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SYMPHALANGISM ASSOCIATED WITH SYNOSTOSIS OF CARPUS AND/OR TARSUS*

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Summary Five Japanese families of symphalangism associated with synostosis of the carpal bones and/or tarsal bones were presented. In four families, the anomaly was also found in other individuals of the family showing autosomal dominant inheritance. Historical aspect, skeletal changes and genetics of symphalangism were discussed.

INTRODUCTION

Symphalangism is an hereditary anomaly manifested by partial or total absence of one or more interphalangeal joints of bilateral hands, with fusion of the involved phalanges. The term "symphalangism" was first coined by Cushing in 1916. He demonstrated the autosomal dominant characteristics of the trait through 7 generations. According to his study, the proband, William B, had migrated to the United States from Scotland in 1700 and settled in Virginia. Among 302 individuals comprised in 72 completed families in four succeeding generations of which records were complete, 78 (25.8%) were affected by symphalangism. Among these 72 families, 44 were from the mating of unaffected parents with 152 children, all of course unaffected. On the other hand, in 28 families with one affected parent there were 150 children, and 78 (52%) of them were found to be affected by symphalangism. In 1917, Drinkwater reported a pedigree of this anomaly under the title of "phalangeal anarthrosis (synostosis, ankylosis) transmitted through fourteen generations." In his report, the anomaly has been traced back fourteen generations to John Talbot, first Earl of Schrewsbery, who died in the battle at Chastillon, near Bordeaux, in 1453. Therefore, the anomaly would appear to have been known from the early 20th century. Reports of this anomaly, however, are relatively meager in literature.

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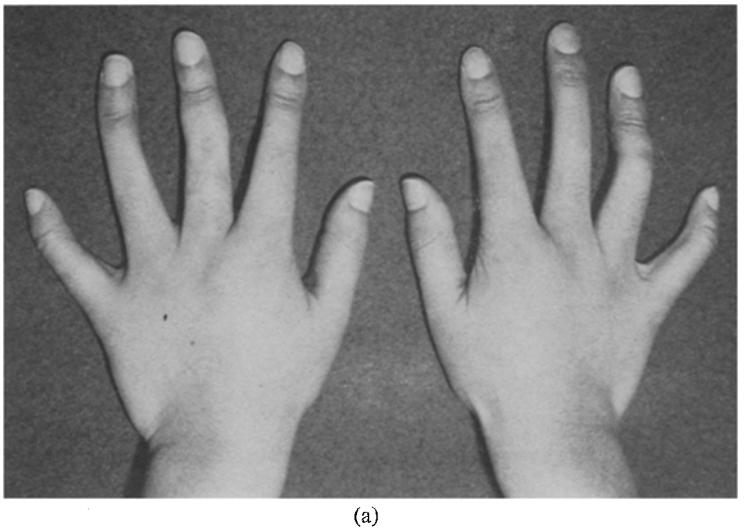
The purpose of this paper is to present five Japanese families of symphalangism associated with synostosis of the carpal and/or tarsal bones.

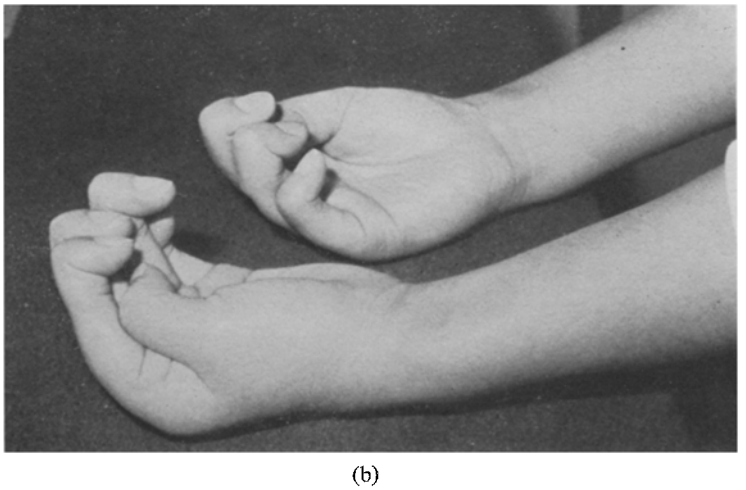
CLINICAL AND ROENTGENOLOGIC FINDINGS

Clinical findings

The history and the clinical findings of the fully developed anomaly were char-



(a)



(b)

Fig. 1a. External appearance of the hands. Transverse cutaneous wrinkles at the proximal interphalangeal joints of the index through little fingers are not seen at all.
Fig. 1b. External appearance of the hands in flexion position. See supernormal flexibility of the distal interphalangeal joints.

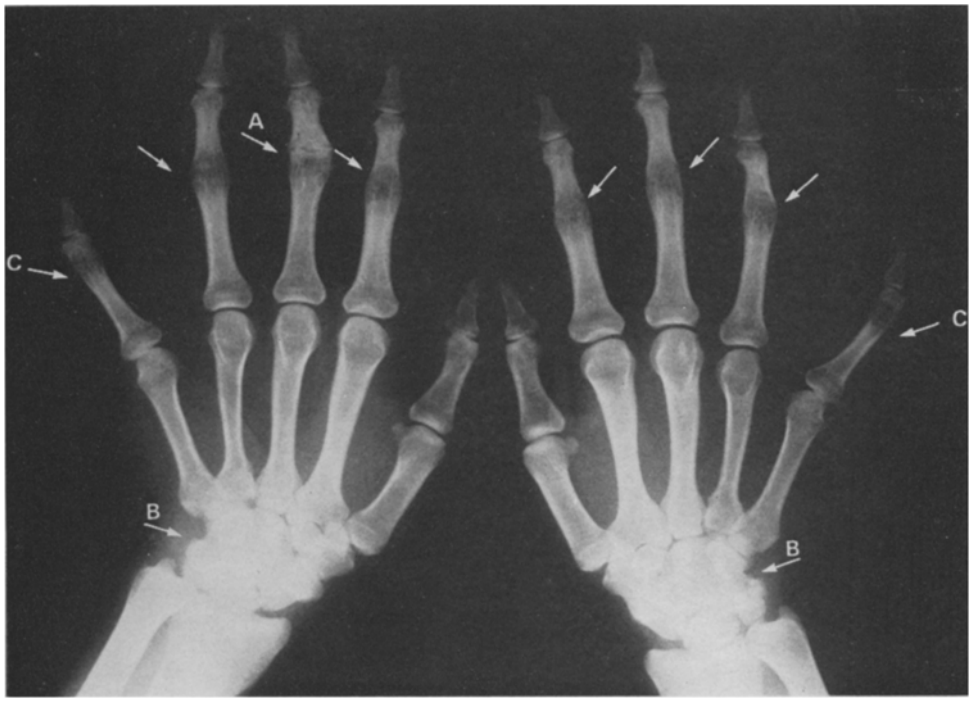


Fig. 2. Dorso-palmar reontegenogram of the hands. ↘ synostosis between basal and middle phalanges. A↘ synchondrosis between basal and middle phalanges. B↘ synostosi samong the os hamatum, os capitatum and os triquetrum. C↘ brachymesophalangy of the little finger.

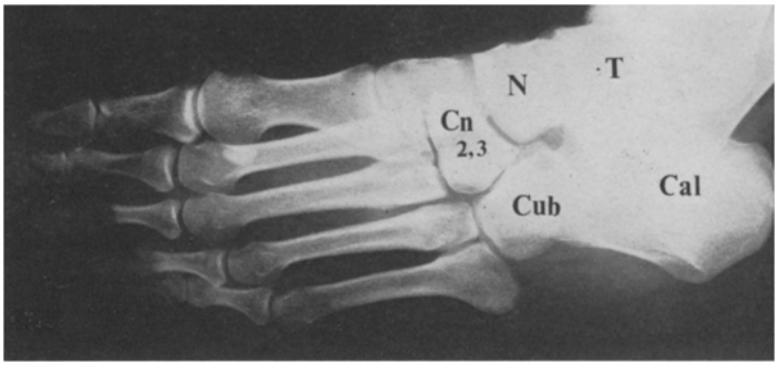


Fig. 3. Oblique roentgenogram of the foot. T, os talus; N, os naviculare pedis; Cal, os calcis; Cub, os cuboideum; Cn 2, 3, os cuneiforme intermedium and os cuneiforme latarale. Synostosis among the os talus, os naviculare pedis, os calcis and os cuboideum, that between the os cuneiforme intermedium and laterale, and that between the middle and terminal phalanges of the 4th and 5th toes are observed.

acteristic. The anomaly appeared bilaterally and almost symmetrically, and was already apparent at birth by the presence of stiff fingers.

In the affected fingers, each proximal interphalangeal (PIP) joint showed ankylosis in extended position. The skin covering the proximal two thirds of the involved fingers was very smooth and neither transverse cutaneous wrinkles nor volar cutaneous creases were seen at the affected PIP joint areas (Fig. 1a). Although the patients were incapable of closing the hand into a fist, they were least handicapped by their "stright fingers" owing to compensative supernormal flexibilities of the metacarpophalangeal (IP) and distal interphalangeal (DIP) joints (Fig. 1b).

Roentgenologic findings

Both in juvenile and adult patients the basal and middle phalanges in each affected finger were ordinarily completely fused at the PIP joint, resulting in a shape of long monophalangy with trabecular continuity of spongy bone. In some PIP joints, however, the basal and middle phalanges were separated by a very narrow joint space indicating synchondrosis of these PIP joints. Brachymesophalangy of the little finger was observed in all cases. In each wrist region, synostosis between

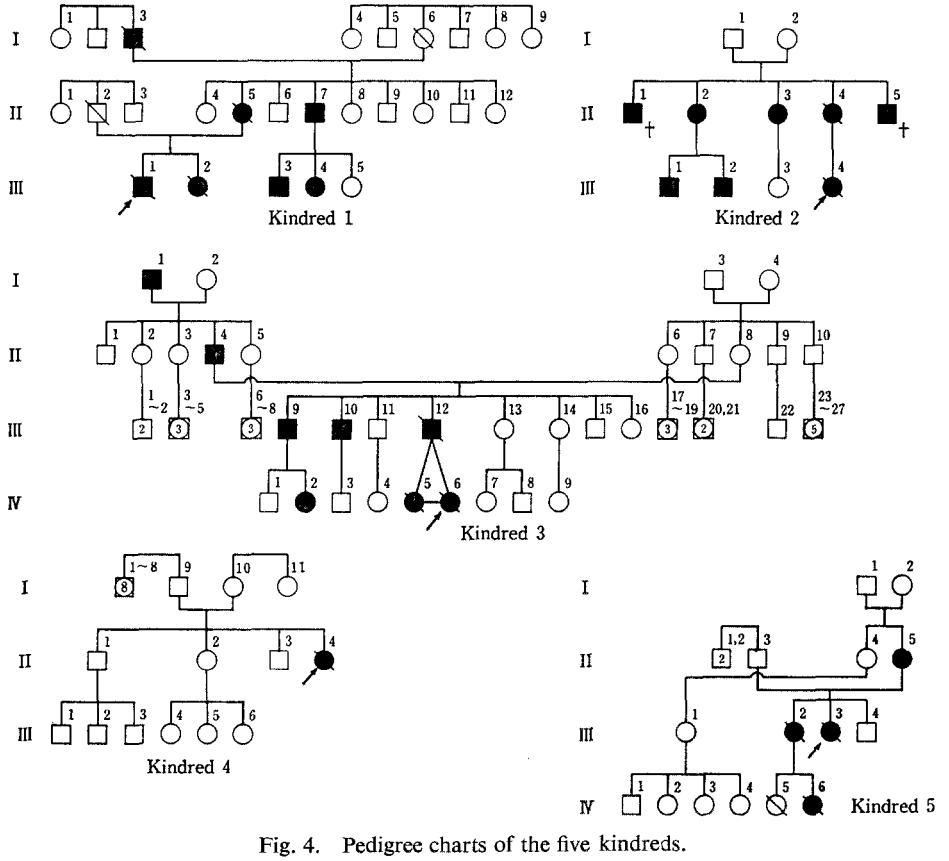


Fig. 4. Pedigree charts of the five kindreds.

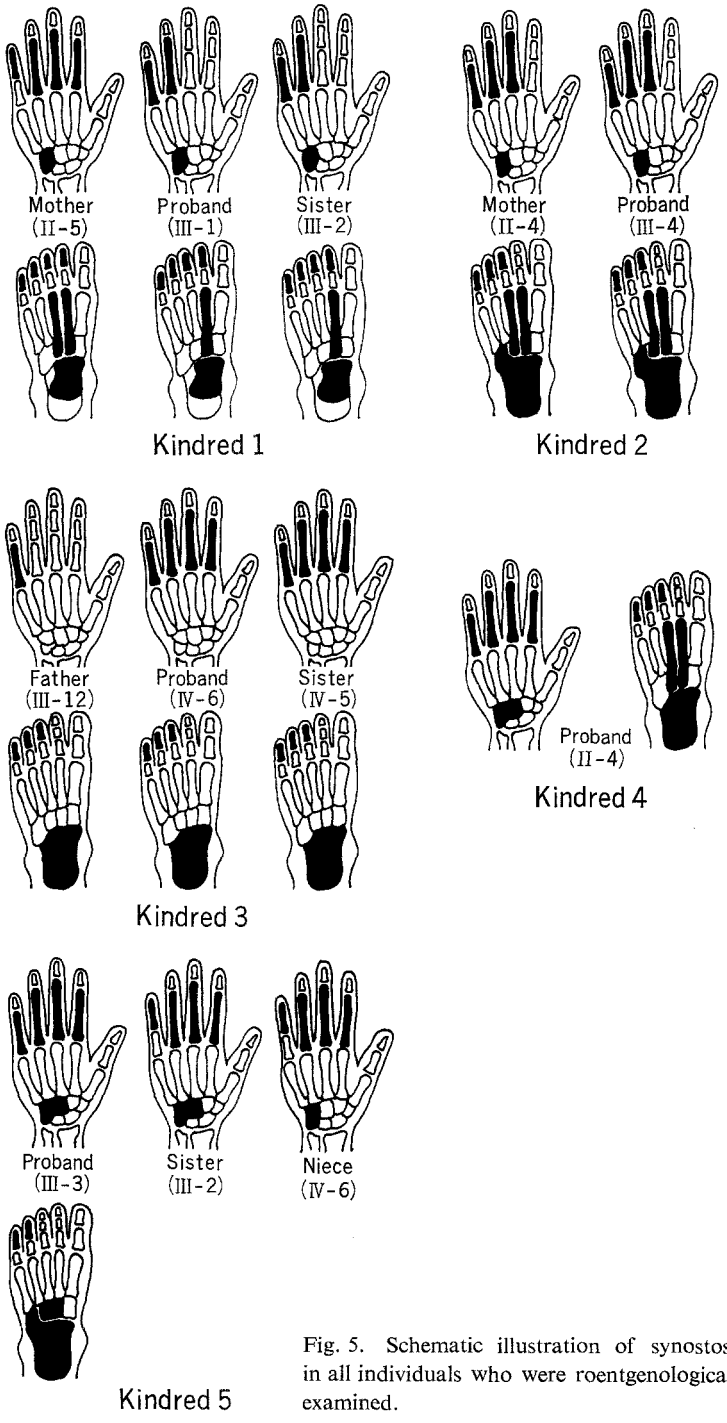


Fig. 5. Schematic illustration of synostoses in all individuals who were roentgenologically examined.

two or among three adjoining carpal bones was observed. In the case shown in Fig. 2, synostosis was clearly observed among the os hamatum, os capitatum and os triquetrum. In each foot, synostosis between two or among three or more adjoining tarsal bones was also observed in all cases. In the case shown in Fig. 3, synostosis among the os talus, os naviculare, os calcis and os cuboideum, and that between the os cuneiforme intermedium and os cuneiforme laterale were observed. Synostosis between each couple of the middle and terminal phalanges of the toes was also observed in all cases.

Developmental process of synostosis at the PIP joint and that at the carpal and tarsal regions during the growing stage of children will be described later in Kindred 1.

RESULTS OF FAMILY STUDY AND THE STATE OF BONE FUSION IN AFFECTED INDIVIDUALS

Family study was performed in all five cases. Pedigree charts obtained by the investigation are shown in Fig. 4, and the state of bone fusion in all of the affected individuals who were roentgenologically examined is schematically illustrated in Fig. 5.

Kindred 1

Proband: A 10-month-old boy (III-I)

The PIP joints of bilateral ring and little fingers were stiff in extended position. Transverse cutaneous wrinkles and volar cutaneous creases over the PIP joints of the affected fingers were not observed. Roentgenologically, narrowing of the PIP joint spaces of the affected fingers was observed as a sign of synchondrosis of the joints. Marked brachymesophalangy of the little finger was also observed (Fig. 6a).

Roentgenologic follow-up study

Wrist and hand: At age 3, the tops of the basal phalanges of the ring and little fingers were fusing with the respective epiphyseal bases of the corresponding middle phalanges, indicating the development of synostosis at their PIP joints. In each wrist region, the proximal margin of the os hamatum showed a small sharp protrusion, indicating the initiation of synostosis to the os triquetrum (Fig. 6b). At age 5, synostosis between each couple of the basal and middle phalanges of the affected fingers became more clear. In each wrist region, synostosis between the os hamatum and os triquetrum was clearly observed (Fig. 6c). At age 13, premature closure of the epiphyseal plates of the middle phalanges of the affected fingers was observed. Ossification of each carpal bone progressed showing almost the same configuration as that in adult (Fig. 6d). At age 20, the narrow PIP joint space was still observable in the ring finger, while the basal and middle phalanges of the little finger were completely fused (Fig. 6e).

Foot: At age 10 months, the base of the 2nd metatarsus was slightly elongated proximally (Fig. 7a). At age 3, the base of the 2nd metatarsus was markedly elongated and showed synchondrosis with the newly ossified ossification center of the

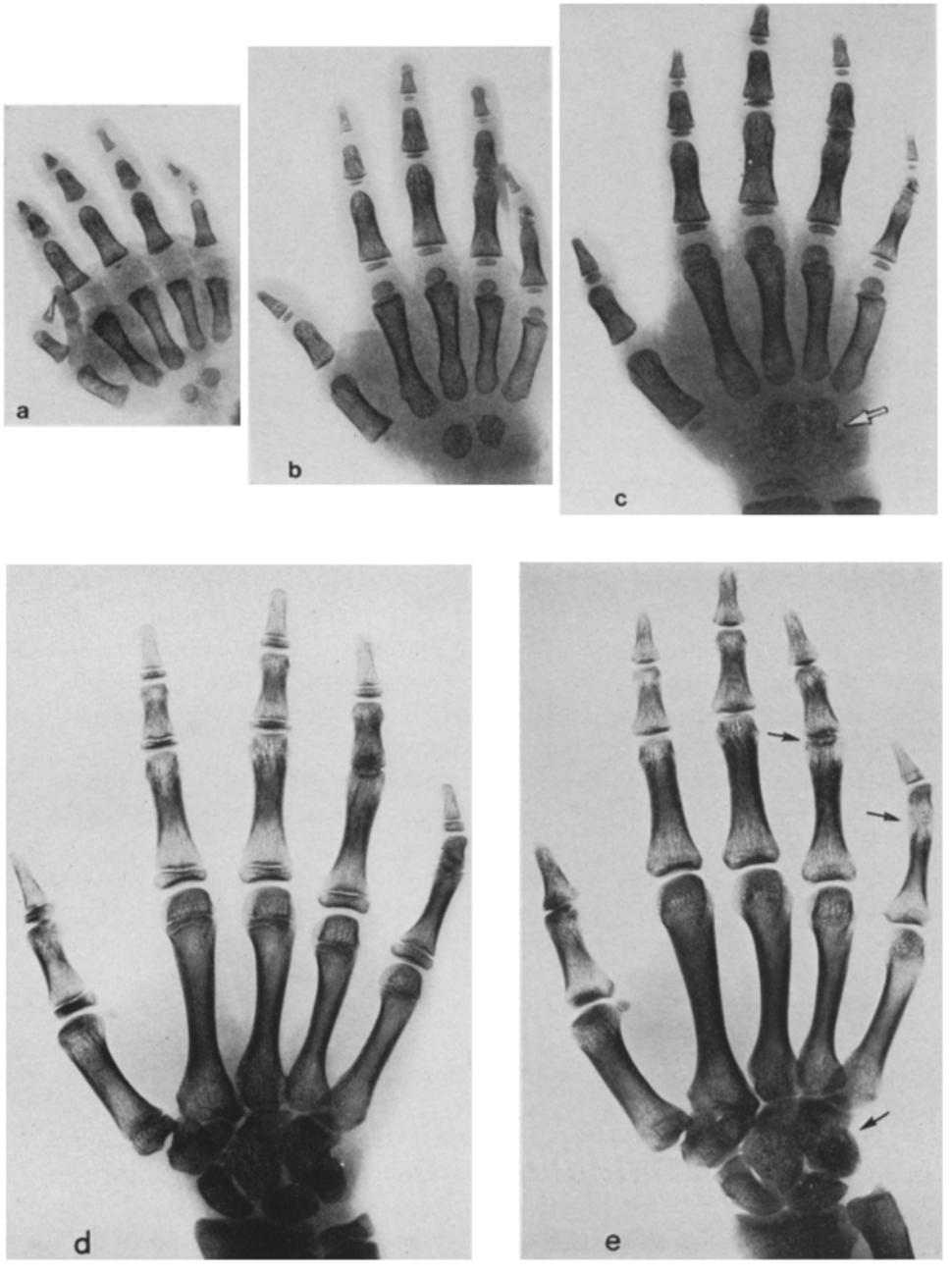


Fig.s 6a–e. Serial roentgenograms of the right hand of the proband of kindred 1. a, at 10 months of age; b, at 3 years of age; c, at 5 years of age; d, at 13 years of age; e, at 20 years of age.

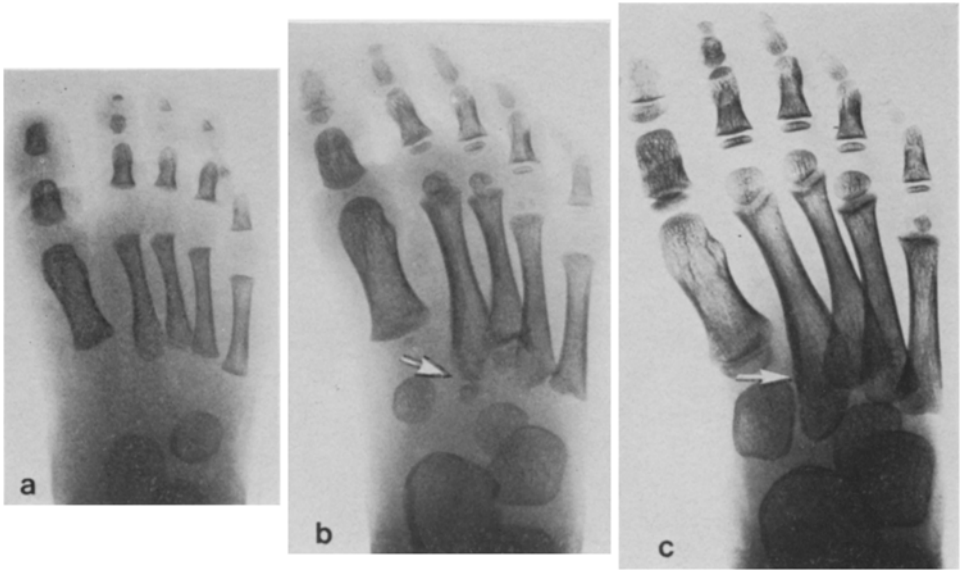


Fig. 7a–c. Serial roentgenograms of the right foot of the proband of kindred I. a, at 10 months of age; b, at 3 years of age; c, at 5 years of age.

os cuneiforme intermedium. The outline of the base of the 3rd metatarsus was markedly irregular (Fig. 7b). At age 5, synostosis between the os cuneiforme intermedium and the 2nd metatarsus was clearly seen. The outline of the base of the 3rd metatarsus showed normal configuration. Synostosis between the os talus and os naviculare was also seen. Joint spaces at the DIP joints of the 2nd through 5th toes were very narrow indicating synchondrosis of the respective joints (Fig. 7c).

Mother (II-5)

In each hand, complete synostosis between the basal and middle phalanges of the index, middle and ring fingers, and that between the middle and terminal phalanges of the little finger were observed. Synostosis between the os hamatum and os triquetrum was also observed (Fig. 8). In each foot, synostosis between the middle and terminal phalanges of the 2nd through 5th toes, that between the os cuneiforme intermedium and the 2nd metatarsus, that between the os cuneiforme laterale and the 3rd metatarsus, and that between the os talus and os naviculare were observed.

Bone changes of the proband at his first visit and those of the mother was reported by one of the authors, Inagaki, in Japanese in 1960.

Sister (III-2)

The sister was born five years after the first visit of the proband. In infancy she showed stiffness of the PIP joints of the ring and little fingers. Roentgenologic study of her over a 15-year-period revealed completely the same process of synostosis as that in the proband.

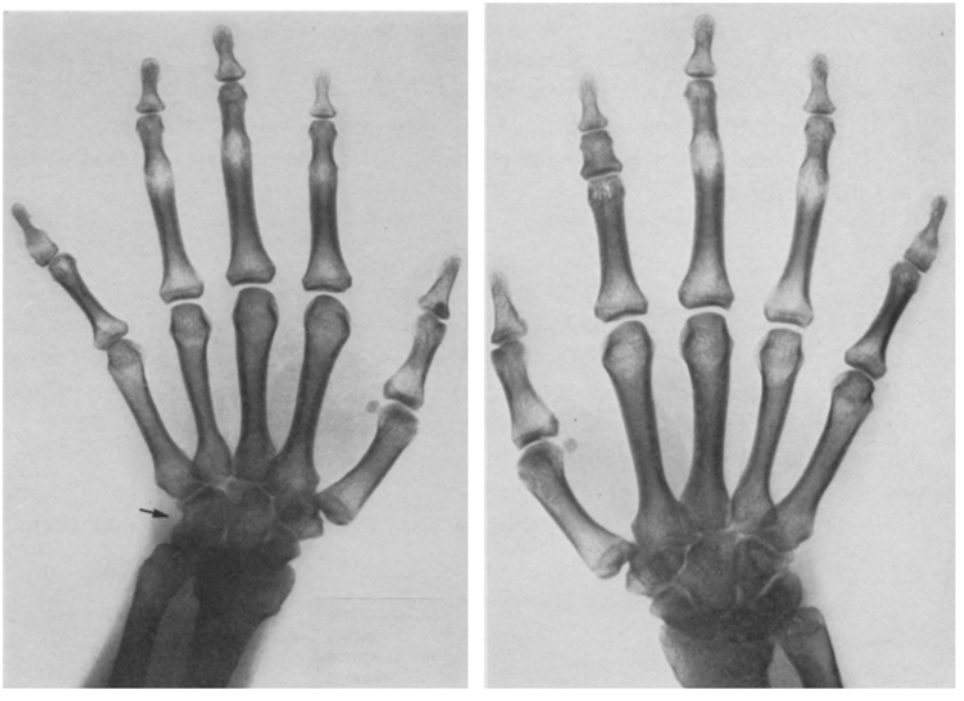


Fig. 8. Roentgenogram of the hands of the mother of kindred 1. Synostosis between the basal and middle phalanges of bilateral index, middle and ring fingers, that between the middle and terminal phalanges of bilateral little fingers, and that between the left os hamatum and os triquetrum (arrow) are observed.

In this family, 7 persons through three generations were affected (I-3, II-5, II-7, III-1, III-2, III-3, III-4).

Kindred 2

Proband: A 7-year-old girl (III-4)

In this family, 8 persons through two generations were affected (II-1, II-2, II-3, II-4, II-5, III-1, III-2, III-4).

Outline of this family was already reported in Japanese by the authors in a previous issue of this Journal in 1960.

Kindred 3

Proband: A 6-year-old girl (IV-6, a monozygotic twin)

In this family, 8 persons through four generations were affected (I-1, II-4, III-9, III-10, III-12, IV-2, IV-5, IV-6). In 6 affected persons other than monozygotic twins only the little finger was affected.

Kindred 4

Proband: A 24-year-old female (II-4)

In this family no other persons were affected.

Kindred 5

Proband: A 24-year-old female (III-3)

Bone changes of this patient were already shown in Figs. 2 and 3.

In this family three persons through two generations were affected (III-2, III-3, IV-6). In the sister and niece of the proband, roentgenologic examination of the feet was not performed.

SEGREGATION ANALYSIS

Segregation analysis was performed on four kindreds in which intrafamilial occurrence of the anomaly was observed. There were 12 sibships born to affected × unaffected couples. Number of affected and unaffected siblings by mating type and sex are shown in Table 1. Same-sexed twins were excluded from the analysis.

Of 36 children, excluding three index cases, 14 were affected and 22 were not, the deviation from the 1 : 1 ratio being statistically insignificant. Ratios of affected to unaffected children were 9 : 19 in sibships born to an affected father × unaffected

Table 1. Segregation analysis.

Mating	Children					
	Males		Females		Both	
Affected father × Unaffected mother	affected	unaffected	affected	unaffected	affected	unaffected
Kindred 1						
I-3	1	3	1	4	2	7
II-7	1	0	1	1	2	1
Kindred 3						
I-1	1	1	0	3	1	4
II-4	3	2	0	3	3	5
III-9	0	1	1	0	1	1
III-10	0	1	0	0	0	1
Subtotal	6	8	3	11	9	19
Affected mother × Unaffected father						
Kindred 1						
II-5	0 (1)	0	1	0	1 (1)	0
Kindred 2						
II-2	2	0	0	0	2	0
II-3	0	0	0	1	0	1
II-4	0	0	0 (1)	0	0 (1)	0
Kindred 5						
II-5	0	1	1 (1)	0	1 (1)	1
III-2	0	0	1	1	1	1
Subtotal	2 (1)	1	3 (2)	2	5 (3)	3
Grandtotal	8 (1)	9	6 (2)	13	14 (3)	22

mother, and 5 : 3 in those born to an affected mother × unaffected father, being also statistically insignificant. Thus, the segregation ratios observed were consistent with the assumption of autosomal dominant inheritance.

DISCUSSION

Historical aspect

According to Strasburger *et al.* (1965), Mercier reported in 1838 the first cases of this anomaly in a French family with only two phalanges in each finger. Since then there were 6 prehistorical reports (Moutard-Martin and Pissavy (1895), Kirmisson (1898), Walker (1901), Goerlich (1908), Drey (1912), Morgenstein (1913)). In 1916, Cushing first coined the term "symphalangism" for this anomaly. Since the publication of a report by Drinkwater in 1917, at least 32 reports have been published by different authors; Bonney (1920), Duken (1921), Brugger (1923), Hefner (1924), Elkin (1925), Stecher (1925), Mouchet and St. Pierre (1931), Pervès (1932), Rochlin and Simonson (1932), Mestern (1934), Bridicza (1938), Stiles and Weber (1938), Schwarzweller (1939), Slater and Rubinstein (1942), Freud and Slobody (1943), Austin (1951), Shiroma (1958), Inagaki (1960), Sugiura and Inagaki (1960), Vessel (1960), Comings (1965), Strasburger *et al.* (1965), Elkington and Huntsman (1967), Harle and Stevenson (1967), Wildervanck *et al.* (1967), Fuhrmann *et al.* (1969), Geelhoed *et al.* (1969), Gorlin *et al.* (1970), Yanagawa *et al.* (1971), Maroteaux *et al.* (1972), Masumi *et al.* (1972), Kato *et al.* (1974).

In 1965, Strasburger *et al.* traced the descendants of the Cushing's pedigree to their latest generations. The anomaly had occurred at least in 151 persons through 10 generations. In 1967, Elkington and Huntsman traced the descendants of the Drinkwater's pedigree to their latest generations, reexamining Drinkwater's insistence saying that transmission of symphalangism had occurred through 14 generations of the Talbot family. After close reappraisal of the historical descriptions, they concluded that sufficient proof for the transmission in such a long term of generations was lacking in the Talbot family.

Skeletal changes

Synostosis between adjoining phalanges: According to the reports of different authors, the main characteristic feature of this anomaly is synostosis of the basal and middle phalanges at the PIP joint of one or more fingers. In most cases, the basal and middle phalanges fuse completely. In some cases the basal and middle phalanges are separated by a very narrow PIP joint space indicating synchondrosis of the PIP joint. In the severest cases, synostosis is observed in 4 fingers, the index through little fingers. In the mildest cases, synostosis is observed only in the little finger. In some cases, the middle and terminal phalanges of the little finger fuse instead of synostosis between the basal and middle phalanges. In the feet, the middle and terminal phalanges of the toes often fuse. No pathological changes have been reported on the thumb or great toe.

Synostosis in the carpal and tarsal regions: In symphalangism, synostosis has also been observed in the carpal and tarsal regions, sometimes in isolation in either of them. In literature, though many authors such as Rochlin and Simonson (1932), Bloom (1937), Bridicza (1938), Schwarzweller (1939), Slater and Rubinstein (1942), Austin (1951), Elkington and Huntsman (1967), Wilderneck (1967), Geelhoed *et al.* (1969), Yanagawa *et al.* (1971), and Maroteaux *et al.* (1972), have reported such association, the remaining authors have not given any comment of this association. On the authors' cases, all of the cases showed synostosis in the carpal and/or tarsal regions. So, the authors would like consider that if the closer roentgenologic investigation had been made on the carpal and tarsal regions, more cases of synostosis in these regions would have been found.

Other skeletal changes: In addition to the above mentioned skeletal changes, one or more following skeletal changes have been noted in families with symphalangism:

1. Brachymesophalangy of the little finger (Bloom, Cushing, Daniel, Drinkwater, Freud, Hann, All of the cases by the authors)
2. Brachymetacarpy of the 5th metacarpus (Kirmisson, Kindred II by the authors)

Brachymetacarpy of the 1st metacarpus (Pervès)

4. Absence of the distal and middle phalanges of the little finger (Walker)
5. Absence of the terminal phalanges of the middle and ring fingers with lack of nails (Drey)
6. Absence of the 1st metacarpus (Mercier)
7. Clubfoot (Kirmisson)
8. Syndactyly (Freud and Slobody)
9. Bone fusion of ear ossicles (Gorlin *et al.*, Masumi *et al.*, Strasburger *et al.*, Vesell).

Associated deafness in symphalangism was concidered to be caused by this bone fusion as a part syndrome of symphalangism.

10. Humero-radio-ulnar synostosis (Mouchet and St. Pierre, Brdicza, Maroteaux *et al.*)

For this association, Maroteaux *et al.* coined the term "la maladie des synostoses multiples."

Genetics

Since the report by Cushing in 1916 and that by Drinkwater in 1917, autosomal dominant inheritance has been ascertained by many researchers. In 1965, Strasburger *et al.* traced the descendants of the Cushing's pedigree to their latest generation. According to their report, the family tree spanned 10 generations including 684 individuals, of whom 351 were affected. From these figures and the pedigree chart, it was confirmed that the affected individuals were heterozygous. X-linkage was ruled out by numerous occurrence of male-to-male transmission of the abnormal

allele. In the affected lines 50% of the individuals had the trait equally distributed between males and females.

Segregation analysis of the present study also revealed that symphalangism has autosomal dominant inheritance.

Other distinct genetic variety of symphalangism

In this type of symphalangism described here, the proximal interphalangeal joints were primarily affected (proximal symphalangism). But several cases with symphalangism of the distal interphalangeal joints (distal symphalangism) have been reported by Inmann (1924), Cole (1935), Bloom (1937), and Steinberg and Reynolds (1948). It is commonly agreed that distal and proximal symphalangisms are two separated hereditary entities (Strasburger *et al.*, 1965, McKusick 1978).

Kassner *et al.*'s case of synostosis at the metacarpophalangeal joints may be classified another type of symphalangism.

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