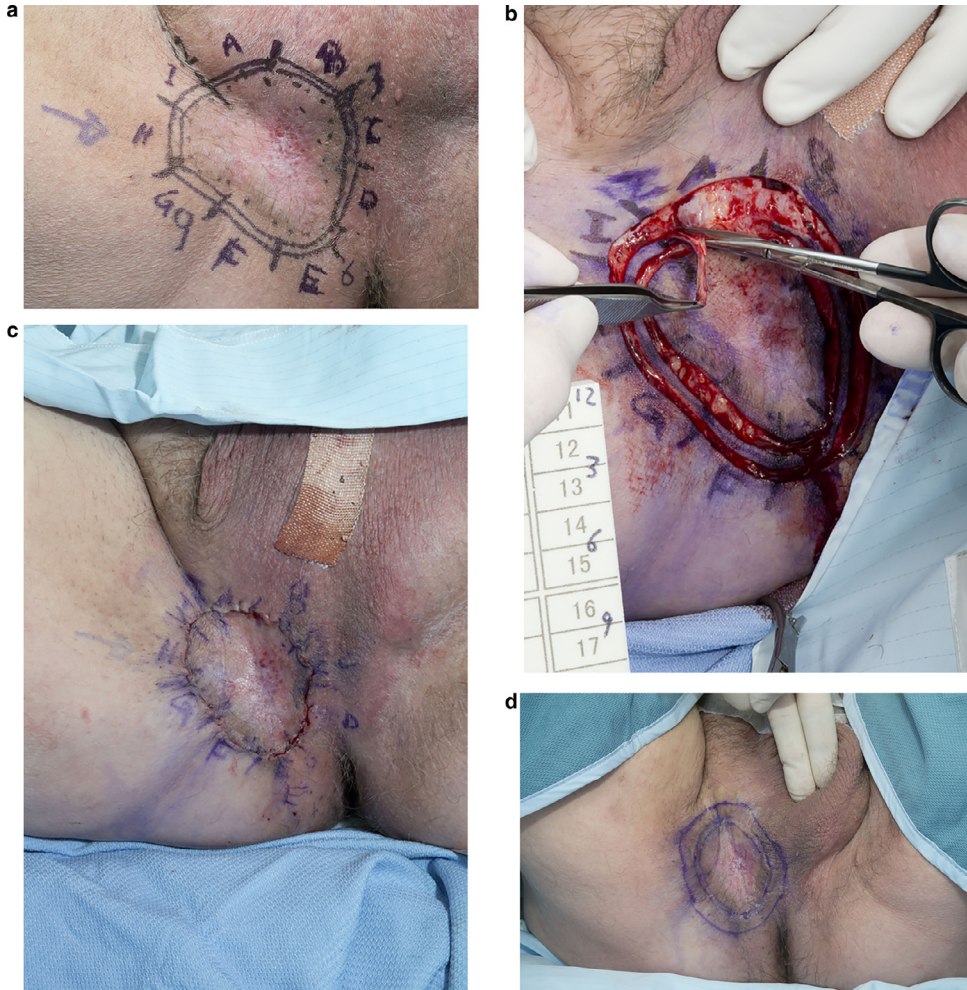
### Clinical cases

#### *Patient 1*

A 49 year-old female was referred to the plastic surgery out-patient department with a biopsy-proven LMM over her left cheek. The Breslow thickness of the invasive component was measured to be 2 mm situated within a wider area of lentiginous change. Peripheral margins were accompanied by areas of patchy pigmentation and were clinically indistinct. A sentinel lymph node biopsy was performed at the same time as the first-stage procedure to identify any potential micro-metastases within draining lymph node basins and the senior author managed to remove an intra-parotid sentinel node through the circumferential strip margin. The pathology report demonstrated clear peripheral margins and a sentinel lymph node biopsy that was negative. During the second stage procedure, a 0.5 cm margin was taken around the previous circumferential strip biopsy scar (ensuring appropriate clearance around the biopsy-confirmed invasive disease) and the central segment was excised. Reconstruction was undertaken using a full thickness skin graft with a good cosmetic result and the lesion was completely excised.

#### *Patient 2*

A 52 year-old diet-controlled diabetic male with a history of recurrent furuncles over his right groin and upper thigh, presented to dermatology colleagues with a long-standing macerated plaque over his right inguino-crural fold. The lesion measured 5 by 4 cm and had been slowly increasing in size over the preceding 18 months. Extra-mammary Paget's disease was suspected and confirmed by punch biopsy. A CT scan and colonoscopy was performed to rule-out accompanying pelvic malignancy but revealed no suspicious change. The patient was referred to plastic surgery for excision of the lesion and due to its location; the senior author felt that the linguine technique should be employed to facilitate peripheral margin assessment and a conservative excision of scrotal tissue. The first-stage

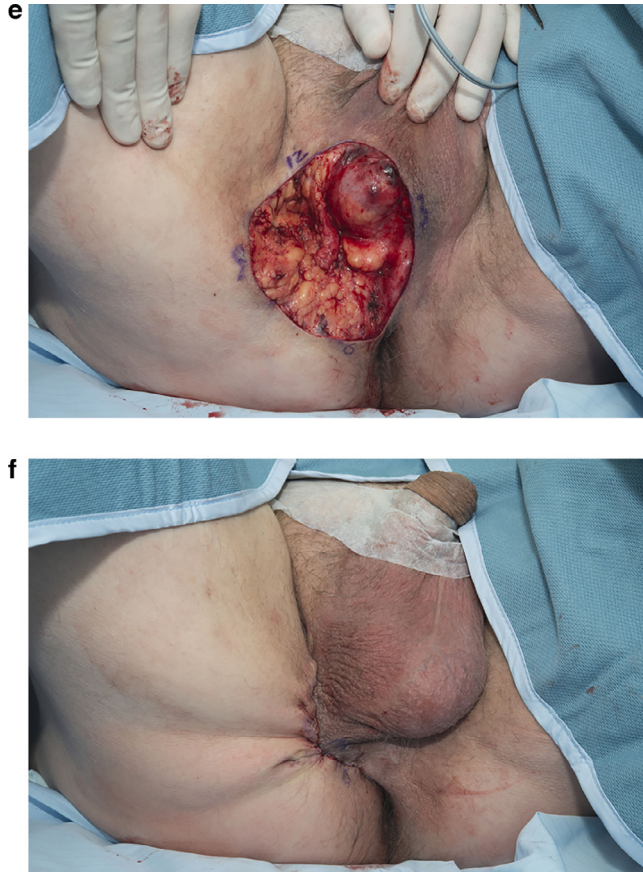


**Figure 2.** Operative Images for Patient 2. (a) Clinical margins of the biopsy-confirmed extramammary Paget's disease over the right inguinocrural fold. The clinical lesion is marked pre-operatively with the dotted line and a 2 mm wide circumferential strip is marked beyond divided into 9 segments. (b) The circumferential peripheral margin (linguine strip) is excised. (c) Central lesional segment left *in situ* and the circumferential strip is closed primarily. (d) Following histopathological confirmation of complete peripheral clearance, a further 1 cm peripheral margin is added beyond the circumferential strip to treat extramammary Paget's disease. (e) Central segment and margin excised taking a small amount of scrotal skin. (f) Defect closed primarily with undermining and advancement of thigh skin.

linguine procedure was performed and demonstrated no evidence of disease in the circumferential strip excision. During the second-stage procedure, the central lesion was excised with a cuff of fat and a further 2 cm peripheral margin beyond the circumferential strip scar whilst the defect was closed primarily. The lesion was completely excised.

### Patient 3

A 62 year-old male with a history of excess sun exposure from a childhood in Africa presented to the dermatology department with a number of slow-growing lesions over his face, back and limbs. Dermoscopic examination revealed Fitzpatrick type 2 skin and a number of characteristic basal cell and squamous cell carcinomas, in addition to a 3 by 3 cm variably pigmented area of dense reticulo-



**Figure 2.** Continued

macular change over his left cheek. An incisional biopsy of the cheek lesion was undertaken at the same time as wide local excision of the trunk and limb lesions. Histology of the cheek lesion confirmed junctional proliferation of mildly atypical melanocytes without any invasive component that was keeping with LM. The patient was offered Mohs micrographic surgery or the linguine technique but opted for the latter based on expediency. Histology from the first-stage procedure demonstrated chronically sun-damaged skin with evidence of Bowen's disease, actinic keratosis but complete peripheral excision of the LM. The second-stage procedure was performed with a further 5 mm margin marked beyond the circumferential strip scar and the residual LM central segment excised with a cuff of deep fat. The defect was reconstructed with a purse suture and a full-thickness skin graft and the lesion was confirmed completely excised.

### Discussion and conclusions

LM, LMM and extra-mammary Paget's disease present therapeutic challenges for the determination of clinical and pathological margins that may be approached in a number of ways. By choosing to pursue the linguine technique we highlight an alternative to existing narrow-margin procedures such as MMS or conventional staged-excision.<sup>4,5</sup> The linguine technique is not new and has previously been described by others,<sup>8,9</sup> however, to the best of our knowledge it does not appear to have been widely adopted despite having a number of advantages compared to alternatives. By facilitating the

evaluation of peripheral margins through longitudinal rather than transverse sections, the linguine technique is a procedure that allows 100% assessment of peripheral margins. It avoids the limitation of transverse bread-loaf histological analysis that may allow peripheral disease between sections to be missed despite apparent complete excision by ensuring full longitudinal peripheral margin assessment.<sup>10</sup> The deep margin is not ignored and always confirmed by bread-loaf histological analysis following the second-stage procedure. The linguine technique is analogous to the peripheral margin assessment undertaken during MMS but in contrast, overcomes the diagnostic uncertainties involved in the pathological interpretation of fresh-frozen sections through the use of paraffin sections. This avoids the problems of histological distortion that can occur with fresh-frozen sections and lead to keratinocytes being misinterpreted as atypical melanocytes, inflammatory cells appearing to be invasive disease and alterations in the epidermal architecture. Such an advantage is particularly pertinent when considering LM often occurs in areas of actinic field change where ephelides, pigmented actinic keratoses and simple lentigines may be present and confident interpretation of histological specimens is paramount. Another advantage is the parsimony of the linguine technique. It is a simple surgical procedure well within the skill set of any plastic surgeon or surgical dermatologist. It negates the need for prolonged specialised training as well as expensive equipment and avoids any demands being placed on a surgeon to interpret histopathology specimens or liaise with pathology colleagues whilst a patient anxiously waits in the outpatient department. In our case-series, we demonstrate that the procedure is versatile and when anatomically feasible can be undertaken simultaneously with other procedures whilst also being suitable for the excision of non-melanocytic skin cancer. We undertook all the cases as general anaesthetic day-case procedures, nonetheless the technique can very appropriately be undertaken using local anaesthetic to minimise hospital stay. Finally, by avoiding the need for a patient to tolerate open wounds that need to be dressed between first- and second-stage hospital attendances, the technique is cosmetically very acceptable and comfortable for patients over the course of their treatment.

When compared to the alternatives of Mohs micrographic surgery and conventional excision and delayed-reconstruction, published work reporting the follow-up of patients who have undergone the linguine technique is limited. Gaudy-Marqueste et al. reported their experience using the analogous spaghetti technique in 21 patients (16 lentigo maligna and 5 acral lentiginous melanoma) with a mean follow-up of 25 months. They found that the mean number of procedures necessary per patient was 1.55 and reported one recurrence after 48 months of follow-up.<sup>9</sup> In a considered paper, Möller et al. reported their outcomes in 61 LM or LMM patients using the same technique but employed 5 mm margins. With a median follow-up of 14 months, they demonstrated no local recurrence but reported that further marginal excision was undertaken in 24% of patients following their first-stage procedure.<sup>11</sup> In an earlier article published in 1997, Johnson et al. reported the results of a very similar procedure, the “square” technique where quadrangular picture-frame markings were taken instead of a contour circumferential strip approach that ultimately resulted in less tissue conservation. In their 2-year follow-up of 35 patients, no instances of local recurrence were demonstrated.<sup>12</sup> Anderson et al. have reported their experiences with the same technique in 150 patients over a follow-up period of “less than 5 years” and identified only one recurrence<sup>13</sup>. Jejurikar et al. reported follow-up using the square technique in 48 patients with 51 lesions and identified no recurrence over a mean period of 31 months<sup>14</sup> and similarly Mahoney et al.<sup>15</sup> reported no recurrence in 11 patients with a short mean follow-up of 4.7 months and 1.9 stages of excision. Agarwal-Antal et al.<sup>16</sup> reported their experiences with a further modification, the polygonal technique, in 92 cases of LM with no recurrences at 4 years and interestingly found that a 5 mm margin only confirmed complete excision in 50% of cases.

When compared to MMS, the linguine technique does have some disadvantages. Firstly, it has been criticized by those<sup>17</sup> that argue the sutured closure of the peripheral margins between the first and second-procedures inevitably means that some degree of inflammatory tissue reaction is elicited and this may increase the difficulty of any subsequent peripheral margin assessment and also facilitate bacterial ingress into a wound. We feel this risk is low if strict aseptic precautions are maintained and non-dissolvable sutures employed. Secondly, unlike MMS if the presence of invasive disease is not suspected but detected following a complete excision of the central segment, the technique may result in invasive disease being left *in situ* for longer than strictly necessary whilst central segment excision is awaited. Finally, the linguine technique is a staged procedure (in similarity to Mohs) so the

expediency of the second-stage is therefore dependent on the speed with which hospital pathology services can paraffin set and interpret specimens. This necessarily means that the complete second-stage excision of a lesion may be delayed for days or weeks – a situation that should be explained to a patient at the outset.

## Conclusion

In conclusion, we report a versatile technique for narrow-margin excision of lesions with poorly defined peripheral margins in 3 cases of LM, LMM and extramammary Paget's disease. The technique is oncologically sound and particularly suitable for the treatment of lesions within the head and neck. It facilitates longitudinal assessment of peripheral margins using gold-standard paraffin sections, negates requirements for MMS expertise and expensive equipment whilst avoiding the need for patients to tolerate an open wound.

## Conflicts of interest

None.

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