THE ETIOLOGY OF QUADRICEPS CONTRACTURE IN CHILDREN

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The clinical histories of six patients with quadriceps contracture seen within the last five years have been analysed and a search has been made for a common cause. Additional information has also been sought from authors who have recently reported this condition, Hněvkovský (1961), Fairbank and Barrett (1961), and Gammie, Taylor and Urich (1963), to whom we are greatly indebted for their help.

Study of our six patients revealed that they had all had injections or infusions into the thigh soon after birth, four of them for serious illnesses. Upon inquiry it transpired that out of twenty-two patients observed by the previous authors six had been similarly treated. This report is concerned with the implications of this observation.

CASE REPORTS

Case 1—A boy, aged five years five months, had a full term normal delivery. His birth weight was 6 pounds 15 ounces. He had abdominal distension at birth and was found to have meconium ileus with underlying fibrocystic disease of the pancreas and lung. Immediate operation was followed by intravenous and nasal drip feeding. He was also given injections of penicillin four hourly for at least three weeks into alternate thighs. Inability to bend the left knee beyond a right angle was first noticed at one year and eight months. This progressed until only 20 degrees of flexion movement remained. When this movement took place tight bands could be felt deep in the quadriceps muscle. A lesser degree of restriction was present in the right knee. Operation was not done on this child because of the fibrocystic changes in the lung.

Case 2—A girl, aged five years 4 months, had flexion from 0–60 degrees in the left knee. The age of onset was obscure. She had suffered from duodenal obstruction in infancy, relieved by operation. This was followed by volvulus, which demanded massive resection of the small gut. Since that time she has been admitted to hospital several times for treatment of a malabsorption syndrome and dehydration. She had received many injections and infusions during her illness but the routes by which they were given are not recorded. On examination, a firm block to flexion and a dimple over the lateral aspect of the thigh were found. The dimple deepened on forced flexion of the knee, and the vastus lateralis and to a lesser extent the tensor fasciae latae, became tight. Surgical division of the tensor and the lateral intermuscular septum did not improve the deformity; so the incision was enlarged and the vastus lateralis and medialis were detached from the quadriceps tendon. This was also unrewarding. Division of the patellar tendon, however, allowed full correction and this was sutured to allow for lengthening. The leg was immobilised at 90 degrees for a month. A severe extension lag was quickly overcome and three months later only 10 degrees lag remained. Flexion of 110 degrees was present—that is to say a gain of 50 degrees.

Case 3—A girl, aged five years four months, was unable to bend the knees fully. The condition had been present for three years and had become more obvious during the six months before examination, during which time she had become unable to kneel. Manipulation under anaesthesia one year before had failed to improve the deformity. She had been born prematurely and received both Synkavit and prophylactic antibiotic injections soon after birth. Movements were restricted to 60 degrees of flexion on the left and 90 degrees on the right. Electromyography
revealed normal motor activity in the quadriceps as a whole, but detailed study could not be made because the patient was not cooperative. At operation on the left knee, there were some adhesions in the suprapatellar pouch and behind the patella but the only barrier to flexion was the quadriceps expansion itself. Division of this improved flexion to 110 degrees, and transverse division of the vastus medialis expansion allowed virtually full movement. This improvement was found to be maintained two years later. Histological examination showed well developed fibrous tissue between the muscle bundles of vastus medialis but no other obvious abnormality.

**Case 4**—A boy, aged eight years. This child was noticed to have limited flexion in the right knee. He was first treated at the age of three weeks for osteomyelitis of the right radius and left femur by primary drainage. He developed a deformity of the left knee, from a disturbance of epiphysial growth. During his early illness he received injections of antibiotics into the thighs. On examination the right knee flexed from 0-110 degrees. There was a puckered scar on the lateral aspect of the thigh which appeared to be tethered to the tensor fasciae latae (Figs. 1 and 2). No treatment was recommended for this patient as his disability was so slight.

**Case 5**—A Eurasian girl, aged two years, had been noticed to kick only with the left leg from the age of six months. At the age of one month she had received daily injections into alternate thighs for seven to ten days "for rickets," and at two months she was treated for dehydration by intravenous infusions into the right long saphenous vein at the ankle. On examination the right knee flexed only from 0-30 degrees. The quadriceps muscle was explored from the front. There were some adhesions from the vastus medialis and lateralis to the vastus intermedius in the lowest third of the thigh. The rectus femoris appeared normal. A Z-lengthening of the vastus intermedius tendon gave some improvement, but full flexion
was not possible until the insertions of the vastus medialis and lateralis into the central tendon were divided. These were sutured to the tendon with the knee held in flexion. Histological examination of the three vasti muscles showed swelling of some fibres, variation in depth of staining, the presence of "many small, pale fibres and focal clumping of sarcolemmal cells—due partly to a moderate degree of atrophy." The rectus femoris was normal. Four months after operation the range of flexion was from 0–100 degrees.

**Case 6**—A girl, aged six years six months had a normal delivery and weighed five pounds at birth. She was found to have a recto-vaginal fistula, which was dilated at birth. She was given injections of penicillin and streptomycin into the thighs for about six weeks and, in addition, she received intramuscular injections of ascorbic acid and vitamin K. These injections were discontinued when induration of the upper third of the thighs was noticed. At this time both knees flexed only from 0–50 degrees. At the age of six months the right knee had a full range of movement, but there had been no change in the left knee. At six and a half years the right knee had again lost some movement, and flexed from 0–80 degrees, and there was the characteristic puckerated dimple on the outer aspect of the thigh. By this time the left knee had become stiff in 25 degrees of flexion.

**ADDITIONAL INFORMATION ABOUT THE PATIENTS DESCRIBED BY PREVIOUS AUTHORS**

Hněvkovský (1961)—Of seventeen cases that this writer has seen up to the present time, he has been able to establish a definite history of injections into the thigh in four. His view now is (Hněvkovský 1963) that there may be two syndromes, the progressive idiopathic type that he described in his article and the type due to injections. A recent article by Dufek (1962) draws attention to the latter type.

Fairbank and Barrett (1961)—Their patients were identical twins born prematurely by breech delivery. Both children received a month's course of intramuscular Crystamycin (which was at the time a prophylactic routine for premature infants) and intramuscular Synkavit. The exact route of these injections is not known.

Gammie, Taylor and Urich (1963)—One of their patients was premature and a binovular twin. The birth weight was 4 pounds 4 ounces. In view of the prematurity she was admitted to hospital, but there is no record of any injections. The second patient was born at thirty-six weeks by breech delivery, and was the first of uniovular twins. She had melena ten hours after delivery and was transfused with blood via the umbilical vein. She also received 6 milligrams of Synkavit in divided doses, but the route is not known.

**DISCUSSION**

In the three previous reports referred to, some suggestions have been made about etiology. Hněvkovský suggested that in his cases there was "a muscular dysplasia of congenital origin of the vastus intermedius and rectus femoris muscles. . . ." Fairbank and Barrett, after describing similar histological appearances to those described by Hněvkovský, suggested a resemblance between this condition, arthrogryposis, and sternomastoid contracture in torticollis; and because their patients were identical twins, they proposed a genetic factor which, because of Hněvkovský's patients and the Polish ancestry of their own patients, they suggested might be more common in Eastern Europeans.

Gammie, Taylor and Urich attempted to dispose of the genetic and racial possibilities by reporting one of a pair of uniovular twins and by describing a patient of "pure British stock." They did not think that birth injury was a likely cause because the lesion had first appeared in one of their own patients at the age of five and in one of Hněvkovský's at the age of seven.

We believe that clinical histories of obstetrical difficulties, twinning and prematurity, common as they may be, are relevant only in so far as these children often have need of treatment soon after birth. In particular we would incriminate injections into the quadriceps muscles.
muscle in very early life. Table I shows that such injections had been administered to all of our patients, and, although the amount injected and the duration of the courses varied, some—notably Cases 1 and 6—received large and prolonged courses of treatment. It is perhaps significant that these patients were among the most affected. In three patients stigmata of injections or infusions have persisted as skin dimples. Case 6 is of particular interest, in that induration had actually been noticed as a result of the injections, which had been stopped because of it. The progressive nature of the condition is also illustrated by this patient, and it is noteworthy that the right knee recovered full movement, only to lose it again, whereas the left knee progressed relentlessly until all movement was lost and a fixed flexion deformity developed.

**Table I**

**Details of Six Patients with Quadriceps Contracture**

<table>
<thead>
<tr>
<th>Case number</th>
<th>Age of onset (months)</th>
<th>Prematurity</th>
<th>Neonatal illness</th>
<th>Neonatal injections</th>
<th>Stigmata</th>
<th>Range of movement (degrees)</th>
<th>Operation</th>
<th>Histology</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>20</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>Right 0–90; left 0–20</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Unknown</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Dimple</td>
<td>Left 0–60</td>
<td>Yes</td>
<td>Fibrous tissue between muscle bundles</td>
</tr>
<tr>
<td>3</td>
<td>28</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>No</td>
<td>Right 0–90; left 0–60</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Unknown</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Dimple</td>
<td>Right 0–110</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>6</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>Right 0–30</td>
<td>Yes</td>
<td>Moderate atrophy</td>
</tr>
<tr>
<td>6</td>
<td>1½</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Dimple</td>
<td>Right 0–80; left fixed at 25</td>
<td>No</td>
<td></td>
</tr>
</tbody>
</table>

It has been fashionable from time to time to give prophylactic injections of various drugs to newborn premature infants. Fairbank and Barrett's patients, for example, received intramuscular Crystamycin for a month. Vitamin K preparations have also been given to newborn infants, not only for haemorrhagic disease but also to those without signs of this condition, as a prophylactic measure.

Other forms of therapy commonly employed include the so-called "subcutaneous" infusion. The subcutaneous space is very small in the newborn and such infusions do not usually run well unless they are, in fact, intramuscular. Nurses who attend these babies are familiar with, and often alarmed by, the bruising and swelling of the thigh that accompany these injections. Further support for our hypothesis comes from the appearances found at necropsies on infants. The muscles into which these injections and infusions have been given are often the site of considerable oedema and haemorrhage. The occasional development of fibrosis followed by contracture after months or years is thought to be a reasonable possibility.

Available histological reports do not suggest a congenital abnormality. Indeed the accounts are remarkable for their variety, and the only feature commonly encountered is fibrosis, either between or within the muscle bundles. Other findings are not sufficiently constant to be significant.

**Summary**

1. Six cases of quadriceps contracture in children are described. All were either premature or suffered severe illnesses soon after birth.
2. Some additional information is given about cases of the same condition previously published by other authors.
3. It is suggested that injections and infusions given to newborn babies are sometimes the cause of the condition.
4. Some of the therapeutic substances and measures which may be responsible are discussed. These include antibiotics, vitamin K preparations and "subcutaneous" fluid therapy.

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REFERENCES


