FAMILY STUDIES AND THE CAUSE OF CONGENITAL CLUB FOOT *

Talipes Equinovarus, Talipes Calcaneo-valgus and Metatarsus Varus

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This paper on congenital club foot in Devonshire is based on a study of 635 patients treated at the Princess Elizabeth Orthopaedic Hospital, Exeter. It deals with the family history and associated congenital abnormalities, and attempts to clarify the causation of club foot. Particular attention is given to talipes equinovarus, and there are short notes on talipes calcaneo-valgus and metatarsus varus. Only the uncomplicated congenital deformity has been investigated—patients with spina bifida, cerebral palsy and arthrogryposis are not included.

The hospital serves the greater part of the largely rural county of Devon (excluding the Plymouth area), whose population is fairly stable, so that it is relatively easy to trace families and it is possible to follow progress over long periods.

INVESTIGATION OF A CONGENITAL DISORDER

There are three main causes of congenital abnormalities.

Genetic causes—The abnormality may be determined purely genetically from the moment of conception, and then exhibits a typical pattern of inheritance. In the case of a dominant gene such as in diaphysial aclasis the affected individual has an affected parent. The gene has come either from the father or the mother, and is apparent in both parent and child unless a fresh mutation has arisen. In the case of a recessive gene the disorder is apparent in the child only if an abnormal gene is contributed by each parent. Thus, although most of the parents appear normal, several of their children are often affected. In haemophilia the abnormality is apparent only in the male, but is transmitted by the female who carries the gene on the X chromosome.

Twin studies are helpful in determining whether or not a disorder is genetic in origin. In a purely genetic disorder, identical twins invariably present with the same abnormality. If this is not so, then the disorder cannot be purely genetic, though it may be partly so. In that case identical twins will more often both be affected than fraternal twins of similar sex.

Environmental causes—The congenital disorder may be unrelated to the genetic constitution. For instance, cerebral palsy can be caused entirely by birth trauma; congenital deafness may be produced by rubella occurring in the early weeks of pregnancy. The nature of the damaging agents is varied—there may be damage from mechanical causes, anoxia, radiation, drugs, bacterial toxins or other causes. The type of damage is less significant than the stage of development at which it occurs. For instance, Duraiswami (1952) found that by injecting insulin into chick embryos on the sixth day of incubation, he could regularly produce congenital dislocation of the hip.

In a retrospective study such as the present one, the evaluation of environmental factors of this nature is not feasible. It is all too easy for a mother who has had a deformed child to think back and remember imprecisely any minor disturbances during pregnancy.

Mixed genetic and environmental causes—The third group of congenital disorders fits neither into the purely genetic nor the purely environmental categories. There is no recognisable

* This paper forms the basis of the first part of the Robert Jones Prize Essay for 1962.

pattern of inheritance, and yet there is clearly some genetic factor. The cause is thought to be of a mixed genetic and environmental nature. It is to this group that congenital club foot belongs.

THE CAUSE OF CONGENITAL TALIPES EQUINOVARUS

There has for centuries been much speculation on the cause of congenital talipes equinovarus. In reviewing the last seventy years' literature in English, one is struck by the contrast of the paucity of available facts with the wealth of theorising. These theories fall roughly into two groups: 1) Compression of the foetus in the uterus (Parker and Shattock 1884, Browne 1934), and 2) "Anatomical" theories.

Intra-uterine compression—There has been little genuine evidence since the investigations of Parker and Shattock (1884). They pointed out that inversion of the foot is normal during the earlier months of foetal life, and that if this inversion "be maintained beyond the normal period of time, the muscles and ligaments will as a consequence be adaptively short on one aspect of the limb and too long on the other. A normal position of inversion will finally become a deformity."

Browne has persisted in the theory of mechanical pressure maintaining the feet out of position. He and Parker and Shattock mention oligamnios with a small "tight" uterus and uncomfortable pregnancy as a cause, yet this is by no means an invariable occurrence, even if talipes equinovarus is occasionally caused in this way. However, a feeling remains that mechanical pressure may well be of some significance: a newborn child with club feet *looks* as though the feet have been pressed into the deformed position. All that can be said with certainty is that undue compression of a foetus does from time to time occur—a child can be born with pressure sores resulting from one part pressing on another, and it remains possible that talipes equinovarus is an allied phenomenon.

"Anatomical" theories—Nearly every structure in the foot and leg has at one time been blamed for the deformity, often with little appreciation that the time of birth is but an incident in the development of a child. If a congenital anomaly occurs early in the development of the foetus, secondary changes are well established by the time of birth. At this time there is no question about the existence of tight fibrotic structures on the medial side of the foot, and of overstretched, weakened peronei on the lateral side.

It has often been said that the cause of the deformity is this thickened, fibrotic mass of tissue on the inverting side, and that there is a primary weakness of the peroneal muscles that allows the invertors to overact. Undoubtedly there is muscle imbalance, yet it is no more likely that this is the primary fault than that it is a result of maintenance of a deformed position from some extrinsic cause.

Brockman (1930) considered that the cause of talipes equinovarus was a dislocation of the talo-navicular joint. This may certainly be present, but it is no more an explanation of why the deformity occurs than it is to say that the cause of congenital dislocation of the hip is congenital dislocation of the hip.

Dittrich (1930) put forward the theory that the primary failure was in the peripheral nerve to the peroneal muscles. He found a sluggish electrical reaction in these muscles in a premature infant with club foot. Durante and Courtilier (quoted by Adams, Denny-Brown and Pearson 1953) found foci of sclerosis in the anterior horn cells of the spinal cord, and considered this to be the primary cause of the deformity. In each case cause and effect are again confused: degeneration of anterior horn cells will lead to paralysis of the associated muscles; but conversely, when a muscle cannot function adequately there will be retrograde degeneration of its anterior horn cells.

The most reliable information comes from dissection of the foetus. Wiley (1959) reviewed forty-two cases and added six more of his own. He concluded: "there is no macroscopic or microscopic abnormal finding which is constant, nor is there a regular pattern

of abnormality in the case of the tendons and their insertions." This material suggests that in talipes equinovarus the primary cause of the deformity is an extrinsic factor maintaining the foot in a deformed position and causing growth deficiencies in the long muscles.

Yet this is not enough: it is well known that a family history of the deformity is not uncommon, and that heredity plays some part.

INHERITANCE OF TALIPES EQUINOVARUS

Material—All the available case notes for the period from the start of the hospital in 1926 up to 1961 were studied, provided that the diagnosis of congenital talipes equinovarus was certain. In all, the notes of 340 patients were studied. The main, near-complete, group for study was that of the 144 patients traced who were born between 1940 and 1961 (Table I). This is the only orthopaedic hospital in the area, and since both private and National Health Service patients were included, it is believed that the notes of every case in the district during this period were available.

Incidence of talipes equinovarus in the general population—The period 1948–60 inclusive has been taken, using the Registrar General's figures for this district (Table II).

McKeown and Record (1960) noted 4.44 cases per 1,000 births in their Birmingham survey, but the type of talipes was not specified. Stewart (1951) noted one case of talipes equinovarus per 1,000 births.

Sex ratio—The ratio of male to female is 2.17 to 1. This ratio of about two to one is noted by all writers on the subject. We noted also forty-six patients with "postural" talipes equinovarus—that is, feet which at birth are in equinovarus but which correct easily within a few weeks and show no tendency to relapse. Girls were so affected more often than boys, in the ratio of 1 to 0.3.

FIG. 1

FIG. 2

First degree relatives of the index patients, with talipes equinovarus. The increased risk to the male relatives of the female patients is shown. Figure 1—First degree relatives of the male patients. Figure 2—First degree relatives of the female patients.

First degree relatives with talipes equinovarus—The 1940–61 group is considered in relation to the first degree relatives—that is, the parents, brothers and sisters of the index patients (Table III); 2.9 per cent of siblings have the same deformity, as against 1.2 per 1,000 cases in the general population. The chances therefore that a second child in a family will have this deformity are increased more than twenty times. The preponderance of boys with the deformity has been noted, and it is of interest to consider the male and female first degree relatives separately (Table IV and Figs. 1 and 2). It is seen that there is an increased risk to the male relatives of the female patient. She suffers from the deformity only half as often as

the male, but her male relatives are far more frequently affected. The female relatives of the male patients seem very unlikely to acquire the deformity.

Metatarsus varus occurred infrequently in these families and talipes calcaneo-valgus only once (Table III).

Other points which could be significant were noted:

Consanguinity—There were no first cousin marriages in the parents of the 1940–61 group.

Age of parents—The ages of the mothers were compared with those shown in the Registrar General's national figures (Table V). There was no significant difference between the ages of the mothers of affected children and the expected ages of the mothers of normal children.

The difference between the mother's and father's ages was also noted. In this country, on average, the father is 2·3 years older than the mother (Penrose 1955). The figures for the parents of children with talipes equinovarus show a difference of 2·8 years (Table VI).

Birth order—The position of the deformed child in the family was noted. When the figures were compared with those of the Registrar General no significant excess among first-born or subsequent children was found (Table VII).

Twin studies—This series is too small to include many twin pairs. Details are recorded in Table VIII. Here the work of Idelberger (1939) is invaluable. He examined 174 pairs of twins with talipes equinovarus, the comparison being between identical (monozygotic) twins, and those of like sex, but not identical (dizygotic) (Table IX).

In other words, one in three of identical twin pairs both have talipes equinovarus, as opposed to only one in thirty-five of non-identical twin pairs. The latter figure (2·9 per cent) is exactly the same as the Exeter finding for the incidence in non-twin siblings.

Thus, there is no recognisable pattern of inheritance to indicate a dominant, recessive or sex-linked gene. The chances of any individual having this deformity are approximately 1 in 800. If one child in a family is affected, the risk that a second will have it is 1 in 35. The risk to the second of identical twins is 1 in 3.

Clearly, the more alike the genetic constitution, the greater the risk—and yet the cause cannot be solely genetic since both identical twins do not invariably have the deformity. In addition to the genetic factor we must look further, therefore, for an environmental factor, which, acting in the uterus, can affect one twin without the other.

INHERITANCE OF TALIPES CALCNEO-VALGUS AND METATARSUS VARUS

Material—Children with these deformities are not always referred to hospital for treatment. Obviously many cases must go unnoticed or ignored. There was a marked rise (not apparent in the case of talipes equinovarus) in the numbers attending hospital after the start of the National Health Service. Hence the period for review is from 1948 to 1961. Only those brought in the first few months of life before walking is established are included, since the deformity usually disappears and a retrospective diagnosis is not possible. Seventy-four children with talipes calcaneo-valgus and seventy-two with metatarsus varus were traced, but only fifty-three of each were seen personally (Tables X and XI). They were, of course, far more difficult to trace than were the patients with talipes equinovarus since many of them only attended hospital for a few months in infancy. The figures are less valuable because of the necessarily incomplete nature of the survey.

Incidence of talipes calcaneo-valgus and metatarsus varus in the general population—As in talipes equinovarus, the incidence is approximately one per thousand live births (Tables XII and XIII).

Sex ratio—Both talipes calcaneo-valgus and metatarsus varus are commoner in girls than in boys. The ratio of male to female is 0·61 to 1 in talipes calcaneo-valgus, and 0·76 to 1 in metatarsus varus.

First degree relatives with talipes calcaneo-valgus and metatarsus varus—Approximately 4.5 per cent of siblings are affected—as against one per thousand cases in the general population (Tables XIV and XV).

Consanguinity—No first cousin marriages were noted.

Age of parents—In the case of talipes calcaneo-valgus there is a significant excess among young mothers (Table XVI). This is perhaps secondary to the birth order effect. There is no evidence of any connection in metatarsus varus (Table XVII). The difference in age between parents is not markedly different from that expected in normal families (Tables XVIII and XIX).

Birth order—In the case of talipes calcaneo-valgus there is a highly significant excess among first-born children, but there is no evidence of any connection in metatarsus varus (Tables XX and XXI).

Twins—Investigations for one twin pair with each deformity are shown in Table XXII.

The chances of any individual having either deformity is approximately 1 in 1,000. If one child is affected, the risk to a second in a family is 1 in 20. Again, we seem to be dealing with a condition which shows no clear pattern of inheritance, but must be partly genetic in origin. The fact that talipes calcaneo-valgus occurs especially often in first-born children rather suggests that the environmental factor is compression. This might well operate in the primigravida with a small "tight" uterus and good abdominal muscles.

FIG. 3

FIG. 4

FIG. 5

Examples of joint laxity in three patients with talipes equinovarus.

CONGENITAL ABNORMALITIES ASSOCIATED WITH TALIPES EQUINOVARUS, TALIPES CALCANEO-VALGUS AND METATARSUS VARUS

It was thought that some more precise information about the nature of the genetic factor in club foot might be obtained from studying the associated deformities in the patients and their relatives (Table XXIII).

In grouping these defects we have followed McKusick's (1960) theories. He suggested that some generalised congenital disorders (osteogenesis imperfecta, Marfan's syndrome, Ehlers Danlos syndrome, gargoylism) could be ascribed to a developmental defect in the formation of collagen producing a variety of disorders, not only of the skeleton, but also of soft connective tissue anywhere in the body—for example hyperextensible joints, herniae, aortic aneurysm and so on. In connection with the present study, it was noted that McKusick observed that these generalised "connective tissue" disorders might be associated with club foot. It was decided, therefore, to approach the question from the other angle, and to try and establish whether or not in the present club foot survey there were any associated soft tissue disturbances. The investigation shows that this is indeed so.

Joint laxity—Hyperextensible joints are a common component of the generalised disorders studied by McKusick, though in gargoylism the joints are noted to be unduly stiff. One is reminded of arthrogryposis, in which in one individual some of the joints are unnaturally stiff, and those which escape this may be abnormally lax. It seems reasonable to think that one

type of mechanism may be at fault, producing a soft tissue disorder giving rise either to abnormally lax or to abnormally fixed joints.

Familial joint laxity without other defect is a condition which appears often to be caused by a dominant gene (Carter and Sweetnam 1960). There have been occasional reports in the literature of an association between familial joint laxity and congenital club foot. Key (1927) described a family in which joint laxity appeared in a father and four sons. Three of these sons had talipes equinovarus, and one had talipes equinus. Kite (1930) noted one case of "congenital relaxed joints" in his series of 100 patients with talipes equinovarus. Carter and Sweetnam (1958) reported one male with joint laxity and bilateral talipes equinovarus in their family with recurrent dislocation of the patella.

In the present survey undue mobility of joints has been looked for in every patient seen who was over the age of two years (Figs. 3 to 5). An arbitrary minimum of three sites in the body was taken as significant. It is realised that the method of testing is imprecise, but since

each individual was examined by the same person, who also examined 200 control patients, it is felt that the figures are worth including. The controls were drawn from hospital staff, visitors and patients admitted because of injuries and were comparable with the club foot patients as regards age and sex.

FIG. 6

FIG. 7

Abnormalities associated with talipes equinovarus. Figure 6—Boy with right talipes equinovarus and left genu recurvatum. Figure 7—Girl with bilateral talipes equinovarus and flexion contracture of the left knee.

RESULTS

Two main groups of associated abnormalities are immediately apparent: 1) Minor defects of connective tissue. These are conditions such as herniae, recurrent dislocation of patella, flexion contracture of joints and genu recurvatum as an isolated phenomenon*, as well as generalised joint laxity (Figs. 6 and 7). 2) Other deformities at the periphery of the limbs (Tables XXIV to XXIX).

Minor defects of connective tissue—In all, 17 to 18 per cent of patients with talipes equinovarus and of patients with talipes calcaneo-valgus have an abnormality of

this nature, and some have more than one manifestation. The patients with metatarsus varus do not show the same tendency. Hernia is a common congenital abnormality affecting about nine in 1,000 males in the normal population (Knox 1959), but in the present survey of males with talipes equinovarus, the figure was 7 per cent.

Another point, remarked on by Browne (1955), is the association of congenital dislocation of the hip with talipes calcaneo-valgus. In this series 4 to 5 per cent of patients with talipes calcaneo-valgus had congenital dislocation of the hip, whereas only one of the children with talipes equinovarus was similarly affected.

Peripheral deformities—Four to five per cent of all patients showed some other deformity at the periphery of the limbs—ring constrictions, syndactyly, missing or extra fingers and toes and so on. In general the two groups did not overlap. The significance of this is not

* Not, of course, including patients with an equinus deformity of the foot, since hyperextension of the knee could be secondary to this.

understood, but clearly these deformities may sometimes be a part of a more generalised disturbance, although they often occur singly.

ASSOCIATED CONGENITAL ABNORMALITIES IN FIRST DEGREE RELATIVES

Enquiry was made regarding other congenital abnormalities in the immediate family of patients. Only 1 to 2 per cent of the relatives were affected, but there was the same tendency towards a "connective tissue" disturbance, or to some other deformity at the periphery of the limb (Table XXX).

DISCUSSION

The intention of this paper has been to marshal a few facts concerning the inheritance of club foot rather than to attempt new theories. It is suggested only that: 1) the genetic factor relates to the formation of connective tissue, which is in some way defective; 2) because of this the feet are abnormally mobile at some stage in intra-uterine development, and are easily pushed out of position; 3) the "environmental" (intra-uterine) factor may well be related to abnormal pressure or position in the uterus. It could be that the deforming force is transient, and pushes the abnormally mobile feet into equinovarus at a vulnerable time in their development. They then become "set" in this position, and subsequent muscle development is inhibited and inadequate to overcome the deformity. Carried further this could result in arthrogryposis.

If a deforming force were to be exerted later in foetal life, it could account for the less severe calcaneo-valgus deformity and "postural" talipes equinovarus, both of which are easily correctible and do not tend to recur.

There is no evidence in this survey to suggest that patients with some other connective tissue disturbance are any more difficult to treat than the otherwise normal individuals; neither is there any evidence to indicate that the deformity is of greater severity when more than one member of a family is affected.

SUMMARY

1. The family history of, and associated congenital abnormalities in, patients with talipes equinovarus, talipes calcaneo-valgus and metatarsus varus living in Devonshire has been studied.
2. The chances of any individual having one of these deformities is approximately one per 1,000 in each case.
3. If one child in a family has the deformity, the chances of a second having it are one in thirty-five for talipes equinovarus and one in twenty for talipes calcaneo-valgus and metatarsus varus.
4. The male relatives of the female patients with talipes equinovarus are at particular risk.
5. It is suggested that the cause of club foot is partly genetic and partly environmental, from a factor acting on the foetus in the uterus.
6. The classification of associated congenital abnormalities leads to the suggestion that the genetic factor in talipes equinovarus and talipes calcaneo-valgus relates to defective formation of connective tissue.

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TABLES

TABLE I
TALIPES EQUINOVARUS. MATERIAL

Number of case notes available for period 1940-61	163
Number of patients traced (133 seen personally; eleven questionnaires received)	144
Untraced	13
Incomplete information (foster children)	2
Refused cooperation	4

TABLE II
INCIDENCE OF TALIPES EQUINOVARUS
PER 1,000 LIVE BIRTHS

Males.	1.62
Females	0.80
Total	1.24

TABLE III
TALIPES EQUINOVARUS. FIRST DEGREE RELATIVES WITH THE SAME DEFORMITY

	Father	Mother	Brothers	Sisters
Male index . 97*	1 of 97	None of 97	4 of 115 (1 with metatarsus varus)	None of 90 (1 with metatarsus varus)
Female index 47†	3 of 47	None of 47 (1 with talipes calcaneo-valgus)	2 of 33	2 of 34 (1 with metatarsus varus)
Total . . 144	4 of 144	None of 144	6 of 148	2 of 124
8 of 272 siblings affected (2.9 per cent)				

* Includes one brother pair. Relatives counted twice.

† Includes one sister pair. Relatives counted twice.

TABLE IV
TALIPES EQUINOVARUS. MALE AND FEMALE FIRST DEGREE RELATIVES WITH TALIPES EQUINOVARUS

97 male index patients .	5 of 212 male relatives affected 2.3 per cent 1 in 42 None of 187 female relatives affected —
47 female index patients	5 of 80 male relatives affected . 6.2 per cent 1 in 16 2 of 81 female relatives affected 2.5 per cent 1 in 40

TABLE V
TALIPES EQUINOVARUS. AGES OF MOTHERS (DEVON BIRTHS 1948 TO 1960)
COMPARED WITH REGISTRAR GENERAL'S FIGURES

	Age of mother (years)			
	15-24	25-29	30-34	35 and over
Observed	29	33	22	10
Expected	31.54	30.64	19.01	12.82
$\chi^2_{(3)} = 1.48. p > .10. \text{ Not significant}$				

TABLE VI
TALIPES EQUINOVARUS
DIFFERENCE BETWEEN MOTHERS' AND FATHERS' AGES

Average age of mother . . .	26.0 years
Average age of father . . .	28.8 years
Difference	2.8 years

TABLE VII
TALIPES EQUINOVARUS. BIRTH ORDER (DEVON BIRTHS 1948 TO 1960)
COMPARED WITH NATIONAL FIGURES

	Birth order			
	1	2	3	4
Observed	45	23	17	9
Expected	37.19	28.75	14.28	13.78
$\chi^2_{(3)} = 4.97. p > .10. \text{ Not significant}$				

TABLE VIII
TWIN PAIRS OF LIKE SEX, ONE OF EACH PAIR WITH TALIPES EQUINOVARUS

	ABO	Rh genotype	MN	Kell	P	Fy ^a
Twin 1 (male: talipes equinovarus)	A ₁	R ₁ R ₂	M	Negative	Positive	Positive
Twin 2 (male: normal)	A ₁	R ₁ r	M	Negative	Positive	Positive
These twins differ in Rh genotype and are therefore non-identical						
Twin 3 (male: talipes equinovarus)	A ₁	CDe/cDE	MN	Negative	Positive	Negative
Twin 4 (male: normal)	A ₁	CDe/cDE	MN	Negative	Positive	Negative
Both twins possess the same red cell antigens, and are almost certainly uniovular						

TABLE IX
IDELBERGER'S TWIN STUDIES

Twins	Concordance	Discordance	Total	Concordance (per cent)
Identical	13	27	40	32.5
Fraternal	4	130	134	2.9

TABLE X
TALIPES CALCANEAL VALGUS. MATERIAL

Number of case sheets available for 1948-61	100
Number of patients traced (53 seen personally, 21 questionnaires received)	74

TABLE XI
METATARSUS VARUS. MATERIAL

Number of case sheets available for 1948-61	111
Number of patients traced (53 seen personally, 19 questionnaires received)	72

TABLE XII
INCIDENCE OF TALIPES CALCANEO-VALGUS
PER 1,000 LIVE BIRTHS

Males	.	.	.	.	0.87
Females	.	.	.	.	1.33
Total	.	.	.	.	1.09

TABLE XIII
INCIDENCE OF METATARSUS VARUS PER
1,000 LIVE BIRTHS

Males	.	.	.	.	1.02
Females	.	.	.	.	1.42
Total	.	.	.	.	1.21

TABLE XIV
TALIPES CALCANEO-VALGUS. FIRST DEGREE RELATIVES WITH THE SAME DEFORMITY

		Father	Mother	Brothers	Sisters
Male index	26	None of 26	None of 26	None of 24	1 of 12
Female index	48*	None of 48	None of 48	1 of 20	2 of 30
Total	74	None of 74	None of 74	1 of 44	3 of 42
4 of 86 siblings affected (4.6 per cent)					

* Includes one sister pair. Relatives counted twice.

TABLE XV
METATARSUS VARUS. FIRST DEGREE RELATIVES WITH THE SAME DEFORMITY

		Father	Mother	Brothers	Sisters
Male index	30	None of 30	None of 30	None of 26 (1 with talipes equinovarus)	2 of 22
Female index	42	None of 42	None of 42	2 of 35 (2 with talipes equinovarus)	1 of 30 (metatarsus varus one foot; talipes equinovarus the other)
Total	72*	None of 72	None of 72	2 of 61 (3 with talipes equinovarus)	3 of 52 (1 with metatarsus varus one foot; talipes equinovarus the other)
5 of 113 siblings affected (4.4 per cent)					

* one brother/sister pair. Relatives counted twice.

TABLE XVI
TALIPES CALCaneo-VALGUS. AGES OF MOTHERS (DEVON BIRTHS
1948 TO 1960) COMPARED WITH REGISTRAR GENERAL'S FIGURES

	Age of mother (years)			
	15-24	25-29	30-34	35 and over
Observed	32	13	10	8
Expected	21.14	20.53	12.74	8.59
$\chi^2_{(3)} = 8.97$. $.05 > p > .025$. Significant excess among young mothers				

TABLE XVII
METATARSUS VARUS. AGES OF MOTHERS (DEVON BIRTHS 1948 TO 1960)
COMPARED WITH REGISTRAR GENERAL'S FIGURES

	Age of mother (years)			
	15-24	25-29	30-34	35 and over
Observed	23	21	10	5
Expected	19.80	19.23	11.93	8.05
$\chi^2_{(3)} = 2.15$. $p > .10$. Not significant				

TABLE XVIII
TALIPES CALCaneo-VALGUS
DIFFERENCE BETWEEN MOTHERS' AND FATHERS' AGES

Average age of mother	.	.	26.4 years
Average age of father	.	.	30.3 years
Difference	.	.	3.9 years

TABLE XIX
METATARSUS VARUS
DIFFERENCE BETWEEN MOTHERS' AND FATHERS' AGES

Average age of mother	.	.	26.9 years
Average age of father	.	.	29.4 years
Difference	.	.	2.5 years

TABLE XX
TALIPES CALCANEAL-VALGUS. BIRTH ORDER (DEVON BIRTHS 1948 TO 1960)
COMPARED WITH NATIONAL FIGURES

	Birth order			
	1	2	3	4
Observed	39	8	5	11
Expected	24.92	19.27	9.57	9.24
$\chi^2_{(3)} = 17.06. p < .001. \text{ Highly significant excess of cases among first-born}$				

TABLE XXI
METATARSUS VARUS. BIRTH ORDER (DEVON BIRTHS 1948 TO 1960)
COMPARED WITH NATIONAL FIGURES

	Birth order			
	1	2	3	4
Observed	21	25	13	—
Expected	23.34	18.04	17.62	—
$\chi^2_{(3)} = 4.13. p > .10. \text{ Not significant}$				

TABLE XXII
TWINs OF LIKE SEX, ONE OF EACH WITH TALIPES CALCANEAL-VALGUS

	ABO	Rh genotype	MN	Kell	P	Fy ^a
Twin 5 (female: talipes calcaneal-valgus)	A(A ₂)	CDe/c(de)	MN	Positive	Positive	Negative
Twin 6 (female: normal)	A(A ₂)	CDe/c(de)	MN	Negative	Positive	Negative
These twins differ in the Kell factor and are therefore non-identical						

TABLE XXIII
PATIENTS WITH ASSOCIATED MALFORMATIONS

	Seen personally	Questionnaire received	Total number traced	Information from medical records only
Talipes equinovarus (340)	175	18	193	147
Talipes calcaneal-valgus (157)	53	21	74	83
Metatarsus varus (138)	53	19	72	66
Total (635)	281	58	339	296*

* Forty-nine of these were evacuees and were treated for only a short time during the war. No attempt has been made to trace them.

TABLE XXIV
TALIPES EQUINOVARUS—ASSOCIATED ABNORMALITIES

Case number	Sex	Joint laxity	Other minor connective tissue deformity	Deformity at periphery of limb	Other abnormalities	Congenital deformity in first degree relatives
Patients aged 20 and over						
1	Male	Present	—	—	—	—
2	Male	—	—	Congenital amputation fingers right hand. Ring constriction left fifth finger. Syndactyly right toes	—	—
3	Male	Present	—	—	Mental defective	—
4	Male	Present	Bilateral inguinal herniae, umbilical hernia, genu recurvatum, recurrent dislocation patella	—	Pigeon chest	Sister: pigeon chest
5	Male	—	Genu recurvatum	—	Bilateral slipped upper femoral epiphyses (16 years)	Brother: talipes equinovarus
6	Female	Present	—	—	—	—
7	Female	Present	Bilateral inguinal herniae	—	—	—
8	Male	Present	—	—	—	—
9	Male	—	—	—	Structural scoliosis	—
10	Female	—	Recurrent dislocation of patella	—	—	—
11	Female	Present	—	—	—	—
12	Male	—	Bilateral inguinal herniae, umbilical hernia	—	—	Brother: talipes equinovarus
13	Male	Present	Recurrent dislocation of patella	—	—	—
14	Male	—	Right inguinal hernia	—	—	Daughter: talipes equinovarus
15	Female	Present	Umbilical hernia	—	—	—
16	Male	—	Right inguinal hernia	—	—	—
Patients aged 10 to 19						
17	Male	Present	—	—	—	—
18	Male	—	—	—	Pigeon chest. Micronathos	—
19	Female	—	—	Absent left fifth ray	—	—
20	Male	—	—	Aplasia of fingers	Möbius syndrome	—

TABLE XXIV—*continued*
TALIPES EQUINOVARUS—ASSOCIATED ABNORMALITIES

Case number	Sex	Joint laxity	Other minor connective tissue deformity	Deformity at periphery of limb	Other abnormalities	Congenital deformity in first degree relatives
Patients aged 10 to 19—<i>continued</i>						
21	Female	—	Recurrent dislocation of patella	—	—	Sister: talipes equinovarus
22	Male	—	Umbilical hernia	—	—	—
23	Male	—	—	—	Mental defective	—
24	Male	Present	—	—	—	—
25	Female	—	—	Ring constrictions left second, third and fourth toes	—	—
26	Female	Present	—	—	—	—
27	Female	—	Flexion contracture left knee	Double nail ring finger. Ring constriction right hallux	—	—
28	Male	Present	—	—	—	Brother: congenital deafness
Patients under 10 years old						
29	Male	—	Right inguinal hernia and undescended testicle	—	Pigeon chest	—
30	Male	Present	Left inguinal hernia, umbilical hernia	—	—	—
31	Male	Present	Right inguinal hernia	—	—	—
32	Male	Present	—	—	—	—
33	Female	Present	—	—	—	—
34	Female	Present	Flexion contracture of elbows	—	Achondroplasia	—
35	Male	—	Umbilical hernia	—	Mongol	Father: talipes equinovarus
36	Female	—	—	Diminutive right thumb	Absent right ear	Sister: diminutive thumb
37	Male	—	Congenital dislocation of hip	—	—	—
38	Male	Present	—	—	—	—
39	Male	—	Interventricular septal defect	—	—	Father: syndactyly toes
40	Male	Present	—	—	—	—
41	Female	—	—	Aplasia left toe tips	—	—
42	Male	Present	—	—	—	—
Total 42 of 175 (25 per cent)		Joint laxity 19 of 154 (children under 2 years excluded) 12 per cent. (Control series 4 of 196. $p < .001$. Highly significant excess among patients with talipes equinovarus.) Total 33 of 175 have joint laxity or other connective tissue disorder (19 per cent)				

TABLE XXV
TALIPES EQUINOVARUS—ASSOCIATED ABNORMALITIES
 Information obtained from medical records only

Case number	Sex	
1	Male	Recurrent dislocation of patella
2	Male	Syndactyly index/middle finger
3	Male	Mentally defective
4	Male	Bilateral genu recurvatum
5	Male	Mentally defective
6	Male	Structural scoliosis
7	Female	Mentally defective
8	Male	Mentally defective
9	Female	Mongolism
10	Male	Pierre Robin syndrome
11	Male	Mentally defective
12	Male	Mentally defective
13	Male	Mentally defective
14	Male	Absent fourth and fifth rays right foot. (Left talipes equinovarus only)
15	Female	Gargoylism
16	Male	Mentally defective
17	Male	Syndactyly second and third toes
18	Male	Blind right eye. ?Congenital heart lesion. (Died age 4 months)
Total 18 of 165 (10 per cent)		

TABLE XXVI
TALIPES CALCANEAL-VALGUS—ASSOCIATED ABNORMALITIES

Case number	Sex	Joint laxity	Other minor connective tissue deformity	Congenital dislocation of hip	Deformity at periphery of limb	Other abnormality	Congenital deformity in first degree relatives
1	Male	Present	Right inguinal hernia, congenital dislocation of knee	Present	—	—	—
2	Female	—	Right inguinal hernia	—	—	—	—
3	Male	—	—	—	Six fingers and toes	Mental defective	—
4	Female	—	—	—	Supernumerary thumb	—	Mother: supernumerary thumb
5	Female	Present	Recurrent dislocation of patella	—	—	—	Mother: congenital dislocation radial heads
6	Female	—	—	Present	—	—	—
7	Female	—	—	Present	—	—	—
8	Male	—	Left inguinal hernia	—	—	—	—
9	Male	Present	Bilateral inguinal herniae and hydroceles	—	—	—	—
10	Male	—	—	—	Syndactyly right first and second toes, absent fourth toe. Syndactyly left second and third toes, aplasia fourth toe	—	—
Sisters							
11	Female	Present	—	—	—	—	—
12	Female	Present	—	—	—	—	—
Total 12 of 53 (23 per cent)		Joint laxity 5 of 35 (children under 2 years excluded) 14 per cent. (Control series 4 of 196. $p < .05$. Significant excess among patients with talipes calcaneo- valgus.) Total 7 of 53 have joint laxity or other connective tissue disorder (13 per cent). Including congenital dislocation of hip 9 of 53 (17 per cent)					

TABLE XXVII
TALIPES CALCaneo-VALGUS—ASSOCIATED ABNORMALITIES
 Information obtained from medical records only

Case number	Sex	
1	Male	Aplasia thumb, absent ears
2	Male	Bilateral congenital dislocation of hips
3	Female	Bilateral congenital dislocation of hips
4	Male	"Resistant rickets"
5	Male	Right inguinal hernia
6	Male	Congenital dislocation right hip, flexion contracture both knees
7	Male	Flexion contracture both knees
8	Female	Cleft palate, exostosis occipital bone
9	Female	? Congenital heart lesion. (Died age 4 months)
10	Male	Bilateral synostosis radio-ulnar joints
11	Male	Absent fibula and outer three rays
12	Female	Marfan's syndrome. Bilateral congenital dislocation of hips
Total 12 of 104 (10 per cent)		

TABLE XXVIII
METATARSUS VARUS—ASSOCIATED ABNORMALITIES

Case number	Sex	Joint laxity	Other minor connective tissue deformity	Deformity at periphery of limb	Other abnormality	Congenital deformity in first degree relatives
1	Female	—	—	Aplasia right thumb	—	—
2	Male	—	—	Aplasia right fingers	—	—
3	Male	Present	—	—	—	—
4	Female	Present	—	—	—	—
5	Female	Present	—	—	—	—
6	Female	—	—	Syndactyly right middle/index finger. Syndactyly right first, second and third toes	—	—
7	Male	—	Congenital heart lesion — ? interventricular septal defect	—	—	—
8	Male	—	Flexion contracture left elbow	Absent left ulna, middle and ring fingers	Micronathos. Cleft palate	—
Total 8 of 53 (15 per cent)		Joint laxity 3 of 44 (children under 2 years excluded) 7 per cent. (Control series 4 of 196. $p > .05$. No evidence of connection.) Total 5 of 53 have joint laxity or other connective tissue disorder (9 per cent)				

TABLE XXIX
METATARSUS VARUS—ASSOCIATED ABNORMALITIES
Information obtained from medical records only

Case number	Sex	
1	Male	Bilateral genu recurvatum
2	Male	Umbilical hernia
Total 2 of 85 (2 per cent)		

TABLE XXX
ASSOCIATED DEFORMITIES IN FIRST DEGREE RELATIVES
(including those shown in Tables XXIV to XXIX)

	Father	Mother	Brothers	Sisters	Sons	Daughters
Talipes equinovarus						
Male index . 134	1 hare lip. 1 syndactyly toes	2 congenital dislocation of hip	(182) 1 hare lip. 1 hare lip/ cleft palate	(139) 1 syndactyly toes*	(18) 1 cleft palate*	(16)
Female index 59	—	—	(53)	(56) 1 diminutive thumb	(8)	(5)
Total . . 193						
Talipes calcaneo-valgus						
Male index . 26	—	—	(24) 1 congenital dislocation of hip	(12)	—	—
Female index 48	—	1 congenital dislocation radial heads.* 1 bilateral flexion contracture medial three fingers.* 1 supernumerary thumb*	(35) 1 coarctation of aorta and patent ductus.* 1 flexion contracture middle and ring fingers*	(39) 1 recurrent subluxation of shoulder	—	—
Total . . 74						
Metatarsus varus						
Male index . 30	—	—	(26) 1 hydrocephalus	(22)	—	—
Female index 42	—	—	(35) 1 congenital hemiplegia.* 1 arrested hydrocephalus and congenital deafness*	(30) 1 metatarsus primus varus. 1 congenital heart lesion	—	—
Total . . 72						

* Confirmed by personal observation or adequate medical records.