Dupuytren's Contracture in the Negro

U. MENNEN

J Hand Surg [Br] 1986 11: 61
DOI: 10.1016/0266-7681(86)90015-X

Dupuytren's Contracture in the Negro

U. MENNEN
From the University of Pretoria.

In 1979 the first Negro patient with Dupuytren's Contracture who had no evidence of Caucasoid Admixture in his genetic make-up was reported on. Another five such cases are presented.

For many years doubt has existed whether Dupuytren's Disease in fact did occur in the pure Negro (Hueston, 1974; Kanavel, 1929; Moorehead, 1956). A number of cases with Dupuytren's Disease in the black man have been reported in the literature (Furnas, 1971; Haeseker, 1981; Plasse, 1979; Zaworski, 1979) but none of these have scientific conclusive evidence of being from pure negro stock.

In 1979 a negro patient who had no evidence of Caucasoid admixture in his genetic make-up was described with bilateral palmar and plantar fasciitis (Mennen, 1979).

Since then five more patients with this disease and without any genetic evidence of Caucasoid admixture have been treated by our unit.

Material and Methods

During a survey of all eighty-seven negro epileptic patients at a Psychiatric Unit in Pretoria, the second case (O.M.) who has palmar fibromatosis on his right hand little finger was discovered. This patient is a twenty-two-year-old epileptic male patient, who has taken anti-epileptic drugs for four years (Figure 1).

At the Kalafong Hospital Hand Unit in Pretoria only four otherwise normal patients with Dupuytren's Disease (A.M., C.P., A.M. and J.P.) were seen in the last four years out of a total of some 6,000 negro hand patients (Figures 2-5). This is an extremely low incidence as compared to a much higher occurrence rate of this disease amongst white patients ranging from 1 to 13 per cent in the literature.

Table 1 summarises the vital statistics regarding the sex, age, work and other aspects of all six patients.

All six cases have been thoroughly genetically investigated and the data are given in Table 2.

Results

Genotyping revealed no admixture with European (that is white or Caucasoid) stock. The presence of certain alleles in the Duffy, second phosphoglucomutase locus (PGM_2) and the transferin systems are of particular significance in Negroes.

Received for publication June, 1985.
Ulrich Mennen, F.R.C.S., FCS (SA) Orth., M.Med(Orth), M.D.(Orth), Dept. of Hand Surgery, P.O. Box 186, Medunsa, 0204 Pretoria, South Africa.

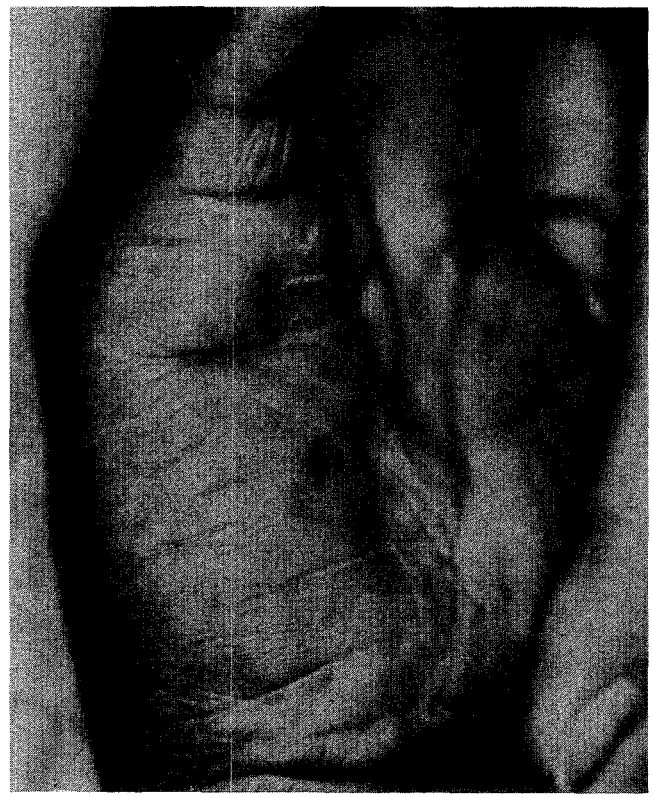

Fig. 1 Palmar aspect of the right hand of patient O.M.

Although the *Fy* allele in the Duffy system, the *cDe* (Ro) allele in the Rhesus system and the PGM_2^2 allele in the phosphoglucomutase (second locus) system may occur in Caucasoid populations their presence in high frequencies in Negroid people is incontestable (Mourant, 1976; Tills, 1983).

The fact that none of the 'classical' Caucasoid genes (P^c in the acid phosphatase system, AK^2 in the adenylate system, ADA^2 in the adenosine deaminase system, EsD^2 in the esterase D system, Tf^B in the transferrin system, *CDe* and *cDE* in the Rhesus system and *K* in the Kell system) occur in any of the six patients with Dupuytrens contracture may be taken as evidence in favour of the claim that there is no Caucasoid admixture in their backgrounds.

Editor's Note: None of the 'classical' Caucasoid genes have a frequency over 10% in Caucasoid populations except CDe (41%) and cDE (14%). Evidence from genotype analysis is therefore not absolute proof of the absence of Caucasoid ancestry.

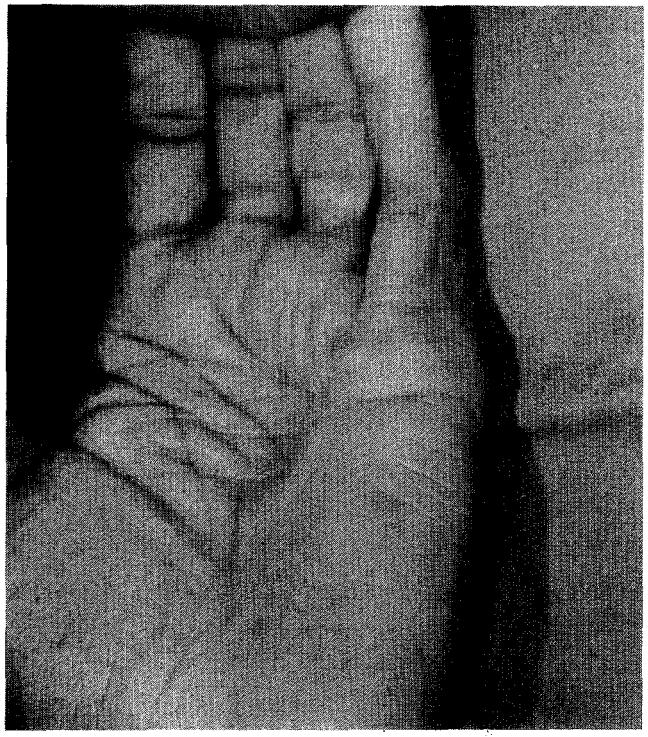

Fig. 2 Palmar aspect of the right hand of patient A.M.

Discussion

Abnormalities in the many different kinds of collagen is the reason for a variety of recognised muskuloskeletal and skin diseases, for example Dupuytren's Disease, scoliosis, club-feet, keloids, osteogenesis imperfecta, non-union of fractures (Weiss, 1982).

The cause for abnormal collagen production may lie in many aetiological factors: for example the Chikungunya virus causes a type of Rheumatoid arthritis (Brighton, 1981) or, unfavourable conditions at a fracture site such as a low oxygen tension and undue movement results in the production of fibrous collagen rather than osteoid collagen which is mineralizable (Weiss, 1982).

The aetiological agent which causes abnormal collagen production in the palmar fascia in Dupuytren's Disease is still unknown. Many influencing factors have in the past been blamed, for example chronic alcoholism, trauma, cardiovascular disease. Since a direct correlation with these diseases and palmar fasciitis has escaped numerous studies the exact role of these factors still remains very uncertain (Hueston, 1974).

Epilepsy, or, more specifically, the use of antiepileptic drugs, notably barbiturates and phenytoin, over a prolonged period of time seems to have a definitive influence in the production of abnormal collagen in the palmar fascia, the plantar fascia and even other areas such as the penis (Viljanto, 1973).

However, it is still an open question as to why Dupuytren's Disease is so very uncommon amongst the Negroes. An answer might lie in a different quality or type of collagen of their deep fascia. This difference in collagen character might explain the well known fact that keloid formation of skin lesions amongst negroes is more common and more severe (Saver, 1980). Another difference in collagen character between Negroes and Caucasoids is the very low incidence of discogenic pathology in the former group (Unpublished data from University of Pretoria).

In order to be able to understand Dupuytren's Disease (and many other collagen-related diseases) we will have to study all the different aspects of collagenous connective tissue: for example origin, production, metabolism, physical appearance, properties, functions,

TABLE 1
Patient information is summarized for clarity.

Patient:	*N.B.*	*O.M.*	*J.P.*	*A.M.*	*C.P.*	*A.M.*
Date first seen	1977	1982	1982	1981	1984	1984
Sex	Male	Male	Male	Male	Male	Male
Age	26 years	22 years	73 years	56 years	66 years	42 years
Nation (i.e. Negro tribe)	Pedi	Tswana	Tswana	Swazi	Pedi	Pedi
Work	Labourer	Labourer	Shoemaker	Watchman	Teacher	Labourer
Dominance	(R) hand	(R) hand	(R) hand	(R) hand	(R) hand	(R) hand
Affected hand	(L) + (R)	(R) little finger	(L) + (R)	(R) little finger	(L)	(L) + (R)
Duration of Dupuytren	Uncertain years	Uncertain years	± 25 years	± 2 years	1 year	Uncertain years
Other affected areas	Both feet	None	None	Knuckle pad	None	None
Family history	None	Uncertain	None	Uncertain	None	None
Other diseases	Epilepsy	Epilepsy	No abnormalities detected	No abnormalities detected	No abnormalities detected	No abnormalities detected
Alcohol consumption	None	None	Moderate	Minimal	Minimal	Minimal
Drugs:						
primidone ("Mysoline")	× on treatment for 5 years		None	None	None	None
phenytoin sodium						
phenobarbital ("Garion")	×	× on treatment				
thioridazine HCL ("Melleril")	×	× for 4 years				
diazepan ("Valium")		×				
carbamezepine ("Tegretol")		×				

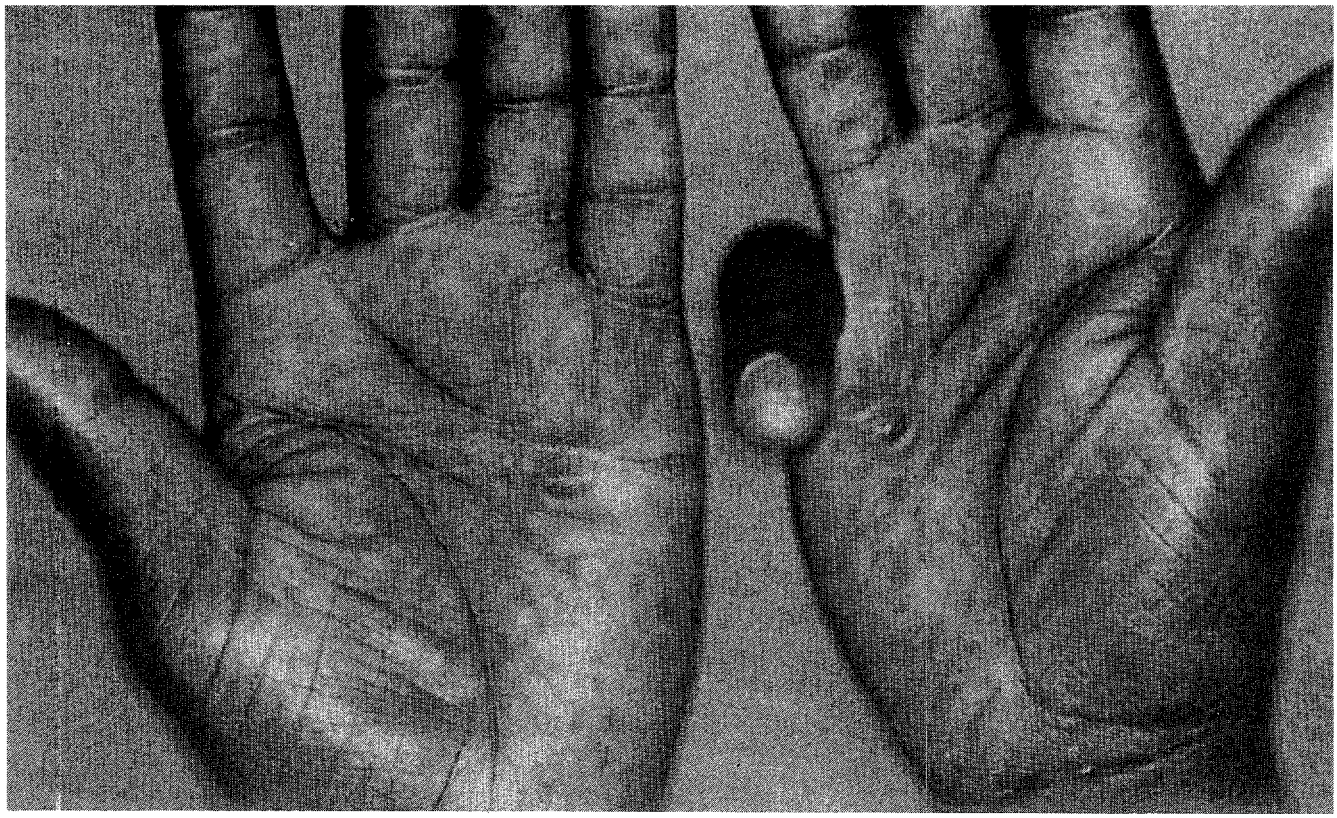

Fig. 3 Palmar aspect of the right and left hand of patient J.P.

TABLE 2
Each patient underwent a battery of genotyping.

Genotyping	*N.B.*	*O.M.*	*J.P.*	*A.M.*	*C.P.*	*A.M.*
BLOOD GROUPS:						
ABO	O	A1	O	A	A_2B	A_2
Rhesus	(R1/Ro)	Ro(cDe/cDe)	r/r^1(cde/Cde)	pos(Ro/Ro)	(cDe)	(cDe)
MNSs		MS/Ns or Ms/NS	MS/Ms		MNs	NSs
Henshaw	neg			neg	neg	neg
P1	pos	pos	pos		pos	pos
Kell	neg	neg	neg	neg	kk	kk
Duffy	Fy(a − b −)	Fy(a b −)	Fy(a − b −)	Fy(a − b +)	Fy(a − b −)	Fy(a − b −)
Kidd					JK(a + b −)	JK(a + b −)
HAEMOGLOBIN TYPE:	$A+A_2$	$A+A_2$	$A+A_2$	$A+A_2$	$A+A_2$	$A+A_2$
RED CELL ENZYMES:						
Glucose 6 Phosphatedehydrogenase	B	B	B	B	B	B
6 Phosphogluconatedehydrogenase	A	A	A	AC	A	A
Acid Phosphatase	B	B	RB	B	B	A
Peptidase A	1-1	1	1	2-1	1	1
Peptidase B	1-1	1	1	1		
Glutamate pyruvate transaminase	1-1	1	1	1	1	1
Glyoxalase	2-2	2-1	2-1	2	2	2-1
Carbonic Anhydrase I	1-1	1	1	1	1	1
Carbonic Anhydrase II	1-1	1	1	1	2-1	2-1
Adenylate Kinase	1-1	1	1	1	1	1
Adenosine Deaminase	1-1	1	1	1	1	1
Esterase D	1-1	1	1	1	1	1
Phosphoglucomutase (1st Locus)	2-1	1 +	1 +	1 +	1 +	1 + 2 +
Phosphoglucomutase (2nd Locus)	2-1	1	1		1	1
SERUM PROTEINS:						
Haptoglobin	2-1	2-1 Mod	1	2	2-1	2-1
Transferrin	CD	C	C	C	C	C
Group Specific Component		1^F	$1^F - 1^S$	1^F	1^F	1^F
Properdin Factor B					SF	SF

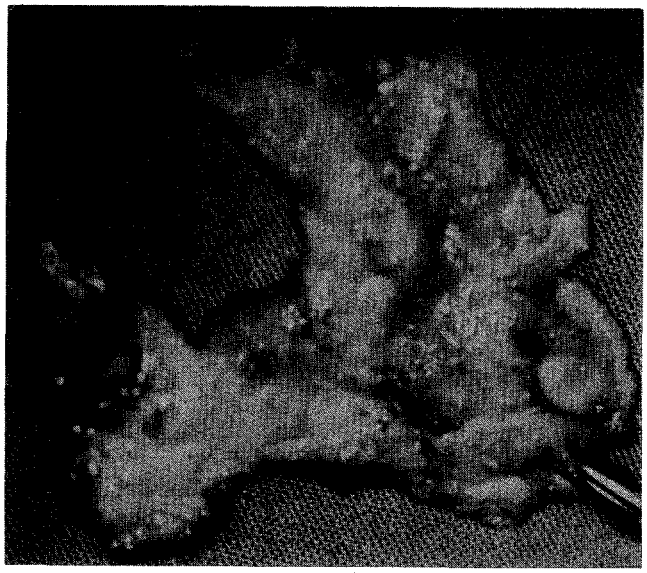

Fig. 4 Resected material from right palm of patient J.P. in Figure 3.

influence on the immuno-system, etc. This is being done more and more as reflected by recent publications (Gelberman, 1980; Hoopes, 1977; Menzel, 1979; Prockop, 1982; Weiss, 1982).

If we understand collagen in this way we will probably be able to manipulate or even better, prevent collagen abnormalities.

Conclusion

In conclusion, another five cases of Dupuytren's Disease in Negro patients have been added to the first ever genetically examined case. All six cases have, by genotyping, been established to be without any evidence of Caucasoid admixture.

The aim of this paper is not only to establish the fact that Dupuytren's Disease does in fact occur in the epileptic and non-epileptic healthy pure Negro, but, also to encourage an intensified focus on the uncovering of the different *characteristics* and behaviour of collagen in general.

Acknowledgement

My sincerest appreciation is expressed to Professor Trefor Jenkins MD (Lond), Professor and Head, Department of Human Genetics, South African Institute for Medical Research and University of the Witwatersrand, Johannesburg, South Africa, who was instrumental in doing and interpreting the genetic studies.

References

BRIGHTON, S. W. (1981). Chikungunya virus infection. (Letter) South African Medical Journal, 59 (16): 552.

FURNAS, D. W. (1979). Dupuytren's Contractures in a Black Patient in East Africa. Plastic and Reconstructive Surgery, 64 (2): 250-251.

GELBERMAN, R. H., AMIEL, D., RUDOLPH, R. M. and VANCE, R. M. (1980). Dupuytren's Contracture: An Electron Microscopic, Biochemical and Clinical Correlative Study. The Journal of Bone and Joint Surgery, 62A(3): 425-432.

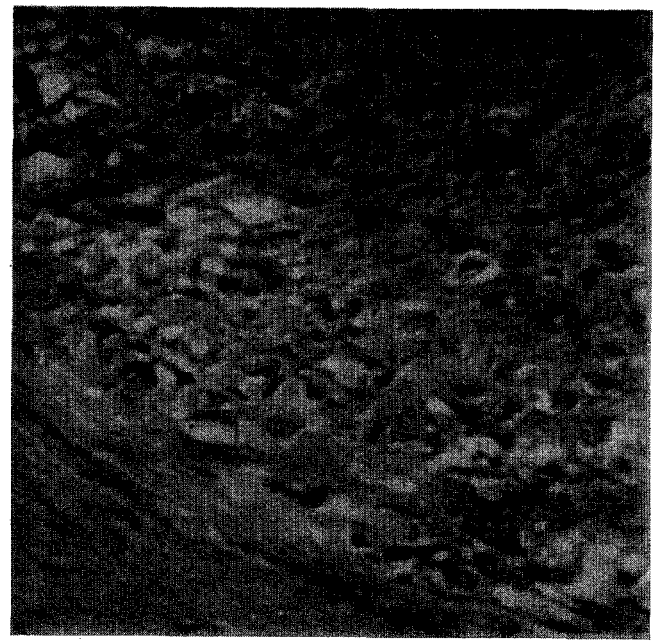

Fig. 5 Histology of material resected from patient in Figure 3 (J.P.) shows fibroplasia and varying degrees of cellularity of the lesion.

HAESEKER, B. (1981). Dupuytren's disease and the sickle-cell trait in a female black patient. British Journal of Plastic Surgery, 34(4): 438-440.

HOOPES, J. E., JABALEY, M. E., Su C-T, WILGIS, E. F. S. and IM, M. J. C. Enzymes of glucose metabolism in palmar fascia and Dupuytren's contracture. The Journal of Hand Surgery, 2(1): 62-65.

HUESTON, J. T. *Dupuytren's Disease*. Eds Hueston, J. T. and Tubiana, R. Edinburgh, Churchill Livingstone, 1974. p29.

KANAVEL, A., KOCH, S. and MASON, M. (1929). Dupuytren's Contracture. With a Description of the Palmar Fascia. A Review of the Literature. And a Report of Twenty-Nine Surgically Treated Cases. Surgery, Gynecology and Obstetrics, 48: 145-190.

MENNEN, U. and GRÄBE, R. P. (1979). Dupuytren's contracture in a negro: A case report. The Journal of Hand Surgery, 4(5): 451-453.

MENZEL, E. J., PIZA, H., ZIELINSKI, C., ENDLER, A. T., STEFFEN, C. and MILLESI, H. (1979). Collagen Types and Anticollagen-Antibodies in Dupuytren's Disease. The Hand, 11(3): 243-248.

MOOREHEAD, G. G. (1956). Dupuytren's contracture: Review of the disputed etiology. New York Journal of Medicine, 56: 3686-3703.

MOURANT, A. E., KOPEĆ, A. C., DOMANIEWSKA-SOBCZAK, K. *The distribution of the Human Blood Groups and other Polymorphisms*. London. Oxford University Press, 1976. 62-69.

PLASSE, J. S. (1979). Dupuytren's Contractures in a Black Patient. Plastic and Reconstructive Surgery, 64(2): 250.

PROCKOP, D. J. *Symposium on heritable disorders of connective tissue*. Akeson, W. H., BORNSTEIN, P., GLIMICHER, M. J. (eds). CV Mosby Company, 1982. p16.

SAVER, G. C. *Manual of Skin Diseases*. J B Lippincott Co. 1980. p281.

TILLS, D., KOPEĆ, A. C., TILLS, R. E. *The Distribution of the Human Blood Groups and other polymorphisms*. Supplement I 1983. Oxford University Press, London.

VILJANTO, J. A. (1973). Dupuytren's contracture: A review. Seminars in Arthritis and Rheumatism. 3: 155-176.

WEISS, J. B. and AYAD, S. An introduction to collagen. In Weiss, J. B., Jayson, M. I. V. (eds). *Collagen in Health and Disease*, Churchill Livingstone, 1982. p1-17.

WEISS, J. B., JAYSON, M. I. V. (eds). *Collagen in Health and Disease*. Churchill Livingstone, Edinburgh 1982. Chap 16, p289, Vitto, J. and Baver, E. A. Chap 19 p362, Sakamoto, S., Chap 28 p472, Baver, E. A. and Vitto, J. Chap 30 p498, Davison, P. F.

ZAWORSKI, R. E. and MANN, R. J. (1979). Dupuytren's Contractures in a Black Patient. Plastic and Reconstructive Surgery, 63(1): 122-124.

Unpublished statistical data from the Department of Orthopaedic Surgery. H. F. Verwoerd and Kalafong Hospitals, University of Pretoria, Pretoria, Republic of South Africa.