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TERATOLOGIC CONGENITAL DISLOCATION OF THE HIP

Report of Two Cases

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The incidence of teratologic dislocation of the hip is about 0.04 per thousand. Teratologic CDH is usually described together with other anomalies, such as arthrogryposis.

Quite different opinions about the diagnostic criteria are found in the literature; some of these are reviewed in this report. Two cases of teratologic CDH with no other anomalies are also reported.

Computer tomography was used to confirm reduction in plaster when conventional radiologic examination gave ambiguous results.

Key words: computer tomography; congenital; dislocation; hip; teratologic

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Congenital dislocation of the hip, CDH, is usually divided into two main groups: typical and teratologic, of which typical is the more common. Von Rosen (1962) in Sweden, found 1.7 per thousand typical CDH, among 24 000 newborns, using the Ortolani sign as diagnostic criterion. In the same material he found one case of teratologic CDH, that is 0.04 per thousand. Artz et al. (1975) also found an incidence of 0.04 teratologic CDH per thousand. Of 38 000 newborns in our district between 1972 and 1981, we have found one case of teratologic CDH, or 0.03 per thousand.

Factors that in general are included in the concept of teratologic CDH are: 1) development in utero, 2) uni or bilateral affection, 3) characteristic resistance to closed treatment, 4) pathologic findings comparable with those in untreated typical CDH after several years of life, 5) frequent association with other anomalies as, Arthrogryphosis Multiplex Congenita, Larsen's syndrome, Goldenhairs syndrome, Hallerman-Streiff syndrome, some vertebral deformities and clubfeet. Some authors even claim that the diagnostic criterion is presence of other anomalies.

According to Jungmichel (1963) Paletta in 1820 demonstrated severe changes in a luxated hip of a child that died soon after birth. Larsen et al. (1950) described CDH in combination with other dislocations and with facial abnormality. Jungmichel (1963) presented 15 cases of CDH, one of which he called teratologic CDH because the child also had other anomalies. As a diagnostic criterion he added normal shape of the acetabulum. Stanisavljevic & Mitchell (1963) described two cases which they called "prenatal" hip dislocation. None of the children had any other anomalies. Hommel (1966) described several cases of teratologic CDH, most, but not all, of them combined with other anomalies. His

criteria were a) impossibility of closed reduction and b) normal shape of the acetabulum. Crasselt (1968) also regarded a normal acetabulum as a criterion in teratologic CDH, and he claimed that the child has to have other anomalies to qualify for the diagnosis. Milgram & Tachdjian (1976) described a dissection of a 10-month-old child with teratologic CDH and other multiple anomalies. Dissection showed severe changes in the hip with a small and shallow acetabulum filled with fibrous tissue and fat. Katz (1980) suggested that teratologic CDH stems from a broad spectrum of medical conditions and that its most characteristic feature is resistance to therapy.

CASE REPORTS

Case 1

A girl with birthweight 3410 g, delivered by Cesarean Section because of breech-presentation. At birth she had a clinically fixed dislocation of the left hip with abduction limited to 60 degrees. The dislocation was also confirmed by radiologic examination. She was treated with traction for 10 days and then the hip was reduced under general anesthesia, reaching a symmetrical abduction of 100°. The control in plaster showed that the left femoral head was located somewhat cranially but it was not thought to be luxated (Figure 1). Computer tomography showed that the hip was luxated dorsally (Figure 2). The procedure was repeated with traction and a new attempt was made to reduce the femoral head into the acetabulum. This time, as well, the hip was thought to be properly reduced but the plain radiologic examination was again doubtful and computer tomography showed that the dislocation remained unchanged. When the girl was 4 weeks old, surgery was performed. Capsulotomy, excision of the hypertrophic ligamentum teres, and lengthening of the iliopsoas were necessery to make the reduction of the femoral head into the acetabulum possible. The acetabulum was small and dysplastic and the caput femoris seemed large in proportion. The hips were immobilized in a plaster-cast for 6 months.

The girl is now 2 years old. She walks and runs in a completely normal way. The hips are normal in physical and radiologic examination (Figure 3) and we have not found any other abnormalities in the child.

Case 2

A girl with birthweight 2530 g, delivered by Cesarean Section because of breech-presentation. Her father has Osteogenesis Imperfecta Tarda. At birth she had fixed bilateral dislocation of the hips. Attempt at closed reduction was performed after a period of traction. Because of uncertainty as to whether the hips were still luxated she was treated at hospital at 4, 6 and 9 weeks of age. After each change of the plaster-cast the hips were felt to be properly reduced but at every radiologic

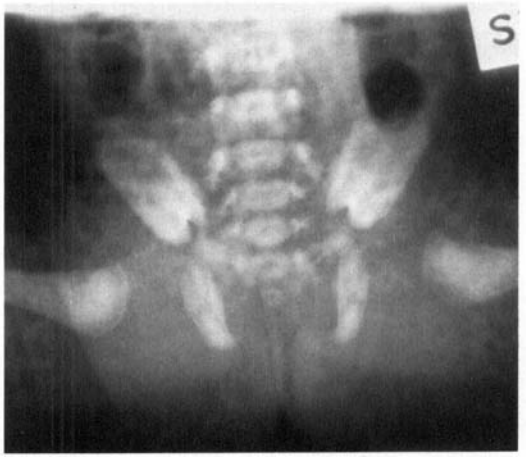

Figure 1. Teratologic congenital dislocation of the left hip in a 10-day-old girl. Examination in plaster after an attempt at closed reduction. The left proximal femur is located somewhat more cranially.

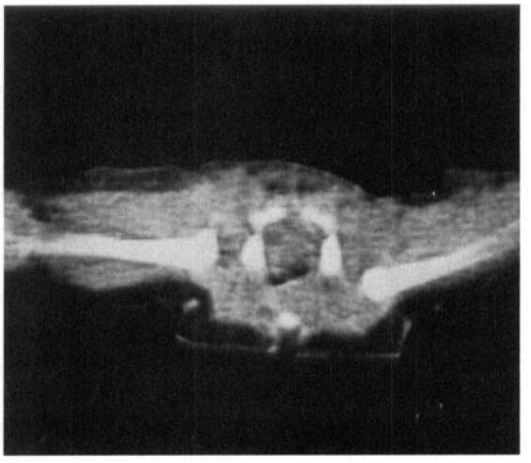

Figure 2. Computer tomography shows that the left hip is luxated dorsally. This examination was done after the radiologic examination in Figure 1, while the patient still had the same plaster.

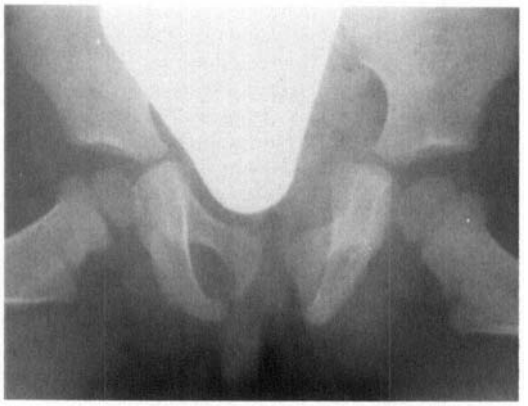

Figure 3. The same patient as in Figures 1 and 2 at 2 years of age. Radiologic examination is now normal and the girl runs and walks normally.

examination doubt arose. At the 9-week control the reduction was accepted.

When the child was 4 months old the family moved to Stockholm and she was admitted to this unit for change of the plaster-cast.

The radiologic examination showed that both hips were luxated. They were completely fixed after a new period of traction. Surgical reduction was performed bilaterally. Capsulotomy, excision of the hypertrophic ligamentum teres, and lengthening of the iliopsoas proved necessary before reduction of the femoral head into the acetabulum was possible. The acetabulum was small and dysplastic, partially filled with fatty tissue and the head seemed too large to fit into the acetabulum. Computer tomography was used to verify the reduction. The hips were immobilized in a plaster-cast for 6 months.

The girl is now 1½ years old. She walks without support and the hips are stable and show normal mobility.

DISCUSSION

In most reports teratologic CDH occurs together with other anomalies. Several authors consider multiple anomalies as a criterion for diagnosis. It is clear that several different medical conditions can result in teratologic CDH but it has also been described as an isolated condition. Two cases of isolated teratologic CDH have been presented.

In the concept "teratologic CDH" we consider a CDH in which reduction requires surgical treatment. According to our experience for reduction of the femoral head into the acetabulum not only division of the capsule but also lengthening of the iliopsoas is needed. A typical CDH can on the other hand be easily reduced up to 2 years of age after pretreatment by traction followed by adductor myotomy. Jungmichel, Hommel and Crasselt suggest that the acetabulum should be "normal" (normale pfannenverhältnisse) for the diagnosis teratologic CDH but we cannot see any motivation for this criterion.

Computer tomography has been shown to be of invaluable diagnostic help in one of our cases. Presumably it can be of great value in many cases when evaluation of the reduction with conventional radiologic examination is difficult, especially when the hips are fixed in plaster. Computer tomography cannot, however, be recommended as a routine method, since the radiation-dose is considerably higher than with conventional radiologic examination.

An estimation of the gonadal dose in an examination with computer tomography gave 3.8 ± 0.6 mGy for boys (= skin dose) and 2.2 ± 0.2 mGy for girls. With conventional radiologic examination (one picture) the gonadal dose was 0.10 ± 0.02 mGy for boys and 0.037 ± 0.007 for girls.

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