Syndactylism of Fingers : A Clinical Study

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Introduction :

Syndactyly (united or webbed digits) is known as the commonest congenital anomaly of hand. It may be bilateral and symmetrical, and sometimes runs in families. Two or more fingers may be involved. There is frequent association of other congenital anomalies in the hand and other parts of the body. A similar deformity of webbed toes is also known and the condition may co-exist with syndactylism of fingers.

In the human embryo, the finger-buds grow much faster than the rest of the hand-plate during the 5th and 6th weeks, so that the fingers become clearly outlined by the 7th week and well-formed by the 8th week. It is to be observed that as a result of this rapid growth of the fingers from the hand-plate, the connecting tissues between the fingers become obliterated and the fingers stand out well-defined. The formed fingers develop at the same rate as the rest of the hand thereafter. An arrest of foetal development at a 'critical period' such as the 5th-8th weeks of embryonic life, interferes with the process of development of the hand. This may be brought about by various factors most of which act in a non-specific manner. The obliteration of the connecting tissues between the fingers may be hampered as a result of the interference. With subsequent growth of the fingers and the webs between the fingers at an uniform rate, the fingers are found webbed together at birth.

Material and Methods :

The present discussion is based on an analysis of 44 cases of syndactylism seen and treated at the Department of Plastic Surgery, S.S.K.M. Hospitals and Institute of Post-graduate Medical Education and Research, Calcutta, between 1958 and 1966, in the course of a study on different aspects of congenital hand anomalies. All the patients were subjected to operative treatment and followed up for a period of at least 1 year since the final operation.

Incidence and other factors :

McCollum (1940) reported that webbed fingers occured once in 2000 to 2500 births. Bunnell (1956) mentioned the incidence of syndactyly as 1 in 1000 to 3000 births. The births in the obstetric unit of our Hospital were scrutinised for a period of three years (1963-65), and among a total number of 4805 births during the period, there were 11 cases of congenital hand anomaly. 3 of these were cases of syndactylism of fingers. Thus, one syndactylism per 1602 births, approximately.

23 out of the 44 cases treated in our department were males and 21 were females. The majority of the patients belonged to the economically backward class, the total income of the family being less than Rs. 250/— per month in 37 out of 44 cases. This appears more significant when we consider the fact that the S.S.K.M. Hospital is essentially a paying Hospital.

Two mothers gave history of vaginal bleeding in the early part of pregnancy (2nd month) and in one of these cases, there was also a history of accidental fall preceding the haemorrhage. Out of the 44 patients, 20 were the eldest children of their parents – an observation consistent with those of Macklin (1929) and Murphy (1939) who noticed that congenital malformations occured more commonly in the 1st issues.

Heredity :

History of some congenital defect in other members of the family were obtained in 10 (22.72%) of our cases. In 6 of these cases, the anomalies were exactly similar to those present in the patient's hand. History of other hand anomaly were present in 2 cases (congenital amputation below elbow in maternal uncle of father of case no. 1, and unilateral hexadactyly in maternal uncle of case no. 4) of our series. In 2 other cases, syndactylism of toes were present in some members of the family. One of the parents were actually involved in 8 of the cases, while in 2 cases anomalies were present in other relatives, the parents being normal (Case No. 1 and 4). In one case (Case No. 31), both the parents as also the three off-springs (the patient being one of them) had identical and bilateral syndactyly of the thumb, index, middle and ring fingers. The anomalies were bilateral and symmetrical in as many as 8 of the 10 patients with history of congenital anomaly in the family.

Grouping of cases :

A classification almost similar to that of Emmett (1963) was followed (Table I).

Group A. — Simple syndactylism of fingers with involvement of skin and subcutaneous tissues only — 24 cases (54.54%).

Group B. — Syndactylism of fingers associated with other congenital anomaly or anomalies of hand — 5 cases (11.38%).

Group C. — Acrosyndactyly. The distal parts of the digits are fused but the proximal region and particularly the depth of web remain unaffected — 7 cases (15.9%).

Group D. — Syndactylism with abnormality of skeletal tissues of the digits — 8 cases (18.18%).

In 22 cases, (50%) there was involvement of both hands (Table I). The involvement was bilaterally symmetrical in all these cases except one where middle, ring and little fingers of the right hand and ring and little fingres of the left hand were involved in simple syndactylism (Gr. A).

Out of the 22 cases of unilateral involvements, as many as 20 were of the right hand. The knowledge of the importance of the right hand from the functional aspect was the probable reason which compelled the patients to seek operative treatment

more readily than those with involvement of left hand only.

Syndactylism was complete in nature in 30 cases (68.18%) and in 5 of these, the nails were also fused. Out of the 14 incomplete cases, fusion was incomplete proximally in 7 cases (all the cases of Acrosyndactyly i.e. Gr. C) while the fusion did not extend distally beyond the level of the proximal interphalangeal joint in 5 cases, and that of the distal inter-phalangeal joint in 2 cases.

Distribution of cases :—

The digitwise distribution of cases has been set out in Table II. The commonest involvement was of two fingers only 23 cases (52.27%), and the fingers most commonly involved were the middle and ring fingers—17 cases. Three fingers were involved in 8 cases (18.18%). In 11 cases (25%), four fingers were involved and in three of these, the thumb was also involved. All the five digits were found to be webbed in 2 cases (4.5%), and in one of these (Case No. 10, Fig. 9), the involvement was bilateral. There was one case of involvement of non-adjacent fingers in 'acrosyndactyly' the index, ring and little fingers being fused at the tip and the middle finger remaining free (Case No. 9, Fig. 15).

Associated anomalies :—

Simple syndactylism of fingers with other associated congenital hand anomaly or anomalies belonged to Group B. In addition to these cases, it was observed that some of the cases belonging to Groups C and D were associated with congenital hand anomaly or anomalies other than acrosyndactyly (essential feature of Gr. C) and skeletal abnormality of digits (essential feature of Gr. D) respectively. Congenital anomalies of remote parts of body were also present in a number of cases.

There were 27 cases in all (61.36%), where one or multiple congenital anomalies were associated The different anomalies encountered in our series were : brachydactyly (short digits) — 2 cases (Case Nos. 28 and 43); clinodactyly (bent digits) — 2 cases (Case Nos. 10 and 37); symphalangism (end-to-end fusion of phalanges) — one case (case no. 8); ipsilateral absence of pectoralis major muscle—one case (Case No. 40); axillary web-one case (Case No. 43); flexion contracture of fingers-one case (Case No. 37); constriction rings—7 cases (all the cases of Gr. C—Case Nos. 3, 4, 5, 9, 13, 14, 16); syndactyly of toes — 15 cases (Case Nos. 2, 4, 9, 10, 11, 13, 18, 20, 22, 23, 30, 32, 36, 41, 42); polydactyly of foot—4 cases (10, 20, 31, 32); cleft lip and/or palate — 4 cases (Case Nos. 17, 18, 19, 23). The association of craniostenosis with syndactyly (acrocephalosyndactyly or 'Apert's syndrome') was observed in 3 cases (Cases Nos. 18, 31, 41) and in one of these cases (Case No. 38, Fig. 2), cleft palate and syndactyly of toes were present as well. All these three cases exhibited some degree of mental retardation also.

Acrosyndactyly (Gr. C) :—

All the 7 cases of acrosyndactyly in our series were associated with proximal ring constrictions. 6 of the cases were also

associated with anomalies other than acrosyndactyly, distal to the ring constriction rings. These were : partial amputation of digits — 3 cases (Case Nos. 3, 4, 13); impairment of sensation — 3 cases (Case Nos. 5, 13, 16); oedematous blobs of tissues — 2 cases (Case Nos. 4 and 5). Acrosyndactyly of toes were associated in 3 of the cases (Case Nos. 4, 9, 13). 2 cases had associated constriction rings in the thigh Case Nos. 5 and 16).

Histopathological study of the ring constrictions (bone deep) showed the structure of skin with dearth of sub-cutaneous tissue.

Skeletal abnormalities (Gr. D) :—

These included : fusion of terminal phalanges — 3 cases (Case Nos. 10, 25, 37); fusion of metacarpals 2 cases (Case Nos. 1 and 18); fusion of metacarpals as also of terminal phalanges — one case (Case No. 36); symphalangism — one case (case no. 8, Fig. 6); and gross deformity of inter-phalangeal joints — one case (Case No. 7 Fig. 19).

Functional aspect :—

There was no functional impairment in the affected hand or hands in 14 cases. 11 cases showed some impairment — a good 'grasp' with a weak 'pinch' — probably due to involvement of the index finger in all these cases. Impairment of both 'grasp' and 'pinch' was noticed in 15 cases. Gross impairment of function was noticed in 4 cases—both the thumb and the index being involved in each of these cases. The associated anomaly or anomalies of hand, when present, modified the functional behaviour of the hand in most of the cases.

Management :—

This was essentially operative. All the cases presented the problem of cosmetic appearance apart from functional impairment in a number of cases. The operative age varied from 1 to 23 years, the majority (28 cases) being operated between 4-6 years of age. Bilateral cases were always taken up in at least two stages, only one hand being taken up at a time. In syndactylism of more than two fingers, separation of the two sides of any involved finger was always carried out in two different stages, with a gap of at least three months.

A total number of 117 webs were separated in 44 patients. In 7 cases of Acrosyndactyly (Gr. C), the depth of the web was not affected (the syndactylism was incomplete proximally) and the problem of reconstruction of the commisure did not arise. As already mentioned earlier, all these cases of acrosyndactyly in our series were associated with proximal ring constrictions. The treatment consisted of Z-plasty, correction of the constriction rings followed by separation of the fingers at the same stage (3 cases) or at a second stage (4 cases) after one month.

The operation :—

All the operations were carried out under general anaesthesia, employing a sphygmomanometer cuff around the arm as a pressure tourniquet, which was never applied for more than 60 minutes at a stretch.

The fingers were separated by zig-zag incisions (Fig. 1) in 32 cases. The incisions on the dorsal and palmar aspects were planned so as to achieve as much closure of raw

areas by skin-flaps as possible without tension. In 12 cases, the separations were achieved with straight incisions. In cases where separation of fingers had to be carried out in more than one stage, the same type of incision (zig-zag or straight) was used for separation at the different stages for any particular case, so that the results could be studied and assessed case wise. The construction of the commisure (depth of web) was carried out in all the 37 cases (this was not necessary in 7 cases of acrosyndactyly) by crossed V–plasty flaps (Fig. 1) with the base of both palmar and dorsal flaps at the level of metacarpo-phalangeal joints.

Damage to the neurovascular bundles were avoided and the flaps were handled with utmost care and gentleness. The defatting' of the flaps were restricted to a minimum. 4/0 – 5/0 silk was used for suture of the skin-flaps. For the raw areas which could not be covered with skin flaps, split skin-grafts (0.01″ – 0.014″) taken from the thigh were applied and secured in place with tie-over dressings.

Each separation was designed to cover one of the separated fingers entirely with skin-flaps and to use full length wide skin-grafts in the other separated finger. We were successful in 13 cases only. In the majority of cases (31 cases—70.46%), skin-graft small and/or full length had to be used for every separated finger, so that cover of the raw surface without tension could be achieved.

Fused terminal phalanges, (Gr. IV) were separated with bone cutter and trimmed smooth. Mild clinodactyly in 2 cases and symphalangism in one case of Gr. IV were left alone without any special treatment. Fused nails present in 5 cases of Gr. I were split and strips of the edges with symmetrical width of matrix were excised. The small gaps created could be filled up easily with adjustment of local skin-flaps.

Post-operatively, the hand was immobilised in a dorsal Plaster-of-Paris cast extending from above the wrist to the finger tips. Tie-over dressings were removed on the 5th day after operation and the cast was discarded as soon as the wound healed completely. This was followed by encircling plasters of the separated fingers in slight flexion of the metacarpo-phalangeal and inter-phalangeal joints for a fortnight. In the meantime, dorsal aluminium splints with rings to hold on to the fingers were made ready according to measurements of each finger, and patients were made to wear splints day and night (except for two periods of exercise when all the movements of the joints involved were encouraged) on removal of the encircling plasters. The hand was allowed free with normal activity usually by 3 months.

Post-operative Results

Primary healing occured in 35 out of 44 cases (79.54%). In 9 cases, there was necrosis of skin-flaps and (20.46%) or failure of graft at some stage or separation necessitating secondary skin-grafting. Some degree of wound infection was present in all these cases, 3 of the 9 cases were of acrosyndactyly (Gr. C). A pertinent observation was that primary healing occured in 3 out of the 4 cases of acrosyndactyly (Gr. C) treated by Z-plasty of ring constriction and separation of fingers at separate stages. Secondary

grafting was necessary in 2 out of the 3 cases of acrosyndactyly where the procedures were carried out in the same stage (Table IV). This proved the necessity of removing the ring constriction for the improvement of blood supply to the distal parts of the fingers.

In long-term follow-up, post-operative deformities (Table IV) were found to occur in 7 cases (15.9%). These were recurrence of web—2 cases (Case No. 6 & 40), longitudinal band on fingers limiting extension—2 cases (Case No. 2 & 25), flexion contracture of inter-phalangeal joints-one case (Case No. 7), and lateral deviation of inter-phalangeal joints—2 cases (Case No. 12 & 38).

The relation between the type of incision (zig-zag or straight) and occurence of some of the deformities post-operatively has been illustrated in Table V. There was only one recurrence of web among the 32 cases where zig-zag incisions were used for separation of digits. On the otherhand, there were 3 cases of post-operative deformity (recurrence of web in one case and 2 cases of longitudinal contracture band on fingers) among 12 cases of separation of digits by straight incisions. As already mentioned, there were 3 cases of deformity with abnormal position of joints. In one of these cases (Case No. 12, Fig. 16), operated at the age of 23, gross flexion deformity of the ring and little fingers developed bilaterally.

Discussion :

Aetiologically, congenital anomalies of hand have been broadly and generally divided into two major groups :—1. Intrinsic or Genetic-originating from abnormality of germ plasm itself and 2. Extrinsic-abnormalities due to 'teratogenic' environmental factors (Duraiswamy, 1952). Nutritional deficiencies, mechanical trauma from uterine malposition, different chemical and toxic factors, infection during pregnancy, and irradiation during pregnancy are among the factors held responsible (Barsky 1958). As already mentioned earlier, these factors act at a 'critical' period of development and differentiation of the part affected.

There has been much discussion regarding the mode of transmission of the genetic factor. A deformity may be transmitted in the pure form or, a tendency towards deformity of a part is inherited and the exact form may be variable (Patterson, 1959). Hereditary factor was positive in 10 cases (22.72%) of our series. This included history of other type of hand anomaly separate from that present in the patient in 2 cases, and anomaly of other part of the body in 2 cases. It seems that the genetic factor may be modified by other aetiological factors, and congenital anomaly may occur in different forms in different generations of the same family, a tendency towards deformity being inherited. It is also to be noted that the hereditary factor may be actually more significant than is statistically apparent because, as pointed out by Barsky (1958), the anomaly in the patient under study may be the first appearance of a trait in the family.

The fact that most of our cases belonged to the economically backward class, seems to amplify the importance of poor hygienic

Fig. 13—Fusion of terminal phalanges of index and middle fingers in a case of syndactylism (Case No. 36)

Fig. 15—An unusual case of Acrosyndactyly. Fusion of non-adjacent fingers (index, ring and little) while the middle finger stands out free. The features help a great deal in discussions on aetiogenesis—Case No. 9

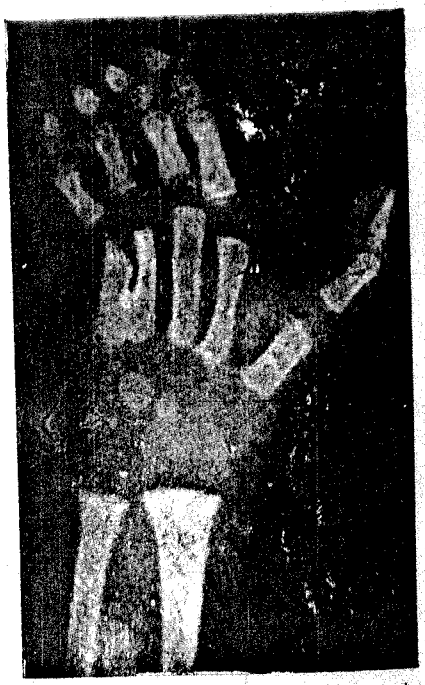

Fig. 14—Fusion of 4th and 5th metacarpals with syndactylism of index, middle, ring and little fingers. (Case No. 1)

Fig. 16—Post-operative joint deformity (ring and little fingers) following operation for syndactylism. (Case No. 12)

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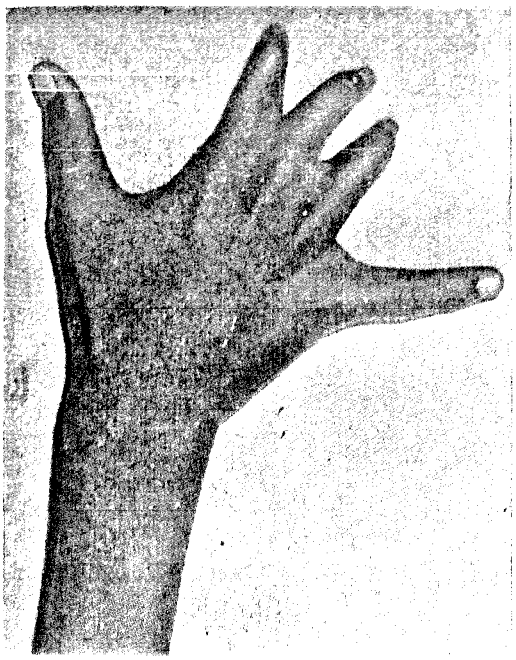

Fig. 17—Recurrence of Web after separation of syndactylism of index, middle and ring fingers (Case No. 40)

Fig. 18—Longitudinal contracture band after separation of syndactylsim in ring and little fingers. Straight incisions were used for separation. (Case No. 25)

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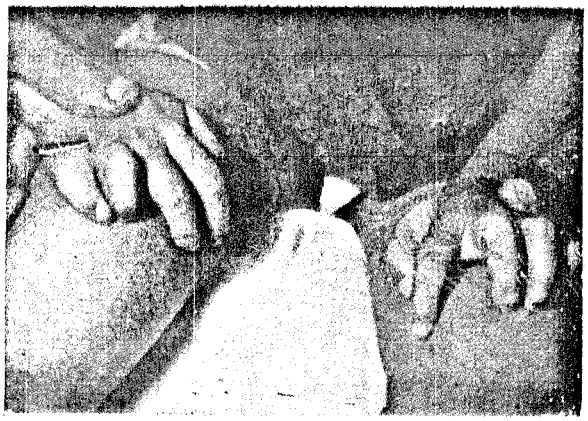

Fig. 19 – Flexion contractures at inter-phalangeal joints following operation for syndactylism of fingers (Case No. 7). Patient neglected use of splint post-operatively.

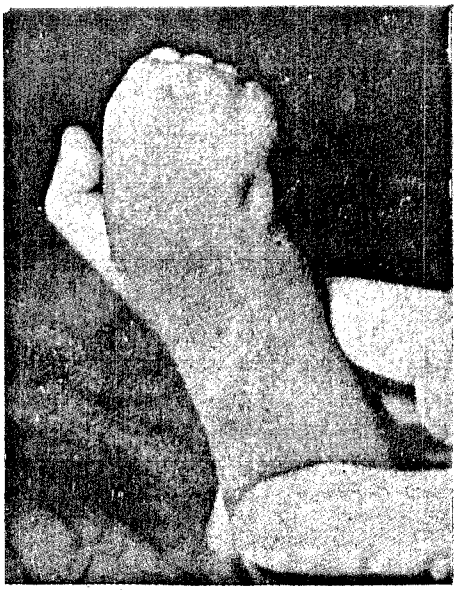

Fig. 5—Complete syndactylism of index, middle, ring and little fingers of right hand (Case No. 8)

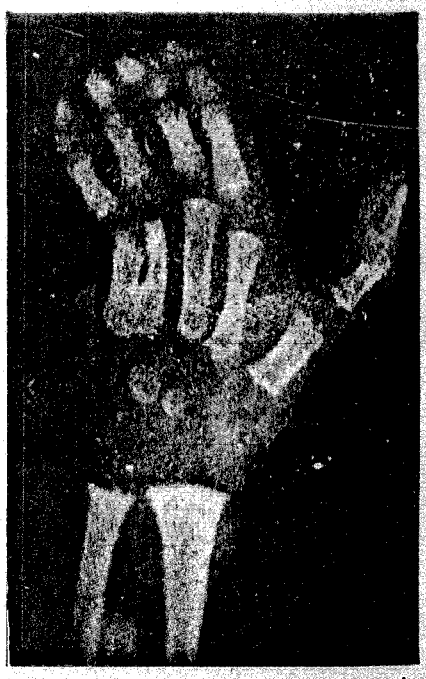

Fig. 6—X-ray of the hand shown in fig. 5-showing symphalangism (fusion of proximal & middle phalanges) of middle and ring fingers and clinodactyly (lateral displacement) of inter-phalangeal joints. (Case No. 8).

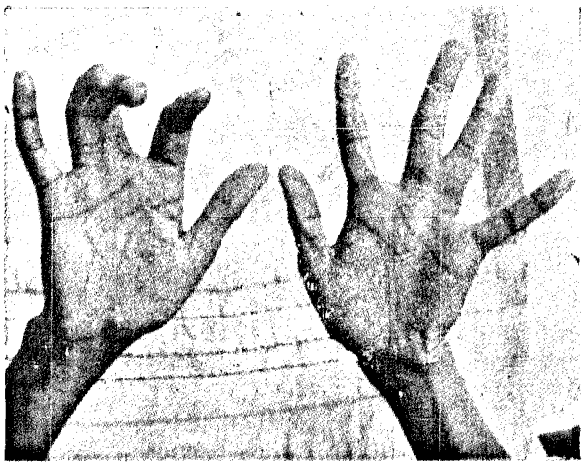

Fig. 7—Bilateral complete syndactylism with flexion contracture of middle and ring fingers of right hand. Incomplete syndactylism middle and ring fingers of left hand. (Case No. 37)

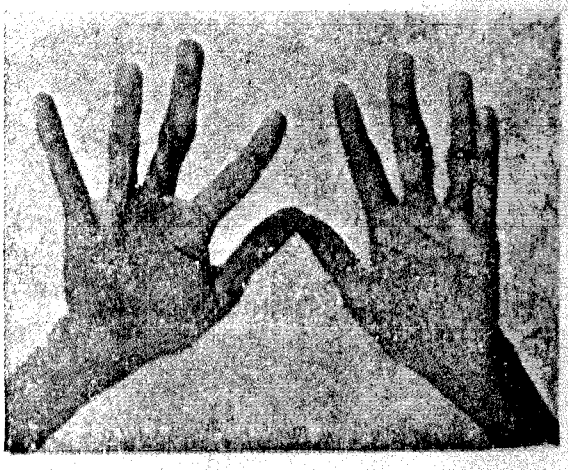

Fig. 8—Post-operative photograph (3 months) of the hands shown in fig. 7 (Case No. 37)

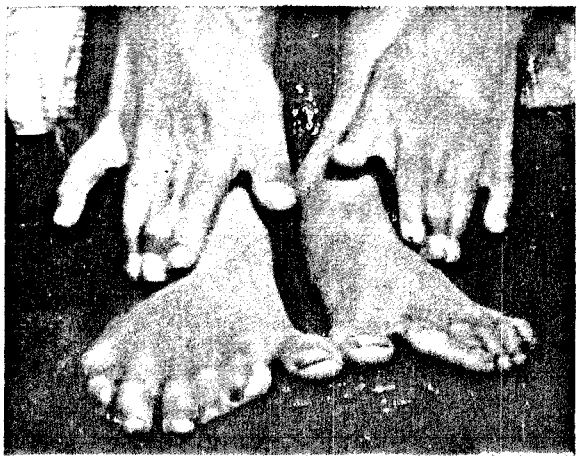

Fig. 9—Bilateral syndactylism of all fingers with associated polydactyly of feet (Case No. 10)

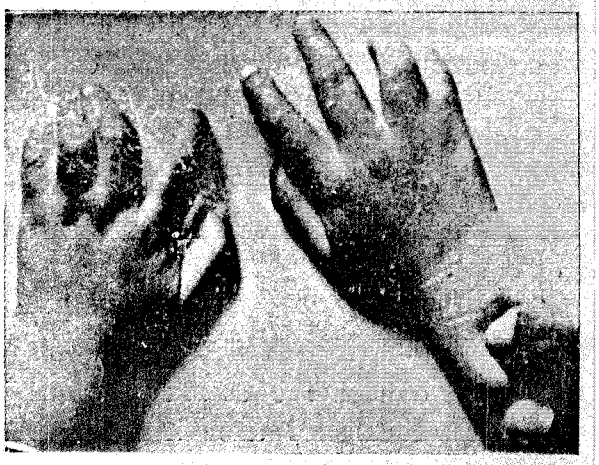

Fig. 10—Bilateral syndactylism involving three fingers of left hand and two fingers of right hand (Case No 7).

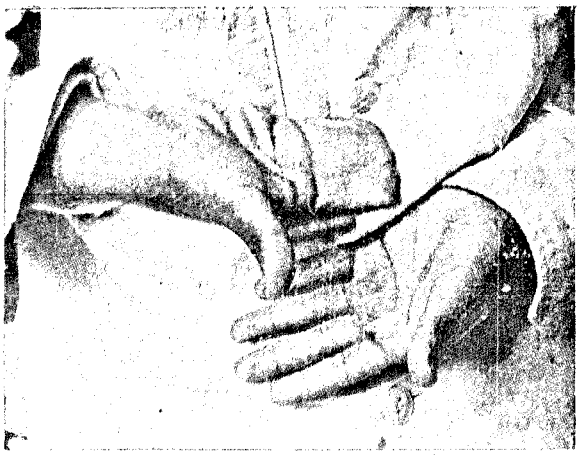

Fig. 11—Complete syndactylism of index, middle, ring and little fingers of right hand (Case No. 34)

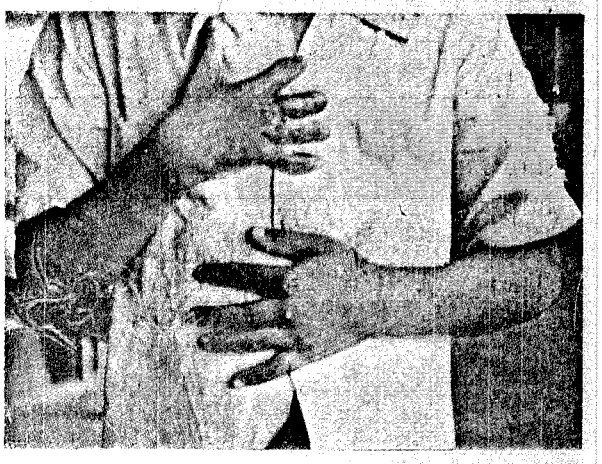

Fig 12—Immediate post-operative photograph (after removal of all dressings) in the case (Case No. 34) shown in Fig. 11. Photo taken after the last stage of separation. The principles illustrated in fig. 1 were followed for separation in all the stages.

and dietary conditions as aetiological factor. Maternal morbidity in the early part of pregnancy, present in 2 cases of our series, may be only a matter of coincidence, but it has to be remembered that unfavourable circumstances of embryonic development during the 5th-8th weeks (critical for development of hand) may be logically related to congenital anomaly of hand in the offspring.

The occurence of associated anomalies, most of which were examples of 'arrest' or 'absence' of development, in as many as 27 cases (61.36%) of our series lends weight to the belief that 'syndactyly' is the result of embryonic development at a 'critical' period of the hand's development. The cases of 'Acrosyndactyly' deserve special discussion. Although the anomaly is very often described as only a particular variety of syndactyly, the aetiological basis seems to be different. In acrosyndactyly, the depth of the web remains unaffected, but the digits are fused distally. This deformity can not be explained by the failure of obliteration of the webbing tissues between the digits, which as has been shown by Arey (1954), is achieved under normal conditions, by rapid elongation of digits from the hand-plate. Hence, in acrosyndactyly, the fingers appear to have developed separately and then got fused distally (Maisels, 1962). Constriction rings proximal to the acrosyndactyly were present in all the cases of Gr. C of our series and in 2 cases, constriction grooves were present in the thigh also. Ring constrictions have been regarded as examples of absence of development of the sub-cutaneous tissue borne out by histopathological evidence (Patterson 1961). It may be argued that the primary defect in these cases of Acrosyndactyly was the absence of development of the subcutaneous tissue (manifested by ring constrictions) including vessels, nerves, and lymphatics. The vascular insufficiency led to ischaemic necrosis of parts of the digits near the tip. The resultant raw areas (ulcers) subsequently got adherent to each other leading to the deformity in acrosyndactyly. Involvement of non-adjacent fingers in acrosyndactyly (Case No. 9 Fig. 15) is also satisfactorily explained by this argument. The associated features in our case like loss of sensation, partial amputations, oedema of tissues distal to the ring constrictions, could be very well explained by absence of development of tissues of different kinds and degree. The absence of primary healing in some of the operated cases are additional evidence.

Syndactyly was found to occur most commonly in the simple form and the middle and ring fingers were the commonest combination in our series. These observations are consistent with earlier reports (Barsky, 1958—39 cases and Emmett, 1963 60 cases) but the remarkable features were the greater incidence of Acrosyndactyly in our series and the constant association of ring constructions in these cases. Emmett. (1963) reported 5 cases of acrosyndactyly, with only one example of associated ring constriction among 60 cases of syndactyly.

The construction of the commisure with crossed V-plasty flaps in all the 37 cases of our series (the 7 cases of acrosyndactyly did not have this particular problem) seem to be quite satisfactory with only 2 cases getting

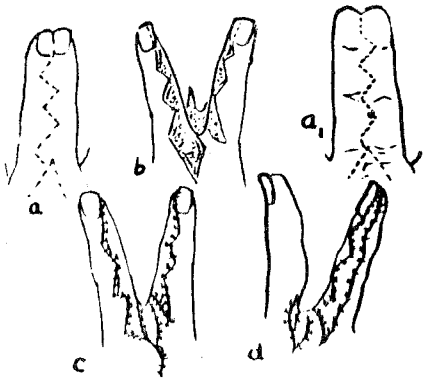

Fig. 1—Showing the steps of operations (aa', b, c, d) as followed in majority of the cases of our series.

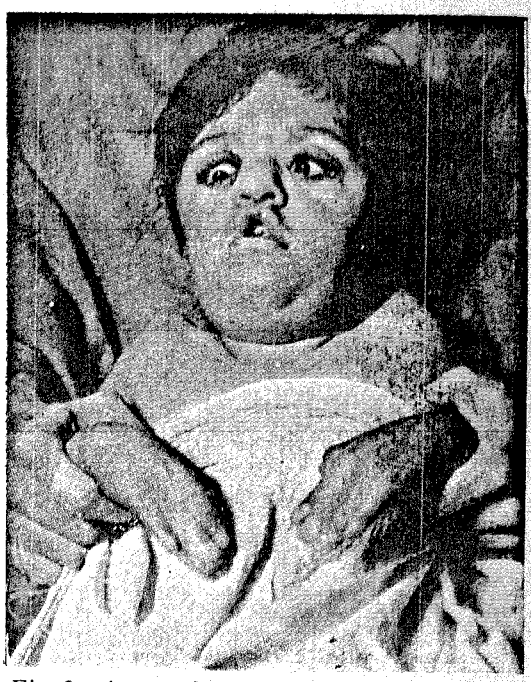

Fig. 2—Acrocephaly with bilateral syndactylism of fingers-Apert's Acrocephalosyndactyly. The child had cleft palate and syndactylism of toes as well. (Case No. 38)

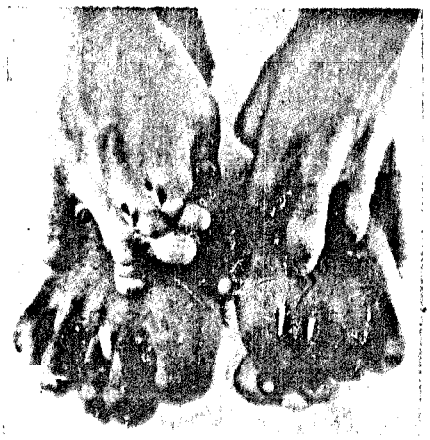

Fig. 3—Acrosyndactyly of hand and feet. Distal oedema and partial amputation of digits are also shown. (Case No. 4)

Fig. 4—Brachysyndactyly (syndactylism in short digits) of Right index, middle and ring fingers. The normal left hand is also shown for comparison. (Case No. 43)

recurrence of web (5.67%). The use of zig-zag incisions for separation of fingers was also satisfactory but we were not happy with the straight incisions—2 out of 12 cases developing longitudinal contracture bands limiting extension. Skin-grafts were necessary for cover in all the 44 cases. It should be emphasised that skin-flaps should always be sutured without any tension and skin-grafts used liberally.

Post-operative management of the cases must be carried out with utmost care. Splintage plays a very vital role. Post-operative movements should also be gradually encouraged and the parents have a vital role to play.

The operative age is yet another important factor. Generally speaking, the recovery appears to be more sound in patients operated at 5 or 6 years of age, the probable reason being the fact that the child learnt newer movements, which were not possible before separation, more rapidly at such age. The older patients, on the otherhand, adopted themselves to trick or improvised movements before treatment, and later found it difficult to discard those and learn correct movements after treatment.

Table I—Showing distribution of cases.

Group of syndactyly	Rt. hand.	Lt. hand.	Bilateral	Total
A. Simple syndactyly	9	1	14	24
B. Syndactyly with other congenital anomaly of hand	4	—	1	5
C. Acrosyndactyly	6	—	1	7
D. Syndactyly with skeletal abnormality.	1	1	6	8
Total :	20	2	22	44

Table II—Showing distribution of cases (digitwise).

	R. hand	L. hand	Bilateral
Group A. (Simple)	MR—5 cases IMR—1 case IMRI—2 cases TIMR 1 case	TI—1 case	MR—9 cases MRL—1 case MRI—RL—1 case IMRI 1 case TIMR—2 cases
Group B. (Syndactyly with other hand anomaly)	IM—1 case MR—1 case IMRL—1 case TIMRL—1 case		IMR—1 case
Group C. (Acrosyndactyly)	IM—2 cases IMR—2 cases IRL—1 case IMRL—1 case		MR—1 case
Group D. (Syndactyly with bony abnormality)	IMRL 1 case	IMR—1 case	MR 1 case RL—2 cases IMRL—2 cases TIMRL—1 case

T, I, M, R, L,—signify thumb, index, middle, ring and little fingers.
Involvement of 2 fingers 23; 3 fingers—8; 4 fingers—11; 5 fingers—2 cases.

Table III

Showing the different methods of separation and skin cover.

Method.	Group A.	Group B.	Group C.	Group D	Total
Zig-zag incision	5	1	2	—	8
a) closure with skin-flap only of one separated finger possible, skin-graft of the other.		—	—	—	—
b) Skin-grafting necessary for every involved finger.	12	3	1	8	24
Straight incision					
a) Closure with skin-flap of one finger possible, skin-graft of other.	—	1	4	—	5
b) Skin-grafting of every involved finger.	7	—	—	—	5

Table IV

Showing post-operative deformities (groupwise).

Post-operative deformity	Group A.	Group B.	Group C.	Group D.	Total
Recurrence of Web	2	—	—	—	2 (4.54%)
Longitudinal band on fingers	1.	1	—	—	2 (4.54%)
Flexion deformity at inter-phalangeal joints.	1	—	—	—	1 (2.27%)
Lateral (side-wise) deviation at interphalangeal joint.	—	—	—	2	2 (4.54%)
Total	4	1	—	2	7 (15.9%)

Table V

Showing relation of some complications with type of incision.

Type of incision	Total	Recurrence of Web.	Longitudinal band
Zig-zag incision	32	1	—
Straight incision	12	1	2

Table VI

Showing healing of Acrosyndactyly (Group C) cases.

Method	Total	Primary healing	Secondary skin-graft necessary
One-stage Z-plasty of ring constriction and separation of digits.	3	1	2
Z—plasty of ring and separation at different stages	4	3	1
Total :	7	4	3

Summary :

Different aspects of 44 cases of Syndactylism seen and treated at Department of Plastic Surgery, S. S. K. M. Hospital and IPGMER between 1958 and 1966, and followed up for varying periods from 1 to 8 years are described. The probable aetiological factors, the clinical variations, and the treatment are discussed.

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