

## Dermal Lymphatic Invasion: A Rare Feature in Benign Intradermal Nevus

### Abstract

The present case describes classical intradermal nevus with intralymphatic nevus cell emboli. A 13-year-old boy presented with black colored macule on the shin 1 cm in diameter which was gradually increasing in size. Histopathology of the lesion was typical of an intradermal melanocytic nevus. The most notable feature of this case, however, was an occasional aggregate of nevus cells within a lymphatic vessel of the upper dermis. The nevus cells within lymphatic lumen had morphological features of type A nevus cells. The cells were round-to-cuboidal, exhibited abundant cytoplasm with well-defined cell borders and formed nests. These nevus cell aggregates were surrounded by flattened endothelial cells. Due to its rarity, a lymphatic nevus cell embolus creates diagnostic issues for pathologists. This observation must not be interpreted as evidence of malignancy but significant as a rare histological feature. In other words, the nevus cells might have been transported through the lymphatic vessels as a “benign metastasis.”

**Keywords:** *Intradermal nevus, intralymphatic nevus cell aggregates, intralymphatic nevus cell protrusion/projection, melanocytic precursor, pseudolymphatic*

### Introduction

Intradermal melanocytic nevi are common, benign, pigmented skin tumors formed by proliferation of dermal melanocytes. Many uncommon changes are noted such as bone formation in nevi, nodular myxoid changes, amyloid deposition, and granular cell changes.<sup>[1]</sup> Intralymphatic emboli in dermal nevi are extremely rare. Earlier nevus cell aggregates in lymph nodes (NALN) was considered a rare phenomena. Presently reported rate of NALN in melanoma is 0.2%–0.54% and is higher in sentinel lymph nodes (3.9% to 13%).<sup>[2-4]</sup> NALN is also observed in individuals without malignancy as nevus cell aggregates into lymph nodes through lymphatics from cutaneous nevus. So far 18 such cases have been reported.<sup>[5-8]</sup> The observation of a lymphatic nevus cell embolus supports the hypothesis that the nevus cells are likely to be transferred, through lymphatics, from a cutaneous nevus to the draining lymph node.<sup>[9]</sup>

### Case Report

A 13-year-old male presented to the department of plastic surgery with a macule on the shin. A physical examination revealed a brown colored pea-sized

cutaneous macule on the shin. The lesion was sent for histopathology.

The excised lesion showed elliptical skin measuring 1 cm × 1 cm × 0.3 cm with the central pigmented nodule. Histologically, dermis showed nests of nevoid cells with characteristic morphological features of types A, B, and C nevus cells, respectively. The type A nevus cells in the upper dermis were round-to-cuboidal, showed voluminous cytoplasm containing variable amounts of melanin granules and formed nests. Overlying epidermis was normal without any junctional activity.

The type B nevus cells in the mid-dermis, which were distinctly smaller than the type A nevus cells, were arranged in well-defined aggregates or cords and contained less cytoplasm and melanin. The type C nevus cells in the lower dermis were elongated and possessed spindle-shaped nuclei. The histopathology showed a decrease in cell size, melanization and the progression from nests to cords to more neuroidal spindle cells with dermal descent, which is referred to as maturation. No mitotic figure was seen. The upper dermis showed the presence of type A nevus cells within the lymphatic vessels. This aggregate was lined by flattened endothelial cells, which was

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highlighted by immunohistochemistry (IHC) (CD34 DAKO QBEnd10) [Figure 1]. Based on this, nevus cell clusters were considered as intralymphatic nevus cell emboli. No evidence of dysplastic change or malignancy was observed. It was reported as intradermal nevus with incidental intralymphatic benign emboli.

## Discussion

Intralymphatic invasion by nevus cells may sometimes be encountered in Spitz nevi, but its occurrence in banal nevi is rarely reported. It is pertinent to exclude artefacts of tissue processing presenting as clefts or pseudovascular or pseudolymphatic spaces before labeling it as lymphatic invasion.<sup>[8]</sup> This is possibly due to shrinkage of tissue and poor intercellular cohesion of nevus cells. So far 18 cases of intralymphatic invasion by nevus cells have been reported.

Intralymphatic nevus cell aggregates (ILNA) were first described in lymph nodes in 1931 by Stewart and Copeland.<sup>[9,10]</sup> Earlier report described lymphatic invasion as either subendothelial hillocks, intralymphatic nevus

cell protrusion/projection (ILNP), or even free nevus cells in vascular spaces. Subsequently, a large series of 10 cases have been reported wherein they categorized them under two heads. ILNA and ILNP utilizing IHC CD31 and/podoplanin.

The criteria for ILNA were – (a) presence of endothelial layer lining the space with aggregate and periphery of aggregates, (b) similar morphology of intraluminal aggregate with extraluminal nevus cells, (c) focal attachment of aggregates to vessel wall, (d) almost all cells of aggregates seen intraluminally, and (e) absence of smooth muscle/elastic tissue in the wall of the vessel. All the nevus cell aggregate which are not in lumen are considered as ILNP. ILNA is possibly preceded by ILNP.<sup>[6]</sup>

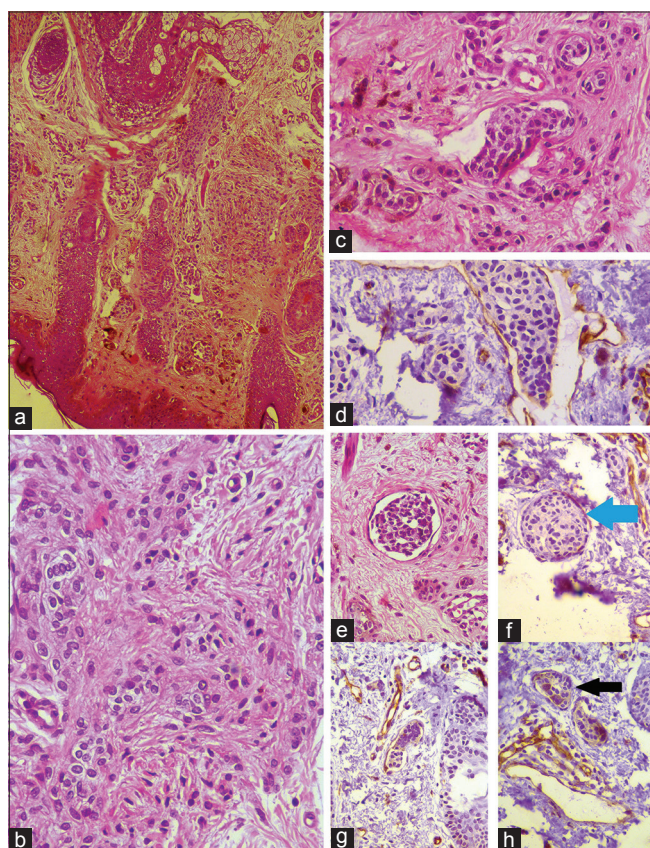
We also utilized CD 31 to highlight the intralymphatic invasion. Similar to the observation of Leblebici, the intralymphatic aggregate were noted at the periphery of the nevus in superficial dermis. The presence of nevus cell aggregate in and around lymph node has been attributed to two theories, i.e., arrested migration and mechanical transport theory. Findings supporting arrested migration theory are – (a) capsular location of nevus cell aggregates with sparing of sinuses, (b) concurrent embryonic migration of melanocytes precursor cells and development of lymphatic system, (c) presence of blue nevi in extracutaneous location, for example, cervix, prostate, seminal vesicle, (d) rare co-occurrence of metastatic melanoma and nevus cells in the same lymph node, and (e) lymphatic nevus cell aggregates more commonly associated with congenital nevi.

The more favored mechanical transport theory on the other hand finds support in – (a) presence of intranodal deposits of other tissue, for example, endometrium, breast, (b) infrequent presence of aggregates within sinuses/parenchyma of lymph node, (c) lack of nevus cells in lymph node draining extradermal site, (d) morphological dissimilarity of arrested melanocytic dermal cells which are bipolar and cuboidal conventional nevi, and (e) presence of nevus cells in afferent lymphatics of lymph node which has no role in embryonic migration.

It has been postulated by Leblebici *et al.* that ILNA can be dislodged by minor trauma and can then be transported along lymphatics to the draining lymph nodes. However, the mechanism of further proliferation of this passive extravasation to lymph node capsule, by escaping immune killing, is still not clear. They also found association of ILNA in 3/15 congenital nevi and found support in mechanical transport theory.<sup>[6]</sup> Polypoidal nevus aggregate projecting into lymphatics in congenital nevi have been attributed to abnormal migration of nevus crest cells.<sup>[3]</sup>

## Conclusion

This is a unique case with benign intradermal nevus presenting with lymphatic emboli. This case is not malignant but has a unique histological finding associated



**Figure 1:** (a and b) Nevus cell aggregate within a lymphatic vessels seen in the dermis showing morphological features type A, B, C Cells (hematoxylin and eosin,  $\times 40$ ,  $\times 100$ ); (c and d) Intralymphatic nevus seen on hematoxylin and eosin and CD 31 (hematoxylin and eosin,  $\times 400$ ; CD31,  $\times 400$ ); (e and f) Intralymphatic emboli identified in the lymphatic vessel lined by flat endothelial cells (solid blue arrow) which is proven by immunohistochemistry CD 31 (hematoxylin and eosin; CD 31,  $\times 400$ ); (g) Positive control for CD 31 showing endothelial cells lining lymphatic channels (CD 31,  $\times 400$ ); (h) Nevus cell aggregate mimicking intralymphatic embolus but negative on CD 31 as shown by black solid arrow (CD 31,  $\times 400$ )

with the lesion. It is essential to educate surgeons and oncologists about the associated intralymphatic emboli in benign intradermal nevus, which is not truly malignant, rather supports the theory of mechanical transport.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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