A Case of Porokeratotic Eccrine Ostial and Dermal Duct Nevus

Young Ran Yoon, M.D., Nack In Kim, M.D., Woo Young Sim, M.D.,
Mu Hyoung Lee, M.D., Choong Rim Haw, M.D.

Department of Dermatology, College of Medicine, Kyung Hee University, Seoul, Korea

We report a case of porokeratotic eccrine ostial and dermal duct nevus in a 28-year old man. The skin lesions, present since birth, were multiple keratotic papules and punctate pits on the palms, soles and heels bilaterally. Histopathologically, the lesion was characterized by dilatation and hyperplasia of the eccrine sweat ducts with parakeratotic plugs related to sweat gland pores. This entity should be considered the differential diagnosis of other types of porokeratosis and comedo-like keratosis on palms and soles. (Ann Dermatol 3:(1) 40—44, 1991)

Key Words: Cornoid lamella, Eccrine duct hyperplasia, Porokeratotic eccrine ostial and dermal duct nevus

Porokeratotic eccrine ostial and dermal duct nevus was first described in 1980 by Abell and Read[1]. They described an epidermal nevus, which pathologically demonstrated gross examples of cornoid lamellae associated exclusively with the eccrine duct and ostia. Since then a number of cases have been reported[2-6]. Clinically, porokeratotic eccrine ostial and dermal duct nevus occures on the plantar or palmar area, with multiple punctate pits and hyperkeratotic plugs arranged in a linear or bandlike fashion. Histopathologically, the lesion is characterized by enlarged sweat ducts with the presence of parakeratotic plugs associated exclusively with the sweat gland pores and ducts.

In this report, we describe an unusual case of porokeratotic eccrine ostial and dermal duct nevus, followed by a review of the literature and discussion of the differential diagnosis of porokeratotic dermatoses.

Received April 28, 1990
Accepted for publication May 26, 1990

Reprint request to: Nack In Kim, M.D., Department of Dermatology, College of Medicine, Kyung Hee University, 1, Hoeki-Dong, Dongdaemun-Ku, Seoul 130-702, Korea

This case was presented at the 42nd Annual Meeting of the Korean Dermatological Association on April 20, 1990.

REPORT OF A CASE

A 28-year-old man was seen at the department of dermatology, Kyung Hee University Hospital with asymptomatic skin lesions on the palms, soles and heels which had been present since birth. Past medical and family history were unremarkable.

On physical examination, multiple small papules with comedo-like keratotic plugs were present on the palms and soles of his feet (Fig. 1-3). The keratotic papules had annular raised margins and atrophic or pitted centers (Fig. 4). Each plug was firmly attached at its base and could not be easily removed. A single keratotic plug extracted from one of the papules, measured 5mm in length and 2mm in breadth. There were no other skin lesions and the rest of the physical examination was unremarkable. Sweat gland function using starch iodine technique demonstrated focal anhidrosis in the area of the lesions after heat induced sweating.

Laboratory studies were all within normal limits. Serologic test for syphilis was negative. The chest radiograph was normal. Histologic examination of three biopsy specimens taken from the left palm, left sole, and heel showed similar abnormalities of the eccrine ostia and ducts. The most distinct feature was a keratotic plug with a characteristic parakeratotic cornoid lamella-like

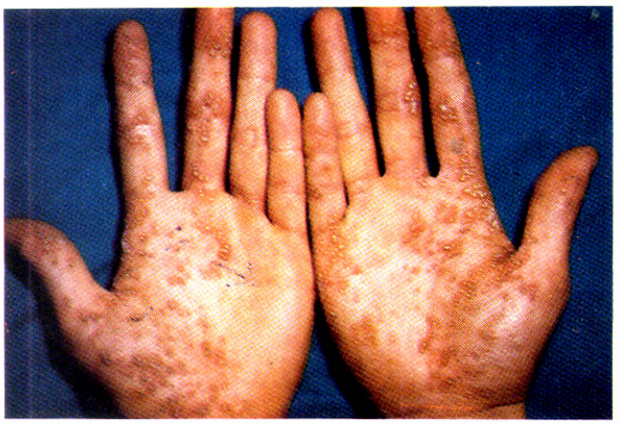

Fig. 1. Symmetric multiple linear or band-like keratotic papules on both palms.

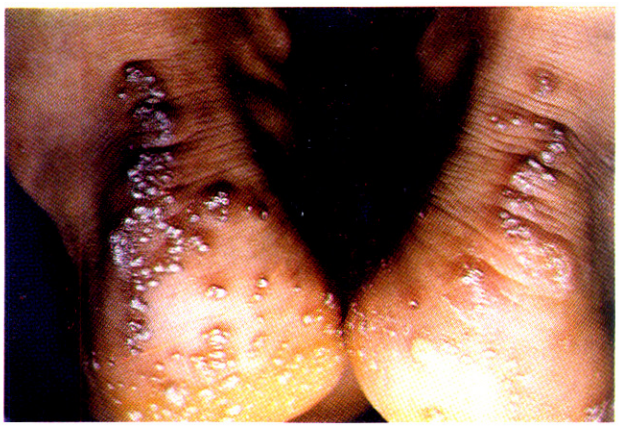

Fig. 3. Symmetrical linear or bandlike keratotic papules on both heels.

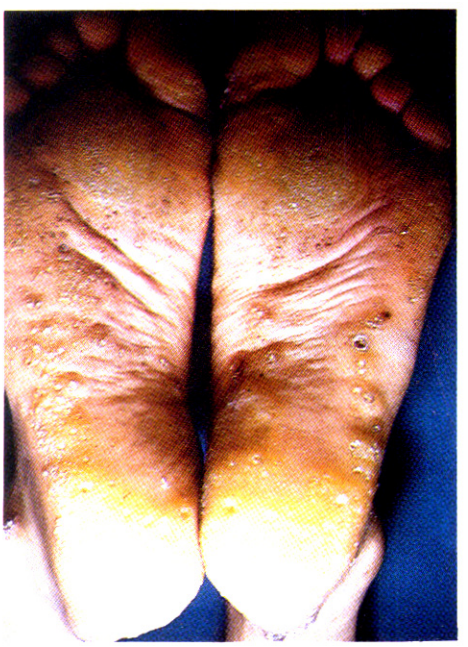

Fig. 2. Marked thickened hyperkeratotic plaque with keratotic papules and punctate pits on both soles

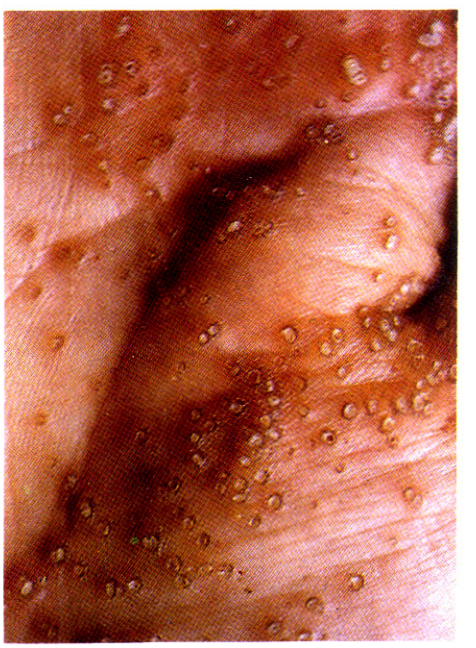

Fig. 4. Close up view of the palm; The keratotic papules are composed of annular margins and atrophic or pitted centers.

center filling an invagination formed by a dilated and hyperplastic acrosyringium and a grossly dilated distal eccrine duct. The glandular portion of the eccrine duct was normal. The granuar layer was reduced focally but no dyskeratotic cells were seen. Adjacent epidermal cells showed vacuolation. There was no inflammatory infiltrate in the dermis (Fig 5-9).

Based on clinical appearance and histologic findings, a diagnosis of porokeratotic eccrine ostial and dermal duct nevus was made.

DISCUSSION

Porokeratotic eccrine ostial and dermal duct nevus (PEODDN) is a recently described disease entity with only nine cases previously reported in the literature[1-6]. Most authors describe this lesion as a variant of porokeratosis[7] or a variant of epidermal nevus[1, 2, 3]. The first case was described in 1980 by Abell and Read in a 3-year-old girl with linear keratotic papules on her left foot. They

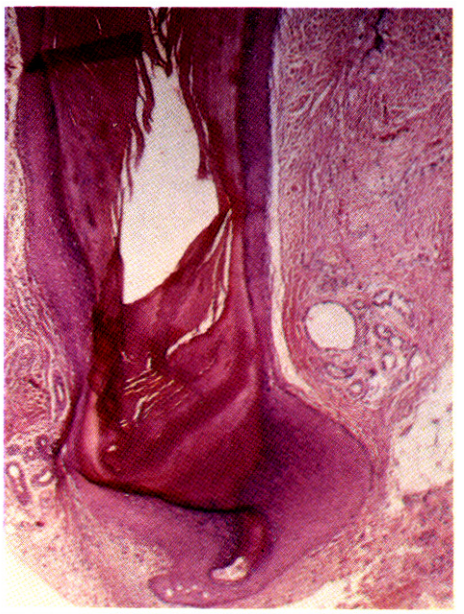

Fig. 5. The histopathologic finding showing a keratotic plug with a cornoid lamella-like center that filled an invagination formed by a dilated and hyperplastic acrosyringium and a grossly dilated distal eccrine duct (H & E stain, ×40).

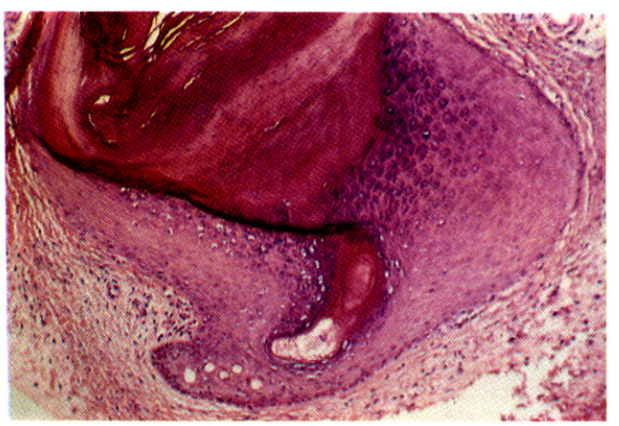

Fig. 6. High-power view of the lower portion of a lesion showing the base of the cornoid lamella, hyperplastic acrosyringium and eccrine ostia (H & E stain, ×100).

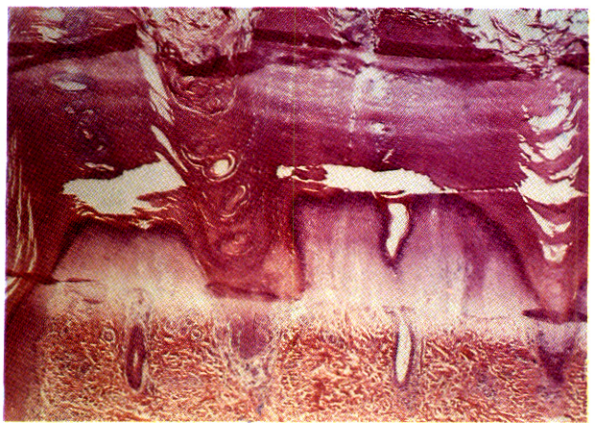

Fig. 7. Showing multiple portion of parakeratotic plugs from deep epidermal invagination associated with eccrine sweat duct (H & E stain, ×40).

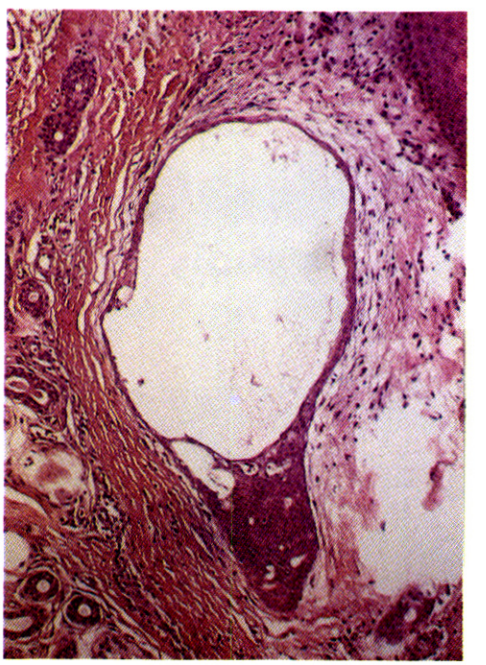

Fig. 8. Upper dermis showes a large dilated eccrine duct (H & E stain, ×40).

considered this disease to be a type of eccrine hamartoma with cornoid lamellation.

Clinically, PEODDN is characteristically seen on the palms and soles as multiple keratotic plugs and punctate pits in a linear distribution. One case has been described with extensive lesions not only on the hands and feet but also involving the extremities, neck and axilla[2]. Almost all cases had been congenital except one case[5] and had no family history[1-6]. Focal anhidrosis at the site of the lesions is a commonly described finding.

Pathologically, PEODDN is characterized by: firstly, cornoid lamedllae that are exclusively associated with the eccrine ostium and duct, and secondly, dilatation and hyperplasia of the acrosyringium and the deeper duct[5].

The differential diagnosis of PEODDN includes punctate porokeratosis, linear porokeratosis,

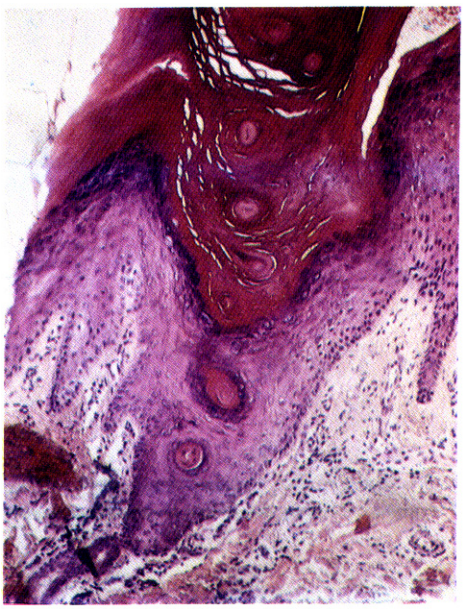

Fig. 9. A parakeratotic plugs arising from acrosyringium and distal eccrine duct (H & E stain, ×100).

porokeratosis palmaris plantaris et disseminata, porokeratotic eccrine duct and hair follicle nevus, nevus comedonicus and punctate keratoses of the palms and soles (PK) and keratotic pits of the palmar creases (KPPC). Clinical and histopathological features of these lesions are listed in Table 1.

The lesions of punctate porokeratosis[10-12], linear porokeratosis[13] and porokeratosis palmaris plantaris et disseminata[14] have a similar distribution and appearance as PEODDN but differ histopathologically. In punctate porokeratosis, there is no eccrine duct hyperplasia and the invagination of the epidermis is more shallow in contrast to PEODDN. In linear porokeratosis and porokeratosis palmaris plantaris et disseminata, cornoid lamella formation is connected not only to the eccrine sweat duct but also to the follicular infundibulum and the epidermis between the adnexa. In 1982, Coskey et al[15] reported a case of porokeratotic eccrine duct and hair follicle nevus, which resembled PEODDN in many respects. But only different histopathological finding is the hair follicle involvement. Nevus comedonicus occasionally shows palmoplantar involvement, which is characterized by malformation of pilosebaceous unit, resulted in cystic dilatation of the follicular canal and keratotic plugging, and there is no cornoid lamella[16]. PK[17] and KPPC[17, 18] show keratotic papules that are scattered diffusely on the palms and soles, or conical depressions confined to the palmar creases. Histopathologically, PK and KPPC are characterized by compact hyperkeratosis without parakeratosis and a normal to slightly increased granular

Table 1. Differential diagnosis of porokeratotic eccrine ostial and dermal duct nevus from other similar dermatoses

	PEODDN	PP	NC	PK & KPPC	Our Case
cornoid lamella	+	+	−	±	+
sweat duct involvement	+	±	−	±	+
hair follicle & epidermis involvement	−	+	+	+	−
eccrine duct dilatation & hyperplasia	+	−	−	−	+
depth of invagination	deep	shallow	deep	shallow/no	deep
anhidrosis	+	−	−	−	+
family history	−	±	−	±	−
age of onset	at birth	later	at birth or later	later	at birth

* PEODDN; porokeratotic eccrine ostial and dermal ductal nevus
PP; punctate porokeratosis
NC; nevus comedonicus
PK; punctate keratosis of the palms and soles
KPPC; keratotic pits of the palmar creases

layer. In some cases reported, cornoid lamella was seen in the center of the hyperkeratoic plug, but recently many authors thought that these cases represent punctate porokeratosis rather than PK and KPPC[8, 10, 11]. And PK and KPPC show no eccrine duct dilatation and hyperplasia, and there is shallow or no invagination of the epidermis. Our case has the characteristics of the bilatesal, linear PEODDN without family history. To our knowledge this is the first report of Korean literature.

REFERENCES

1. Abell E, Read SI: *Porokeratotic eccrine ostial and dermal duct nevus. Br J Dermatol 103:435-441, 1980.*
2. Aloi FG, Pippione M: *Porokeratotic eccrine ostial and dermal duct nevus. Arch Dermatol 122:892-895, 1986.*
3. Driban NE, Cavicchia JC: *Porokeratotic eccrine ostial and dermal duct nevus. J Cutan Pathol 14:118-121, 1987.*
4. Morena A, Pujol RM, Salvatella N, Alomar A, de Moragas JM: *Porokeratotic eccrine ostial and dermal duct nevus. J Cutan Pathol 15:43-48, 1988.*
5. Stoof TJ, Starink TM, Nieboer C: *Porokeratotic eccrine ostial and dermal duct nevus. J Am Acad Dermatol 20:924-927, 1989.*
6. Marsden RA, Fleming K, Dawber RPR: *Comedo nevus of the palm-a sweat duct nevus? Br J Dermatol 101:717-722, 1979.*
7. Arnold HL, Odom RB, James WD: *Diseases of the skin. 8th ed, W.B. Saunders Co, Philadelphia, 1990, p668.*
8. Lever WF, Schaumberg-Lever G: *Histopathology of the skin, 7th ed, JB Lippincott Co, Philadelphia, 1990, p525.*
9. Wade T, Ackerman AB: *Cornoid lamellation: A histologic reaction pattern. Am J Dermatopathol 2:5-15, 1980.*
10. Brown F: *Punctate keratoderma. Arch Dermatol 104:682-683, 1971.*
11. Herman PS: *Punctate porokeratotic keratoderma. Dermatologica 147:206-213, 1973.*
12. Lestringant GG, Berge T: *Porokeratosis Punctata Palmaris et plantaris. Arch Dermatol 125:816-819, 1989.*
13. Rahbari H, Cordero AA, Aires B, Mehregan AH: *Linear porokeratiosis. Arch Dermatol 109:526-528, 1974.*
14. Shaw JC, White CR Jr: *Porokeratosis plantaris palmaris et disseminata. J Am Acad Dermatol 11:454-460, 1984.*
15. Coskey RJ, Mehregan AH, Hashimoto K: *Porokeratotic eccrine duct and hair follicle nevus. J Am Acad Dermatol 6:940-943, 1982.*
16. Harper KE, Spielvogen RL: *Nevus comedonicus of the palm and wrist, Case report with review of five previous cases. J Am Acad Dermatol 12:185-188, 1985.*
17. Rustard OJ, Vance JC: *Punctate keratoses of the palms and soles and keratotic pits of the palmar creases. J Am Acad Dermatol 22:468-476, 1990.*
18. Coupe R, Usher B: *Keratosis punctata palmaris et plataris. Arch Dermatol 87:91-95, 1963.*