

Screening of neonatal instability and of developmental dislocation of the hip

A SURVEY OF 132 601 LIVING NEWBORN INFANTS BETWEEN 1956 AND 1999

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etween 1956 and 1999, 132 601 living children Between 1750 and 1777, 202 instability of the hip. All late diagnosed patients have been followed and re-examined clinically and radiologically. During the first years of screening, less than five per 1000 living newborn infants were treated. This figure increased to 35 per 1000 in 1980, but later diminished again to about six per 1000 annually after 1990. The number of referred cases decreased from 45 per 1000 in 1980 to between 10 to 15 per 1000 from 1990. During the period of high rates of referral and treatment a larger number of paediatricians were involved in the screening procedure than during the periods with low rates of referral and treatment. Altogether 21 patients (0.16 per 1000) with developmental dislocation of the hip were diagnosed late, after one week. At follow-up, 18 were free from symptoms and 15 considered to be radiologically normal.

J Bone Joint Surg [Br] 2002;84-B:878-85. Received 3 April 2001; Accepted after revision 8 August 2001

The recommendations for the early diagnosis and treatment of neonatal instability of the hip (NIH) made by Putti^{1,2} and Ortolani³ have been widely accepted. In Sweden, the early results were presented by von Rosen^{4,5} and Palmén.⁶ Fredensborg^{7,8} re-examined von Rosen's material and found an incidence of 0.07 per 1000 of late diagnosed hips between 1956 and 1972 (58 759 newborn infants).

Early screening and treatment appeared to have solved the problem of developmental dislocation of the hip (DDH). Unfortunately, the incidence of late diagnosed hips increased in Malmö in 1980. It therefore seemed appropriate to start a comprehensive, prospective study of the screening procedures, the incidence of referred, treated and

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late diagnosed hips and for some years also, the ethnic origin of the newborn infants. The late diagnosed hips which presented between 1956 and 1999 have also been re-examined.

The health-care and follow-up systems for hips in Malmö are such that it is unlikely that failures have escaped detection. Detailed census data make an incidence study highly reliable.

Patients and Methods

In this study we have defined NIH as a condition in which the hip is dislocated, dislocatable or unstable at examination during the first five days after delivery. No cases were diagnosed between days 5 and 7. Teratological and neuromuscular cases were excluded. Detailed census data, maternity records and radiographs were available. Late-diagnosed DDH is defined as a dislocated or dislocatable hip diagnosed after the age of one week.

Screening for NIH started in Malmö in 1956 (Fig.1). The routine has changed somewhat over the years (Table I). Before 1990, patients with differing degrees of instability, as well as those with dislocatable hips, were referred to the orthopaedic clinic where it was decided whether or not to treat the child. Since 1990, those with hips which were clearly dislocated or dislocatable were still referred directly to the orthopaedic clinic and treated for three months. Patients whose hips were judged to be clinically unstable, but not dislocatable, were examined using a dynamic anterior ultrasonic technique.^{9,10} This was carried out in nearly all infants on the day of referral. The radiologist and the paediatric orthopaedic surgeon measured the degree of instability together, and the orthopaedic surgeon carried out the dislocation-reduction manoeuvre. If the femoral head was dislocatable on sonography the child was treated in a von Rosen splint for three months. If the femoral head was not dislocatable, but the sonographic displacement of the head exceeded one quarter (3 mm) of its diameter, the patient was treated for six weeks in a von Rosen splint. If the displacement of the femoral head, found at sonography, was less than one quarter (\leq 3mm) of its diameter, the patient was not treated. Before 1989, nearly all patients whose hips were unstable or dislocatable, were treated in a von Rosen splint for three months. In late 1959 and early

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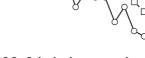




Fig. 1

Graph showing the number of hips with suspected NIH per 1000 treated between 1956 and 1972 and 1980 and 1999, and the number of hips with suspected NIH per 1000 referred from 1980 to 1999. Figures on the horizontal axis represent the number of late-diagnosed DDH patients annually

Years	Hip examination	All newborn infants examined	Tests	
1956 to 1963	Twice a week	Once (by an experienced doctor)	Ortolani's test	
1964 to 1979	Every day except on Sundays and holidays	Most twice within 24 to 48 hours and at discharge from the maternity ward (at least once by an experienced doctor)	Barlow's dislocation- reduction test, sometimes with the pelvis stabilised with one hand	
1980 to 1989	Every day except Sundays and holidays. Special hip examination on Tuesdays and Fridays	At least once. Most twice (those not discharged before Tuesdays and Fridays)	Barlow's dislocation- reduction test, sometimes with the pelvis stabilised with one hand	
1990 to 1999	Every day except Sundays. Thus also on holidays if not a Sunday	At least once. Most by experienced neonatologist or experienced paediatrician	Barlow's dislocation- reduction test, sometimes with the pelvis stabilised with one hand	

Table I. Examination for neonatal hip instability in Malmö during different periods

1960, the period of splintage was shortened. After one case of redislocation, however, the three-month policy was resumed. During 1989, the duration of treatment was changed. Unstable hips were treated for six weeks only, whereas the dislocatable hips were treated for three months.

55 50

45

40

Number of hips per 1000

-0- Treated · □ - Referred

All patients, whether treated or untreated, were examined clinically one week after the first orthopaedic examination and again after three months and at one year. Radiological examination was also carried out at least at three months and one year. Patients showing avascular necrosis or acetabular dysplasia at the one-year radiological examination, continued with regular follow-up. Initially, radiological examination was routinely repeated at three and six years. This was discontinued when it was found that those who were normal when aged three years were normal when aged six, and those who were normal at one year were normal at three years. After 1984, once the dislocation-



Fig. 2

Case 5. Radiograph of a 24-year-old woman with a left-sided DDH showing type-IV avascular necrosis. Closed reduction had been performed at the age of two months, followed by treatment in a von Rosen splint. Neither skin traction nor tenotomies preceded the reduction.



Fig. 3

Case 4. Salter innominate osteotomy was carried out at the age of 13 years for persistent acetabular dysplasia.

reduction manoeuvre had been carried out, plain radiographs were not used as a complement to screening.

Between 1998 and 1999, newborn infants were classified with regard to the ethnic origin of their parents. The figures were added to those from 1990 to 1997 and the group with both parents of Swedish origin were compared with those with parents of foreign origin.¹¹

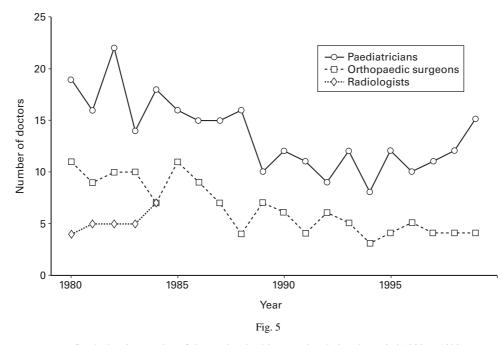
The treatment of children who were diagnosed late has varied somewhat over time. During the early years treatment with skin traction and adductor tenotomy before closed reduction was rare. Since then the treatment has been uniform¹² as described by Morel.¹³ In late diagnosed hips, at the last follow-up examination, the leg length was measured from the anterior superior iliac spine to the medial malleolus and, after Salter innominate osteotomy,

from the umbilicus to the medial malleolus. We used the criteria of Salter, Kostuik and Dallas¹⁴ and Kalamchi and MacEwen¹⁵ for the diagnosis and classification of avascular necrosis. Anteroposterior radiographs of the pelvis and both hips, with the legs parallel and in the neutral position, were used for radiological measurement.¹⁶ Only radiographs with an obturator index between 0.56 and 1.8, in patients up to five years of age and with a symphysis-os ischium angle within the defined lines were used.¹⁷ The percentage of migration was calculated, with $\geq 20\%$ being considered to be abnormal.¹⁸⁻²⁰ We calculated the acetabular angle according to Almby and Lönnerholm,²¹ the centre-edge (CE) angle according to Wiberg²² and the spherical index according to be normal.²³ The suggestion by Lempicki,





Case 3. In this patient with bilateral involvement the left hip redislocated. Four attempts at open reduction followed by two proximal femoral osteotomies failed and arthrodesis was necessary.



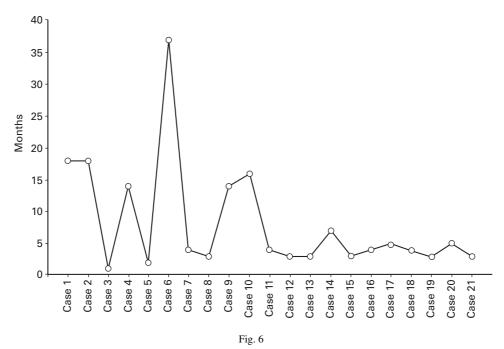
Graph showing number of doctors involved in screening during the period 1980 to 1999.

Wierusz-Koslowska and Kruczynski²⁴ that an angle of 15° to 20° should be considered to be uncertain, was taken into consideration. A spherical index <35 was considered to be abnormal.^{7,8} We used Severin's²⁵ definition concerning the CE angle. For patients aged six to 13 years >19^{\circ} is normal and 15^{\circ} to 19° is uncertain, while for patients aged ≥14 years >25° is normal and 20° to 25° is uncertain. The size of the femoral head was defined as the maximum width of the head multiplied by its maximum height measured from

this line.^{7,8} We classified the radiological assessments of the results according to Severin.²⁵

We used Student's *t*-test, the chi-squared test and Wilcoxon's matched pairs for statistical analyses. A p value of less than 0.05 was considered to be significant.

Complications of primary splintage. To our knowledge there were only two failures of initial treatment.⁷ One hip redislocated after two weeks. The failure was discovered after a further two weeks and plaster immobilisation con-



Age at diagnosis of late diagnosed DDH between 1956 and 1999.

tinued for ten months because of persistent instability. Radiological examination at the age of 11 years showed normal hips. One hip was noted to be subluxed at follow-up at one year and was treated in a von Rosen splint for three months. Radiographs at the age of eight years showed normal hips.

Avascular necrosis occurred in three hips, one type-IV (case 5) and two type II. In one patient (Fig. 2) closed reduction was not preceded by skin traction and adductor tenotomy, resulting in leg shortening due to avascular necrosis. Since this patient had left Malmö epiphysiodesis was not done at the correct time. As a result femoral lengthening was necessary at the age of 22 years to correct 4 cm of femoral shortening.

In one patient (case 4) a Salter innominate osteotomy was carried out at the age of 13 years (Fig. 3) and in another at the age of 5 years 10 months, for acetabular dysplasia. In one patient with bilateral involvement (case 3) the left hip redislocated and four attempts at open reduction were made, followed by two proximal femoral osteotomies, hip arthrodesis, supracondylar femoral osteotomy and the removal of the plate (Fig. 4). A shelf operation was carried out on the right hip followed by distal femoral epiphysiodesis and subsequent removal of staples.

Results

Early diagnosis and treatment. During the early years of the screening programme one paediatrician and one orthopaedic surgeon were largely responsible for the screening procedure. Later, the number of doctors involved in the screening increased dramatically so that in 1982, 22 paediatricians were involved (Fig. 5). Between 1956 and 1999, 132 601 living children were born in Malmö and screened for NIH. The number treated in a von Rosen splint has varied over the years (Fig. 1). During the early years of screening, <5 per 1000 newborn infants were treated. This figure increased to 35 per 1000 in 1980. It then diminished and has remained about 6 per 1000 since 1990. The number of referred patients has also decreased from 45 per 1000 in 1980 to between ten and 15 per 1000 from 1990.

Since 1990, all newborn infants with unstable, but not dislocatable, hips have been examined using a dynamic anterior ultrasound technique.²⁵ The number referred from the paediatric to the orthopaedic clinic, the number of treated as well as those not treated, and also the number of late diagnosed patients were analysed and a comparison made between two groups, namely, those treated or referred between 1980 and 1989 and those between 1990 and 1999 (Table II). The number of referred patients decreased from 31.7 per 1000 to 12.9 per 1000, the number of treated patients from 16.9 per 1000 to 6.6 per 1000 and the number of late diagnosed patients from 0.51 per 1000 to 0.07 per 1000.

Newborn infants of entirely Swedish parentage were compared with those with both parents of foreign extraction (Table III). There was a higher incidence of referred, treated, dislocatable, unstable and untreated hips in the Swedish group, but this was only statistically significant for the referred hips (p = 0.001).

The incidence of avascular necrosis in hips which were splinted during the period between 1956 and 1964 was 1 in 111 and in the period 1990 to 1999, 1 in 198. Both hips

 Table II.
 Number of newborn infants in Malmö between 1980 and 1989 and 1990 and 1999, referred to the orthopaedic clinic with suspected neonatal hip instability and late diagnosed DDH

Years	Number of newborn infants	Suspected instability		
		Referred	Treated	Late diagnosed DDH
1980 to 1989	25 468	807	432	13
1990 to 1999	29 799	386	198	2
			Dislocatable 151	
			Unstable 47	

 Table III.
 Comparison of 18 035 newborn infants with both parents of Swedish extraction and 7543 with both parents of foreign extraction

	Total (per 1000)	Parents of Swedish extraction (per 1000)	Parents of foreign extraction (per 1000)	p value
Referred	13.3	14.8	9.8	0.001
Treated	7.0	7.7	5.2	0.026
Dislocatable	5.4	5.9	4.2	0.094
Unstable	1.5	1.8	0.9	0.114
Not treated	6.4	7.1	4.6	0.024

recovered spontaneously. A total of 21 patients (20 girls, 1 boy, 8 right, 13 left, 2 bilateral) with DDH were diagnosed late, i.e. after the first week (0.16 per 1000). Most (12 children, 0.6 per 1000) were diagnosed in the period between 1980 and 1986. One patient was considered to be unstable and one dislocatable by the paediatrician at the neonatal examination and referred to the orthopaedic clinic where the diagnosis could not be confirmed by the paediatric orthopaedic surgeon and as a result these two children were not initially treated. Both patients were examined twice by the paediatric orthopaedic surgeon, and one was also examined by ultrasound on the day of referral and after one week. These two patients were later diagnosed at the routine follow-up examination at three months of age.

In five patients instability, click or 'crepitations' were noted by the paediatrician at the first examination, but they were not referred to the orthopaedic clinic. In two of these, the hips were considered to be normal at a second examination on the second and third days after birth. In the other three, no second examination was done.

For the 21 patients with late diagnosis, the first paediatric examination was during the first day after delivery in 15 patients, within two days in five and within three days in one. The second examination was between the first and sixth days in 15 patients and further examinations in four patients on the third to the fifth day. The first paediatric examination was by different doctors in all except four patients. Two doctors had examined two patients each; one of the most experienced had examined five of the missed cases either on the first or second examination. Long experience does not, therefore, guarantee 100% successful screening. No conclusion could be drawn with regard to the day of the week in which the child was born or whether the child was born immediately before or during a holiday period.

Late diagnosis and treatment. The mean age of late

VOL. 84-B, No. 6, AUGUST 2002

diagnosis for the whole study period was eight months (1 to 37). Two-thirds of patients were diagnosed before the age of six months. Before 1983, the mean age at diagnosis was 13 months (1 to 37) and after 1982, four months (3 to 7) (Fig. 6).

Using Severin's method²⁵ for radiological classification, 16 hips were rated as group 1A, two as 2A, two as 3, two as 4 and one as 6. One previously undislocatable hip should be considered as group 1B.

Discussion

Between 1977 and 1979 the number of patients with NIH referred from paediatric to orthopaedic departments in Sweden varied from 2 to 46 per 1000 live births.²⁶ In Malmö it decreased from 45 per 1000 in 1980, when 19 paediatricians were involved in the diagnostic procedure, to 40 in 1000 in 1994, when eight paediatricians were concerned, two of whom had the main responsibility. In spite of the high number of referred patients with suspected NIH between 1980 and 1989, the number of late diagnosed hips during the same period increased dramatically to 0.51 per 1000, the same level as in the whole of Sweden between 1973 and 1976 (0.53 per 1000).²⁶ This should be compared with 0.07 per 1000 in Malmö between 1956 and 1972 and 0.07 per 1000 between 1990 and 1999.

A total of 24 paediatricians and two paediatric orthopaedic surgeons was involved in the initial screening of the 21 patients with late diagnosed DDH. Were some of them inexperienced? Retrospectively, it is very difficult to define the term 'experienced'. Even if the paediatrician or orthopaedic surgeon is experienced in hip examination today this is not a guarantee of his or her skill 20 years ago. Even if some of the late diagnosed hips were a result of inexperience, this does not explain the fact that several experienced paediatricians and paediatric orthopaedic surgeons were involved in the early examination of hips which were subsequently diagnosed late.

The screening programme has altered somewhat over the years and it is impossible to state that it has always been rigorously followed. The routine described in Table I is based on several interviews with paediatricians who worked during the time of the study and a memorandum by one of the authors who has worked in the orthopaedic clinic since 1959. The difference in overdiagnosis as well as in late diagnosed hips cannot be explained by how early the examination was done, whether it was done twice a week or daily, or whether once or twice. Statistically, late diagnosis was neither more common if the child was born on a particular day of the week or during or immediately before a holiday period, nor was the first examination especially late. Neither will the Barlow test, carried out with the pelvis stabilised with one hand (a stricter test), explain it (Table I). Although the date and exact time of delivery were well documented, usually only the date of the hip examination was noted. It was thus not possible to study the exact interval between delivery and examination.

Was the greatly increased number of late diagnosed hips related to sleeping? From the middle of 1960 to 1982, most newborn infants in Malmo slept in a prone or lateral prone position. Between 1992 and 1994 this was changed to the supine position. Even although the dramatically increased number of late diagnosed hips during the period 1980 to 1986 occurred within the period between 1965 and 1992, no definite correlation could be established. Furthermore, it was not possible to determine, retrospectively, the child's sleeping position and no reliable conclusion could be drawn concerning any correlation between sleeping posture and late diagnosed DDH.

The dramatic increase in the rate of splintage (overtreatment) can be correlated with the retirement of an experienced examiner. The incidence increased from \leq 5 per 1000 before to about 20 per 1000 when the responsibility for screening was divided among several individuals.^{7,8} Von Rosen's retirement from the orthopaedic clinic in 1965 probably also accentuated the increase.^{7,8} During 1980 and 1981, we decided to treat only those patients in whom the orthopaedic surgeon found the hip to be dislocatable or unstable. The number treated was subsequently reduced from 35 per 1000 to about 15 per 1000. When an anterior dynamic ultrasound examination was introduced in 1990, the incidence of treated hips reduced to about five per 1000.

The reason for the very high rate of diagnosis (over diagnosis) with up to 45 per 1000 newborn infants referred to the orthopaedic clinic with the diagnosis of NIH was probably the result of the policy that all doctors should be involved in the screening programme; in one year there were 22 doctors (Fig. 5). The incidence of referred cases diminished when this policy was changed so that the screening procedure was mainly undertaken by only two doctors.

In spite of the markedly increased incidence of late diagnosed cases between 1980 and 1986, the incidence over the entire period of 45 years was only 0.16 per 1000.

The good results of screening for NIH in Malmö cannot be explained by a low incidence among Swedes.¹¹

In a survey of the literature concerning splintage of DDH in newborns and infants, Williams, Jones and Bishay²⁷ found the incidence of avascular necrosis to be between 0% and 30%. Avascular necrosis in the initially normal hip was also described. The incidence of 0.6% in our study is one of the lowest published, particularly when both hips recovered spontaneously unlike avascular necrosis in late diagnosed and treated hips.

Missed cases were diagnosed earlier towards the end of the study, i.e., after 1982 at the age of four months (3 to 7). This is in agreement with the figures for the whole of Sweden,²⁶ when, between 1978 and 1979, 71% of late diagnoses were made between the age of one and four months old and, in 1979, 90% were before the age of one year. This is to a large extent due to the untiring efforts nationwide of doctors such as Palmén and von Rosen.

The treatment of late diagnosed hips has varied during the study, but has been homogeneous since 1977. The age at re-examination varied between two and 39 years, but all except one were ten years or older at re-examination. The results are thus medium-term and not long-term, and both clinical and radiological deterioration can be expected with time.²⁸ The earlier treatment starts, the better the results. A regimen of skin traction, adductor tenotomy, closed reduction and hip spica, open reduction when closed reduction fails and a Salter pelvic osteotomy to stabilise openly reduced hips or to improve the acetabular angle, can give excellent results.¹¹ Open reduction after the age of two years, without shortening varus osteotomy, as in case 6, is not recommended.^{11,28} Surgical procedures in the treatment of DDH should be undertaken by a few specialists and the first operation must be successful; repeated surgery or operation by inexperienced surgeons may lead to unsat-isfactory results (Fig. 4).^{11,29} Avascular necrosis occurred in three hips, one of type IV (Fig. 2). This is in agreement with most other studies. Unfortunately, differences in definition as well as variations in treatment and age at the start of treatment lead to marked differences in the incidence; 0% to $73\%^{14}$ and 0% to $98\%^{24}$.

The treatment of late diagnosed patients has not always been optimal partly because for long periods there were no late diagnosed cases and as a result no surgeon had personal experience of open reduction. The results, have, however, gradually improved because of concentration of experience, centralisation of treatment, and an increased number of immigrants and refugees with late diagnosed DDH. The results in 21 late diagnosed cases showed one failure, two with radiological and clinical signs and a further three with a radiological abnormality, but no clinical symptoms. The remainder were considered to be clinically and radiologically normal. No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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