Ind J Dermatol Venereol Leprol 1993; 59 : 302-3

HEREDITARY CAMPTODACTYLY ASSOCIATED WITH PALMOPLANTAR HYPERHIDROSIS

Sandipan Dhar, Amrinder J Kanwar, Surrinder Kaur

Hereditary camptodactyly can be asociated with several other developmental anomalies. A case of harediterary camptodactyly in a 14-year-old girl with asociated palmoplantar hyperhidrosis is reported.

Key Words : Hereditary camptodactyly, Hyperhidrosis

Introduction

Hereditary camptodactyly is a rare type of fibromatosis with autosomal dominant inheritance affecting both sexes equally. It begins in childhood involving little fingers (bilateral) producing persistent flexion of proximal interphalangeal joints with sparing of metacarpo-phalangeal joints.[1] Rarely other fingers are also affected.[2] The resulting deformity closely mimics clawhand due to leprosy with which it is often confused with.[3,4] It has been reported to be associated with several other developmental abnormalities. We here report a case of camptodactyly associated with palmoplantar hyperhidrosis.

Case Report

A 14-year-old girl presented with excessive sweating of palms and soles of 3 years duration. It was continuous in nature and used to become worse in summer. There was no history of aggravation of sweating by emotion, anxiety and mental tension. No history of excessive sweating over any other part of the body including axillae was obtained. No other family member had similar problem.

On examination, both her palms and soles were cold and wet with sweating. Starch-iodine test demonstrated individual sweat droplets. There was no acrocyanosis, Raynaud's phenomenon or aglodystrophy. Additional feature on examination was fixed flexion deformity of proximal interphlangeal joint of little finger of both hands (Fig. 1) which

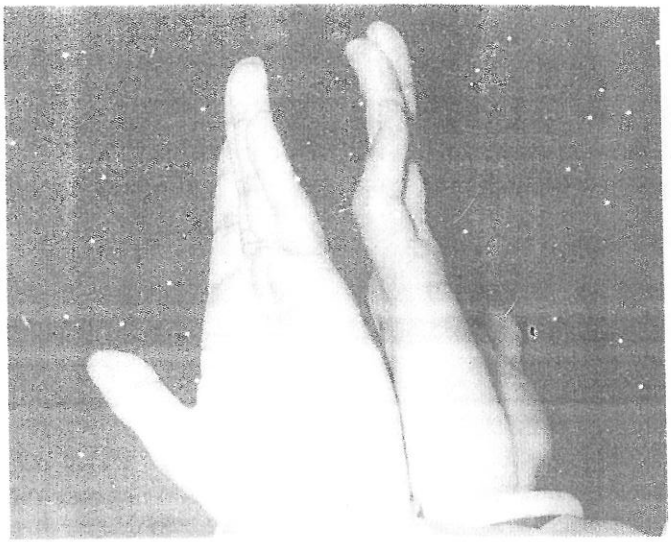

Fig.1. Bilateral fixed flexion deromity of proximal interphalangeal joint

according to her was present since birth. Her mother also had similar deformity since birth. All modalities of sensation were preserved. There was no wasting or weakness of small muscles of hands, no thickening of ulnar, median or radial cutaneous nerves or palmar fascia. Systemic examinations and relevant

From the Department of Dermatology, Venereology and Leprology, Postagraduate Institute of Medical Education and Research, Chandigarh-160 012, India.

Address correspondencde to : Dr A J Kanwar

laboratory investigations were within normal limits.

The patient was diagnosed to have camptodactyly with associated palmo-plantar hyperhidrosis. Hyperhidrosis was treated with daily application of 20% aluminium chloride in absolute alcohol. The nature of deformity was explained to her and her father and they were reassured. When last seen, there was 60% improvement in sweating after 6 weeks application.

Discussion

Several developmental and systemic abnormalities have been reported in association with camptodactyly e.g., scoliosis, pectus excavatum, high arched palate, Dupuytren's contracture, Marfan's syndrome, trisomy-13, occulodento-digital, oro-faciodigital and cerebro-hepatovenal syndromes.[1,5] Camptodactyly has also been reported to be associated with knuckle pad[6] and taurinuria.[7] However, to the best of our knowledge, hyperhidrosis of palms and soles has not been reported so far as an association of camptodactyly. Since palmoplantar hyperhindrosis is thought to be hereditary in origin,[7] there could be some genetic interrelation between these two entities.

References

1. Burton JL. Disorders of connective tissue. In Textbook of Dermatology (Champion RH, Burton JL, Fbling FJG, eds), 5th edn. Oxford : Blackwell Scientific Publications, 1992 ; 1803.
2. Welch JP, Temtany SA. Hereditary camptodactyly. J Med Genet 1966 ; 3 : 104-7.
3. Pavithran K. Camptodactyly simulating clawhand in a patient with indeterminate leprosy. Ind J Leprol 1991 ; 63 : 232-4.
4. Singh G, Kaur V. Hereditary camptodactyly masquerading leprosy. Ind J Dermatol Venereol Leprol 1993 ; 59 :103-4.
5. Jobe MT, Wright II PE. Congenital anomalies of hand. In: Campbell's operative orthopaedics (Crenshaw AH, ed), 8th edn. Missouri : Mosby Year Book, 1992 ; 3416.
6. Pavithran K. Camptodactyly with knuckle-pad. Ind J Dermatol Venereol Leprol 1986 ; 52 : 222-3.
7. Nevin NC, Hurwitz LJ, Neill,DW. Familial camptodactyly with taurinuria. J Med Genet 1966 ; 3 : 265-8.
8. Herxheimer A. Excessive sweating : a review. Trans St John's Hosp Dermatol Soc 1958 ; 40 : 20-5.