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CONGENITAL FLEXION-ADDUCTION DEFORMITY OF THE THUMB
(CONGENITAL "CLASPED THUMB") *

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Five patients with congenital flexion-adduction deformity of the thumb have been seen during the past five years. Three of these patients, seen shortly after birth, responded well to plaster immobilization; the fourth patient, seen at age two, responded fairly well; the fifth patient, seen at age nine, required surgery.

Congenital absence of the extensor pollicis longus tendon was reported by Zadek in a child two and one-half years of age. This child was successfully treated by tendon transfer. Pollex varus was reported by Miller in two patients. One of the patients, eight weeks of age, was treated by casts and manipulation. More recently White and Jensen reported eight cases of this deformity, which they called "the infant's persistent thumb-clutched hand." Three of these patients were treated by tendon transfer. It is our opinion that the condition described by these authors is of the same nature as that seen in the five patients in our Clinic.

DIAGNOSIS

Congenital flexion-adduction deformity of the thumb is characterized by extreme flexion of the digit at the metacarpophalangeal joint and by adduction into the palm.


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(Fig. 1-A). The digit is held in the palm by the grasp of the overlying fingers. Hypermobility exists at the metacarpophalangeal joint. In the infant, when the grasp of the overlying fingers has been released, passive extension is carried out with ease. Active extension, however, is not manifest (assuming extensor tendons are present) until the thumb has been held out of the palm by the splint for several months (Cases 1, 2, 4, and 5). The condition should be differentiated from others which are similar.

Chronic stenosing tendovaginitis (trigger thumb): Passive extension of the distal joint is limited or associated with snapping. Treatment is tendovaginotomy.

The thumb in spastic paralysis: Here the thumb is caught beneath the tightly flexed fingers. Flexion at the proximal thumb joint is usually not so marked as it is in flexion-adduction deformity. When the tightly flexed fingers have been opened, the thumb exhibits active extension.

The thumb in arthrogryposis: This condition is manifested by limitation of active and passive joint motion. Usually there is resistance to passive extension of the thumb rather than hypermobility of the metacarpophalangeal joint, as seen in cases of flexion-adduction deformity.

CASE REPORTS

Case 1. C. M., one month old, was born with deformities of both hands and feet and with mild flexion contracture of both knee joints. There was no known familial history of congenital deformities.

The thumbs were severely flexed and adducted into the palms (Fig. 1-A), where they were held tightly clasped by the fingers. There were flexion contractures of the fingers: all the digits were long. The cleft between the thumb and index finger was tight bilaterally when the thumbs were passively extended. There was abnormal mobility at the metacarpophalangeal joint of each thumb. There was no observable active extension of the thumbs, although passively they could be brought into normal functional alignment with ease.
Plaster casts were applied, holding the thumbs in functional alignment and the fingers in extension. To allow for growth, the casts were changed at intervals of six to eight weeks. The right thumb was immobilized for five and one-half months and the left, for seven and one-half months. When the casts had been removed, the patient spontaneously held his thumbs in good alignment, and at once it was apparent that there was active extension.

Figure 1-B shows the results at follow-up examination two and one-half years later. Subsequently, plastic procedures of the Z type were carried out on the tight band between the thumbs and index fingers. At present, the child is five years old and uses his thumbs and fingers quite normally.

Case 2. C. F., six weeks of age, was unable at birth to extend the thumbs. No other abnormality was noted. The mother exhibited abnormal mobility of her proximal thumb joints but had no other abnormalities. One sibling was subsequently born with a club foot.

Examination showed both thumbs flexed and adducted into the palm (Fig. 2-A), where they were held in this hyperflexed position by the overlying fingers. There were no associated flexion contractures of the thumbs.
fingers. No active extension was observed in the thumbs, but passively they could be readily extended. Active flexion was present but did not seem to be excessively strong. When the thumb had been brought into extension, a band of soft tissue was found between the base of the thumb and the base of the index finger, bilaterally.

Bilateral plaster casts, changed at intervals of six to eight weeks, held the thumbs in functional alignment for seven and one-half months. The child then extended her thumbs actively and normally. Figure 2-B shows normal alignment of thumbs at age five.

Case 3. W. M., nine years of age, had been born with congenital deformities of both hands and torticollis and later developed severe scoliosis. The left upper extremity showed absence of the thumb and flexion contractures of the fingers and elbow. The right upper extremity showed flexion-adduction deformity of the thumb and flexion contractures of the fingers, which were held tightly flexed over the adducted thumb. Figure 3-A (obtained from an old hospital record) shows the child at fifteen months.

Figure 3-B shows the condition of the hands when the patient was first seen at nine years of age. The flexion contractures of the fingers and the flexion-adduction deformity of the right thumb remains. There were tight soft-tissue bands on the volar surfaces of the fingers and between the base of the thumb and the index finger on the right hand.

The proximal thumb joint was fused surgically to hold the thumb in functional alignment, and free full-thickness skin grafts were applied to the volar surface of the thumb and fingers to overcome the soft-tissue contractures. Figure 3-C shows the result. The thumb was made functional, although active extension remained
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The flexion contractures of the fingers were improved but not completely overcome.

Partial failure in this instance is attributed to the late stage at which correction was carried out. Permanent joint changes had already occurred in the fingers, and it is felt that the overstretched extensor tendons of the thumb had been permanently damaged.

Case 4. W. M., two years of age, had been born with congenital deformities of both hands, club feet, large inguinal herniae, and pyloric stenosis. Family history was negative for previous congenital anomalies. Examination showed flexion-adduction deformity of the thumb, bilaterally, associated with flexion contractures of the fingers. The position of the left thumb was more extreme than that of the right.

Casts were worn for three months. The right thumb shows good correction, but the left is still rather unstable.

Case 5. M. W., eleven weeks of age, was born with the thumbs clasped in the palms. The only associated abnormality was a slight limitation of extension of the left elbow. There was no known familial history of congenital anomalies.

The thumbs were adducted into the palms and held there by the flexed fingers. No active extension of the thumbs was observed. Passively, the thumbs could be extended normally. No contractures of the fingers and no particular tightness between the thumb and the index finger existed.

Plaster casts were worn for one month on the right hand and three months on the left (Fig. 4-A). The result, six months later, is shown in Figure 4-B.

DISCUSSION

Normal newborn infants hold their hands closed much of the time. They also clasp their thumbs. Among thirty-six newborn infants observed in the nursery at University Hospitals, fifteen (42 per cent.) held their fingers clasped over the thumbs part of the time. It was also noted, however, that the grasp of the fingers was released periodically and that the thumb then moved freely out of the palm.

Bieber states that the thumb in the newborn infant plays no role in grasp and that it usually lies dormant in the palm surrounded by the other digits. The studies of Gesell indicate that the infant begins to use its thumb in grasp at about three to four months of age.

In the five cases presented here, it appears that either the reflex clasp of the fingers upon the thumb was greater than normal or that weakness of the extensor tendons of the thumb allowed it to remain beneath the clasp of the fingers. Duchenne has shown that the proximal phalanx of the thumb becomes more flexed when the short extensor loses its action. White and Jensen ascribe the deformity in their cases to absence of the extensor pollicis brevis tendon. In our cases, with the possible exception of Case 3, the deformity was caused by weakness rather than by absence of the thumb extensor. Weakness of the extensors is undoubtedly accentuated by the over-pull of the flexors and the clasp of the fingers.

Associated flexion contractures of the fingers might have been an additional factor in three of our cases; in the other two they were not present. Some tightness in the web space between the thumb and the index finger, which was present in three of the cases, may also have been a factor. All the patients exhibited abnormal mobility of the metacarpophalangeal joints. The possible role of the grasping reflex when the thumb is in the

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palm is interesting. The mother of C. F. (Case 2) exhibited abnormal mobility of her proximal thumb joints but had no other abnormality of her hands.

The period of immobilization in the first two cases was five and one-half to seven and
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one-half months. In the most recent case, one to three months gave a good result. It is thought that plaster immobilization is most desirable in that it gives the most constant support. The casts must be changed each six to eight weeks to allow for growth. Associated flexion contractures of the fingers, when present, are treated simultaneously.

A trial period of plaster immobilization will show whether or not extensor tendons are present. If no extension returns after several months of plaster immobilization, tendon transfer to restore active extension is indicated.

SUMMARY AND CONCLUSIONS

Of the five cases of congenital flexion-adduction deformity presented in this report, all were bilateral except Case 3. The patient in Case 3 had a congenital absence of the left thumb. In each instance of the deformity, the thumb was firmly clasped beneath the flexed fingers and did not exhibit active extension. Three patients also had an associated tightness of the web space between the thumb and index finger. All the patients had abnormal mobility of the metacarpophalangeal joints of their thumbs.

In three of the patients, seen one to three months after birth (Cases 1, 2, and 5), plaster immobilization gave excellent results. In the fourth patient, seen at the age of two years (Case 4), a fair to good result was achieved after three months' immobilization. In another patient, seen at age nine (Case 3), a fair result was obtained by fusion of the metacarpophalangeal joint of the thumb.

Congenital flexion-adduction deformity of the thumb should be corrected by plaster immobilization in functional position at the earliest possible time, before permanent joint and tendon changes occur. If active extension does not return, tendon transfer is indicated to restore active extension to the thumb.

REFERENCES


DISCUSSION

DR. GEORGE S. PHALEN, CLEVELAND, OHIO: I was very interested to hear of Dr. Weckesser's experiences in treating five cases of congenital clutched thumb. He is most fortunate to have had the opportunity of seeing so many cases with this unusual congenital deformity. I am sure that very few of us here at this meeting can duplicate his experience.

One wonders why this anomaly is not observed more often. For the first few months of life, the thumb actually serves no useful purpose in the infant's existence. It is not until he learns to use the thumb in grasping that he has any good reason to raise his thumb out of his palm. All of us have observed how much of the time a normal infant keeps his thumbs tightly clasped in his palms.

It is easy to see how congenital adduction of the first metacarpophalangeal joint might be part of a generalized arthrogryposis. Four of the author's five cases might fall into this group. The ease and permanency of the correction of the deformity, however, would lead one to believe that congenital clasped thumb is not similar to an arthrogryposis.

I would like to ask Dr. Weckesser if he observed the fate of the soft-tissue bands in the first web in those cases in which an adequate correction of the deformity was obtained. Did these bands soften, stretch, or disappear entirely? Also, in the two cases requiring operative correction, was the contracture in the first web found to be due to a subcutaneous fibrous band or due to skin contracture only? If a subcutaneous fibrous band was found at operation, was it observed to be a thickened part of the normal palmar fascia, such as we
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see in Dupuytren's contracture? If any fascial bands were removed in these two cases, were any histologic
studies made of this tissue?

I would certainly agree with Dr. Weckesser that plaster immobilization is the best method of treating
this deformity. The good results obtained with early treatments serve again to emphasize the importance of
correcting all soft-tissue contractures as soon as possible before permanent joint contractures or bony de-
formities develop.

DR. ARTHUR J. BARSKY, NEW YORK, N. Y.: Dr. Weckesser stated that in four of his five cases flexion
or extension deformities were present in joints other than those of the thumb. The cases in which the condi-
tion was not limited to the thumb joints alone belong, I believe, in the category of what is variously called
arthrogryposis multiplex congenita, multiple articular rigidities, or, to use a term that is perhaps better,
amyoplasia congenita.

Curiously enough, this same condition has been found in lambs, a circumstance which has provided
additional data. There appears to be present a hereditary factor of a recessive nature.

Gilmour had the opportunity of autopsying a baby with hand involvement and found that no extensor
pollicis longus was present. The muscles affected in this baby were much smaller than the normal due to an
actual decrease in the number of fibers. There was a definite increase in the number of collagenous fibers in
the endomysium and perimysium. In another case, Middleton found that the extensor pollicis longus was
replaced by adipose tissue containing fibrous strands and remnants of muscle fibers.

Weak electrical response has been reported but not reaction to degeneration.

The belief has been expressed that stiffness of the joints is the primary cause of the limitation of motion.
Although this fixity of the joints sometimes has the clinical appearance of a fibrous ankylosis, there is no
definite evidence of inflammatory change. The joint fixity is probably largely due to disuse arising out of a
lack of active and passive motion; with disuse one would expect the periarticular tissues and even the skin
to become tight.

Middleton's observation on lambs indicates that the process is one of degeneration of formed and
differentiated muscle fibers. There is no definite evidence as to the nature of the abnormality which causes
the atrophy and degeneration of the already formed muscle fibers, but it is thought (by Gilmour) that the
process is not dependent on external influences but is possibly dependent on an intrinsic genetic abnormality
centered in the muscle cells.

We have had four cases of amyoplasia, three of which give a good idea of the gradations of the condition.

[Note: A lantern slide was shown of the mildest case, in which involvement of only the index fingers of both
hands was apparent. A second slide showed a case in which the wrist and elbow joint were affected, and a
third slide showed involvement of practically all the joints of all four extremities.]

Dr. Weckesser has made an excellent point, one that bears emphasis, namely, that very early treatment
by splinting will probably result in improvement if enough muscle fibers are left to function.

DR. WECKESSER (closing): In answer to Dr. Phalen's questions about the thumb-web contractures, I
wish to state that the tight bands between the thumb and index finger seemed to remain essentially un-
changed. In the two cases requiring surgery, the tight band was found to be due to skin contracture only.
No subcutaneous bands were present.

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(Continued from page 986)

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