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### **Authors**

Malik, Sara Erickson, Christof P Calame, Antoanella et al.

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# Acquired giant plantar fibrokeratoma: case report and review

Sara Malik<sup>1</sup> BA, Christof P Erickson<sup>2</sup> MD, Antoanella Calame<sup>2</sup> MD, Philip R Cohen<sup>3,4</sup> MD

Affiliations: <sup>1</sup>Northwestern University Feinberg School of Medicine, Chicago, Illinois, <sup>2</sup>Compass Dermatopathology, San Diego, California, USA, <sup>3</sup>Touro University California College of Osteopathic Medicine, Vallejo, California, USA, <sup>4</sup>Department of Dermatology, University of California, Davis Medical Center, Sacramento, California, USA

Corresponding Author: Philip R Cohen MD, 10991 Twinleaf Court, San Diego, CA 92131, Email: mitehead@gmail.com

## **Abstract**

Acquired digital fibrokeratoma is a benign fibrous tumor usually located on the toes and fingers. A 63year-old man with an acquired giant plantar fibrokeratoma is described. He presented with an asymptomatic exophytic nodule of ten years duration; there is no history of trauma to the site. It measured 15x10x5mm and was located on the plantar foot proximal to the third toe. Excisional biopsy established the diagnosis of fibrokeratoma. Giant acquired fibrokeratoma has been described in 16 patients including ours: three women and 13 men. They are located on either the upper extremity (one man) or the lower extremity (15 individuals). Acquired plantar fibrokeratoma is rare. Including our patient, it has been reported in 11 patients: one woman and ten men. The woman was 13 years of age and the men ranged from 15 to 77 years old. Plantar acquired fibrokeratomas are located on either the plantar aspect of the toes, the sole of the foot, or the heel. An excisional biopsy provided adequate treatment without subsequent recurrence of both giant and plantar fibrokeratomas.

Keywords: acquired, CD34, digit, fibrokeratoma, finger, foot, giant, heel, periungual, plantar, subungual, toe

## Introduction

Acquired digital fibrokeratoma is a benign fibrous tumor [1,2]. It typically presents as an asymptomatic, filiform papule on the side of a finger or toe. However, albeit uncommon, giant acquired

fibrokeratomas develop that are greater than 1cm in size or are located on the plantar foot, or both [3-19]. A man with a giant acquired plantar fibrokeratoma is described and the features of other patients with either a giant and/or plantar fibrokeratomas are reviewed.

# **Case Synopsis**

A 63-year-old man presented for evaluation of an asymptomatic nodule on the bottom of his left foot. There has been no trauma to the site. The lesion has been present for at least ten years; it initially increased to its current size and there had been no additional growth during the past several years.

Cutaneous examination revealed a painless, flesh-colored exophytic nodule that measured 15x10x5mm (**Figure 1**). The lesion was located on the plantar surface of the left foot, just proximal to the third toe. Based on the morphologic presentation, the clinical differential diagnosis included acquired digital fibrokeratoma, angiofibroma, cellular digital fibroma, and superficial acral fibromyxoma.

An excisional biopsy using the shave technique was performed. Microscopic examination of the tissue specimen showed a dome-shaped nodule with epidermal hyperplasia and slight papillomatosis (**Figure 2**); there was also fibroplasia of the dermal collagen. The tumor cells were negative for cluster of differentiation 34 (CD34) stain (**Figure 3**). However, the dermal blood vessels were stained with the





**Figure 1**. Clinical presentation of an acquired giant plantar fibrokeratoma. **A)** Frontal and **B)** lateral views of the 15x10x5mm flesh-colored, exophytic nodule extending from the left plantar surface (proximal to the third digit) of a 63-year-old man's foot.

antigen. Correlation of the clinical presentation and the pathologic findings established a diagnosis of acquired giant plantar fibrokeratoma. Follow-up examination, four weeks later, showed complete healing of the biopsy site. Subsequently, there has been no recurrence of the tumor.

### **Case Discussion**

Acquired digital fibrokeratoma usually develops on the lateral digit of a patient's upper or lower extremity. Less frequently, it has been described at ectopic sites such as the elbow, heel, lower lip, nailbed, nose, periungual nailfold, or prepatellar leg. However, rarely a fibrokeratoma has been observed on the plantar foot [5].

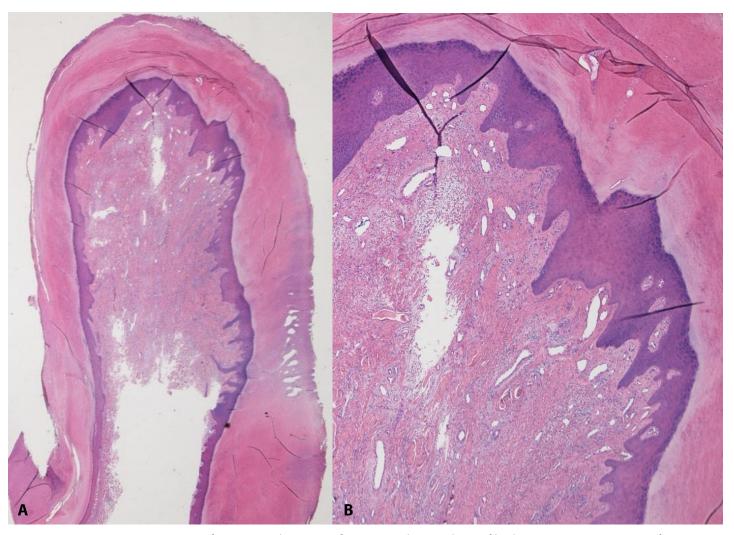
Acquired digital fibrokeratoma are typically less than 5mm in size. Yet, larger fibrokeratomas have been observed. The benign tumor is described as giant when it is greater than 1cm in length [6,9,19].

Giant fibrokeratoma have been described on the upper and lower extremity (Table 1), [3-19]. Including our patient, giant fibrokeratomas have been described in 16 individuals: three women and 13 men. Of all, the onset age ranged from 15 to 77 years (median, 46 years). The onset age ranged from 18 to 41 years (median, 33 years) in women, and from 15 to 77 years (median, 50 years) in men.

A giant fibrokeratoma located on the upper extremity was described in one individual. The patient was a 51-year-old man. To the best of our knowledge, a giant fibrokeratoma has not been reported on the upper extremity of a woman (Table 1), [3].

The duration of the giant fibrokeratoma on the upper extremity was 15 years prior to diagnosis. The patient did not have preceding trauma to the site. The benign tumor was located on the lateral surface of his thumb and presented as flesh colored, solitary, and round lesion; the size of the lesion was  $40\times25\times21$ mm.

In contrast, including our patient, a giant fibrokeratoma of the lower extremity was observed in three women and 12 men (Table 1, cases: 2-7 and 9-17), [5-10,12-19]. The age at diagnosis ranged from 18 to 41 years in women (median, 33 years). The diagnosis age in men ranged from 15 to 77 years (median, 49 years).



**Figure 2.** Microscopic examination of H&E-stained sections of an acquired giant plantar fibrokeratoma. **A)** Lower magnification view shows compact orthokeratosis, acanthosis and mild papillomatosis of the epidermis. **B)** Higher magnification view demonstrates collagen bundles oriented perpendicular to the overlying epidermis and numerous blood vessels in the dermis. H&E, **A)** x2; **B)** x10.

The fibrokeratomas were present for four months to 17 years (median, five years); seven of the patients had their fibrokeratoma for five or less years whereas the tumor was present for more than ten years in four individuals. Only one patient had prior history of trauma at the site: a 33-year-old woman whose tumor had been present for eight years and was located on the nailfold of her great toe [6]. The most common location of the lesion was the heel (four patients), sole of the foot (three patients), on a toe (three patients), and the plantar surface of a toe (two patients) and the lateral side of a toe (one patient).

The giant plantar fibrokeratoma had a similar morphology to the tumor when presenting on the

upper extremity; it usually appeared as a flesh-colored, solitary, and round nodule. Some of the giant plantar fibrokeratomas were hyperkeratotic and pedunculated. The length of the lesions ranged from 1cm to 4cm (median, 3cm) in length. Surgical excision is generally curative without recurrence.

Acquired plantar fibrokeratomas are rare (**Table 1**, cases: 1, 5, 7, 8, 10-14, 16 and 17), [4,8,10,11,13-17,19]. To the best of our knowledge, including our patient, plantar fibrokeratomas have been described in 11 individuals: one woman and ten men. Overall, they ranged in age from 13 to 77 years (median, 48 years). The duration of the acquired plantar fibrokeratoma ranged from several months to 17 years (median, seven years). No patient had prior trauma to the site.



**Figure 3.** Microscopic examination of cluster of differentiation 34 (CD34) stained sections of an acquired giant plantar fibrokeratoma. The view shows positive CD34 staining of the endothelial-lined blood vessels. The fibroblast tumor cells in the dermis do not stain with CD34 x4.

The size of the tumor ranged from 7mm to 4cm (median, 3cm in length). Most of the plantar fibrokeratomas were greater than 1cm in length. Including our patient, plantar fibrokeratomas of greater than 1cm (and therefore considered to be giant) were noted in nine patients.

The reported acquired plantar fibrokeratomas were surgically excised. No recurrence of the tumor was observed.

Kint et al. classified the pathologic features of acquired digital fibrokeratomas into three histological variants: type I, type II, and type III. Type I acquired digital fibrokeratoma, which is the most common, was observed in our patient. Type I acquired digital fibrokeratomas are dome-shaped papules that consist of thick and densely packed

collage bundles, with fibroblasts in between the bundles; fine elastic fibers are also present in the dermis [2].

Type II acquired digital fibrokeratomas have similar pathological features to those of Type I; however, there is an increased number of fibroblasts and number decreased of elastic fibers. Prior investigators have suggested that the type II histologic variant of acquired digital fibrokeratoma has features in common with cellular digital fibroma and superficial acral fibromyxoma. The type III histological variant of acquired digital fibrokeratoma is uncommon; few irregular collagen bundles are present, the edematous dermal stroma is less cellular, and there are no elastic fibers observed [2,20,21].

The clinical differential diagnosis for acquired digital fibrokeratoma may include angiofibroma, cellular digital fibroma, and superficial acral fibromyxoma (**Table 2**), [20-24]. Similar to an acquired digital fibrokeratoma, both angiofibroma and cellular digital fibroma can also occur on the periungual nail fold; in addition, cellular digital fibroma can occur on the side of the digit. In contrast to an acquired digital fibrokeratoma, both angiofibroma and superficial acral fibromyxoma can present as tumors that are either subungual or originate from the periungual nailfold [20-24].

Immunoperoxidase staining may be helpful to differentiate acquired digital fibrokeratoma from cellular digital fibroma and superficial acral fibromyxoma. Cellular digital fibroma and superficial acral fibromyxoma both show diffuse and strong positive CD34 staining. In contrast, acquired digital fibrokeratoma are either negative or only show focally positive staining with CD34. The normal appearing blood vessels in angiofibroma will stain positive with CD34 [20,22,24].

Cluster of differentiation 99 (CD99) stains strongly positive in superficial acral fibromyxoma. Only two cellular digital fibromas have been evaluated with this stain; one was negative and the other only showed focal positivity. We are not aware of any reports evaluating CD99 staining in acquired digital fibrokeratoma or angiofibroma [20,24].

**Table 2**. Differential diagnosis of fibrokeratoma<sup>a</sup>

Condition	Acquired	Clinical presentation <sup>b</sup>	Pathology	CD34 staining	Tx
Acquired digital fibrokeratoma	+	Filiform papule on the side of a finger or toe	Collagen bundles perpendicular to the overlying epidermis	Not studied	SE
Angiofibroma <sup>c</sup>	+	Under the proximal nail fold or subungual	Dilated vessels and collagen fibers	Blood vessels + but collagen fibers -	OS <sup>d</sup>
Cellular digital fibroma	+	Lateral digit or nail fold	Myxoid stroma with less cellularity and increased vascularity	+	SE
Superficial acral fibromyxoma	+	Subungual or nail fold	Short intersecting fascicles of spindle-shaped fibroblasts, dense dermal collagen, subtle vascularity and minimal to no myxoid component	+	SE

Abbreviations: OS, observe site; SE, surgical excision; Tx, treatment; -, absent; +, present.

Recently, a superficial CD34-positive fibroblastic tumor was described that originated from an unusual location, the proximal nail folds of a 63-year-old man's left third finger. This tumor typically occurs on the lower extremities, particularly the thighs. However, the investigators considered this patient's neoplasm to morphologically resemble fibrokeratoma-like Bowen disease [25].

Our patient's fibrokeratoma was not only giant in size but also plantar in location. Only eight other patients with a similar acquired fibrokeratoma, both plantar and giant in size, have been described. Three men had a fibrokeratoma of 4cm in size: a 15-vearold man whose tumor was on the sole of his left foot for three years, a 35-year-old man whose tumor was on his left heel for 1.5 years, and a 54-year-old man whose tumor was on the left plantar region for ten years. Three men had a fibrokeratoma that was 3cm in size: a 48-year-old whose lesion was on the plantar aspect of his great toe for 17 years, a 50-year-old man whose lesion was on his right heel for 12 years, and a 77-year-old whose lesion was on his heel for several years. Additionally, a 43-year-old man had an 11mm lesion on his right heel for a year, and a 51-year-old man had a 15mm lesion on the medial and plantar aspect of his left big toe for 12 years. Similar to the reported patient, none of these patients had a

recurrence of the fibrokeratoma after surgical excision of the fibrokeratoma.

## **Conclusion**

Acquired digital fibrokeratoma usually appears as a small tumor on the lateral finger or toe. However, we encountered a man with a giant acquired fibrokeratoma on his plantar foot. Including our patient, giant acquired fibrokeratomas have been observed in 16 patients; most of these fibrokeratomas (15 of 16 tumors) occurred on the lower extremity. A plantar acquired fibrokeratoma, including our patient, has been observed in 11 individuals: nine of the tumors, including our patient's fibrokeratoma, were greater than 1cm in size and therefore classified as giant. Removal of the tumor during biopsy not only provides treatment of the tumor but also an adequate specimen for microscopic evaluation. Hematoxylin and eosin staining can usually establish the diagnosis. However, negative CD34 staining on the tumor cells typically observed in an acquired digital fibrokeratoma can be helpful to differentiate these benign lesions from other tumors that demonstrate strongly CD34-positive staining, such as cellular digital fibroma and superficial acral fibromyxoma.

<sup>&</sup>lt;sup>a</sup>The most common lesions in the differential diagnosis are summarized in table.

<sup>&</sup>lt;sup>b</sup>The most common and frequent location of the lesion is listed. However, other sites have been observed.

<sup>&</sup>lt;sup>c</sup>Angiofibromas can be observed in patients with tuberous sclerosis; in the setting, they have been referred to as Koenan's tumors. In patients with tuberous sclerosis, these tumors commonly appear between the age of two and five years.

<sup>&</sup>lt;sup>d</sup>Often the tumor is removed during the biopsy; if residual lesion is present, it can be monitored clinically.

## **Potential conflicts of interest**

### Dr. Cohen is a consultant for ParaPRO.

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**Table 1**. Characteristics of patients with acquired fibrokeratoma of the extremity<sup>a</sup>.

С	A S	Site	Size (cm) Duration	Morphology	Tx Rec	Ref
1	13 W	Right heel	0.7 Months	Firm, circular, pedunculated	SE -	[4]
2	18 W	Lateral side of left great toe	2.5×1.6×1.4 2 years	Flesh-colored, solitary, round, protruded nodule	SE -	[5]
3	33 W	Nailfold of right great toe	3.2×3.8x1.5 8 years	Solitary, red, pedunculated nodule	SR -	[6]
4	41 W	Nailbed of right third toe	2.0x2.5x1.0 7 years	Red, hyperkeratotic, hook-shaped	SR -	[7]
5	15 M	Sole of left foot	4x1.5 3 years	Flesh-colored, clustered, hyperkeratotic, verrucous papules	SE -	[8]
6	17 M	Dorsum of right third toe	1.7 0.33 years	Flesh-colored, exophytic round nodule	SE -	[9]
7	35 M	Left heel	4 1.5 years	Flesh-colored, firm, protruded, hyperkeratotic	SE -	[10]
8	35 M	Plantar region of left second toe	0.7x1.8 10 years	Solitary, projecting, keratotic mass, polypoid papule	SE -	[11]
9	40s M	Left great toe	1.5×1.5x1 4 years	Yellow, firm, solitary hyperkeratotic	SE -	[12]
10	43 M	Right heel	1.1x7.0x5.0 1 year	Flesh-colored, fungiform, hyperkeratotic	SE -	[13]
11	48 M	Plantar region of left great toe	3x1.6×1.2 17 years	Firm, elongated mass	SE -	[14]
12	50 M	Right heel	3x2.2x1.0 12 years	Flesh-colored, firm, pedunculated nodule	SE -	[15]
13	51 M	Medial and plantar region of left great toe	1.5x0.5x0.5 7 years	Flesh-colored, pedunculated nodule	SE -	[16]
14	54 M	Left plantar region	4 10 years	Erythematous plaque w/ hyperkeratotic, grooved surface	SE -	[17]
15	62 M	Left fifth toe	1x1x3 5 years	Hyperkeratotic, protruded	SE -	[18]
16	63 M	Sole of left foot	1.5x1.0x0.5 10 years	Flesh-colored, exophytic nodule	SE -	CR
17	77 M	Heel	3.0x1.5 Years	Flesh-colored, pedunculated nodule	SE -	[19]
18	51 M	Lateral thumb	4×2.5× 2.1 15 years	Flesh-colored, solitary, round	SE -	[3]

Abbreviations: A, age (in years); C, case; cm, centimeters; CR, current report; M, man; Rec, recurrence; Ref, reference; S, sex; SE, surgical excision; SR, surgical resection; Tx, treatment; W, woman; -, absent; +, present.

<sup>a</sup>The acquired fibrokeratoma as giant in 15 patients (cases 2-7, 9-17) and it was located on the plantar surface of the foot in 11 patients (cases 1, 5, 7, 8, 10-14, 16, 17) and the upper extremity in one man (case 18).