

A rare triad of Dupuytren's contracture, plantar fibromatosis, and knuckle pads in a young man

Dear Editor,

Fibromatoses represent a group of disorders characterised by fibroblast and myofibroblast activation, including palmar fibromatosis Dupuytren's contracture (DC), Ledderhose disease, penile fibromatosis (Peyronie's disease), and knuckle pads. While these conditions are all classified as fibromatoses, the simultaneous presence of DC, plantar fibromatosis, and knuckle pads is rare, with only a few reports in the literature. Herein, we report this rare triad in a young male.

A 32-year-old man presented with progressive hardening of the palmar and plantar skin. He reported difficulty extending his fingers and toes for the last 6 months. The patient had no addictions or co-morbidities like diabetes, liver disease, HIV, hyperlipidaemia, or epilepsy. His mother had similar complaints on her palms since her forties. There was no history of painful erection or abnormal curvature of the penis on erection.

Examination revealed fixed flexion contracture involving the middle, ring finger, and the adjoining palm of the left hand, as well as the thumb, little finger, and adjoining palm of the right hand with multiple palmar pits and areas of puckering secondary to fibrosis involving both palms [Figure 1a]. Knuckle pads were noted on the dorsum of both hands at the metacarpophalangeal joints and interphalangeal joints [Figure 1b]. Foot examination revealed severe overriding of the toes, flattening of the plantar arch, and puckering of the skin secondary to fibrosis in feet [Figure 1c]. The penile examination was normal. No keloids were noted on cutaneous examination.

Dupuytren's diathesis coexisting with Ledderhose disease and knuckle pads was diagnosed. The patient was treated with dermatofasciectomy with a full-thickness skin graft for palmar lesions. He also underwent left subtalar fusion, calcaneocuboid fusion with screw fixation, and hallux varus corrections for soles under the Departments of Plastic surgery and Orthopedics. At follow-up, the patient was doing well,



Figure 1a: Fixed flexion deformity of the middle and ring fingers of the left hand, and the thumb and little finger of the right hand with profound palmar pitting.



Figure 1b: Knuckle pads on the metacarpophalangeal and interphalangeal joints.



Figure 1c: Severe overriding of the toes, flattening of the plantar arch, and pitting involving feet.

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with improved functional status and range of movements following surgical correction.

Dupuytren's disease is a fibroproliferative disorder affecting the palmar aponeurosis. It usually presents as firm, painless nodules firmly adherent to the skin and fascia, most commonly involving the ring and middle fingers. There may also be pitting and thickening of the palmar skin. It may be associated with concurrent disorders like diabetes, alcoholism, liver disease, HIV infection, hyperlipidaemia, and epilepsy. The mode of genetic transmission in DC is not yet fully understood and appears to be heterogeneous. It is most commonly inherited in an autosomal dominant pattern with variable penetrance, autosomal recessive, and in rare cases, maternal (matrilinear) inheritance, suggesting mitochondrial heredity. Genome-wide studies have identified genetic associations with chromosomes 6, 11, and 16. Among the genomic variations observed in DC, alterations in gene copy number have been identified in chromosomal regions 10q22, 16p12.1, and 17p12. Additionally, an association with the *HLA-DRB1*15* allele has been reported.¹ In cases with matrilinear inheritance, a mutation in the 16S rRNA gene has also been identified.¹ Dupuytren's diathesis is a condition seen in a subset of patients with DC with clinical presentation similar to the present patient, characterised by more severe and rapidly progressive contractures, bilateral involvement, male sex, early age of onset, a positive family history, and

knuckle pads. The condition typically worsens over time, leading to impaired hand function due to a fixed flexion deformity affecting one or more digits. In some cases, slight improvement may occur after many years without treatment.²

Ledderhose disease, which is another type of fibromatosis disorder, is characterised by slow-growing nodules in the central part of the plantar fascia, without flexion deformity of the toes. Knuckle pads/Garrod's pads are well-circumscribed, smooth, and firm plaques over the joints of the hand and feet, manifesting in the second and third decades. It can be primary (in DC) or secondary to trauma. Knuckle pads have the highest association with DC.²

Despite their classification as fibromatoses, there are few cases in the literature reporting fibromatosis presenting with a triad/tetrad in the same individual, as summarised in Table 1.³⁻⁷ In the present patient, the presence of fibromatosis triad, as well as Dupuytren's diathesis, likely indicated a more aggressive disease course with recurrence. Treatment options for DC include segmental fasciectomy, percutaneous needle fasciotomy, radiotherapy, and chemical cleavage by collagenase injections. This highlights the need for awareness among clinicians about conditions classified as fibromatosis, and it reinforces the need for thorough screening of patients presenting with any of the conditions classified as fibromatosis to rule out coexisting fibromatoses.

Table 1: Summary of the reported cases in literature with two or more conditions classified as fibromatoses in the same individual

Age and sex	Onset	Family history	Spectrum and Associations	Treatment
32/M (present case)	6 months	Yes	DC, knuckle pads, and plantar fibromatosis.	Dermato-fasciectomy with a full-thickness skin graft for palmar lesions. Left subtalar fusion, calcaneocuboid fusion with screw fixation and hallux varus corrections for soles.
18/F	12 years	No	DC and plantar fibromatosis. Nail dystrophy. ³	Intralesional triamcinolone.
49/M	Long period	No	DC, plantar fibromatosis, and knuckle pad. Alcohol abuse. ⁴	Surgical exploration of nodules in his hands and left foot sole.
34/M	10 years	Not available	DC, knuckle pads, Peyronie's disease and plantar fibromatosis ⁵	Total aponeurotomy and multiple Z-plasties using the Skoog approach
24/F	17 years	Present	DC, plantar fibromatosis. ⁶	Partial dermo-fasciectomy with full-thickness skin graft from the volar aspect of the left elbow.
42/M	6 months	No	DC (Dupuytren's diathesis), plantar fibromatosis, and Peyronie's disease. Alcohol abuse. ⁷	Partial fasciectomy with intra-operative correction.

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References

1. Michou L titia, Lermusiaux J-L, Teyssedou J-P, Bardin T, Beaudreuil J, Petit-Teixeira E. Genetics of dupuytren's disease. *Joint Bone Spine* 2012;79:7-12.
2. Boe C, Blazar P, Iannuzzi N. Dupuytren contractures: An update of recent literature. *J Hand Surg Am* 2021;46:896-900.
3. Kathuria S, Khanna N. Palmoplantar fibromatosis with nail dystrophy. *Indian J Dermatol Venereol Leprol* 2012;78:646-8.
4. Gafurođlu T , Taşdelen  Y, Eser F, D zg n S, Kadan E, Duran S, *et al.* Palmar-plantar fibromatosis and knuckle pads associated with alcohol consumption. *J Phys Med & Rehabil Sci* 2016;19:50-4.
5. Gonz lez-Mart nez R, Mar n-Bertol n S, Amorrortu-Velayos J. Association of knuckle pads and palmo-plantar and penile contracture as clinical manifestations of polyfibromatosis. *E J Plastic Surg* 1998;21: 101-2.
6. Couto-Gonzalez I, Brea-Garcia B, Taboada-Su rez A, Gonz lez- lvarez E. Aggressive dupuytren's diathesis in a young woman. *BMJ Case Rep* 2010;2010:bcr1220092592.
7. Mbuva R, Kigera J, Maru M, Mogire T, Oburu E. Dupuytren's diathesis in an African male. *Ann Afr Surg* 2016;13:84-5.