

SHORT COMMUNICATION

Subungual Angiokeratoma Presenting as a Longitudinal Pigmented Band in the Nail

Michiko Hasegawa and Atsushi Tamura*

Department of Dermatology, Isesaki Municipal Hospital, 12-1 Tsunatori-machi, Isesaki, Gunma 372-0817, Japan. E-mail: atsushitamura@gunma-u.ac.jp

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Nail pigmentation due to haemorrhage usually shows an irregular shape. However, it may appear as a longitudinal pigmented band, which needs to be differentiated from subungual melanoma (1). Fortunately, a pigmented nail due to haemorrhage gradually grows out and is replaced by non-pigmented nail during follow-up (2). Here, we described a patient with a longstanding longitudinal pigmented band in a toe nail, revealed as intraungual haemorrhage from a subungual angiokeratoma arising in the nail matrix.

CASE REPORT

A 74-year-old man was referred to us with suspected diagnosis of subungual melanoma. He had noticed a brownish streak on the left 4th toenail 10 years previously, and it has increased in depth of colour in the recent 3 months. He had also noticed venous dilatation of his legs 15 years ago. His past history is notable for oesophageal cancer resection 8 months previously. Physical examinations revealed a dark brown longitudinal band of 3 mm in width on the left 4th toenail (Fig. 1a). Dilated veins were observed near the pigmented band (Fig. 1a, b), and varicose veins were also seen on bilateral legs. Dermoscopic examination disclosed a streak composed of red-brown to dark brown globules and dots with a light brown background (Fig. 1c). On the basis of clinical and dermoscopic features, a diagnosis of intraungual haemorrhage was made. We suspected that persistent intraungual haemorrhage lasting for 10 years might be related to the increased subungual venous pressure due to varicose veins of the lower leg. Surgical excision of the lateral nail unit including a pigmented band was performed. Histopathological examinations revealed the presence of erythrocyte aggregates in the middle layer of the nail plate from its root to the distal edge (Fig. 2a). Beneath the nail root, numerous ectatic vessels surrounded by hyperplastic nail matrix epithelium protruded into the nail plate. Red blood cells within thin-walled ectatic vessels were extravasated into

the nail matrix epithelium and overlying cornified layers (Fig. 2b). Although there were thick-walled tortuous vessels in subdermal layer below the nail matrix (Fig. 2c), thin-walled ectatic vessels were not present beneath the papillary layer. Based on these histopathological features, we diagnosed the lesion as a subungual angiokeratoma. There has been no recurrence of the nail lesion during a follow-up period of 10 months.

DISCUSSION

Small haemorrhage under the nail plate is divided into 2 types: nail bed haemorrhage and nail matrix haemorrhage (3). The former occurs in the region beyond the lunulae and haemorrhages remain subungual. In contrast, matrix haemorrhages occur within the lunulae or under the nail root and are incorporated into the nail plate (3). Both types of haemorrhages show distal migration and eventually disappear as the nail grows (3). Longitudinal red dark band similar to our case may occur in patients with monodactylous longitudinal erythronychia when it accompanies single or double interrupted lines of splinter haemorrhages (4). Moreover, partially blanchable red longitudinal streaks named “red comets” has recently been demonstrated in patients with tuberous sclerosis complex, frequently associated with splinter haemorrhages (5). These haemorrhagic lesions exhibit thin longitudinal dark-red lines, whereas our patient exhibited a long-standing longitudinal pigmented band due to persistent intraungual haemorrhage within the nail matrix. Histopathological examination revealed a vascular lesion consistent with angiokeratoma in which intraepidermal haemorrhage and thrombosis often occurs, in our patient lasting for 10 years.

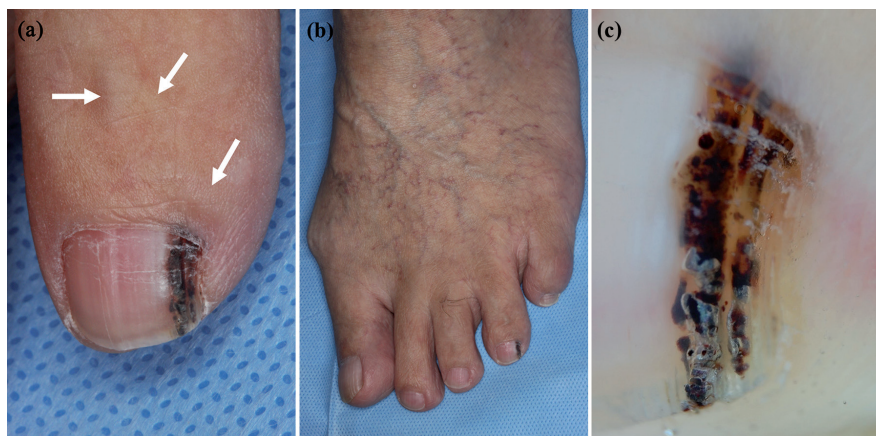


Fig. 1. Clinical and dermoscopic features. (a) A longitudinal pigmented band of the left 4th toenail and dilated veins of the toe (arrows). (b) Dilated tortuous veins of the left dorsal foot extending to the left 4th toe. (c) Dermoscopy showing a streak composed of red-brown to dark brown globules and dots with a light brown background.

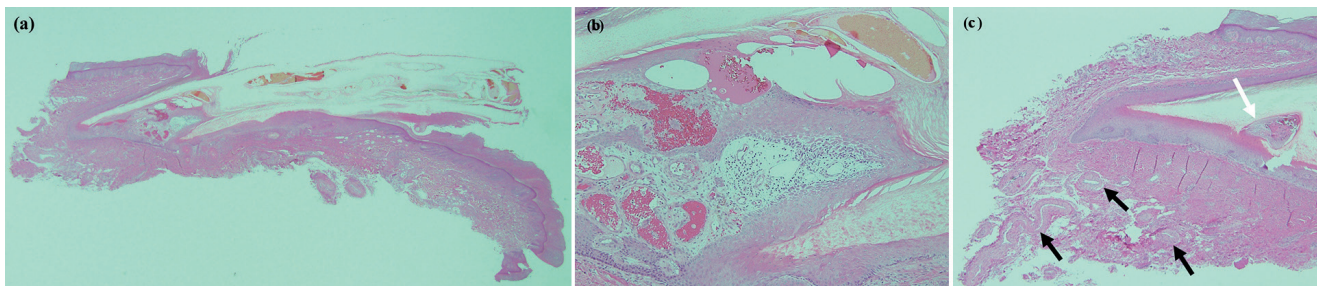


Fig. 2. Histopathological features. (a) Low-power view of the resected specimen showing erythrocytes aggregates in the middle layer of the nail plate. (b) Beneath the nail root, numerous ectatic vessels surrounded by hyperplastic nail matrix epithelium protrude into the nail plate. Red blood cells within thin-walled ectatic vessels are extravasated into the nail matrix epithelium and overlying cornified layers. (c) Thick-walled tortuous vessels (black arrows) lie in subdermal layer beneath the nail matrix on the other section. A white arrow indicates angiokeratoma in this section. Haematoxylin and eosin stain, original magnification: (a) $\times 12.5$; (b) $\times 100$; (c) $\times 40$.

In angiokeratoma the epidermis usually shows a proliferative reaction such as acanthosis and rete ridge elongation with or without hyperkeratosis (6, 7). Many types of angiokeratomas have been described in the literature: solitary angiokeratoma (as in our case), angiokeratoma corporis diffusum, angiokeratoma Mibelli, angiokeratoma of Fordyce, angiokeratoma circumscriptum naeviforme, and angiokeratoma of the tongue (8). The pathophysiology of angiokeratomas remains uncertain except for angiokeratoma corporis diffusum, which is usually associated with a lysosomal storage disorder (9–11); caused by an enzyme defect resulting in a weakness of the capillary wall (6, 12). Some reports suggest that increased venous pressure proximal to the site is responsible for the development of angiokeratomas (12, 13). Our patient had noticed varicose veins on his legs prior to occurrence of the nail lesion.

The development of angiokeratoma in subungual region has not been known until now. Moreover, longitudinal pigmented band in the nail caused by vascular anomalies, including haemangiomas and vascular malformations have never been reported. There are few reports describing dermoscopic studies of nail haemorrhages (14, 15); but none of these studies have mentioned intraungual haemorrhages. Mun et al. (15) reported that longitudinal streaks alone were found in only 3% of 90 investigated lesions of subungual haemorrhage. Dermoscopic features of intraungual haemorrhage have never been reported before.

The authors declare no conflict of interest.

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