Verrucous carcinoma of the foot - A series of 4 cases

Yogesh Bhatt *, Sumer Singh, Sanjay Vaghani and Piyush Doshi

Department of Plastic and Reconstructive Surgery, Sumandeep Vidyapeeth University, Vadodara, Gujrat, India

*Correspondence Info:
Dr. Sumer Singh,
Department of Plastic and Reconstructive Surgery,
Sumandeep Vidyapeeth University, Vadodara, Gujrat, India
E-mail: sumeryadav2004@gmail.com

Abstract
Most of patients having soft tissue lesion on heel/foot referred to plastic surgeon because reconstructive options for heel/foot are difficult for general surgeons/orthopedicians. So as a plastic surgeon we should be aware of this rare but local tumor of foot, so that we can diagnose it early and manage properly.

We present a case series of 4 patients of verrucous carcinoma of the foot. This is a rare, locally invasive, well differentiated, low-grade squamous cell carcinoma, with HPV as a possible causative agent. It follows a chronic course and mimics a variety of skin lesions, delaying diagnosis by up to 15 years. The definitive diagnosis is made histologically, and treatment by wide local excision is recommended. Our patients underwent wide local excision and reconstructed. Three patients had lesion on heel, were managed using reverse peroneal artery flap and fourth patient had lesion on forefoot, was managed using reverse medial planter artery flap. There were no postoperative complications. There was no functional impairment.

Keywords: verrucous carcinoma, foot tumors, reverse peroneal artery flap, reverse medial planter flap

1. Introduction
Most of patients having soft tissue lesion on heel/foot referred to plastic surgeon because reconstructive options for heel/foot are difficult for general surgeons/orthopedicians. So as a plastic surgeon we should be aware of this rare but local tumor of foot, so that we can diagnose it early and manage properly.

Verrucous carcinoma is a rare, locally invasive, well differentiated, low-grade squamous cell carcinoma, with HPV as a possible causative agent. It has a variety of different names; each is distinguished by its differing location but represents the same pathological condition.[1] These terms include: epithelioma cuniculatum plantare, giant condylomata acumina and of the anorectal region (Buschke-Loewenstein tumour), verrucous carcinoma of the oropharynx, papilloma cutis carcinoids, and cutaneous squamous carcinoma.[2-4] It follows a chronic course and mimics a variety of skin lesions, delaying diagnosis by up to 15 years. The definitive diagnosis is made histologically, and treatment by wide local excision is recommended. Most patients with verrucous carcinoma have a good prognosis.

Local recurrence is not uncommon, but metastasis to distant parts of the body is also rare.

Verrucous carcinoma may occur in several locations in the head and neck, gingiva, buccal mucosa, hard palate, floor of the mouth, larynx, oesophagus, penis, vagina, scrotum. The oral cavity is the most common site of this tumor.

Verrucous carcinoma of the foot is very rare.

2. Material and method
All the cases in this series were chronic smoker males and in their 4th to 5th decade of age. 3 patients had lesion on the heel (figure 1,4, 5) and one over forefoot (figure 6). The patients were given history of 2 – 5 years slowly growing lesion with discharging some whitish material. The lesion was increased gradually in size, was painless and slowly growing, non-tender, with irregular surface and everted margins. There is no inguinal lymphadenopathy. Wedge biopsy sent on opd basis and report was suggestive of verrucous carcinoma. Morphologic picture showed a biopsy occupied by an ulcerated, polypoid mass characterized superficially by hyper and parakeratosis, and acanthosis; deeply
the tumor invaded with broad strands that often contained keratin-filled cysts in center. Mitotic activity was low and confined in basal layer. The fibrous stroma surrounding the lesion revealed ectatic vessels, moderate inflammatory infiltrate with neutrophils and focal necrosis (Figure 8).

The definitive histopathological diagnosis was verrucous carcinoma, that is, low-grade squamous cell carcinoma. The patient was posted for wide local excision (1 cm margin) (figure 6) and flap surgery. Pedicled Reverse peroneal artery flap (figure 2) with skin grafting for the donor site was done for the patient with lesion on heel. All margins and base were free from tumor in final excisional biopsy report. After 7-10 days patient discharged and followed after 18 days for final inset. Patient was discharged after 3 days. On 3 month and 6 month follow up flap was healthy (figure 3). Patient satisfaction and functional outcome was good. The patient with lesion on forefoot was treated using reverse medial planter flap and skin graft (figure 7) and discharged after 7 days. A summary of all these 4 patients given in underlying table 1. Summary of previous publications with their references given in Table 2.

| Age/s | location | Size of lesion | Size of defect after wide local excision | Primary surgery | Additional surgery | Biopsy | Flap status | Total inpatient days |
|-------|-----------|----------------|----------------------------------------|-----------------|--------------------|--------|-------------|---------------------|
| 1     | 45/m      | Heel           | 6.5x7 cm                               | 7.5x8 cm        | WLE + RPAF         | As above | As above    | Healthy             | 12 days             |
| 2     | 42/m      | Heel           | 6x7 cm                                 | 7x8 cm          | WLE + RPAF         | As above | As above    | Healthy             | 13 days             |
| 3     | 48/m      | Heel           | 6.5x7 cm                               | 7.5x8 cm        | WLE + RPAF         | As above | As above    | Healthy             | 10 days             |
| 4     | 40/m      | forefoot       | 4.4x4 cm                               | 5.4x5cm         | WLE + Reverse medial planter flap | no      | As above    | Healthy             | 7 days              |

Figure 1. verrucous carcinoma of heel

Figure 2. reverse peroneal artery flap with peroneal artery
Yogesh Bhatt et al / Verrucous carcinoma of the foot - a series of 4 cases

Figure 3. final result with RPAF

Figure 4. 2nd case of verrucous carcinoma heel

Figure 5. 3rd case of verrucous carcinoma heel

Figure 6: 4th case of verrucous carcinoma, lesion on forefoot. Wide local excision with 1 cm margin

Figure 7: Reverse medial planter flap with skin graft

Figure 8: Microscopic view of verrucous carcinoma foot
3. Discussion

Verrucous carcinoma is a subtype of a low-grade SCC, not an SCC with a verrucous presentation. The pathogenesis of VC is unclear, but all arise de novo in the weight-bearing areas of the foot. VC has histological similarities to plantar warts, and HPV may be a causative agent.[2][17-19] Indeed, the deoxyribonucleic acid of HPV types 6, 11, 16, and 18 have been identified in VC specimens.[18] VC typically occurs in men in their 4th to 6th decades, although it has been seen in patients as young as 16 years.[1] It follows a chronic course, evolving from a discrete focal lesion to a large fungating deep penetrating mass. The slow growth and confusing early appearances can lead to delays in diagnosis of 8 to 15 years.[17], and hence under-treatment. Differential diagnoses include viral warts, pseudocarcinomatous hyperplasia, and mycosis.[19][20] Although the clinical and macroscopic findings can be marked (the formation of a bulky, exophytic mass, which may ulcerate with numerous sinuses from which foul-smelling purulent keratinous debris is expressed), the definitive diagnosis is made pathohistologically. Specimens exhibit both endophytic and exophytic growth patterns.[21] Proliferations are usually composed of large pale-staining well-differentiated keratinocytes, with the presence of pronounced hyperkeratosis, parakeratosis and papillomatisis. Unlike SCC, keratin pearl formation is little uncommon. Tumour strands may extend deep into the dermis and forming keratin-filled intra-epidermal abscesses and sinuses connecting with the surface. These sinus tracts are the ‘rabbit burrow’-like spaces from which epithelioma cuniculatum derives its name. The surrounding stroma may demonstrate an infiltration of lymphocytes, eosinophils, histiocytes and plasma cells. VC is a low-grade, locally invasive tumour, which generally never metastasises, and thus has a favourable prognosis.[4] The recommended treatment is wide local excision, rather than marginal excision, as VC often causes structural distortion of adjacent tissues, and the margins are not always apparent intra-operatively. The residual defect can then be covered with a skin graft or radial forearm free flap.[20][22] Other therapeutic modalities include topical chemotherapy, electrocautery, and cryotherapy but all have high recurrence rates.[2][22] Mohs microscopically controlled surgery has reported good results for the less invasive VC. Radiotherapy is not recommended, despite being curative in some reports, because of the possibility of malignant change.[2][19][20] Partial (ray or 5th metatarsal)[1],[17] or radical (foot or below-knee)[4][17][21] amputations are occasionally required for aggressively invasive disease, and in the presence of poor vascular status, massive skin defects, postoperative infections, or in tumour recurrence secondary to incomplete excision. The long-term prognosis for definitively treated VC is good, with cure rates of up to 98%. Patients should be reviewed annually as recurrence and metastasis remain a possibility. Initially amputation was the most commonly used treatment for verrucous carcinoma but now the wide local excision is the standard. We underwent WLE in all of our 4 patients.

4. Conclusion

Most of patients having soft tissue lesion on heel/foot referred to plastic surgeon because reconstructive options for heel/foot are difficult for general surgeons/orthopedicians. So as a plastic surgeon we should be aware of this rare but local

---

Table 2: data of previous publications with their references on verrucous carcinoma of foot

| Age/sex | Site and size of lesion | Management | Reference |
|---------|------------------------|------------|-----------|
| 1 63/m  | Forefoot, 3x1.7 cm     | Amputation of forefoot | [5]       |
| 2 44/m  | Interdigital folds of both feet | Bilateral forefoot amputation | [6]       |
| 3 74/m  | Heel                   | Below knee amputation   | [7]       |
| 5 34/m  | Forefoot, 3x4 cm       | Wide local excision with 5th metatarsal amputation | [8]       |
| 6 66/m  | forefoot               | Wide local excision with skin graft | [9]       |
| 7 54/m  | forefoot               | Amputation of 3,4,5 metatarsals | [10]      |
| 8 37/m  | forefoot               | Wide local excision with 2,3 toe amputation | [11]      |
| 9 50/m  | forefoot               | Wide local excision     | [12]      |
| 10 46/m | -                      | Wide local excision     | [13]      |
| 11 66/m | sole                   | Wide local excision     | [14]      |
| 12 77/m | 3,4 webspace           | -                       | [15]      |
| 13 62/m | sole                   | -                       | [16]      |
| 69/m    |                        |                          |           |

---

Yogesh Bhatt et al / Verrucous carcinoma of the foot - a series of 4 cases

594

www.ssjournals.com
tumor of foot, so that we can diagnose it early and manage properly.

We present a case series of 4 patients of verrucous carcinoma of the foot. This is a rare, locally invasive, well differentiated, low-grade squamous cell carcinoma, with human papilloma virus (HPV) as a possible causative agent. It follows a chronic course and mimics a variety of skin lesions, delaying diagnosis up to 15 years. The definitive diagnosis is made histologically, and treatment by wide local excision is recommended. Our patients underwent wide local excision and reconstructed. Three patients had lesion on heel, were managed using reverse peroneal artery flap and fourth patient had lesion on forefoot, was managed using reverse medial planter artery flap. There were no postoperative complications. There was no functional impairment. (Summary: Table 1)

Conflict of interest statement: Authors declare that they have no funding and no conflict of interest.

Ethical standard: The manuscript does not contain clinical studies or patient data.

References

[1] Brownstein MH, Shapiro L. Verrucous carcinoma of skin: epithelioma cuniculatum plantare. Cancer 1976; 38:1710–6.
[2] Schwartz RA. Verrucous carcinoma of the skin and mucosa. J Am Acad Dermatol 1995; 32:1–21.
[3] Yip KM, Lin-Yip J, Kunta S, Leung PC. Subcutaneous (“inverted”) verrucous carcinoma with bone invasion. Am J Dermatopathol 1997; 19:83–6.
[4] Dogan G, Oram Y, Hazneci E, Ozen S, Karincaoglu Y, Ciralik H. Three cases of verrucous carcinoma. Australas J Dermatol 1998; 39:251–4.
[5] Sarma D, Wang J, Bewtra C, Lee L. Verrucous Carcinoma Arising In A Chronic Non-Healing Ulcer Of The Foot Of A Diabetic Patient. The Internet Journal of Dermatology, Volume 5 Number
[6] Kary Suen, Shiran Wijeratne and John Patrikios. An unusual case of bilateral verrucous carcinoma of the foot (epithelioma cuniculatum). JSCR 2012; 12 (3 pages) doi:10.1093/jscr/rjs020
[7] Pempinello C., Bova A., R. Pempinello, Luise R., and Iannacci G. Verrucous Carcinoma of the Foot with Bone Invasion: A Case Report. Case Reports in Oncological Medicine. Volume 2013 (2013), Article ID 135307, 3 pages
[8] Lesic A, Nikolic M, Sptoa J, Starcevic B, Bumbasirevic M, HDE Atkinson. Verrucous carcinoma of the foot: a case report. Journal of Orthopaedic Surgery 2008;16(2):251-3
[9] Ophir Schein MD1, Ari Orenstein MD2 and Eran Bar-Meir MD2. Plantar Verrucous Carcinoma (Epithelioma Cuniculatum): Rare Form of the Common Wart. IMAJ 2006;8:885
[10] J Garcia-gavin, d Gonzalez-vilas, Rodriguez-pazos, d sanchez-agular, j toribio. Verrucous carcinoma of the foot affecting the bone: utility of computed tomography scanner. Dermatology online journal 16 (2):8
[11] 11Jean Bentley, Dr R Sriramman, Dr Neill Hepburn. The remarkable case of verrucous carcinoma: a rare find. Practical Diabetes Vol. 30 No. 4
[12] Robert allen Schwartz, Gordon h burgess. Verrucous carcinoma of the foot. Journal of surgical oncology 14:333–339 (1980)
[13] Diandra k Gordon, erinn n ponder et al. verrucous carcinoma of the foot, not your typical planter wart: a case study. The foot 06/2014 DOI: 10/2016/j.foot.2014.03.009
[14] Nakamura, Yoshitaka; Kashiwagi, Keisuke; Nakamura, Akiko; Muto, Masahiko. Verrucous Carcinoma of the Foot Diagnosed Using p53 and Ki-67 Immunostaining in a Patient With Diabetic Neuropathy. American Journal of Dermatopathology: March 2015; 37 (3): 257–259.
[15] Catherine McKay, Penelope McBride, James Muir. Planter verrucous carcinoma masquerading as toe web intertrigo. Australian Journal of Dermatology DOI: 10.1111/j.1440-0960.2010.00721.x
[16] Carlos r. milo, ivanir s. milo and lenita p. souza. Epithelioma cuniculatum, a verrucous carcinoma of the foot, report of two cases. Dermatologica 1981; 163: 338 –342.
[17] Green JG Jr, Ferrara JA, Haber JA. Epithelioma cuniculatum plantare. J Foot Surg 1987; 26:78–83.
[18] Schell BJ, Rosen T, Rady P, Arany I, Tschen JA, Mack MF, et al. Verrucous carcinoma of the foot associated with human papillomavirus type 16. J Am Acad Dermatol 2001; 45:49–55.
[19] Smith PJ Jr, Hylinski JH, Axe S. Verrucous carcinoma: epithelioma cuniculatum plantare. J Foot Surg 1992; 31:324–8.
[20] Wright PK, Vidyadharan R, Jose RM, Rao GS. Planter verrucous carcinoma continues to be mistaken for verruca vulgaris. Plast Reconstr Surg 2004; 113:1101–3.
[21] McCann JJ, Al-Nafussi AI. Epithelioma cuniculatum plantare. Br J Plast Surg 1989; 42:79–82.
[22] Spyrounis P, Tentis D, Sparveri I, Arvanitis T. Planter epithelioma cuniculatum. A case report with review of the literature. Eur J Plast Surg 2004; 27:253–6.