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BRIEF REPORT

Clinical and histopathological features of onychopapilloma in an Australian setting: A case series of 50 patients

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Abstract

Onychopapilloma is an uncommon tumour of the nailbed and the distal nail matrix. To date, only 19 case reports and case series have been reported in the literature. This article includes literature review on all reported cases and provides the first case series of onychopapilloma in an Australian population, evaluating the clinical features and histopathological diagnosis of patients with onychopapilloma in an Australian subspecialty nail clinic.

KEYWORDS

nail, nail dermoscopy, nail matrix, nail pathology, nail tumour, onychopapilloma

INTRODUCTION

Onychopapilloma is an uncommon tumour of the nailbed and the distal matrix. The condition was first described as 'localized multinucleate distal keratosis' in 1995, and the term 'onychopapilloma' was coined by the same authors in 2000.[1] It has been known to be a benign tumour, but recently, Haneke et al. reported a case of onychopapilloma with malignant features.[2] Onychopapilloma most commonly presents with subungual hyperkeratosis, longitudinal erythronychia and distal onycholysis. It is also less frequently associated with melanonychia, leukonychia, splinter haemorrhages and distal fissures.[1] To date, 19 case reports and case series have been published in the literature, a total of 204 cases (Table 1). To the best of our knowledge, this is the first case series of onychopapilloma in an Australian population. The objective of this article is to review the clinical features and histopathological diagnosis of patients with onychopapilloma in an Australian subspecialty nail clinic.

METHODS

Between March 2016–December 2020, 50 patients with a clinical or histopathological diagnosis of onychopapilloma were identified at the Skin Health Institute, Carlton Victoria, from the electronic medical record. A database

was used to collect demographic information including age and gender together with clinical details such as reasons for referral, sites affected, presenting features, management, follow-up duration, recurrence and complications. A dermatopathologist with experience in diagnosing nail unit conditions reviewed the histologic slides of the patients who had an excisional biopsy and transversal nail clippings done (SP). Macroscopic and dermoscopic images were captured by professional photographers using a Heine non-polarised contact dermoscope on a Nikon SLR camera. Only cases that had photographic data and whose clinical diagnosis was confirmed by two independent investigators (JSK, JY) were included.

RESULTS

Literature review

A total of 19 case series/reports have been identified in the literature, outlining a total of 204 cases to current date. This is summarised in Table 1, in addition to the data from Tosti et al.[1]

Clinical and dermoscopic features

We identified 50 patients with diagnosis of onychopapilloma – 35 patients were diagnosed on typical clinical features and managed conservatively, and 15 patients were managed surgically with tangential excision and histopathologic evaluation confirmed the diagnosis. Of the 35 patients who were managed conservatively, five patients had transversal nail clippings for histological confirmation of the clinical diagnosis. In four out of five nail clippings, histopathology was consistent with onychopapilloma. (Table 2).

The patients' average age was 54.5, and 68% were female and 32% male. Most of the patients were Fitzpatrick phototype 1 to 3 (86%). The most frequently affected digit was the thumb (n = 28, 56%), followed by middle finger (n = 7, 14%), first toe (n = 5, 10%) and ring finger (n = 4, 8%). The most common clinical features of onychopapilloma were subungual hyperkeratotic mass (n = 29, 58%). This was followed by distal fissures (n = 23, 46%), erythronychia (n = 21, 42%) and longitudinal ridge (n = 21, 42%). It was less frequently associated with onycholysis, leukonychia, short splinter haemorrhage, melanonychia and long splinter haemorrhage. Approximately 65% of cases with subungual hyperkeratosis also had coexisting chromonychia and nearly half of the cases with distal fissures had coexisting chromonychia. Only 30% of the cases with onycholysis had distal fissures.

In addition to the fifty cases described here, there were nine further cases where onychopapilloma had been initially considered as a differential diagnosis based on clinical features. Histopathology on excisional biopsy, however, led to alternative diagnoses. Clinically, these cases were most frequently associated with erythronychia (n = 6, 66.7), longitudinal ridge (n = 5, 55.5) and less frequently subungual hyperkeratosis (n = 1, 11.1) and distal fissures (n = 2, 22.2).

Pathology

The histological features can be seen in Figure 1g–j. The diagnosis is best made on an oriented longitudinal tangential excision. Given nail biopsy specimens are often not orientated, the interdigitating rete ridges in the nail bed epithelium in transverse sections may mimic epidermal papillomatosis seen in onychopapilloma, which may lead to wrong diagnosis. Histopathology from transversal nail clippings can also be suggestive of the diagnosis. In this case series, excisional specimens of onychopapilloma from 15 patients showed the following: papillomatosis (86.7%), nail matrix metaplasia of nail bed (60%), subungual hyperkeratosis (53.3%) and haemorrhage (20%). Other features included parakeratosis, acanthosis and keratinocyte multinucleation. In four patients who had nail clippings performed, all cases showed subungual hyperkeratosis and papillomatosis, consistent with onychopapilloma.[1] Features of squamous cell carcinoma and recently described malignant onychopapilloma were not identified.[2]

DISCUSSION

This is the first case series of onychopapilloma in an Australian setting. As previously reported in the literature,[1] onychopapilloma is most frequently seen on the thumb, and the presence of subungual hyperkeratotic mass is highly suggestive of the diagnosis. These findings are also seen in pathologically verified cases in our series. Subungual hyperkeratosis is proposed to arise from the tumour in the distal nail matrix creating a longitudinal defect of the nail plate and subsequently the affected nail-bed filling up its undersurface.[1] The next most common findings are longitudinal erythronychia and distal fissures. It is proposed that longitudinal erythronychia and associated splinter haemorrhage develop by onychopapilloma compressing on the nail bed, thinning the ventral nail plate, which allows an enhanced view of corresponding streak of engorged nail bed. This makes the nail more fragile, with tendency to split distally.[1] Furthermore, half of onychopapilloma cases in this case series showed distal

TABLE 1 Literature review

Authors	Patients	Clinical presentation	Management
Baran R, Perrin C 1995[1]	4	Longitudinal erythronychia, splinter haemorrhage, distal subungual keratosis	Longitudinal excision
Baran R, Perrin C 2000[1]	14	Longitudinal erythronychia, splinter haemorrhage, distal subungual keratosis	Punch biopsy Longitudinal excision
Gee BC, Millard PR, Dawber RP 2002[1]	1	Longitudinal erythronychia, distal keratosis	Longitudinal excision
Criscione V, Telang G, Jellinek NJ 2010[1]	1	Longitudinal leukonychia	Lateral nail plate curl avulsion
Jellinek NJ 2011[1]	Not specified, at least 10	Longitudinal erythronychia or one or more of the following: splinter haemorrhage, distal subungual keratosis distal onycholysis	Longitudinal excision
Miteva M, Fanti PA, Romanelli P, Zaiac M, Tosti A 2012[1]	1	Longitudinal melanonychia	Nail clipping Longitudinal excision
Beggs S, Butala N, Heymann WR, Rubin AI 2015[1]	1	Longitudinal erythronychia, subungual hyperkeratosis	Excision
Ito T, Uchi H, Yamada Y, Oda Y, Furue M 2015[3]	1	Longitudinal melanonychia, distal onycholysis	Excision
Tosti A, Schneider SL, Ramirez-Quizon MN, Zaiac M, Miteva 2016[1]	47	Longitudinal erythronychia, longitudinal leukonychia, longitudinal melanonychia, long splinter haemorrhages without erythronychia, leukonychia or melanonychia, short splinter haemorrhages without erythronychia, leukonychia or melanonychia, subungual keratosis, distal fissures	Longitudinal excision
Jellinek NJ, Lipner SR 2016[4]	41	Longitudinal erythronychia	Longitudinal excision, tangential excision, shave biopsy
Kim M, Sun EY, Jung HY, Cho BK, Park HJ 2016[5]	3	Various chromonychia, including longitudinal erythronychia, longitudinal reddish-yellow longitudinal chromonychia and multiple yellowish chromonychia	Nail extraction and curettage
Sarkissian L, Fattouh K, Kanitakis J, Villani AP 2017[6]	1	Longitudinal erythronychia, longitudinal haemorrhage, subungual keratosis	Longitudinal excision
Halteh P, Magro C, Scher RK, Lipner SR 2017[3]	1	Longitudinal leukonychia, subungual keratosis	Tangential biopsy
Delvaux C, Richert B, Lecerf P, André J. 2018[7]	68	Longitudinal erythronychia, distal subungual hyperkeratosis, distal onycholysis, splinter haemorrhage, punctiform haemorrhage, longitudinal crest, notch in the lunula	Longitudinal excision or tangential longitudinal excision
Ramos Pinheiro R, Cunha N, Lencastre A. 2018[8]	1	Longitudinal erythronychia	Longitudinal excision
Tambe SA, Ansari SMM, Nayak CS, Chokkar R, Patil PD. 2018[9]	1	Longitudinal melanonychia, subungual keratosis	Excision
Haneke E, Iorizzo M, Gabutti M, Beltraminelli H. 2020[2]	1	Longitudinal erythronychia, distal onycholysis, subungual keratosis	Tangential excision
Baltz JO, Telang GH, Jellinek NJ. 2020[10]	1	Longitudinal erythronychia, distal onycholysis	Tangential excision
Starace M, Ferrari T, Pezzetta S, Savoia F, Zengarini C, Maria Piraccini B, Alessandrini A. 2020[7]	6	Longitudinal melanonychia	Longitudinal excision

TABLE 2 Clinical and histopathological features of onychopapilloma

Onychopapilloma cases (n = 50)								
Age (avg)	**Gender (n, %)**	**Referral diagnosis**	**Digit (n, %)**	**Clinical features (n, %)**	**Management (n, %)**	**Histopathology features (n, %)**	**Follow-up (Avg)**	**Recurrence (n, %)**
54.5	F (34, 68) M (16, 32)	Rule out malignancy (n = 26) Inconvenience (n = 19) Pain (n = 5)	Thumb (28, 56) Index (3, 6) Middle (7, 14) Ring (4, 8) Little (1, 2) First toe (5, 10) Fourth toe (1, 2) Multiple digits (1, 2)	Subungual hyperkeratosis (29, 58) Distal fissures (23, 46) Erythronychia (21, 42) Longitudinal ridge (21, 42) Onycholysis (10, 20) Leukonychia (7,14) Short splinter haemorrhage (7,14) Melanonychia (4, 8) Long splinter haemorrhage (2, 4)	Watch and wait (30, 60) Watch and wait with transversal nail clipping for histology (5, 20) Surgical (15, 30)	N/A Transversal nail clippings (n = 5) • Consistent with onychopapilloma (4, 80) • Nail plate only (1,20) Subungual hyperkeratotic mass (8, 53.3) Papillomatosis (13, 86.7) Matrix metaplasia (9,60) Splinter haemorrhage (3, 20)	4.9 months (n = 9) Lost to follow up/ discharged from clinic (n = 26) 7.8 months (n = 15)	N/A N/A Yes (n = 2, 13.3) No (n = 13, 86.7)

Abbreviation: N/A, not applicable.

fissures and coexisting chromonychia. Longitudinal leukonychia and longitudinal melanonychia were seen less frequently than longitudinal erythronychia.

Onychopapilloma can often be diagnosed clinically if the above typical features are present including non-specific subungual hyperkeratosis or haemorrhage.[1] In cases where the clinical presentation is typical for onychopapilloma, and there is no recent or rapid change in the appearance we regard a watch and wait approach as sufficient. Transversal nail clippings can also be very easily performed in the office setting if subungual hyperkeratosis is present. It is assessed in Haematoxylin and Eosin stains (H and E) and with good collaboration with the dermatopathologist, it can be helpful to suggest the diagnosis of onychopapilloma. Definite diagnosis does, however, require histopathological confirmation from a longitudinal biopsy including the distal nail matrix. Therefore, if tissue sampling is required to exclude malignancy or desired due to functional and/or cosmetic impact of the lesion, a full tangential excision of the entire lesion including distal nail matrix is recommended in all cases. This has the highest diagnostic yield and chance of the onychopapilloma with good long-term cosmetic outcome and relatively low recurrence rate. Histopathological characteristics include epithelial papillomatosis, nail matrix metaplasia of nail bed, subungual hyperkeratosis and subungual haemorrhage.[1]

In the presenting case series presented, with an average of 7.8 months follow-up, the recurrence occurred in two out of fifteen cases managed surgically and another two out of fifteen had subsequent nail dystrophy. As with any nail unit surgery, there is of course a real risk of (potentially severe) complications and nail dystrophy and this has to be discussed in detail with the patient on a case-by-case basis.

Onychopapilloma has been considered a benign condition. However, a case of a malignant onychopapilloma has been recently reported.[2] The patient presented with a linear longitudinal lesion that has been present over a few years, becoming painful over a few months. The surgical excision showed malignant onychopapilloma. Whether the malignant transformation was primary or secondarily as malignant degeneration of a previously existing benign onychopapilloma is unclear. Yet, this case report highlights the importance of ongoing follow-up. Therefore, we recommend self-monitoring and representation in case of clinical changes, or annual reviews, for example in context of general skin surveillance. In our experience, malignant tumours clinically presenting as typical onychopapilloma are exceedingly rare.

Common differential diagnoses of onychopapilloma include glomus tumour, Darier disease, lichen planus, traumatic splinter haemorrhages, melanoma and recently described malignant onychopapilloma.[2] In this case series,

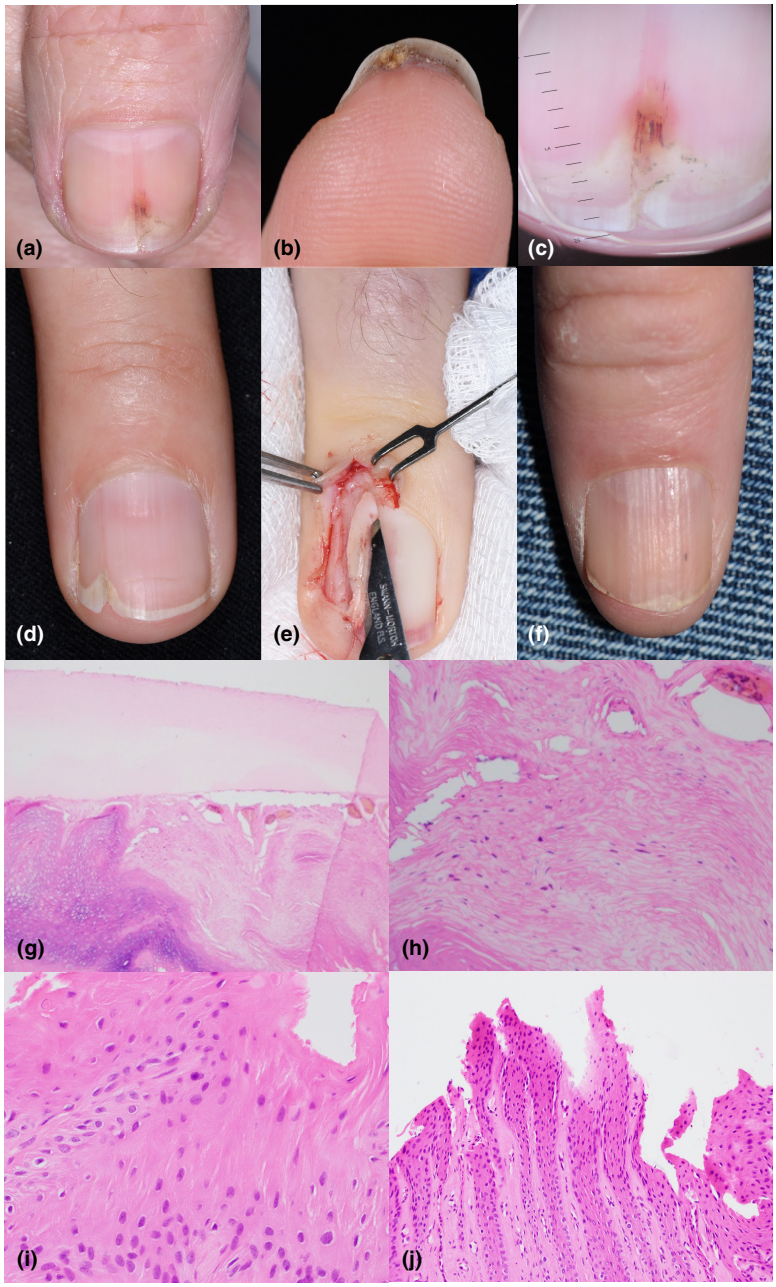

FIGURE 1 Onychopapilloma. (a) Longitudinal erythronychia, distal fissure, short splinter haemorrhage. (b) Subungual keratotic mass under the free edge of the nail plate. (C) Dermoscopy of the short splinter haemorrhage: 5 interrupted linear parallel bands extending to the proximal nail bed. (d) Onycholysis, subungual keratotic mass, longitudinal erythronychia. (e) Excisional biopsy of the onychopapilloma. (f) Post-surgical management of onychopapilloma, onycholysis. (g) Transverse nail clipping: Subungual papillomatous hyperkeratosis and parakeratin, with altered blood (H&E ×200). (h) Transverse nail clipping: Parakeratin with blood at top right (H&E ×400). (i) Excisional biopsy: Nail matrix metaplasia (H&E ×400). (j) Excisional biopsy: Nail bed papillomatosis (H&E ×200).

we also identified patients who were diagnosed with nail eczema, onychomycosis, Bowen's disease, squamous cell carcinoma (SCC) on histopathologic evaluation, where onychopapilloma had been an initial differential diagnosis based on clinical features. The distinguishing features of onychopapilloma include involvement of single digit, presence of subungual keratotic mass and the absence of preceding trauma.

CONCLUSION

This case series of patients with onychopapilloma is the first in the Australian setting. Onychopapilloma has a wide range of presentations, but the presence of subungual keratotic mass is highly suggestive of the diagnosis. Surgical management is possible where the onychopapilloma causes pain, inconveniences the patient or to rule

out malignancy. Yet, biopsy to achieve the diagnosis may be avoided in many cases by careful clinical and dermoscopic assessment or with non-invasive transversal nail clippings for histology.

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CONFLICT OF INTEREST

The authors have no conflict of interest to report.

ETHICAL APPROVAL

As per the National Statement on Ethical Conduct in Human Research (paragraphs 5.1.22 and 5.1.23), this case series is exempted from ethics review.

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