BILATERAL TRIGGER THUMB IN INFANTS

BY
THEODORE JAMES
From Pinelands, The Cape of Good Hope
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'Trigger thumb' or 'snapping thumb' in infants is a curious digital restriction in the very young and perhaps is best referred to as a tendovaginitis stenosans of the sheath of the flexor hallucis longus muscle as it is found in infants. The characteristic feature of this condition is the sudden overcoming of involuntary resistance occurring in the normal range of movement of the interphalangeal joint of the thumb, after which movement is again free. Sometimes the sudden overcoming of the resistance is accompanied by a snap. When the physician first sees the infant the interphalangeal joint of the thumb is usually actively inextensible beyond an angle of about 150°. Frequently flexion from full extension is blocked at a slightly more obtuse angle in the same child but the resistance is more readily overcome voluntarily by the stronger flexor tendon.

The recognition of this entity has been mentioned in one or two American text-books of orthopaedic surgery and it has been written about in the continental European literature but there is scarcely a mention of it in the British medical press other than two case reports of the bilateral condition by Rose (1946) in Australia. For this reason a case of bilateral trigger thumb in an infant of 14 months is submitted. Also, it clearly fixes the manner and time of its clinical appearance.

Case Report

T.S. was just 14 months old when her father, who was fond of playing with his young children, noticed for the first time that her left thumb seemed fixed in flexion ('bent at the knuckle'). He believed that had this persistent digital flexion been present earlier he would have observed it. No other member of the family group, in which there were five adults (including two grandparents), had seen or remarked upon the state of the thumb. The father, therefore, was inclined to believe that the limitation of movement had come on suddenly. At the time of his initial observation he immediately compared the two thumbs but the active and passive ranges of movement of the other thumb were normal as far as he could make out.

When the parent brought the infant for advice it was noted that the affected thumb's interphalangeal joint could not extend beyond about 135°, to which angle both active and passive movements were painless. Forced passive extension to the maximal normal limit was sudden and painful enough to make the infant cry. Soon after, within five minutes of the forced passive extension, the joint spontaneously resumed its position of persistent restricted extension. Palpable over the flexor pollicis longus tendon and at the head of the first metacarpal was a lentil-sized nodule in the subcutaneous tissue. The diagnosis was straightforward. However, no comparable nodule in the other thumb was noted at the time. An open operation to split the tendon-sheath only was successful and the skin sutures were removed eight days later. At this time comparison again of the two thumbs showed no functional difference; but 10 days after the removal of the sutures, or 18 days after the operation, the observant father saw with interest and some little distress that an identical limitation of extension of the other thumb had taken place.

Review of the Literature

In 1933 Compere collected notices of 40 cases of trigger thumb and established its rarity in infancy. His collection included bilateral cases in adults and these were comparatively frequent. There were only two reports of cases in children. His case of bilateral snapping thumb came to him when she was 20 years old and she was able to recall that she had had the condition without any real disability resulting since the age of 4 years. His other case he saw when the baby was 9 months old but the mother informed him that both thumbs snapped into extension soon after birth.

Compere (1933) quotes Hauck reporting an infant girl with bilateral involvement. So up to the time of Compere's article only three instances of bilateral trigger thumb appear to have been reported, of which two were in the infant age group. Lipscomb (1944) reviewed 190 cases of tendovaginitis stenosans and found among them only 11 of the thumb, none of these unilateral and none in children. In 1936 Jahss made a point of the fact that because of the rarity of trigger thumb in infants he had not had a
case referred to him for treatment that had been correctly diagnosed. Most of the diagnoses made had been 'congenital contracture of the thumb'. He reported nine cases. None of the mothers of his cases could recall exactly how long the condition had been present in her child. Jahss states that he actually saw 15 cases but confirmed only 10 by operation and their ages ranged from 3½ months to 15 years. Three were bilateral and their ages 15, 2 and 14½ years when he saw them. In 1946 Rose in Australia published two more instances of trigger thumb in both hands, the ages of the infants being 9 months and 2 years respectively. Sprecher (1949) in America reported 12 cases, in none of whom was there an audible snap. In four, locking of the thumb had been noticed immediately or soon after birth, and in the others before the age of 3 years. In three of Sprecher's cases both thumbs were affected and in two cases with one thumb showing the limitation the other thumb was functioning normally although a palpable nodule was present in the tendon. Sprecher refers to Beck who reported six cases but no details are given. Zadek (1942) also described two unilateral cases in whom there was symptomless thickening on the tendon of the other thumb. In 1953 White and Jensen discussed a series of nine cases of trigger thumb, whose ages ranged from 15 months to 6 years. Two cases had a family history and five patients came from the same island in the Hawaiian Group. All cases were unilateral. It was in 1953 also that Chiari was able to give the results of his own method of a subcutaneous operative approach to the stenosed part of the tendon sheath in 100 children. Although Chiari admits to the stenosis being found for the first time in later infancy and childhood, he nevertheless rather unhappily uses the term congenital contracture of the thumb (angeborenen Daumenkontraktur). There is no doubt from his article that his 100 cases were all stenosing tendovaginitis. His contribution would have had an added value if Chiari had analysed his remarkable series into age groups, sex, time of onset, whether uni- or bilateral and so on. It is to be hoped that he will do so. That Chiari was able from his own practice to collect 100 cases on whom to apply his surgical technique suggests the probability that there were others differently treated and that in his part of the world trigger thumb is not so rare an entity as we are inclined to regard it.

Discussion

I have been able to collect nine undoubted cases of bilateral trigger thumb in infants from my review of the available literature. About 25% of the cases manifesting trigger thumb in infancy have been bilateral. When one attempts an aetiological hypothesis which excludes trauma, the causative factor for trigger thumb in adults, one inclines to wonder why it is that many more, if not all inlines, do not sooner or later become bilateral. Perhaps closer follow-up observation of unilateral trigger thumb is desirable. Despite Chiari's big series which leaves an impression that tendovaginitis stenosans is not uncommon, all available evidence emphasizes the rarity of the bilateral state.

Clinical recognition of the stenosed sheath of the long flexor tendon of the thumb in infants is made by the fact that there is a persistent, painless limitation of extension of the digit to an angle of about 150°. Passive attempts to extend the interphalangeal joint fully are somewhat painful but possible and sudden, when a snap may be heard. Once the joint is fully extended passive flexion is neither painful nor will it produce a snap. Some parents have given a history of a gradual increase of resistance to full extension and the production of the trigger thumb effect, and eventually a complete locking of the joint at a definite angle. The thumb, when first seen in the locked state or when no attempt has been made passively to overcome the resistance to extension, has been confused with a true congenital contracture of the thumb. In this latter disability, however, there is hyperextension of the metacarpophalangeal joint and flexion of the interphalangeal joint, both of which are fixed. Compere mentions rheumatism, neuritis, periostitis, tenosynovitis, tuberculous osteitis, and recurring dislocation at the interphalangeal joint as some of the mistaken diagnoses in his history.

Whereas trigger thumb in adults is of fairly frequent occurrence and can usually be associated with a story of preceding repeated minor trauma, bilateral trigger thumb in adults is rare and unrelated to infancy. In infants there has been no history forthcoming of any sort of trauma excepting one case of Zadek's in which there had been definite injury to the thumb so that it was treated at first as a traumatic dislocation and only later did an antecedent trigger thumb show up. In my case the onset of the bilateral manifestation could be looked on as a simultaneous happening.

The stronger pull of the flexor tendon keeps the nodule on the proximal side of the stenosis, the primary pathology being in the tendon sheath, and Zadek regards the tendon thickening as a reaction to pressure on the tendon by the stenosing band. In one case he states that the tendon was oedematous. Jahss described a pathological study of the sheath in which there were hyalinized conne-
tive tissue and fibrous villi but there were no adhesions and no free fluid.

Because trigger finger and trigger thumb in adults can almost always be associated with trauma, Compere postulates that in infants there is a predisposing factor and he proposes that it is the acuteness of the angular change of course and pull of the flexor pollicis longus tendon at the metacarpophalangeal joint; but if this were the factor or even a factor it has been stated that the condition would be much more common, certainly in its bilateral form. It has also been postulated that a congenital anomaly of muscle insertion initiates the condition in the foetus which might account for those cases discovered at or soon after birth. The lateral head of the flexor pollicis brevis muscle and the adductor pollicis muscle have been incriminated. Possible additional bands between these two muscles and passing over the tendon sheath of the long flexor, or an abnormally close relationship between these three muscles may be extra factors, but surgical enquiry has not supported these ideas. And yet a variation of the short flexor muscle of the thumb has been described in which a part of the muscle may be inserted separately into the ulnar side of the proximal phalanx of the thumb with the adductor muscle, when it is known as the first volar interosseous muscle. To reach this point of insertion the shorter muscle would have to cross the long flexor tendon of the thumb.

Sprecher sectioned a piece of the involved part of the tendon and reported evidence of trauma indicated by numerous lymphocytes and monocytes in the stroma of the tendinous tissue. So Sprecher believes that the child’s tendency to grasp its thumb into its palm produces a sharp kinking of the tendon over its ligamentous attachment to the bone and that this is enough to provide the necessary trauma. He was also of the opinion that because the newborn infant will grasp its thumb in this way it could hold it in the same way in utero.

Whether any ethnic influence plays a role is unknown, but a hint of some such possible factor has been put out by White and Jensen who recorded a family history in two of their cases and five others of Filipino ancestry who came from one island in the South Pacific. Chiari’s series was collected in Germany. My case came of allegedly pure Germanic and Sicilian stock.

Summary

An account is given of a curious affection of the sheath of the long flexor tendon of the thumb in infants. Although the incidence of the unilateral condition is by nearly all reports rather uncommon, that of the bilateral state is a rarity. It goes colloquially by the names of ‘trigger thumb’ or ‘snapping thumb’ but is perhaps better described terminologically as tendovaginitis stenosans. A report of a case is followed by a review of the literature and a discussion of possible aetiological factors in its development.

References