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YOUNG

THE YEAR BOOK
of
ORTHOPEDICS
and
TRAUMATIC SURGERY
1973

EDITED BY

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CONGENITAL AND HEREDITARY DEFORMITIES

Congenital Absence of Cervical Vertebral Pedicle. Twelve cases of congenital absence of a cervical vertebral pedicle have been previously reported. The lesion is usually of little or no clinical significance, although it predisposes to a degree of instability and resultant degenerative changes. It is important, however, to make a definitive diagnosis; incorrect diagnosis is common. Dermot P. McLoughlin (Barrie, Ont.) and George Wortzman[1] (Toronto, Ont.) describe 4 patients with congenital absence of the pedicle. The absent cervical pedicle is considered to be due to a defect in the cartilagenous phase of development rather than in ossification. The defect is of the neural arch chondrification center and the single defect explains absence of the pedicle and the associated anomalies of the articular processes, lamina and transverse processes. The defect is best visualized on oblique views. The intervertebral foramen is enlarged in its vertical diameter and the anteroposterior diameter of the foramen is also increased. The ipsilateral lamina was displaced posteriorly in 2 patients and absent in 2. Associated anomalies were present in all 4 patients. Abnormal articulations or instability with degenerative changes may occur secondarily. Myelography in 1 patient showed no outpouching of the dura or arachnoid.

The differential diagnosis of this lesion includes neurofibroma, vertebral artery erosion, fracture of the neural arch and neoplastic disease. Usually the pedicle of C5 or C6 is absent. Two of the present cases were initially misdiagnosed. Eight previously described patients and 1 present patient had neurologic complaints. A fifth patient was recently seen in whom C2 was affected.

Complete Agenesis of Lumbosacral Spine: Case Report. Few reports of this congenital abnormality have appeared since the first in 1910. Maurice Mongeau and Richard Leclaire[2] (Rehabilitation Inst. of Montreal) describe a patient who was born with total absence of the lumbosacral spine and partial absence of the thoracic spine. The last vertebra

(1) J. Canad. A. Radiologists 23:195–200, September, 1972.
(2) J. Bone & Joint Surg. 54-A:161–164, January, 1972.

present was the 9th. This is the highest absence of spinal segments in all reported cases.

Girl, aged 9 months, had weighed 1.6 kg. at birth after an uneventful 40-week gestation. Three other children were normal. When first seen, she weighed 4.5 kg. She lay supine with hips in flexion, external rotation and abduction; knees were in flexion. A wide popliteal web was present bilaterally. The right foot was in supination and the left in pronation. The lower limbs and buttocks were hypotrophic with no voluntary, involuntary or reflex movements. No sensation was present below about the T10 dermatome. The genital region was hypoplastic; T8 and T9 were prominent. The child never had sat; she had always been kept supine or prone. She had normal function of the upper extremities. There was no evidence of mental retardation.

X-rays showed complete absence of lumbar spine, sacrum and coccyx and hypoplasia of the iliac bones; T7 was transitional, T8 bipartite and T9 partial. Two ribs arose from T8. The 9th right rib was hypoplastic; the 10th ribs were without bone attachment. Bilateral hydronephrosis was present. Urine cultures showed a heavy growth of Escherichia coli and Bacillus proteus. The chromosome constitution was normal.

At admission, the child was developing tolerance of the upright position by use of a bucket with back support and with partial weight-bearing by the lower part of the thoracic cage. She will soon require an ileal conduit. Colostomy may eventually be necessary for megacolon. She will benefit from a bilateral subtrochanteric amputation and will be trained with bilateral modified Canadian hip disarticulation prostheses. Spinal fusion by the method of Perry *et al.* (1970) may be tried if it can be done as high as the 7th thoracic vertebra.

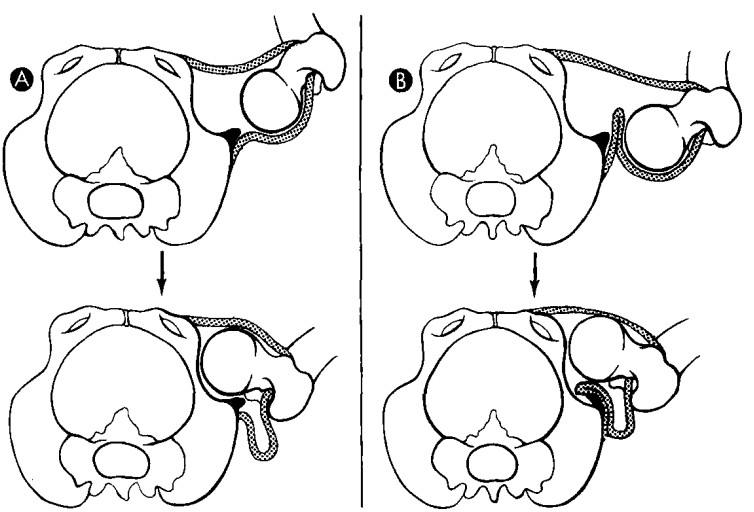
A Postnatal Survey for Congenital Displacement of the Hip was carried out by J. A. Wilkinson[3] in Southampton, England, over a 12-month period starting in June, 1968. Of 6,272 live births, 75% were in hospitals. There were 3,368 births in a central maternity unit. A total of 147 breech births occurred, representing 2.6% of all live births. Varying degrees of arrest of leg-folding were found in 65% of 123 single breech babies and in over 80% of first-born girls. Three of the 147 breech infants had plagiocephaly, all left sided, and 1 of these had postural torticollis. Prenatal molding was more marked than in normal births.

(3) J. Bone & Joint Surg. 54-B:40-49, February, 1972.

There were 18 female and 5 male infants with reducible hip displacements. Nineteen were first born, and 19 had breech presentations. Both hips were unstable in 9 cases. More of these infants were born in the summer months. Intrauterine molding was slight in this group. Sixteen infants were folded in the locked-leg posture. Five had plagiocephaly. Twenty infants were nursed supine on Barlow and then von Rosen splints for 8–12 weeks, and 3 were nursed prone in the frog position with double diapers. Twenty infants responded excellently, including those who were not splinted. Three girls had minor dysplasia persisting beyond 12 months and extreme persistent joint laxity.

Fourteen infants had all the features of hip displacement but a negative Barlow sign. They had above-knee shortening and limited abduction of one or both hips. Six were diagnosed at birth and 6 at age 3–4 months. Those diagnosed at birth were breech born and severely molded in an extended knee posture. Three had plagiocephaly. Three hips responded to neonatal splinting. Another infant later had excision of a limbus. Two of the 6 infants seen at age 3–4 months were

Fig. 1.—Diagrams illustrating concentric and eccentric reductions. *A*, reducible hip displacement without limbus; *B*, irreducible hip displacement with limbus. (Courtesy of Wilkinson, J. A.: J. Bone & Joint Surg. 54-B:40–49, February, 1972.)



born by breech presentation. Hip displacements in this group were confirmed by Andrén's radiologic test. Adductor tenotomy was performed, and Denis Browne splints were applied for 3 months. Two infants recovered fully. Two have acetabular dysplasia, and 2 have fragmentation of the femoral epiphysis. The two infants seen at a later age had limbus excision and obtained concentric reduction and stability.

Concentric and eccentric reductions are illustrated in Figure 1. Eccentric reduction of the femoral head secondary to incarceration of the limbus leads to pressure on and persistent hypoplasia of the acetabular root and femoral epiphysis. Both types of reduction are more likely to affect first-born girls, with a history of breech malposition culminating in breech birth. Spontaneous recovery from reducible hip displacement occurs extremely often, with or without splintage. Irreducible hip displacement does not respond to splintage in the first 6 months of life. Even with adductor tenotomy there is a great risk of acetabular and femoral epiphysial damage.
► [This is an excellent survey of congenital displacement of the hip on a large series of live births. – Ed.]

Problems in Early Diagnosis and Management of Congenital Dislocation of the Hip are discussed by George P. Mitchell[4] (Royal Infirm., Edinburgh). The treatment from birth of congenital hip dislocation should give excellent results in nearly all cases, but difficulties still arise despite more regular routine examination of newborn infants.

In a large maternity unit in Edinburgh, four dislocations were missed among 31,961 births in 1962–68 and were diagnosed after walking began. When a "clunk" sign was found soon after birth and was confirmed on re-examination a few days later, the condition was classified as a luxation and the hips were held by a Malmö splint. Where the "clunk" sign was not confirmed but radiographs showed displacement, the Malmö splint was applied. There were 100 cases of luxation with a positive "clunk" sign at re-examination, for an incidence of about 3 per 1,000 births. There were 126 unstable hips, whereas 123 suspect hips were considered normal.

Ninety-two of 100 luxated hips became anatomically normal with routine splintage. Two showed femoral head de-

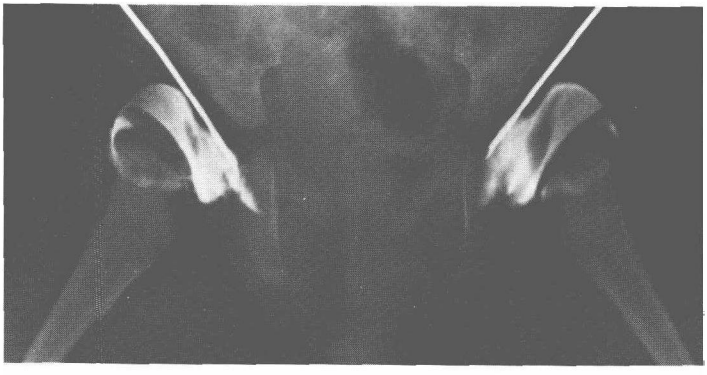
(4) J. Bone & Joint Surg. 54-B:4–12, February, 1972.

formity, 5 had to be reduced surgically and 1 required derotation osteotomy. In the unstable group there were no complications. The 4 missed dislocations were successfully reduced by operation at a later age. In 1 case the hips could not be held reduced in the Malmö splint (Fig. 2). Excision of the limbus and a bilateral Salter osteotomy were carried out, providing stable reductions and functionally normal joints, but residual femoral head deformity was found 6 years later.

Reduction with the Malmö splint may appear satisfactory on radiographs, but displacement may occur on mobilization. Reduction by abduction in the presence of an inverted limbus may result in deformity of the epiphysis or metaphysis, particularly in the early months of life. Anteversion sometimes corrects slowly. Prolonged splintage should never be used to correct anteversion, which almost always will respond eventually to weight-bearing. Increased anteversion corrected itself by age 3 years in all cases but 1, in which bilateral derotation osteotomy was performed.

Excision of an inverted limbus should be a relatively minor procedure. Excision of the limbus has been combined with frog position plasters to correct anteversion. Deformity of the upper femur has not occurred unless there was a previous period of splintage in wide abduction. Eighteen infants under age 1 year had excision of the limbus followed by a plas-

Fig. 2.—Arthrograph in case in which hips could not be held reduced in Malmö splint. Inversion of the limbus and a fold of capsule were present on the left side. (Courtesy of Mitchell, G. P.: J. Bone & Joint Surg. 54-B:4-12, February, 1972.)



ter spica in the frog position for 4 months to correct anteversion; in 4 cases the anteversion did not correct and derotation was carried out. A longer period in the frog position might have been more effective.

Particular care in examination of the newborn should be taken where there is a strong family history of dislocation of the hip. Minor degrees of displacement warrant treatment.

► [I agree with the author that early management of the congenitally dislocated hip gives the best prognosis. The author points out some of the difficulties that still exist in early diagnosis, and perhaps review of this article will be of value in early detection of this deformity.—Ed.]

Difficulties of Early Diagnosis and Treatment of Congenital Dislocation of the Hip in Northern Ireland. Present efforts at eradicating congenital hip dislocation are based on tests for neonatal laxity of the joint. The Ortolani and Barlow tests are difficult to standardize and are not always easy to perform. Jean Williamson[5] (Belfast) reviewed experience gained in an area where about 30,000 live births occur annually, 90% of them in the hospital. The maternity units are widely scattered, and mothers stay for variable periods after delivery. A film of the Ortolani test is shown, and positive signs are demonstrated whenever possible. The importance of complete relaxation cannot be overemphasized. Tight adductors also present a problem. Errors in technic, such as failure to steady the pelvis adequately, may produce false negative results.

Data were reviewed on 783 children treated from birth and on 189 treated after a later diagnosis in 1960–70. Eleven infants were referred for instability not confirmed at the first clinic; subsequent examinations showed instability or slight displacement requiring splintage in abduction. In some cases the sign was unilateral, but the displacement proved later to be bilateral or on the unsuspected side. Many of the cases missed by clinical testing belonged to groups considered to be at high risk. As experience has increased, many more newborns are being referred to the orthopedic clinics with unstable hips, but toddlers with established dislocations are still found.

All children should be rescreened during the 1st year.

(5) J. Bone & Joint Surg. 54-B:13–17, February, 1972.

Those born by breech presentation and those with a family history of dislocation should have radiographs taken in the early months even if clinical tests are negative. Early splintage has failed to prevent established dislocation in about 2.4% of the hips treated.

Congenital Dislocation of the Hip: Development of a Regional Service. Ian G. MacKenzie[6] (Aberdeen, Scotland) reviewed experience gained, in 1960–69 with 1,671 infants with abnormal hips who were followed at least to age 1 year. Barlow's modification of Ortolani's test was used in clinical examination. Radiographic examination is not necessary and may be misleading. In 241 children in whom the Andrén-von Rosen technic was used, 37% of those with positive x-rays had stiff hips without instability on clinical examination, and 70% of those with negative x-rays had definite clinical instability. Some of the negative x-ray results may have been due to faulty positioning.

Of every 100 children with abnormal hips at birth, 78 have unstable hips and 22 have only limited hip abduction. If instability or limited hip abduction is present at 3 weeks, a splint is fitted. Over two thirds of the infants are normal by age 6 weeks. A larger splint usually is needed at this time. The first radiograph is taken at age 3 months, and this is repeated at age 6 months if the femoral epiphyses have not appeared. Clinical abnormality at age 3 months is rare. The Begg splint currently is used. It has been applied in over 2,000 cases. No failure of treatment has been attributed to the splint.

Instability of the hips was found in 813 children at 3 weeks and these children were splinted, as were 348 having limited abduction without clinical instability. Eighty-six children (5%) required inpatient operative treatment, usually because the diagnosis had been missed at birth. Follow-up to age 1 was possible in 93% of the children. In children who were clinically normal at age 3 weeks, delayed appearance of the epiphyses was not associated with late walking. In the group with limited abduction, persistent stiffness was not associated with late walking. Instability was bilateral in 53% of the 813 cases, and 36% of the children had limited abduc-

(6) J. Bone & Joint Surg. 54-B:18–39, February, 1972.

tion as well as instability. Instability persisted at 6 weeks in 13% of the children; this was not associated with late walking. Female infants predominated in the group having operations.

The findings at the 3-week examination were the same as those at 2 days in only 27% of the children. Delayed appearance of the upper femoral epiphysis was not a reliable sign of congenital dislocation. In only 2 children was there delayed appearance of epiphyses beyond age 1 year. The latest time that splintage can be successful is uncertain. Eight children required operation because splints were left off too early. Eight required operation because the mothers refused treatment or removed the splint. Other congenital abnormalities were present in 2% of the children with abnormal hips.

There is almost certainly more than one etiologic factor in congenital hip dislocation. Both genetic and environmental factors may be operative. The condition is not associated with the birth process. Children with familial joint laxity or genu recurvatum should be examined particularly carefully for hip abnormality.

Congenital Dislocation of the Hip: 12-Year Survey. A. R. McKenzie[7] (Auckland) conducted a retrospective survey of patients treated over 12 years for congenital hip dislocation; 97 hips in 61 patients were treated during 1953–65. Diagnosis was most frequent at age 18–24 months. Twenty-three hips were subluxated and 54 dislocated at diagnosis. Initial treatment of 90% of patients over age 6 months was on an Oxford frame modified for rotation of the abducting portions about the long axes. An extra tractional device or "cross pull" was sometimes also used to aid in reduction. Arthrography was done only 4 times. Open reduction was done for inability to obtain or maintain complete reduction of the femoral head into the acetabulum; it was necessary for 30 hips, from 21 of which a limbus was removed. Subluxated hips were treated using a derotational osteotomy only after reduction was achieved on the frame; this was done on 63 hips. In 5 hips the osteotomy was repeated after 1–3 years. Pelvic osteotomy was also done in 2 patients. All patients were in

(7) Australian & New Zealand J. Surg. 41:219–226, February, 1972.

a plaster spica for at least 6 weeks and then in an abduction plaster with the hips and feet free for a variable period. Eighteen patients (27%) had a positive family history. Over half were affected bilaterally.

X-rays were evaluated in respect to a spherical femoral head, the center-edge angle of Wiberg and the neck-shaft angle of the femur. Results were good in 31 hips, satisfactory in 27 and unsatisfactory in 39. Five radiologically satisfactory hips were clinically unsatisfactory. About half the subluxated hips became radiologically good, compared with less than one third of dislocated hips. Clinical results were good in 26 hips, satisfactory in 42 and unsatisfactory in 29. Five patients with clinically good hips had avascular necrosis, as did 6 with satisfactory results and 13 with unsatisfactory results.

Results were satisfactory or better in 60% in this series. Clinical and radiologic assessments did not correspond closely. Better over-all results will be achieved in the future by earlier diagnosis and the observance of extreme gentleness in dealing with the hips of young children. The modified gallows and the Alvic-type of traction-abduction appear attractive alternatives to the Oxford frame, which holds the hip in an extreme extended position, thereby possibly increasing the risk of vascular insufficiency.

► [This is an excellent review of 12 years of experience with congenital dislocation of the hip.—Ed.]

Infantile Coxa Vara is a relatively uncommon congenital hip anomaly of obscure etiology. Joseph D. Calhoun and Guy Pierret[8] (Arkansas Children's Hosp., Little Rock) reviewed 19 cases seen in 1954–69 in which coxa vara was not acquired or associated with other congenital anomalies. The 13 females and 6 males were aged 2–14 years. The mean age at onset of symptoms was 2 years. Both hips were affected in only 2 cases. All children but 2 presented with a painless limp; the other 2 had hip pain. The trochanter on the affected side was usually elevated. Some limitation of motion frequently was encountered; decreased internal rotation was usual in very young patients. The affected limb was always shorter, the average discrepancy being 1.2 in.

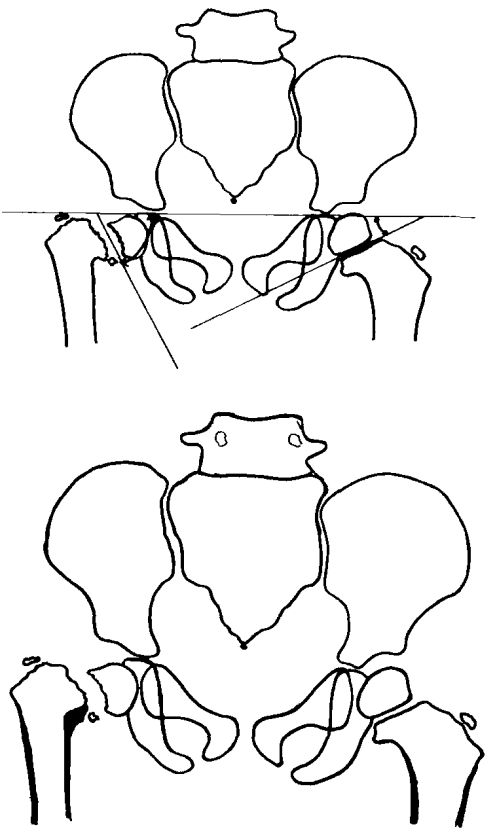
(8) Am. J. Roentgenol. 115:561–568, July, 1972.

The most consistent radiographic finding was bending of the neck into a varus position. The epiphysial line was in a vertical position and widened in relation to the unaffected side (Fig. 3). A triangular fragment was present at the medial inferior portion of the femoral neck in all patients under age 9. In older untreated patients the femoral head and ace-

Fig. 3 (top). – Bending of femoral neck into varus position. The severity is directly related to the verticality of the epiphysial plate. Right epiphysial angle from the horizontal plane is 62 degrees; left angle is within normal at 25 degrees.

Fig. 4 (bottom). – Signs of infantile coxa vara include slender proximal femoral shaft, thickened compression system medially, parallel medial and lateral cortex of proximal shaft and compression system protruding into femoral neck with neck bent over compression system.

(Courtesy of Calhoun, J. D., and Pierret, G.: Am. J. Roentgenol. 115:561–568, July, 1972.)



tabulum were severely deformed. All patients but 1 exhibited a slightly shallow acetabulum. The 2 oldest patients had CE angles indicative of dysplasia. The femoral shaft on the affected side was usually more slender than normal. At the junction of the medial cortex of the upper shaft and the inferior cortex of the neck the compression system was thickened. The medial and lateral cortices of the proximal shaft paralleled each other and the thickened medial cortex protruded into the femoral neck (Fig. 4). The epiphysis of the greater trochanter tended to sit on top of the upper femur rather than on the sloping superolateral surface of the femoral neck.

Pseudarthrosis may occur in untreated cases, and early surgery is indicated to achieve a painless, fully mobile hip without shortening. The goal is to correct the varus deformity and re-establish the proper length tension of the abductor muscles. If the epiphysial plate can be placed in a horizontal position, the neck defect should heal. Indications for surgery include an angle of 100 degrees or less, a vertical neck defect or progressing varus. Seventeen patients underwent subtrochanteric osteotomy with metallic internal fixation. Although results were generally satisfactory, 1 patient had bilateral osteotomies without apparent benefit. Three had recurrence; 2 were reoperated on but only 1 had a good result. Another patient acquired pseudarthrosis at the osteotomy site and needed reoperation and bone grafting.

Apparent Congenital Absence of the Tibia with Lethal Congenital Cardiac Disease. Congenital absence of the tibia has been well documented, but its occurrence with polydactyly of the lower extremity is extremely rare. Avery D. Pratt, Jr.[9] (Univ. of Cincinnati) has encountered 2 cases of absence of the tibia representing the common and rare forms.

Female infant, aged 5 days, the product of an uneventful pregnancy in which the mother had taken only aspirin during early pregnancy, had bilateral talipes equinovarus with shortening and external rotation of the right leg below the knee. Six digits were present on each foot, with partial syndactyly of two big toes bilaterally. A grade 3/6 systolic murmur was heard along the left sternal border and an ECG indicated left ventricular hypertrophy. X-rays showed absence of the right tibia with bilateral duplication of the first ray, synmeta-

(9) Am. J. Dis. Child. 122:452–454, November, 1971.

tarsia of the first ray on the left and polymetatarsia of the first ray on the right. Ten ribs were present on both sides, with deformity of both 10th ribs. The upper lumbar vertebral bodies were of increased height and reduced anteroposterior diameter, and there was a hemivertebra at the L3 level. A right aortic arch was present, with a high ventricular septal defect and filling of the pulmonary arteries from the aorta. The child was admitted with cyanosis and respiratory distress at age 6 weeks and died in 48 hours. Postmortem x-rays showed the interval development of a distal right femoral epiphysis and development of a rudimentary tibial ossification center in the midportion of the right lower leg. Autopsy showed a truncus arteriosus with a mirror image, branching right aortic arch. A four-cusp aortic valve and a high ventricular septal defect were present.

The second patient was a male infant whose mother had taken an antinausea medication in the 3d trimester. He had complete absence of ossification of the right tibia and absence of a right metatarsal (probably the first ray) and the navicular ossification center. The thumbs were absent and lobster-claw deformity and four rays were present bilaterally in the upper extremities.

Absence of the tibia may be defined as a paraxial hemimelia. If a foot anomaly exists, it is always in the form of diminution or absence of a corresponding vertically oriented limb bud. Only 6 cases of combined paraxial deficiency and a more peripheral paraxial duplication were found in the literature. Both heritable and sporadic cases of congenital absence of the tibia have been reported. No previous reports of associated congenital cardiac disease were found.

► [This article, in addition to recording a rare instance of complete paraxial intercalary tibial hemimelia with polydactyly, is noteworthy because it reemphasizes the need to search for multisystem anomalies whenever a congenital abnormality is found.—H. A. Peterson.]

Early Operative Treatment of Congenital Club Foot. Although opinions regarding optimal primary treatment of congenital club foot remain divergent, most authors recommend conservative treatment at an early age. Late results of early operative treatment have not been reported. E. Somppi and M. Sulamaa[1] (Univ. of Helsinki) made a follow-up study of 54 patients who, during 1959–66, were operated on before age 2 weeks for club foot; 69% of the patients were males. There were 33 bilateral cases; thus 87 feet were operated on.

Medial release combined with achillotenotomy was carried out. The talonavicular joint was widely opened. The deltoid

(1) Acta orthop. scandinav. 42:513–520, 1971.