Palmoplantar epidermoid cysts: two cases and brief review of the literature

Jorge Arandes-Marcocci MD, Gemma Melé-Ninot MD, Mònica Quintana-Codina MD, Maribel Iglesias-Sancho MD, Montse Salleras Redonnet MD PhD.

Affiliations: Department of Dermatology, Hospital Universitari Sagrat Cor, Barcelona, Spain

Corresponding Author: Jorge Arandes MD, Department of Dermatology, Hospital Universitari Sagrat Cor, Viladomat Street 288, 08029, Barcelona, Spain, Tel: 34-933221111, Email: jorge.arandes@hotmail.com

Abstract

Palmoplantar epidermoid cysts can range in clinical presentation from an asymptomatic slowly enlarging mass to a painful nodule. We report two cases: an epidermoid cyst on the sole and another on the palm. This article reviews the possible etiology, diagnosis, and prognosis of palmoplantar epidermoid cysts.

Keywords: epidermoid cysts, palmoplantar, human papillomavirus, eccrine ducts, ultrasonography, metaplasia

Introduction

Epidermoid cysts (ECs) are firm intradermal or subcutaneous tumors that develop from the epithelium of the infundibular portion of the hair follicle [1]. Most cases have their onset in middle age and show a male predominance [1, 2]. The face, neck, scalp, and trunk are the most commonly affected regions [1]. Currently, the etiology and pathogenesis of ECs remain unclear. We herein provide two cases of palmoplantar ECs and a review of the literature, emphasizing two atypical locations.

Case Synopsis

Case 1

An 82-year-old woman was referred for a painful nodule on her left sole, which had gradually enlarged over the prior 6 months. She denied a history of trauma . On physical examination, she exhibited a unique well-defined, skin-colored nodule on her left sole (**Figure 1A)**. Ultrasonography revealed a hypoechoic, well-circumscribed oval nodule measuring 21mm in maximum diameter. On color

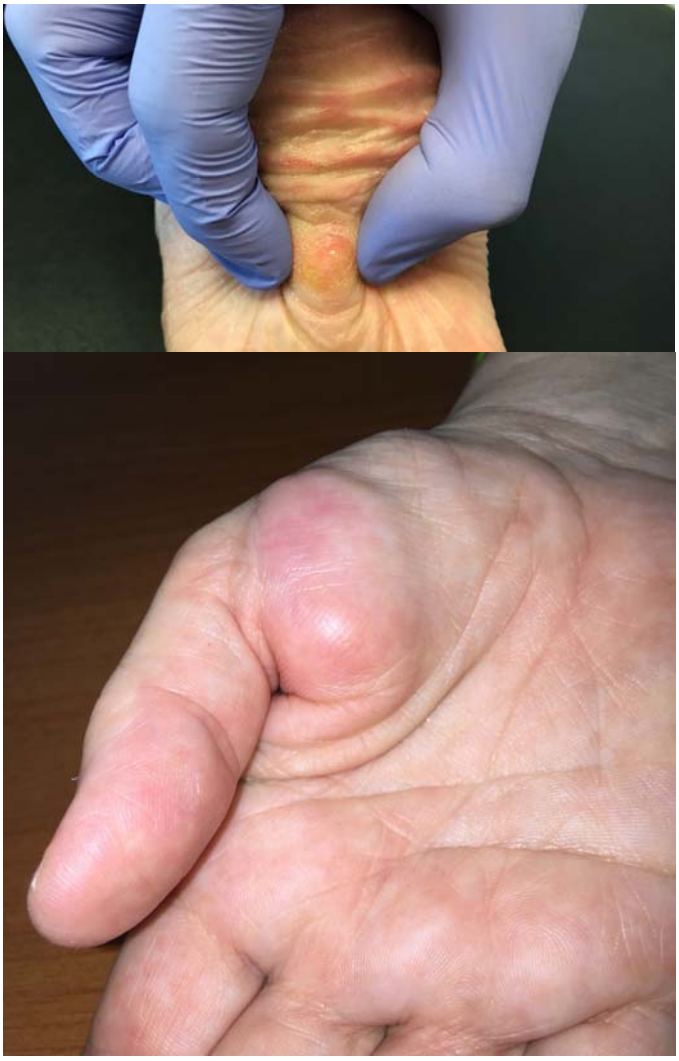

Figure 1. A) *Well-defined, skin-coloured and indurated 20×15mm nodule on her left sole.* **B)** *Solitary tender 30×30mm nodule, with inflammatory signs on the right thenar eminence.*

Table 1: Non-palmoplantar location reports revealing the presence of HPV.

Author	Year	Type of study	Patient #	Diagnosis	HPV-type
Lee S et al.	2003	Retrospective study	6	PCR	60
Jeon J et al.	2005	Correspondence letter	2	Histology and PCR	60
Ramagosa R et al.	2008	Case report	1	PCR	6, 8
Kawase M et al.	2018	Concise communication	1	Immunochemistry and DNA-DNA *in situ* hybridization	6, 11

doppler sonography, blood flow was not present within the lesion. Surgical excision was subsequently performed. Histopathology examination showed laminated fibrillar keratin covered by several layers of squamous and granular cells (**Figure 2**). Consequently, the diagnosis of epidermoid cyst was confirmed.

Case 2

A 63-year-old woman presented with complaints of swelling on her right palm for the last three weeks. She noted a ten-year history of a painless enlarging nodule for which she had consulted a physician on multiple occasions. No history of trauma was recalled. Physical examination showed a solitary tender nodule with inflammatory signs on the right palm (**Figure 1B)**. Ultrasonography revealed a well-circumscribed, oval lesion located in the dermis and subcutaneous tissue (**Figure 3**). Blood flow was not observed within the lesion on color doppler imaging. An incisional biopsy was performed. Histopathology examination showed laminated keratotic material lined with a squamous epithelium including a granular layer, all findings compatible with an epidermoid cyst. The patient refused excisional intervention.

Figure 2*: Histological examination shows a cystic wall filled with eosinophilic keratin, lined by a stratified squamous epithelium and granular layer. H&E, 10×.*

Case Discussion

Occasionally, ECs occur in areas devoid of hair follicles, such as palms and soles. Although implantation of epidermal fragments into the dermis as a result of a traumatic injury had previously been thought to be the main cause of palmoplantar ECs, Yanagihara et al. reported human papillomavirus (HPV) as another etiology [1, 2]. HPV that have been detected in palmoplantar ECs include HPV 57 and HPV 60 [3].

Currently, there are at least two main alternative explanations for the association between palmoplantar ECs and HPV infection; a superinfection by a blunt penetrating injury (wart penetration located on the weight-bearing surfaces) versus metaplasia of the epithelium of eccrine ducts in response to the infection [4, 5]. Egawa et al. speculated that HPV initially infects the upper parts of the eccrine duct, where ridged warts develop, and then migrates into dermal portions of the eccrine ducts, where the virus-associated ECs develop [5].

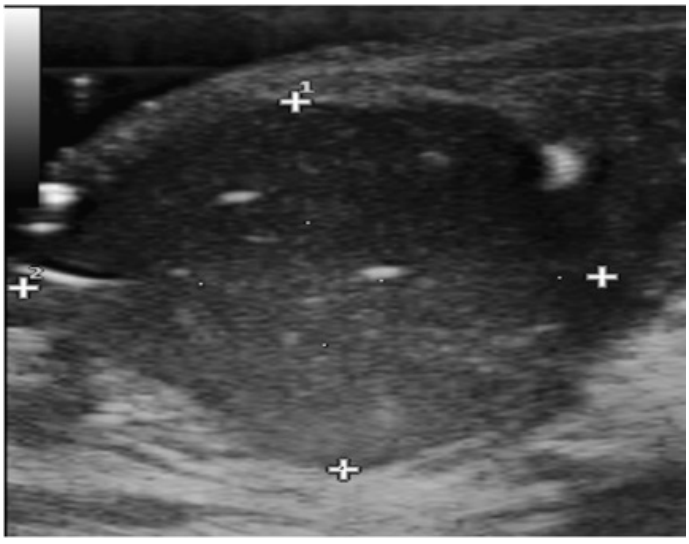

Figure 3*: Ultrasonography shows a 15×16mm well-circumscribed, heterogeneous, oval lesion located in the dermis and subcutaneous tissue.*

PCR has been demonstrated to be a more sensitive tool than immunochemistry for the detection of HPV [4]. However, histological examination in our two cases did not show specific HPV changes such as vacuolated cells, intracytoplasmic inclusion bodies, nor eccrine glands [3, 5]. Therefore, tests for HPV were not performed.

In addition, an association between HPV 6, 8, 11 and 60 with non-palmoplantar ECs has also been reported. (**Table 1**) In these sites, histological findings did not usually show the characteristic morphology as in palmoplantar locations [6, 7].

Sato et al. described ultrasonography and magnetic resonance imaging findings in patients with HPV-associated plantar ECs. By ultrasound, these lesions presented as several adjacent globular cysts with posterior acoustic enhancement and with no vascularization on color doppler sonography [8]. Owing to the lack of these characteristic imaging and histological features in our cases, we question the theory of HPV in an etiological role in palmoplantar ECs.

Epidermoid cysts of the sole constituted 7.2% of plantar lesions in a large histopathological study, being the third most common type of lesion in this location [1]. In a recent review by Nigam et al, of a total of 103 body ECs, 19 were located on the feet; the heel was the most common site followed by the lateral border of the sole [9]. The differential diagnosis of plantar ECs includes neuromas, ganglion cysts, lipomas, warts, and appendageal tumors [1].

In a review by Lincoski et al., ECs of the palms represented an incidence of 16% among 623 tumors of the hand from 1977 to 2004. Patients typically complained of a painless enlarging mass and the majority (71%) were located on the palmar aspect of the fingers, unlike our case [10]. Contrary to ECs of the sole, more than half occurred in manual laborers and 48% recalled a traumatic incident that preceded the cyst. The differential diagnosis should include ganglion cyst, foreign body granuloma, gouty tophus, fibroma of tendon sheath, lipoma, mucoid cysts, glomus tumor, neurofibroma, schwannoma, and malignant neoplasms [10].

In most cases of ECs of the sole, the presence of parakeratosis and a focal absence of granular layer were found at least at the upper portion of the cyst wall [11]. When ECs rupture, the cyst contents are released into the dermis, with resultant keratin granuloma formation that exhibits foreign-body giant cells and chronic inflammatory cells [12].

Treatment consists of surgical excision of the whole lesion to avoid recurrences, especially on palms and soles, but the recurrence rate is high [4, 9, 10]. However, further studies are needed to establish if HPV is a cause and if ECs related to HPV present a higher risk of recurrence.

Conclusion

In summary, we describe ECs in two unusual sites. Considering that this is an uncommon finding, we would like to make practitioners aware of this entity.

Acknowledgments

The authors would like to acknowledge Luis Novoa Lamazares MD for his assistance in medical writing, and María Teresa Fernández-Figueras MD, PhD for her contribution of the histological information.

Conflicts of interest

The authors declare no conflicts of interests.

References

1 Fisher BK, Macpherson M. Epidermoid cyst of the sole. *J Am Acad Dermatol*. 1986;15:1127–1129. [PMID: 3771865].

2 Yanagihara M, Sumi A, Mori S. Papillomavirus antigen in the epidermoid cyst of the sole. *J Cutan Pathol*. 1989;16:375–281. [PMID: 2559111].

3 Kitasato H, Egawa K, Honda Y, et al. A putative human papillomavirus type 57 new subtype isolated from plantar epidermoid cysts without intracytoplasmic inclusion bodies. *J Gen Virol*. 1998;79:1977–1981. [PMID: 9714246].

4 Park H-S, Kim W-S, Lee J-H, et al. Association of human papillomavirus infection with palmoplantar epidermal cysts in Korean patients. *Acta Derm Venereol*. 2005;85:404–408. [PMID: 16159731].

5 Egawa K, Honda Y, Inaba Y, Ono T, Villiers E- MD. Detection of

human papillomaviruses and eccrine ducts in palmoplantar epidermoid cysts. *Br J Dermatol*. 1995;132:533–542. [PMID: 7748742].

6 Lee S, Lee W, Chung S, et al. Detection of human papillomavirus 60 in epidermal cysts of nonpalmoplantar location. *Am J Dermatopathol*. 2003;25:243–247. [PMID: 12775987].

7 Kawase M, Egawa K, Ishiji T, Nakagawa H. Human papillomavirus type 6/11 identified in an epidermoid cyst of the scrotum. *J Dermatol*. 2018;45:224–227. [PMID: 24057441].

8 Sato Y, Nozaki T, Matsusako M, et al. Human papillomavirus-associated plantar epidermoid cysts: MR and US imaging appearance. *Skeletal Radiol. 20*14;43:257–261. [PMID: 24057441].

9 Nigam JS, Bharti JN, Nair V, et al. Epidermal Cysts: A Clinicopathological Analysis with Emphasis on Unusual Findings. *Int J Trichology*. 2017;9:108–112. [PMID: 28932061].

10 Lincoski CJ, Bush DC, Millon SJ. Epidermoid cysts in the hand. *J Hand Surg Eur* 2009;34:792–796. [PMID: 19786411].

11 Shimizu Y, Sakita K, Arai E, et al. Clinicopathologic features of epidermal cysts of the sole: comparison with traditional epidermal cysts and trichilemmal cysts. *J Cutan Pathol*. 2005;32:280–285. [PMID: 15769277].

12 Harish S, Jan E, Ghert M, Alowami S, Finlay K. Pseudotumoral appearance of a ruptured epidermal cyst in the foot. *Skeletal Radiol. 20*06;35:867–871. [PMID: 16794809].