

Longitudinal Erythronychia Manifesting With Pain and Cold Sensitivity

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A 38-year-old woman presented to our nail specialty clinic with a red line and associated pain on the left fourth fingernail of 2 and 3 years' duration, respectively. The patient described the pain as throbbing, with sensitivity to pressure and cold. She noted that the nail grew slowly and would sometimes split at the distal edge. She did not recall any discrete trauma to the digit or nail. The patient was right-handed, making the symptoms less likely to be due to overuse from daily activities. She had received no prior treatment for these symptoms.

The patient's medical history included iron deficiency as well as acne and eczema. She had no personal or family history of skin cancer. Physical examination of the affected digit and nail revealed a longitudinal red line and distal onycholysis. With contact dermoscopy, the red line blanched. Pressure applied using a #11 scalpel blade elicited pinpoint tenderness (positive Love test), and application of an ice pack caused pain (positive cold test). A radiograph of the left hand was negative for bone erosions, and magnetic resonance imaging showed a 0.3-cm subungual lesion at the level of the fourth distal phalanx. An excision of the nail unit was performed.

WHAT'S YOUR DIAGNOSIS?

- a. eccrine spiradenoma
- b. glomangiomyoma
- c. leiomyoma
- d. myxoid cyst
- e. neuroma

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THE DIAGNOSIS: Glomangiomyoma

The nail unit excision specimen showed collections of cuboidal cells and spindled cells within the corium that were consistent with a diagnosis of a glomangiomyoma, a rare glomus tumor variant (Figure).

Glomus tumors are benign neoplasms comprising glomus bodies, which are arteriovenous anastomoses involved in thermoregulation.¹ They develop in areas densely populated by glomus bodies, including the fingers, toes, and subungual areas. Glomus tumors most commonly develop in middle-aged women.² Clinically, they manifest with a characteristic triad of intense pain, point tenderness, and cold sensitivity and may appear as reddish-pink or blue macules under the nail plate and/or longitudinal erythronychia.²⁻⁶ The presence of multiple glomus tumors is associated with neurofibromatosis type 1.⁷

Advanced imaging including ultrasonography and magnetic resonance imaging (MRI) may help confirm the diagnosis but may not be cost effective, as excision with histopathology is needed to relieve symptoms and render a definitive diagnosis. Radiography is highly insensitive in identifying bone erosions associated with glomus tumors.⁸ With ultrasonography, glomus tumors appear hypoechoic; with Doppler ultrasonography, they appear hypervascular. With MRI, glomus tumors appear as well-defined nodular lesions with hypointense signal intensity on T1-weighted sequence and hyperintense signal intensity on T2-weighted sequence, with strong enhancement using gadolinium-based contrast.^{9,10} On histopathology, a glomus tumor appears as a nodular tumor with sheets of oval-nucleated cells arranged in multicellular layers surrounding blood vessels and are immunoreactive for α -smooth muscle actin, muscle-specific actin, and type IV collagen.^{11,12}

There are several glomus tumor variants. The most common is a solid glomus tumor, which predominantly is composed of glomus cells, followed by glomangioma, which mainly is composed of blood vessels. Glomangiomyoma, which mostly is composed of smooth muscle cells, is the rarest variant.¹³

While glomus tumors are common in the subungual areas, it is an uncommon location for glomangiomyomas, which have been reported in the nail unit in only 7 prior case reports identified through searches of PubMed and Google Scholar using the terms *glomangiomyoma*, *glomangiomyoma nail*, and *subungual glomangiomyoma* (Table).¹³⁻¹⁹ Glomangiomyomas more commonly are described in solid organs, including the stomach, kidney, pancreas, and bladder.¹⁶ The mean age of patients with subungual glomangiomyomas, including our patient, was 40.4 years (range, 3-61 years), with the majority being female (75.0% [6/8]). Most patients presented with fingernail involvement (75.0% [6/8]), nail dystrophy (eg, nail plate thinning, longitudinal grooves, splinter

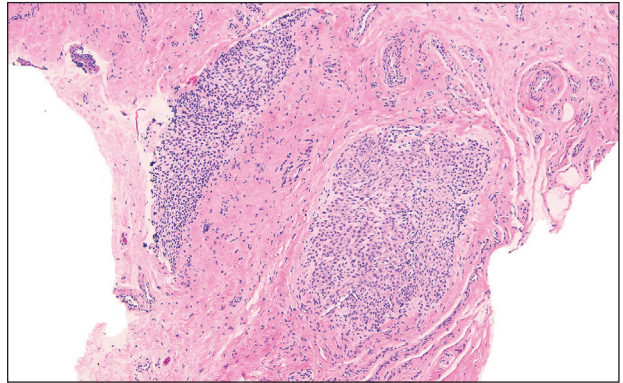


FIGURE. Glomangiomyoma. Collections of cuboidal cells and spindled cells within the corium (H&E, original magnification $\times 100$).

hemorrhages, longitudinal erythronychia)(62.5% [5/8]), and intermittent pain and/or point tenderness in the affected nail (75.0% [6/8]).¹³⁻¹⁹ Notably, only our patient had longitudinal erythronychia as a clinical feature, and only one other case described MRI findings, which included a lobulated mass with intense contrast and distal phalanx destruction.¹⁸ One patient was a 3-year-old girl with a family history of generalized multiple glomangiomyomas. Although subungual glomangiomyoma was not confirmed on histopathology, the diagnosis in this patient was presumed based on her family history.¹³ On histopathology, glomangiomyomas are composed of oval-nucleated cells surrounding blood vessels. These oval-nucleated cells then gradually transition to smooth muscle cells.²⁰

A myxoid cyst is composed of a pseudocyst, which lacks a cyst lining, and is a result of synovial fluid from the distal interphalangeal joint entering the pseudocyst space.² It typically manifests with a longitudinal groove in the nail plate. A flesh-colored nodule may be appreciated between the cuticle and the distal interphalangeal joint.² The depth of the longitudinal groove may vary depending on the volume of synovial fluid within the myxoid cyst.²¹ In a series of 35 cases of subungual myxoid cysts, none manifested with longitudinal erythronychia. Due to their composition, myxoid cysts can be distinguished easily from solid tumors of the nail unit via transillumination.²² Pain is a much less common with myxoid cysts vs glomus tumors, as the filling of the pseudocyst space with synovial fluid typically is gradual, allowing the surrounding tissue to accommodate and adapt over time.²¹ In equivocal cases, MRI or high-resolution ultrasonography may be used to distinguish myxoid cysts and glomus tumors.⁸ Histopathology shows accumulation of mucin in the dermis with surrounding fibrous stroma.²³

TABLE. Summary of the Literature on Glomangiomyomas of the Nail Unit

Reference (year)	Patient sex (age, y)	Site	Cold sensitivity	Intermittent pain	Point tenderness	Nail dystrophy	Radiologic findings
Calduch et al (2002) ¹³	F (3)	Right 3rd toenail	No	No	Yes	No	N/A
Mentzel et al (2002) ¹⁴	M (39)	Fingernail (unspecified)	N/A	N/A	N/A	N/A	N/A
Kang et al (2008) ¹⁵	F (38)	Right 1st fingernail	N/A	Yes	Yes	Yes; longitudinal groove	Radiograph negative
Wollstein and Wollstein (2012) ¹⁶	F (56)	Right 1st fingernail	Yes	Yes	Yes	Yes; thinning of nail plate	Radiograph negative; bone scan negative
Aqil et al (2018) ¹⁷	F (37)	Right 4th fingernail	Yes	Yes	Yes	No	N/A
Demirdag et al (2020) ¹⁸	M (31)	Right 3rd toenail	No	No	No	Yes; overlying nail dystrophy and enlargement of the distal segment	Lobulated mass with intense contrast and distal phalanx destruction on MRI
Vega et al (2022) ¹⁹	F (61)	Right 1st fingernail	N/A	Yes	Yes	Yes; longitudinal grooves, splinter hemorrhages	N/A
Current case	F (38)	Left 4th fingernail	Yes	Yes	Yes	Yes; longitudinal erythronychia	Radiograph negative; MRI: 0.3-cm subungual lesion at the level of the 4th distal phalanx

Abbreviations: F, female; M, male; MRI, magnetic resonance imaging; N/A, not available.

Subungual neuromas are painful benign tumors that develop due to disorganized neural proliferation following disruption to peripheral nerves secondary to trauma or surgery. In 3 case reports, subungual neuromas manifested as painful subungual nodules, with proximal nail plate ridging, or onycholysis.²⁴⁻²⁶ Since neuromas have only rarely been described in the subungual region, reports of MRI and ultrasonography findings are unknown. Histopathology is needed to distinguish neuromas from glomus tumors. Histopathology shows an acapsular structure consisting of disorganized spindle-cell proliferation and nerve fibers arranged in a tangle of fascicles within fibrotic tissue.²⁵ On immunohistochemistry, spindle cells typically are positive for cellular antigen protein S100.²⁶

Leiomyomas are benign neoplasms derived from smooth muscle, typically localized to the uterus or gastrointestinal tract, and have been described rarely in the nail unit.^{27,28} It is hypothesized that subungual leiomyomas originate from the vascular smooth muscle in the

subcutaneous layer of the nail unit.²⁸ Like glomus tumors, leiomyomas of the subungual region often manifest with pain and longitudinal erythronychia.²⁷⁻³⁰ Subungual leiomyomas may be distinguished from glomus tumors via advanced imaging techniques, including ultrasonography and MRI. Cutaneous leiomyomas have been described with mild to moderate internal low flow vascularity on Doppler ultrasonography, while glomus tumors typically reveal high internal vascularity.²⁸ Biopsy with histopathology is needed for definitive diagnosis. On histopathology, leiomyomas demonstrate bland-appearing spindle-shaped cells with elongated nuclei arranged in fascicles.²⁷ They typically are positive for α -smooth muscle actin and caldesmon on immunostaining.

Eccrine spiradenomas are benign adnexal tumors likely of apocrine origin with limited case reports in the literature.^{31,32} Clinically, eccrine spiradenomas involving the nail unit may manifest with longitudinal nail splitting of the nail or as a papule on the proximal nail fold, with associated tenderness.^{31,32} In a report of a 50-year-old

woman with a histopathologically confirmed eccrine spiradenoma manifesting with longitudinal splitting of the nail and pain in the proximal nail fold, the mass appeared hypoechoic on ultrasonography with increased intramass vascularity on Doppler, while MRI showed an intensely enhancing lesion.³¹ These imaging features, combined with a classically manifesting feature of pain, make eccrine spiradenomas difficult to distinguish from glomus tumors; therefore, histopathologic examination can provide a definitive diagnosis, and surgical excision is used for treatment.³¹ On histopathology, these tumors are well circumscribed and composed of both small dark basaloid cells with peripheral compact nuclei and larger cells with central pale nuclei, which may be arranged in tubules.^{31,32}

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