



CASE REPORTS

The use of selective lymphadenectomy in malignant blue naevus of the scalp

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SUMMARY. Malignant blue naevus (MBN) is a rare cutaneous tumour with a close biological resemblance to malignant melanoma. MBN spreads to regional lymph nodes, creating a dilemma in managing patients with clinically negative nodal basins. Sentinel lymph node (SLN) biopsy has evolved as a powerful staging tool by identifying occult metastatic nodal disease in patients with cutaneous malignancies. Here, we report a patient with MBN of the occipital scalp who underwent wide local excision together with preoperative lymphoscintigraphy and intraoperative radiolymphoscintigraphy and vital dye injection to identify all draining SLNs. No occult nodal disease was identified. This report adds to the growing body of literature supporting the role of SLN biopsy in staging individuals with cutaneous malignancies, including MBN. © 2003 The British Association of Plastic Surgeons. Published by Elsevier Science Ltd. All rights reserved.

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Malignant blue naevus (MBN) is an aggressive melanocytic neoplasm that arises in association with blue naevus.¹ The common blue naevus is a slowly growing benign skin lesion that appears as a blue–black dome-shaped well-circumscribed papule, generally less than 0.5 cm in diameter.² There have been fewer than 50 well documented cases of MBN reported in the Western literature, with the largest series including only 12 cases.³ MBN occurs predominately on the scalp, but may also appear in other sites, such as the trunk and feet. MBN occurs more commonly in men, with up to 2:1 male to female ratio. It is usually diagnosed in the fourth or fifth decade of life.⁴

MBN poses a diagnostic challenge to histopathologists. Typically, the tumour will involve the dermis and/or subcutis without epidermal involvement. These tumours have densely cellular fascicles of spindle cells containing variable amounts of cytoplasmic melanin. Cellular pleomorphism and mitotic activity are always present, with occasional foci of necrosis.⁵

The exact relation between MBN and melanoma remains somewhat controversial. Many pathologists consider MBN to be a malignant subtype of melanoma that develops in association with a pre-existing common blue naevus (commonly the cellular subtype). The opposing view is that MBN represents the malignant transformation of a previously benign blue naevus. Nevertheless, MBN is considered to be a virulent cutaneous lesion, which should be treated in a similar fashion to malignant melanoma.⁶

The natural history of MBN is one of spread to regional lymph nodes and distant sites. While the literature warns of its aggressive behaviour, there are currently no guidelines regarding adequate resection margins and prophylactic lymphadenectomy in patients with clinically negative lymph nodes.⁷

Sentinel lymph node (SLN) biopsy is a relatively new technique that enables the mapping and identification of lymph node(s) draining the site of a primary cutaneous malignancy.⁸ The SLN is defined as the first node draining a cutaneous territory. Biopsy of the SLN provides crucial staging information regarding the spread of tumour cells from the primary site, obviating the need for a full nodal dissection if biopsy findings for the SLN are negative. SLN biopsy was first popularised for malignant melanoma and has since been used in other malignancies, such as cutaneous squamous cell carcinoma, Merkel cell carcinoma, and breast and vulvar carcinoma, where an orderly lymphatic drainage is expected.^{9–13} We report a patient with MBN with clinically negative lymph nodes who underwent SLN biopsy using preoperative and intraoperative lymphatic-mapping techniques.

Case report

A 21-year-old white female with no significant past medical history presented with a rapidly enlarging 2 cm wide nodular pigmented lesion over the right parieto-occipital scalp. Follow-

ing a non-diagnostic incisional biopsy, the lesion was excised and submitted for evaluation by a dermatopathologist specialising in melanoma (M.C.M.). The diagnosis of MBN was made, but a tumour thickness could not be determined. The patient was referred to the University of Louisville Brown Cancer Center for further surgical care.

On physical examination, she was a healthy well developed white female with a 2.5 cm well healed linear scar over the right parieto-occipital region with no residual cutaneous pigment. Further examination of the head and neck revealed no other suspicious lesions. There was no parotid, occipital, cervical, clavicular or axillary lymphadenopathy bilaterally. Her remaining physical examination was unremarkable.

On the morning of the day of the operation, preoperative lymphoscintigraphy was performed using technetium 99 m antimony trisulphate in the Department of Nuclear Radiology. This is routinely performed to identify the location and number of SLNs draining a given anatomic area. The radioactive tracer was injected intradermally around the biopsy scar in four equal parts. Imaging was performed using a large field-of-view E-CAM gamma camera (Siemens Corp, Chicago, IL, USA) immediately after injection and again at 10 min, 1 and 2 h after injection (Fig. 1). Multiple lymph nodes were identified in the ipsilateral mastoid and cervical areas.

Once in the operating theatre, the biopsy scar was injected intradermally with a total of 1.0 ml of lymphazurin 1% isosulphan blue dye (Ben Venue Labs, Bedford, OH, USA) in a circumferential fashion. Intraoperatively, a hand-held gamma detector (Neoprobe Corp, Dublin, OH, USA) was used to assist in locating the previously identified SLNs.

Surgical exploration through two separate incisions revealed six SLNs (one in the ipsilateral occipital area, one in the ipsilateral postauricular region and four in the ipsilateral cervical region (level two and three cervical nodes)). The background count following excision of the SLNs was less than 10% of the count of the hottest lymph node, indicating removal of all significantly radioactive nodal tissue. Following removal of the SLNs, the excisional biopsy scar was excised with a 2 cm margin, following the guidelines established for intermediate and thick melanoma. Reconstruction was performed using a full-thickness skin graft from the right groin crease.

The postoperative course was unremarkable, with complete take of the skin graft and uncomplicated wound healing. Histological examination of the SLNs revealed no malignant foci on haematoxylin and eosin (H&E) or S-100 and HmB45 immunohistochemical staining. The patient remains free of disease 20 months postoperatively. No adjuvant chemotherapy or radiation therapy was used.

Discussion

MBN occurs so rarely that every confirmed case should be reported for better understanding of its epidemiology, natural history and clinical management.¹⁴ What is available in the literature indicates that MBN has an aggressive natural history with spread to regional lymph nodes and distant sites. Current management includes surgical resection of the primary tumour and clinically involved lymph nodes. Distant metastases are managed with a combination of resection and/or chemotherapy, generally with an unfavourable outcome.^{3,7,15} Management of those patients with clinically negative lymph node basins remains poorly defined, with unclear guidelines. In one series, four out of 11 patients developed local recurrences, ten developed nodal metastases and eight died

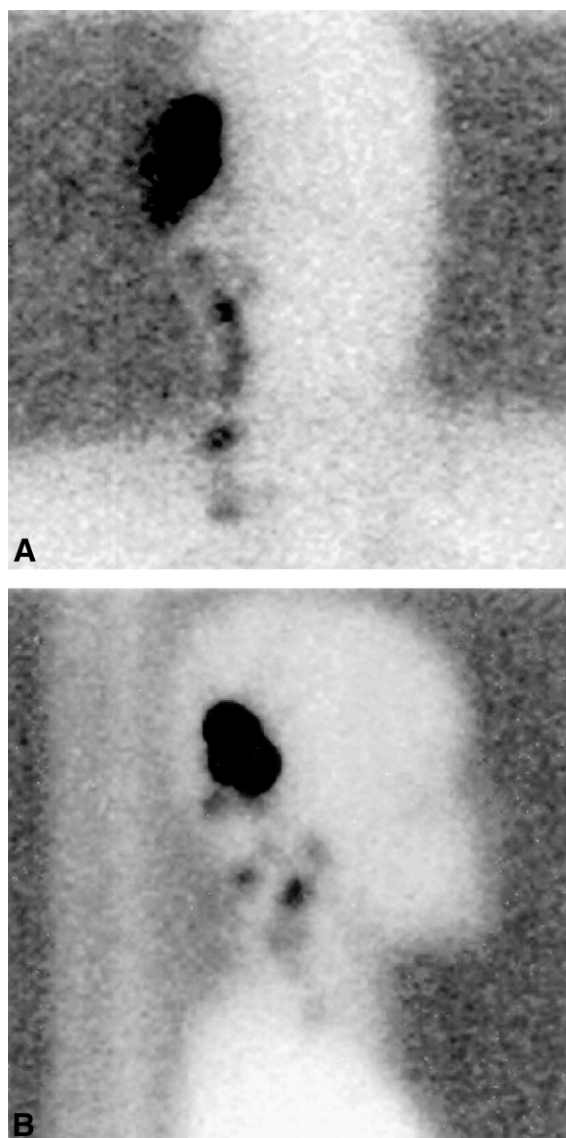


Figure 1—Preoperative radiolymphoscintigrams demonstrating multiple sentinel lymph nodes: (A) anterior–posterior view, (B) lateral view.

from their disease.³ With such an aggressive tendency to spread to regional nodal basins, it seems oncologically reasonable to use SLN technology to rule out occult nodal disease.

Morton et al first proposed the SLN technique in the management of cutaneous melanoma.^{8,9} Melanoma nodal metastases have been shown to occur in an orderly and sequential manner, with the SLN being the first lymph node in a lymphatic basin to be involved.¹⁶ Similar patterns of nodal spread have been demonstrated for other cutaneous carcinomas.^{9–11,13} While there are currently no studies that establish an orderly sequential lymphatic spread for MBN, this tumour's close biological relationship to malignant melanoma suggests that it may metastasise in similar orderly manner.^{5,17,18}

The primary utility of the SLN technique is as a staging tool to identify occult metastatic disease in clinically node negative lymph node basins. If a SLN is

found to harbour metastatic disease, a formal nodal-basin resection is recommended. If the SLN is found to be free of cancer on H&E and immunohistochemical staining, no further surgical intervention is recommended and close clinical observation is instituted. Therefore, the SLN technique allows the identification of occult disease while avoiding formal lymphadenectomy in all patients with clinically negative lymph-node basis.¹⁹

The use of preoperative lymphoscintigraphy and intraoperative vital dye radiolymphoscintigraphy to isolate the SLN is highly successful and reliable. Furthermore, Reintgen et al have demonstrated that the probability of the SLN being free from disease while another node in the same nodal basin harbours disease (i.e. false negative) is less than 1% for malignant melanoma.¹⁶ Finally, the SLN technique will allow identification of the exact nodal basin(s) draining an anatomical site where multiple basins may be involved (e.g. trunk, head and neck). The SLN technique can therefore identify the presence and exact location of occult lymph node disease with a high level of reliability.

In this patient, we identified six SLNs draining the primary tumour. This is consistent with the literature on head and neck melanoma, where the average number of nodes retrieved per SLN procedure is 2.2, but often many more.^{20,21} No occult disease was identified in any of the specimens, obviating the need for further surgery, chemotherapy or radiation therapy.

In conclusion, the SLN technique may serve as a useful staging tool for identifying patients with MBN who have occult nodal disease in the lymphatic basin at risk for metastases. Individuals with positive SLNs would be offered a formal completion lymphadenectomy, while sparing those with local disease only.

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