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OBSERVATIONS IN CHILDREN WITH CONGENITAL DISLOCATION OF THE HIP

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In 111 children who were diagnosed and treated for CDH, certain variables concerning their vital statistics, the laxity of their joints and circumstances at birth were recorded. It was observed that breech presentation, birth rank one and female preponderance are common traits in children with CDH, who also become taller and heavier than control children. Anomalies found at birth or later were more common among these children, in particular inguinal hernia. Also, joint laxity was more common in these children. An increased incidence of CDH was found among siblings.

Key words: congenital; dislocation; hip; inguinal hernia; joint laxity; foetal presentation

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In a report on the effect of early diagnosis it was established that in the city of Malmö almost all cases of congenital dislocation of the hip (CDH) could be diagnosed at birth. In a follow-up study on the late results of early treatment it was proved that treatment using the von Rosen splint resulted in completely normal hips. Some information included in the case histories and the vital statistics of the children involved in the study on the effect of early treatment will be presented briefly.

MATERIAL AND METHODS

During the years 1956–1964 a total of 30,280 live births were recorded in the city of Malmö. The incidence of CDH and of undiagnosed cases appears in Table 1.

Of 119 cases diagnosed within the first few days of life and treated using the von Rosen splint, there was one child with arthrogryposis, one with meningomyelocele, one with Bonnevie-Ullrichs syndrome and one with cerebral palsy. Another child died later on of cardiac failure and three had emigrated leaving 111 typical cases of CDH for the follow-up examination. The information was collected at the re-examination, from the maternity records and from interviews with child and parents.

For the study of joint laxity the same number of age and sex matched control children, taken from the city schools in Malmö, were investigated. Joint laxity was estimated according to five variables (Figure 1). It was established whether or not the children were able to:

1. Hyperextend the elbow joint more than 10°.
2. Hyperextend the thumb and flex the wrist until the thumb touched the forearm.
3. Dorsiflex the fingers and the wrist to make the fingers parallel to the forearm.
4. Hyperextend the ankle joint more than 45°.
5. Hyperextend the knee joint more than 10°.

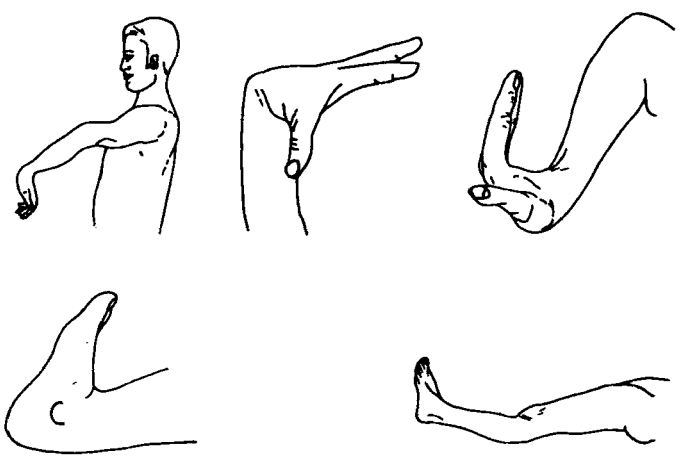

Figure 1. Demonstration of joint laxity.

RESULTS

Sex ratio: The ratio girls:boys in this series was 5.2:1. The average for Malmö is 1.0:1.0.

Foetal presentation. Thirty-one per cent of the girls and 33 per cent of the boys were born in the breech presentation as compared with the average which is 3.7 per cent in Malmö. Of the children born in the breech presentation, 78 per cent were first-born. One girl was delivered by Caesarean section. One girl was number one of a pair of dizygotic twins (number two, a boy, was unaffected).

Birth rank. Sixty per cent of the patients were first-born whereas the average for Malmö is 45 per cent. The difference was significant ($0.01 > P > 0.001$).

Birth weight. The birth weight in CDH girls was 3,457 ± 530 g. In a random control sample from Malmö, the birth weight of 100 girls was 3,352 ± 465 g. The difference was not significant ($0.2 > P > 0.1$).

Height and weight. In this study only girls were included. The height and the weight were compared with a control sample (Karlberg et al., in preparation). There were significant differences between CDH girls and controls in that CDH girls were somewhat taller and heavier (Figures 2 and 3). Since Willner (1974) has demonstrated that heights in groups of children from various parts of Sweden agree well, the comparison with Karlberg et al. is justified.

Menarcheal age. The menarcheal age in the oldest 19 girls in the CDH series was 12.3 ± 1.4. This means, however, that in most girls the menarcheal age is not known and that a late menarche must be expected in some of the remaining individuals. Therefore, the difference between these data and data on menarcheal age from a contemporary material (Andersen 1968), 13.3 ± 1.3 years, is not necessarily significant.

Seasonal variation. The incidence of diagnosed CDH in newborns was increased in the period August–November but the deviation in this series was not significant (Figure 4).

Table 1. The incidence of diagnosed and missed cases of CDH in Malmö during the period 1956–1972.

	Number	Incidence/1000
Total live births	30,280	
Children with CDH	121	4.00
Missed cases	2	0.07

Heredity. An attempt was made to estimate the prevalence of CDH in the families of the CDH children. Only an interview with one of the parents was

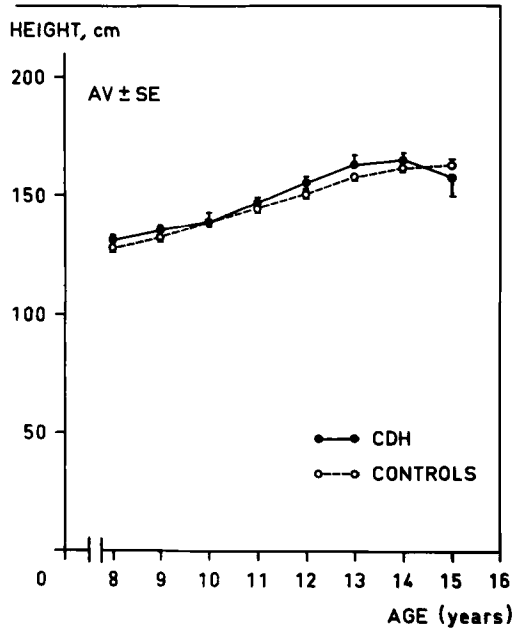

Figure 2. The height of 91 CDH girls compared with 554 controls.

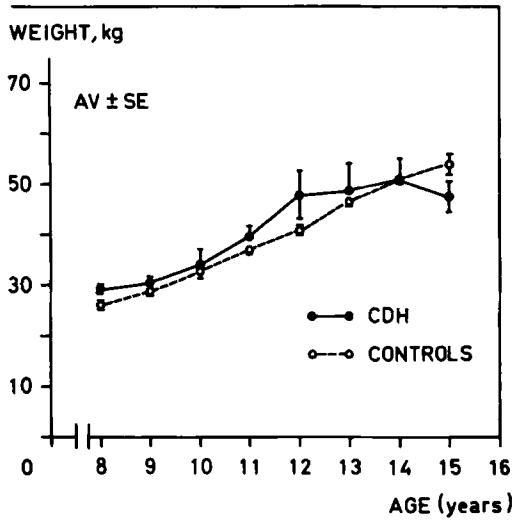

Figure 3. The weight of 91 CDH girls compared with 554 controls.

made. A number of children of the same generation had been treated after early diagnosis in these families: Five siblings, four first cousins and five second cousins. In the previous generation CDH was known in one mother and suspected in a few more cases who had obviously had hip conditions. The CDH children were known to have 136 siblings with an expected risk of CDH of less than one case. Therefore, the five cases observed indicate an increased risk, whereas the observations in more distant relatives add

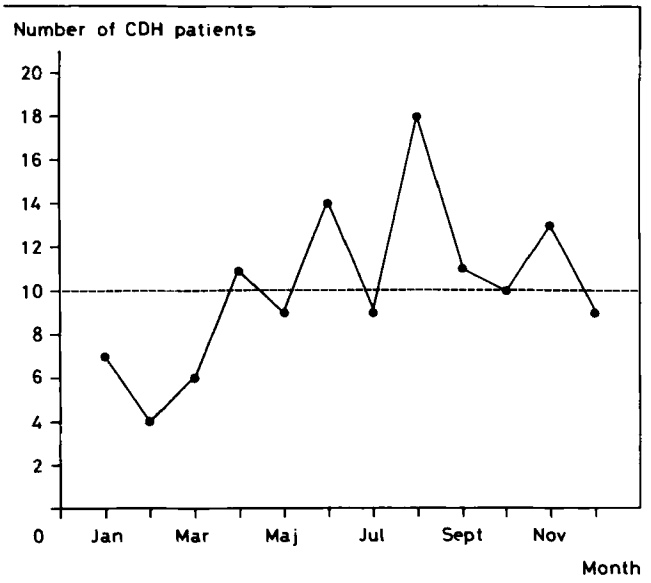

Figure 4. Seasonal variation in birth of CDH children.

no further proof to the hypothesis of heredity.

Concomitant anomalies. During the years 1956–1964 a total of 121 children were diagnosed as having CDH. Among these a series of anomalies were recorded (Table 2).

Table 2. Concomitant anomalies in 121 children with CDH.

Anomaly	Number Boys	Girls
Congenital heart failure	1	
Testes not descended	1	
Meningomyelocele		1
Arthrogryposis		1
Bonnevie-Ullrichs syndrome		1
Torticollis		1
Pubertas praecox		1
Inguinal hernia		
bilaterally		2
unilaterally	1	2

None of the conditions listed in Table 2 is expected to occur in more than one per thousand of the population. Therefore, even excluding meningomyelocele, arthrogryposis and Bonnevie-Ullrichs syndrome the number of anomalies significantly exceeds the expected. One diagnosis, inguinal hernia, is *per se* more frequent than expected.

Joint laxity. In all but one of the variables there was a highly significantly increased incidence of signs of joint laxity in the CDH children (Table 3).

DISCUSSION

The sex ration in this study, 5.2:1, is in agreement with that of earlier investigators. Idelberger (1951) reported a ratio of 5.4:1 based on 45,611 cases and Palmén (1961) a ratio of 4.1:1 in 894 neonatal cases and 5.8:1 in 1,486 mainly late-diagnosis cases. A similar difference in sex ratio between neonatal and late-diagnosis was reported by Wynne-Davies (1970) and Bjerkreim (1974).

Breech presentation has been referred to as an environmental factor in the aetiology of CDH (Record & Edwards 1958, Andrén 1961). Several investigators (Wilkinson 1963, 1972, Carter & Wilkinson 1964 a, Fettweiss 1973) claimed that the risk of CDH is particularly increased in breech malposition. The association of breech presentation and birth rank was reported by Record & Edwards (1958), von Rosen (1959) and Andrén (1962). If, in the present series, cases born in the breech presentation are excluded, no primogeniture effect remains. This has also been reported by Bjerkreim & van der Hagen (1974).

A few investigators have observed deviations from the average in birth weight in CDH children (Record & Edwards 1958, Bjerkreim & van der Hagen 1974, Artz et al. 1975). However, the investigators do not agree as to the direction of this deviation. In the present study the difference was not significant.

As for the concomitant conditions the findings in this study are in agreement

Table 3. Joint laxity in patients with CDH and controls.

Joint	CDH children		Controls		
	Positive	Negative	Positive	Negative	
Thumb	65	45	52	58	$P > 0.5$
Wrist	49	61	22	88	$P < 0.001$
Elbow	40	70	6	104	$P < 0.001$
Knee	51	59	15	95	$P < 0.001$
Ankle	34	76	0	110	$P < 0.001$

with those of previous investigators. Concomitant malformations have been reported as occurring in CDH children to an increased degree by Record & Edwards (1958), Wynne-Davies (1970) and Bjerkreim & van der Hagen (1974). The data of the present study are not conclusive but taken together with the observations of other investigators they seem to indicate such an increased prevalence. The prevalence of inguinal hernia is of special interest. Inguinal hernia is extremely rare in girls (Knox 1959). In this series the expected number is very close to zero but four cases of hernia were observed. Similar small increases in the prevalence of hernia in CDH children have been observed by Wynne-Davies (1970) and Bjerkreim & van der Hagen (1974). Taken together these studies indicate a significantly increased risk of inguinal hernia in CDH girls. Wagh et al. (1974) found a decreased amount of collagen in the anterior rectus sheath in patients with direct inguinal hernia. Similarly Fredensborg & Udén (unpublished data) have demonstrated changes in the collagen of the umbilical cord in children with CDH.

It has in the past been demonstrated that there is a relationship between a general joint laxity and CDH. Massie & Howorth (1951) were the first to report the combination of general joint laxity and CDH. This has been confirmed by Wilkinson (1963), Carter & Wilkinson (1964 b) and Howorth (1965). Wynne-Davies (1970) demonstrated the occurrence of joint laxity also in CDH relatives. The previously suggested relationship between general joint laxity and CDH is strongly supported by the findings in the children of the follow-up study.

Most of the data on CDH children in the present study are in agreement with those of other investigators and merely confirm these findings as well as the identity of the sample. One observation, however, has not been previously presented, namely, the fact that CDH girls are somewhat taller and heavier than their controls. These findings cannot be interpreted at present but might be caused by an earlier maturation in girls born with CDH.

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