Onycholemmal carcinoma: A morphologic comparison of 6 reported cases

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Background: We report 6 new cases of onycholemmal carcinoma, a rare, often misdiagnosed, subcategory of squamous cell carcinoma. All reported cases to date have been treated with amputation of the affected digit.

Objective: The purpose of this study was to present the clinical and pathological features of each new case and to discuss treatment options that spare digit functionality.

Methods: Hematoxylin-eosin stains were performed on tumor sections and examined using light microscopy. In situ hybridization using probes against human papillomavirus were examined in 1 case.

Results: The female to male ratio was 1:1 with involvement of fingers in 3, thumb in 1, and toe in 1. Among the symptoms were onycholysis, periungual erythema, and pain; symptom duration ranged from 6 months to 2 years. Histologically, all cases showed a well-differentiated atypical infiltrative squamous proliferative lesion exhibiting a lobulated and cystic pattern of growth in the dermis. Abrupt keratinization reminiscent of trichilemmal keratinization was noted. Mohs micrographic surgery and radiation therapy were used as primary treatment modalities, maintaining digit functionality and achieving remission.

Limitations: Limitations of this study included the small number of cases, the infrequency with which this tumor has been reported in the literature, and the inability to obtain follow-up on an older archival case.

Conclusions: Onycholemmal carcinoma is a distinct type of squamous cell carcinoma arising from the nail isthmus; its natural clinical course is indolent. In this regard less aggressive digit-sparing treatment modalities such as radiation or Mohs micrographic surgery should be considered. (J Am Acad Dermatol 2013;68:290-5.)

Key words: dermatopathology; Mohs micrographic surgery; nail isthmus; onycholemmal carcinoma; radiation therapy; squamous cell carcinoma; trichilemmal keratinization.

Onycholemmal carcinoma is a rare malignant epithelial tumor that originates from the nail bed epithelium. A few cases have been reported in the literature describing this slow-growing, subungual infiltrating neoplasm.[1-4] Clinically, these cases have been described as warty,

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discolored, crusted, or ulcerative lesions that progress slowly until the patient presents with pain, swelling, or onycholysis at times associated with boney involvement.[1,2,4] An initial trial of antibiotic therapy fails to improve the symptoms.

Histopathologically, onycholemmal carcinoma demonstrates solid collections of atypical keratinocytes and small keratin-filled cysts lined by atypical squamous epithelium devoid of a granular layer. This type of abrupt keratinization is reminiscent of the pattern of trichilemmal keratinization described in the proliferating pilar tumor.[1,5]

We report a series of 6 cases of onycholemmal carcinoma presenting the clinical and pathological features of this distinctive but uncommon variant of acral squamous cell carcinoma (SCC).

CAPSULE SUMMARY

- Onycholemmal carcinoma is a rare malignant tumor that originates from nail bed epithelium and is characterized by solid collections of atypical keratinocytes and small, keratin-filled cysts.
- This article further delineates histopathologic features of onycholemmal carcinoma and provides additional treatment options that spare digit functionality.
- This case series brings awareness regarding the presentation and diagnosis of onycholemmal carcinoma, in addition to putting forth Mohs micrographic surgery and radiation therapy as treatment modalities.

CLINICAL PRESENTATIONS

See Table I. The patients comprised 3 women and 3 men ranging in age from 39 to 84 years (mean age of 62 years). The known sites of involvement were left fourth finger, right fourth finger, left great toe, left third finger, and right thumb. Among the clinical presentations were a nonhealing progressive periungual ulcer of the nailfold (1) (Fig 1, *A*), pain and swelling of the nail resembling paronychia (2), pain and swelling of the toe (1), painless keratotic plaque of the nail (1), and onycholysis (1) (Fig 2, *A*), with erythema of the right thumb (1). In 1 case radiographic studies disclosed a soft-tissue defect and slight osteopenia but no lytic and or destructive lesion of the distal phalanx. The symptoms ranged in duration from 6 months (1 patient) to 5 years (2 patients); in 3 patients the symptoms were extended over a long period of time that was unspecified. Among the treatment modalities were Mohs micrographic surgery (1), radiation (1), excisional biopsy and debridement of the nail plate and distal nail bed (1), excision of the nail apparatus (1), and amputation (1). In the patient who underwent Mohs micrographic surgery, the final defect was closed by a V-Y advancement flap from the palmar side of the fingertip. At 1-month follow-up, the wound was well healed and the patient had full function of her finger. There was no evidence of metastatic or recurrent disease in any of the patients regardless of the treatment received.

LIGHT MICROSCOPIC FINDINGS

All biopsy specimens appeared similar showing an infiltrative squamoid lesion characterized by variably sized lobules of squamous epithelial cells showing abrupt central keratinization without any interposed keratohyaline granules; an incomplete pattern of keratinization was not seen (Fig 2). The keratinization was most reminiscent of that seen in the setting of proliferating pilar tumors but with enhanced atypia sufficient to warrant categorization as carcinoma. In 1 case, there was dystrophic calcification. In each case there were foci of high-grade squamous atypia manifested by cells exhibiting high nuclear to cytoplasmic ratios and marked hyperchromasia. In 1 case, assessment of human papillomavirus (HPV) was negative.

DISCUSSION

We have presented 6 patients in whom a diagnosis was made of onycholemmal carcinoma. In our series there was no sex predilection. The patients were typically middle-aged to older adults with most lesions occurring on the fingers. The lesions exhibited an indolent clinical course with variable therapeutic maneuvers designed to extirpate the lesion comprising Mohs micrographic surgery, amputation, and radiation. There are 5 other case reports that have been previously published, reporting a predominance of men with a mean age of 69 years and a finger site predisposition. In these previously published cases a more aggressive clinical approach being that of amputation was performed. At least in our experience, a less aggressive approach was equally as effective in achieving complete removal without recurrent tumor, distant metastatic disease, or both.

Onycholemmal carcinoma originates in the nail isthmus located in the proximal nail bed immediately distal to the lunula. The sterile matrix/nail isthmus plays a critical role in the effective sealing of the nail bed by a very thin compartment of cornified cells strongly adherent to the inferior border of the nail plate. This semirigid keratin produced through

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