CONGENITAL SPLIT FOOT (LOBSTER CLAW)
AND TRIPHALANGEAL THUMB

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Split foot (lobster claw) is not a common congenital anomaly. Its occurrence in association with triphalangeal thumb, an even rarer abnormality, justifies the description of the pedigree of a family in which these deformities occurred.

CLINICAL MATERIAL

Although the pedigree (Fig. 1) is described over three generations it has not been possible to examine all members of the family because of difficulty in locating them and, in some cases, due to lack of cooperation. The latter factor reflects the embarrassment which the lobster clawing can cause to the sufferer.

![Pedigree Diagram]

Little is known of the first generation of this family other than by repute. The subject in Case II, 4 could not recall any deformity of hands or feet in this generation and certainly none in her own parents. There was no evidence of consanguinity throughout.

Cases II, 1 and 2—These were healthy undeformed males in whose children the congenital abnormalities had not occurred.

Case II, 3—This individual, a female, refused examination because of the embarrassment that her deformities caused. She admitted that she had marked splitting of the forefeet and that her thumbs were crooked. Although married for several years she and her husband had refrained from having children because of their awareness of the familial disorder.

Case II, 4—This patient, a forty-four-year-old woman, was examined in detail. There were deep clefts of the forefoot. Both great toes showed marked valgus deformity. The lateral two toes of each foot showed syndactyly (Fig. 2).

Radiographs of the left foot showed that the calcaneus, talus, cuboid and navicular bones were normal (Fig. 3). The deep cleft was due to total absence of the second ray and hypoplasia of the third metatarsal with absence of its phalanges. The fourth and fifth metatarsals were present but shared a conjoined proximal phalanx. The middle and distal phalanges were
normal. Although the first metatarsal and its proximal phalanx were normal, the distal phalanx had a boss of bone on the medial aspect near its base, suggestive of an attempt at replication. In the radiographs of the right foot the hindfoot seemed normal although naviculo-cuboid fusion could not be excluded (Fig. 3). In the forefoot the fourth and fifth metatarsals were present but only the fourth had a group of three phalanges. These deviated medially. The first metatarsal was broad and squat and the fact that it was completely fused to the intermediate cuneiform suggested that it represented fused first and second metatarsals. At the medial side of its neck there was a small ossicle. Although this may have represented a sesamoid, it may have been a first metatarsal element. The two phalanges of the great toe were normal except for a little boss of bone on the medial side of the base of the distal one.
Again, this may have represented an attempt at replication. This patient had only a mild ache on the dorsum of both feet. For comfort she wore broad surgical shoes.

Clinical examination of the left hand revealed a complete extra phalanx in the thumb with marked ulnar deviation of the terminal phalanx (Fig. 4). Radiographs (Fig. 5) confirmed the presence of an additional phalanx in the thumb. The nail of the index finger was rather thin and atrophic and had always been so, but its skeleton and that of the other fingers were normal.

In the right hand the thumb had two phalanges, yet the terminal one deviated ulnarwards (Fig. 4). There was no evidence that there had ever been a thumb nail. Radiographs (Fig. 5) showed a boss of bone on the radial aspect of the terminal phalanx near its tip. This may have represented an attempt at replication. The remaining fingers and their metacarpals were normal.
This patient had no treatment for her hand deformities. Apart from their unsightliness she had not been troubled by them.

Case II, 5—This patient, a thirty-nine-year-old woman, lived in Northern Ireland. She was not examined personally but her general practitioner described her deformities and submitted radiographs of her hands and feet.

Radiographs of the right foot (Fig. 6) showed no cleavage in the hindfoot but the cuboid and the lateral cuneiform appeared fused. There was an accessory navicular bone. The forefoot was deeply cleft. The fifth metatarsal and its phalanges were normal. Near the base of this metatarsal there was an osseous mass representing metatarsal elements. The base of
the first metatarsal articulated with the medial and intermediate cuneiforms suggesting conjoined adjacent metatarsals. The phalanges of the hallux were normal in number and shape but valgus displacement was obvious.

In the left foot (Fig. 6) the hindfoot bones were normal except for cuneiform fusion and the presence of an accessory navicular. The fifth metatarsal and its phalanges were normal. Abutting its base was an irregular osseous mass fused to the cuneiforms. The first metatarsal and its phalanges, too, were normal but valgus deviation was noted at the distal segment.

In the left hand the thumb was absent and the index finger was grossly abnormal and distorted. In the right hand the radial digit (not a thumb) was very short; the skin and fat were much thickened over it. The ring and middle fingers were completely webbed. Apart from a mild flexion deformity the little finger was normal.

Radiographs (Fig. 7) showed that on the left side there was no thumb. The three ulnar rays were normal. The index metacarpal was well developed. The distal end of the proximal phalanx was bifid. From its ulnar horn there hung two hypoplastic phalanges. It may be that the radial horn was the extreme rudiment of a thumb. In the right hand the skeletal framework of the three ulnar rays was normal despite syndactyly of the middle and ring fingers. The index metacarpal was normal but was surmounted by a stunted phalangeal element whose distal end was bifid—remnants perhaps of phalanges of index finger and thumb.

This patient had married rather late in life and had no children.

Cases II, 5, 6 and 7—These patients, two men and a woman, were reputed to be normal in all respects, as were their offspring.

The third generation to be described comprised the offspring of Case II, 4 and a normal father in whose family there had been no history of congenital abnormality.

Case III, 1—This patient, a twenty-one-year-old male, was examined in detail. He had deep forefoot clefts but the great toes were not displaced into valgus.

On the right side, radiographs revealed a normal hindfoot but the cuboid and lateral cuneiform were fused (Fig. 8). There was a large lateral metatarsal with a medial boss near its base. This metatarsal may have represented fused third, fourth and fifth metatarsals. It articulated with three normal phalanges. The medial metatarsal was short and squat, articulating with two cuneiforms, thus representing fused first and second metatarsals. Its metatarso-phalangeal joint already showed osteoarthritic changes. There were two complete phalanges to the great toe but at the lateral side of the base of the distal one there was a separate ossicle, articulating with both phalanges. This may have represented an attempt at replication.

On the left side, radiographs showed a normal hindfoot (Fig. 8). The two lateral rays were intact. The third metatarsal was hypoplastic. It had no phalanges. The first metatarsal articulated with two cuneiforms and may therefore have represented the medial two metatarsals. The great toes had two phalanges but, again, an ossicle was present at the lateral side of the base of the terminal phalanx articulating with both.

A previous unsuccessful attempt had been made in infancy to close the cleft in the left foot. Now he had no symptoms in his feet. He was an active football player.

In both thumbs he had an additional interphalangeal joint with associated skin creases. The distal segment in each deviated ulnawards. The nails were normal. All other digits were healthy. Radiographs of the hands (Fig. 9) showed an additional middle phalanx. This was very short and markedly wedge-shaped, particularly on the left side. Function of the hands was normal.

Case III, 2—This patient, an eighteen-year-old boy, when seen in June 1969, complained of painful deformed feet (Fig. 10). Clearly, the prominent bilateral hallux valgus, with broadening of the forefoot, was making the wearing of normal shoes difficult. Pain was localised to a bursa overlying the first metatarso-phalangeal joint.

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In the left foot there was a deep forefoot cleft. The first metatarsal was broader than usual but it was surmounted by two normal phalanges. On the lateral side of the foot there were two metatarsal rays but their toes showed syndactyly.

![Fig. 8](image1)

![Fig. 9](image2)

Case III, I. Figure 8—Radiographs of the feet. Figure 9—Radiographs of the hands in the same patient.

On the right side the deformity was similar but the cleft seemed broader and more shallow, being partially filled by an irregular bony mass.

The right thumb showed an extra set of volar joint creases and an additional interphalangeal joint (Figs. 11 and 12). The terminal segment of the thumb deviated ulnarily. Other digits were normal but the nail of the left index finger was somewhat dystrophic with an incomplete
median split. Radiographs showed no abnormality in the bones of the left hand (Fig. 13). On the right, however, there was an additional slightly wedge-shaped bone between the two phalanges of the thumb—the third phalanx. Hand function was normal.

This boy was admitted to hospital for surgical correction of the feet. Suitable skin flaps were raised around the margins and at the base of the cleft in the right foot. Metatarsal remnants were excised. A basal osteotomy of the first metatarsal was performed to narrow the cleft and to ease tension in the sutured skin flaps. A similar procedure was done on the left foot one week later. The feet were immobilised in plaster boots for four weeks. Primary skin healing occurred. Within two months the patient was wearing shoes of average breadth and had returned to full activity. He was well pleased with the functional and cosmetic result achieved (Figs. 14 and 15).

**Cases III, 3, 4 and 5**—A boy aged sixteen years, a girl of fifteen years and a boy of nine years were found to be normal in all respects.

**Case III, 6**—This seven-year-old girl had normal feet but in both thumbs there was an additional interphalangeal joint, with associated skin creases. The distal thumb segments were deviated ulnarwards. Radiographs (Fig. 16) confirmed the presence of additional middle phalanges in both thumbs. They were stunted and somewhat wedge-shaped. The remaining digits were healthy in all respects. Hand function was excellent.

**DISCUSSION**

**Split foot**—Barsky (1964) in describing cleft hand, or ectrodactyly, ascribed to Lange (1936) the recognition of two types—the one, atypical, was a unilateral deformity in which there was no associated foot deformity or evidence of familial inheritance; the other, typical, was always a bilateral deformity. Often, too, the feet were split; a family history of deformity in this type was invariable. Clearly, if such a classification is applicable to split foot (lobster claw) then the family herein described can be reckoned to show the typical deformity.

Although bilateral lobster clawing of the feet may be an isolated deformity, numerous associated skeletal and soft-tissue abnormalities have been described. Walker and Clodius (1963) noted its occurrence with cleft lip and palate. It has been claimed by Potter and Nadelhoffer (1947) that split foot is frequently associated with abnormalities of the hand. These include lobster clawing itself (Stiles and Pickard 1943), reduction in number and size of the phalanges (Potter and Nadelhoffer 1947), duplication of the thumb and other digits (Soeur 1959), and syndactyly and polydactyly (Lewis and Embleton 1908). Duplication of toes in lobster claw feet was noted by Cowan (1965). An associated triphalangeal but hypoplastic thumb was described in one case by Lewis and Embleton (1908) in their large series of split foot. Stiles and Pickard (1943), although not claiming triphalangism, did describe five cases in a family with split foot deformities in which the thumb was elongated and its distal segment deviated ulnarwards.

Most modern authors (Stiles and Pickard 1943, Potter and Nadelhoffer 1947, Barsky 1964) believed that the lobster claw deformity was not sex linked, sex influenced or sex limited in its inheritance but that the defects of feet and the numerous hand deformities were caused by the effects of a single dominant gene in which expression was incomplete. In the present
Case III, 2. Figure 11—Dorsal view of the hands. Figure 12—Palmar view. Note the additional skin creases on the thumbs and the ulnar deviation of their terminal segments. Figure 13—Radiographs of the hands.
family it would seem to be so too. Although expression is somewhat incomplete, that is, three of eight affected in generation II, and two of six in generation III, it compares so favourably with the classical description of dominant inheritance that no other interpretation is tenable. Genetic mutation may well have occurred in generation I.

![Figure 14](image1.png)

**FIG. 14**

Case III, 2. Figure 14—Dorsal view of the feet after operation. Figure 15—Plantar view of the feet.

![Figure 15](image2.png)

**FIG. 15**

Case III, 6—Radiograph of the hands.

It is not intended to discuss at length the surgery of correction of cleft foot. Indications for closure of the foot cleft are cosmetic or for pain due to callus formation or hallux valgus. Frequently, however, the latter symptoms can be controlled by the use of adaptive footwear. Surgery should consist of excision of useless bony remnants and the creation of suitable skin flaps to allow closure of the cleft. Metatarsal osteotomies may be required to narrow the feet. It has been stated that a deep suture around adjacent metatarsals is necessary to reduce tension on the skin flaps (Peet and Patterson 1963). This was not found to be so in the present instance. **Triphalangeal thumb**—Triphalangism of the thumbs is believed to be very rare. Lapidus, Guidotti and Coletti (1943) found six examples in 75,000 men drafted to an American Army...
Centre. Abramowitz (1959) is to be commended for his erudite writings on this abnormality. Typically, there is an extra set of skin creases overlying the additional interphalangeal joint. The additional phalanx is invariably the middle one. It may be complete but it may be no more than a stunted wedge-shaped ossicle. The terminal phalanx always deviates ulnarwards. Inheritance of this defect is by a dominant gene of variable expressivity.

The embryological notion of causation of the triphalangeal thumb as postulated by Lapidus et al. (1943) and substantiated by Abramowitz (1959, 1967) is of interest in that it may have a bearing on the pathogenesis of split hand and foot. These authors suggest that the initial embryological defect is a bifid thumb, that is, two digits arising from a common metacarpal. Resorption of one of these is a reversion to normal. Partial resorption can lead to persistence of a phalanx of one in whole or in part, as a complete bone or merely a wedge-shaped ossicle. Should the radial thumb atrophy in part and the ulnar persist completely, then ulnar deviation of its terminal phalanx is inevitable. Such a theory, though plausible, is somewhat speculative, yet where the additional phalanx is wedged it becomes more tenable.

Perhaps the buttress or boss of bone seen on the radial side of the tip of the terminal phalanx of the right thumb in Case II, 4 (Fig. 5) represents a more distal fusion of an accessory phalanx. In the feet persistent bony bosses were seen in the terminal phalanges of both great toes of Case II, 4 (Fig. 3). In Case III, 1 extra ossicles were noted in both great toes—a rather more rudimentary form of triphalangism (Fig. 8).

If the evidence of duplication as a cause of triphalangeal thumb, great toe or any other digit is acceptable, then the association of these deformities in the family described above, with the principal deformity of split foot, may assume some etiological significance. In the cases described in the literature, the cleft in the lobster claw deformity has never involved the carpus or the tarsus. The distal portion only of the limb was involved. It may be that duplication may involve all metatarsal or metacarpal rays in whole or in part (e.g., extreme forms of polydactyly). The tendency to revert to normal may be incomplete, as has been shown in the thumb. Complete absorption of the lateral duplicated elements of the medial two or three rays and absorption of the medial duplicated elements of the lateral two or three rays
may result in a normal limb but persistence in part may prevent soft-tissue fusion with consequent deep splitting of the hand or foot. Absorption of the duplicated elements may be more complex and haphazard so that fat broad metatarsals—the fused duplicated elements of one metatarsal—may result. Partial persistence of their phalanges leads to triphalangism (Figs. 17 to 19).

In the embryological development of the limbs constrictions appear in the limb buds at positions which correspond to the future major joints during the second month (Cunningham 1964). Terminally grooves appear as the first indication of digits. Excessive activity in the pre-axial and post-axial borders of the terminal portion of the limb buds may lead to duplication of all rays—activity, it is suggested, brought about by an abnormal gene. Genes responsible for continuance of normal growth of the limb buds may not be able to reverse completely the damage done by the defective mutant. Thus split foot and triphalangeal thumb, or indeed any of the numerous digital defects, including polydactyly, may be the outcome.

SUMMARY
1. The pedigree of a family in which lobster claw foot and triphalangeal thumb occurred together is presented. The affected members of two generations are described. The clinical appearances and radiological abnormalities are described in detail. Results of surgical treatment of the forefoot cleft are presented.
2. Lobster clawing of the foot is discussed with particular reference to associated hand and finger abnormalities. Triphalangeal thumb is discussed. The current and probably acceptable theory of the etiology of duplication is expanded. A hypothesis is formulated of excessive activity at the pre- and post-axial borders of the distal limb bud, followed by variable resorption, to explain not only the cause of lobster claw foot but also its occurrence in association with triphalangeal thumb.

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