



Hair follicle nevus occurred on toe nail bed

Min-Jeong Kang, MD; Seung-Hee Loh, MD; Bark-Lynn Lew, MD, PhD* ;
Woo-Young Sim, MD

Department of Dermatology, Kyung Hee University College of Medicine, Seoul, Korea

To the editor:

Hair follicle nevus (HFN), also known as congenital vellus hamartoma, is a rare congenital hamartoma that typically presents as a solitary, asymptomatic, indistinguishable papule, plaque, or nodule [1]. Follicular differentiation is often found in HFN, and it is characterized by a perifollicular fibrous sheath thickening. Small mature hair follicles are also present in fibrotic stroma histological findings [2]. HFN typically occurs at birth in areas with a plethora of hair follicles, especially on the face.

A 65-year-old male presented to the clinic with a chief complaint of longitudinal nail ridge with slightly brown colored pigmentation on the right 1st toe nail. (Fig. 1) There was no history of trauma. The patient roughly remembered that he discovered the lesion about one year before this visit. The tumor was completely removed by local excision. Upon nail plate extraction of the toenail, a rice grain sized subcutaneous nodule that was slightly erythematous was found in the nail bed. Histopathological examination revealed vellus hair follicles along with randomly distributed bulbs located high in the dermis. (Fig. 2) Although a small deformity of the nail plate was noted after operation, the longitudinal ridge along with signs of recurrence were absent at the six-month follow-up.



Fig. 1 (A) A longitudinal brown colored nail ridge on the right 1st toe nail (B) After nail plate extraction, a rice-head sized slightly erythematous nodule on nail bed was shown.

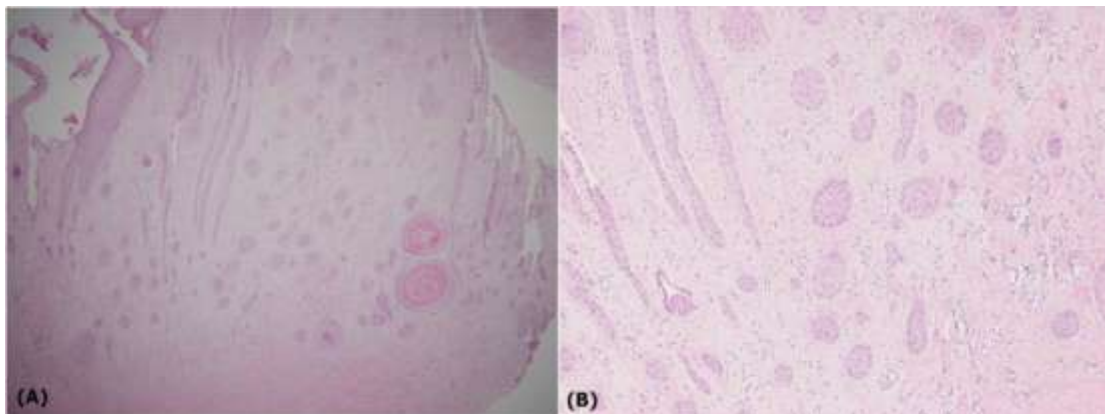


Fig. 2 (A)(B) Histopathological examination revealed numerous vellus hair follicles with several keratin filled cysts. (H&E, respectively, X40, X100)

HFN is a rare disease and there have only been 30 reported occurrences or less [3]. To our knowledge, the present case is the first report of HFN occurring in the nail bed [4,5]. Because there are no apparent hair follicles in the subungual region, the nature of the tumor observed in the nail bed may be debatable. However, hair and nails are analogous differentiation products of the ectoderm, therefore, they have similarities in their anatomical structures and they are associated with each other in many other congenital and acquired diseases [6-8]. So, this could be used to explain our case.

Additionally, although the exact etiology of HFN is unknown, some authors suggest that it may result from random somatic point mutations during fetal development which was supported by subsequent findings of HFN in infants at birth or early childhood years. However, it is thought that other etiologic factors besides abnormalities in fetal development may be involved in our case. This case is valuable in that it shows an atypical presentation of HFN compared to previous encounters in terms of its unusual location and late onset in age. Given the new evidence, we may need to expand the spectrum of clinical presentation of HFN and reexamine the conventional diagnostic criteria.

References

1. Nagarajan P, Bartholomew TS, Allen CM, Peters SB. Hair follicle nevus with features of comedo nevus: an expanding spectrum. *AM J Dermatopathology*. 2016, 38(6):e81-86
2. Karabulut YY, Şenel E, Karabulut HH, Dölek Y. Three different clinical faces of the same histopathological entity: hair follicle nevus, trichofolliculoma and accessory tragus. *An Bras Dermatol*. 2015, 90(4):519-22
3. Motegi S, Amano H, Tamura A, Ishikawa O. Hair follicle nevus in a 2-year old. *Pediatric Dermatol*. 2008, 25:60-62
4. Nagase K, Nagase K, Misago N, Narisawa Y. A preauricular hairy papule in an infant: hair follicle nevus closely similar to accessory tragus. *Arch Dermatol*. 2012, 148:266-268
5. Jedrych J, Akilov O, Gehris R, Ho J. Hair follicle nevus of the abdominal skin: an unusual extracephalic presentation. *Pediatr Dermatol*. 2014, 31:e85-86
6. JH Park, DY Lee, KT Jang, et al; Samsung Medical center. CD13 is a marker for onychofibroblasts within nail matrix onychodermis: comparison of its expression patterns in the

nail unit and in the hair follicle. *J Cutan Pathol.* 2017; doi: 10.1111/cup.13006

7. Baran R, Dawber P.R R, Heneke E. Hair and Nail Relationship. *Skinmed.* 2005 Jan-Feb;4(1):18-23
8. Kumiko M, Hiroyuiki H, Tadashi T. Subungual trichoadenoma showing differentiation toward follicular infundibulum. *J Dermatol.* 2011, 38(11):1118-1121

Received: September 18, 2017; **Accepted:** October 14, 2017; **Published:** October 15, 2017

Competing Interests: The authors have declared that no competing interests exist.

Copyright: 2017 Lew BL *et al.* This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

***Correspondence to:** Bark-Lynn Lew, Department of Dermatology, Kyung Hee University hospital at Gang-dong, Korea

E-mail: bellotte@hanmail.net