Neonatal screening in Vancouver for congenital dislocation of the hip

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After 20 years' experience throughout the Western world the effectiveness of screening newborns for congenital dislocation of the hip remains controversial. Is the clinical test for hip instability (the Ortolani or "jump" sign) reliable? Are other equally important physical signs frequently missed by inexperienced examiners? Do some dislocations develop after the newborn period when no abnormality was identifiable during the first week of life?

In Vancouver, screening for this condition was initiated in 1964. In the 5-year period 1967–1971 an orthopedic surgeon screened all the infants in the newborn nursery of one hospital, while orthopedic residents screened all those at another hospital. Their rates of neonatal and late (after the first month of life) diagnosis of congenital hip abnormalities were, respectively, 6 and 0.3/1000 live births for the surgeon and 5 and 0.8/1000 for the residents. In contrast, at 20 hospitals that did not have regular screening the average rates were 1.2 and 1.4. Systematic screening and early treatment have great potential for reducing the need for immobilization and surgical treatment of infants whose congenital dislocation of the hip is missed in the first month of life. It may also prevent the arthritic sequelae that in adult life afflict many patients whose treatment was begun after the newborn period.

Après 20 ans d'utilisation à travers le monde occidental, l'efficacité du dépistage de la luxation congénitale de la hanche chez le nouveau-né demeure sujette à controverse. Le test clinique servant à mettre en évidence l'instabilité de la hance (le signe d'Ortolani) est-il fiable? Les autres signes physiques également importants échappent-ils fréquemment à l'examinateur inexpérimenté? Des luxations apparaissent-elles après la période néonatale, alors qu'aucune anomalie n'est identifiable durant la première semaine de la vie?

A Vancouver le dépistage de cette affection a débuté en 1964. Sur une période de 5 ans s'étalant de 1967 à 1971 un chirurgien orthopédiste a effectué un examen de dépistage sur tous les bébés qui ont passé dans la pouponnière d'un hôpital, alors que des résidents en orthopédie faisaient un dépistage semblable dans un autre hôpital. Les taux de diagnostic néonatal et tardif (après le premier mois de la vie) de la luxation congénitale de la hanche ont été respectivement de 6 et 0.3/1000 naissances vivantes pour le chirurgien et de 5 et 0.8/1000 pour les résidents. En comparaison, dans 20 hôpitaux qui ne faisaient pas de dépistage régulier les taux moyens furent de 1.2 et de 1.4/1000. Le dépistage systématique et le traitement précoce offrent d'énormes possibilités lorsqu'il s'agit de réduire la nécessité d'une immobilisation et d'une opération chez les bébés dont la luxation congénitale de la hanche a échappé au diagnostic durant le premier mois de la vie. Ceci peut également éviter les séquelles arthritiques qui, pendant la vie adulte, affligent plusieurs patients dont le traitement fut amorcé après la période néonatale.

In the early 1960s von Rosen[1,2] and Barlow[3] stirred worldwide renewed interest in congenital hip disease with reports suggesting a dramatic improvement in early diagnosis and management. They described the rewards of organized screening of newborn infants for the clinical sign of hip instability. Seddon[4] compared this advance to the conquest of poliomyelitis, where the epidemiologic features were equally fascinating and seductive, and the orthopedic treatment was also technically entertaining but sometimes heavy-handed, and was largely forgotten with perfection of the vaccine.

The advantages of early diagnosis of congenital hip disease were stressed in the past, particularly in Italy, where the condition is said to be common.[5,6] However, prior to the institution of screening, congenital dislocation of the hip in most countries was usually not recognized until the affected child began to stand and walk.[7]

Many observers have found more instability of the hips in newborns than would be expected at a slightly later age. Depending upon the examiner, the incidence of hip instability at birth in most Western countries varies from 5 to 10/1000; after the first week of life the rate is half that, and the prevalence of persisting cases in unscreened populations is between 1 and 2/1000.[2,3,8] In British Columbia a retrospective survey of three large Vancouver obstetric departments for congenital dislocation of the hip in children born from 1957 through 1961 (before there was any screening for this condition) revealed a rate of 1.2/1000 live births.[9] The health surveillance registry of the Division of Vital Statistics, Province of British Columbia[10] reported a rate of 0.7/1000 for the whole province for those years. The limitations of ascertainment in smaller communities at that time probably account for the discrepancy in these figures.

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Barlow[3] observed that when treatment was deferred until the infant was 2 months old, secondary adaptive shortening of the adductor muscles occurred by this age, and these children proved almost as difficult to treat as those whose hips were discovered to be dislocated when they were older than 1 year. Seddon[4] reported that treatment started after the newborn period is often followed eventually by osteoarthritis, even though the hip in childhood and early adult life seems to be perfect clinically and almost normal roentgenographically. Since the early management is safe, simple and brief, it is common practice to start treatment in the newborn nursery most cases of hip instability identified there. When the dislocation persists or the diagnosis is made only after the first few months of life, more aggressive treatment becomes necessary. This often involves hospitalization for traction, general anesthesia, invasive investigations (such as arthrography), several months of immobilization in plaster hip spicas, repetitive roentgenography and, in some instances, surgical treatment.

Von Rosen[1,2] and Barlow[3] reported that in their screened populations practically no cases of congenital dislocation of the hip were diagnosed after the newborn period. This indicated to them that the condition is always recognizable and correctable in the newborn. Barlow declared that the concept of a predislocation phase should be abandoned. He felt that cases of hip instability were missed only because of failure to make the diagnosis in the newborn child. He believed that the type of roentgenographic appearance that has been described as a predislocation phase (a sloping acetabular roof and a small capital epiphysis that is poorly covered) is common among children who have had a dislocation treated, and represents a stage of recovery.

Within a few years of these reports, however, other observers were less consistent in describing eradication of persisting dislocation. In the United Kingdom there were differences in the results of screening between the districts studied and failures in early treatment by abduction splintage.[11-16] Reports from Scandinavia,[17] Australia[18] and New Zealand[19] were enthusiastic, but false-positive and false-negative results of examination continued when a variety of family doctors, obstetricians, pediatricians and orthopedic surgeons (often junior staff) acted as screeners. In some studies the number of late diagnoses approximated what would be expected in an unscreened population.[20]

It would be unfortunate if reports of indifferent results dampened the enthusiasm of the 1960s and led to disenchantment with this exacting but potentially very rewarding enterprise. In this paper we wish to demonstrate our observation in Vancouver that impressive results can be achieved in city hospitals when the screening is conducted by experienced examiners.

Examination for hip instability

There is no need to repeat here a description of the examination to elicit the Ortolani or "jump" sign for hip instability. Barlow's modification of it is well known and widely accepted.[3] If the hip joint is sufficiently lax to permit only subluxation rather than dislocation, the sign becomes more difficult to elicit and interpret.

During the screening maneuver other signs may be observed. An occasional soft, innocent click at the hip is palpable and sometimes audible. It is not associated with signs of instability. Its cause is uncertain. Asymmetry of the flexor skin creases in the thighs is usually seen when a hip is in the dislocated position, but it is also present in some normal individuals and is therefore not diagnostic. Finally, there may be unilateral or bilateral restriction of passive hip abduction. In our experience this is much less common than the signs of instability, but we have no record of its frequency. It may be part of a transient generalized muscular hypertonicity and resolve within a few hours or days. When it is associated with a reducible dislocation its presence suggests that prenatally the hip was in the dislocated position sufficiently long for the development of secondary adaptive shortening of the adductor muscles (sometimes referred to as a "tight" dislocation). When abduction is limited unilaterally there is usually a very generous range of adduction of the affected side. This may, when the two limbs are compared, give the impression that there is an abduction deformity in the opposite (normal) hip.[21] Occasionally restriction of abduction is associated with an irreducible dislocation. In our experience this has almost always been an expression of arthrogryposis multiplex.

In 1970 Wynne-Davies[22] expressed her belief that there are two etiologic types of congenital dislocation of the hip: the type with primary acetabular dysplasia, which is responsible for a large proportion of the cases diagnosed late; and the type with joint laxity, which is responsible for most neonatal cases. This concept is now widely accepted, but the possibility remains that the dysplasia is a result rather than the cause of the dislocation.[3] Dysplasia is much more easily defined roentgenographically in a 6-month-old infant than in a newborn, whose hip is essentially a cartilaginous structure.

Finlay and colleagues[23] in 1967 presented a classification of hip abnormalities clinically recognizable in newborns. In Table I these abnormalities are set out in a somewhat simplified form. Some of the physical signs are difficult to define with words or diagrams, and are best appreciated by repetitive experience in the newborn nursery. Film strips are a helpful adjunct in teaching the screening examination.

We have observed that some individuals are unable to elicit the more subtle physical signs. It is understandable that the more successful screeners tend to be persons who are involved and interested in the whole field of congenital hip disease.

Roentgenographic examination of the unstable hip in a newborn is not often helpful. Von Rosen[1] described a method, but the technique is exacting and usually adds no information not available from physical examination. At 6 months of age an ossific centre is usually present in the femoral head, and the acetabular roof is beginning to ossify. Anteroposterior roent-

genography of the pelvis at that age can be useful in confirming or refuting that development is normal, although care must be taken not to misinterpret normal variations.

Vancouver experience with screening

Organized screening of newborns for congenital hip abnormalities was started in Vancouver in 1964. A retrospective study of the records for 1957 through 1961 at the city's three largest obstetric units produced three conclusions: (a) the diagnosis of such an abnormality was rarely made in the newborn nursery at that time, being recorded on only eight occasions in a newborn population of 45 000; (b) the average age at the time of diagnosis was 10½ months, which suggested, as expected, that it was the parents who were finding an abnormality when the children started to stand and walk; and (c) the incidence of congenital dislocation of the hip in the Vancouver population, as estimated from the numbers that presented for treatment (usually as toddlers), was 1.2/1000 live births.[9] After 5 years of screening at one of the hospitals, St. Paul's, the same institutions were studied again: the incidence of persisting dislocation had dropped from 1.2 to 0.6/1000 live births for the infants born at St. Paul's Hospital, whereas it was unchanged for the infants born at the other two hospitals, where screening had not been done.

Screening was gradually adopted in other hospitals, and at present approximately one third of all children born in British Columbia are given a special examination for congenital hip disease (in addition to the examinations done by the obstetric staff).

Further statistical studies were made for the years 1967 through 1971. This 5-year period was chosen in order to compare the data from screening by an orthopedic surgeon (Dr. Michael Bell at Vancouver General Hospital) with those from screening by a changing group of orthopedic residents (at St. Paul's Hospital) and to compare both sets of data with those for the rest of the province, in which essentially no screening was done in those years.

Method

Twenty-two acute care hospitals with obstetric units were visited for medical record studies. The hospitals selected were all those served by an orthopedic surgeon. It was assumed that, except in cases recognized during the newborn period, almost all children with congenital dislocation of the hip would come under the care of an orthopedic surgeon and would be admitted to hospital (however briefly) for treatment. Although the period under study was 1967 through 1971, the records for 1972 were also viewed in order to include patients born at the end of the study period whose hip abnormality was not identified in early infancy. By this method we expected to find almost all cases of dislocation persisting beyond the newborn period in children born in British Columbia.

First the number of cases in which a diagnosis of hip abnormality was made in the newborn nursery was recorded for each hospital. This figure was somewhat imprecise because the diagnosis in many cases was based on no more than a suspicion of instability or a slight asymmetry of hip posture or movement that was difficult to document. In many cases there was no follow-up information available in the small hospitals to confirm or discount these suspicions. A record was then made at each hospital of all the "late" cases, which were arbitrarily defined as those in which the diagnosis was made after the first month of life. For each such case a record was made of the date and hospital of birth, the date of diagnosis and the hospital(s) where treatment was administered. Patients born outside British Columbia were excluded, as were those with multiple congenital anomalies, paralytic dislocation (meningomyelocele) and arthrogryposis multiplex.

It was felt that there were four main sources of error in this method: (a) a few children may have moved away from British Columbia before the diag-

Table I—Types of hip abnormality in the newborn

Type of abnormality	"Jump" sign	Movement	Comments
Reducible dislocation	Present	Full range	These are the common types of neonatal hip instability. Many resolve without treatment.
Dislocatable hip	Present	Full range	
Subluxable hip	Present but not obvious	Full range	
"Tight" dislocation	Present but not obvious	Restricted abduction	Uncommon.
"Tight" hip without dislocation	Not present	Restricted abduction	Uncommon. May be an expression of primary dysplasia. Deserves close follow-up.
Irreducible dislocation	Not present	Restricted abduction	Rare. Usually an expression of arthrogryposis.
Paralytic dislocation	Usually not present	Full range	Rare. Usually an expression of meningomyelocele. Often develops after the newborn period.

nosis was made or may have travelled out of the province for treatment; (b) a few patients may have escaped identification even though the diagnosis usually becomes obvious with ambulation; (c) a few parents may not have accepted treatment for their children; and (d) in a few late cases the child may have been treated without hospital admission or in a small hospital without the involvement of an orthopedic surgeon. Each type of error would reduce the number of cases.

Results

During the study period the population of British Columbia grew by an estimated 311 000 to 2 185 000. Immigration accounted for about half of this increase, and there were 176 000 children born. According to the incidence discovered in the earlier studies 176 000 live births would be expected to yield 211 cases of persisting congenital dislocation of the hip in an unscreened population. In fact, we found 195 cases (1.1 per 1000 births). The average age at the time of diagnosis was 7 months, as opposed to 10½ months in the earlier study. This suggested only a very modest improvement. The records of the hospitals where there was no regular screening were then compared with the records of the hospitals that conducted screening.

The greatest success occurred when screening was carried out by one orthopedic surgeon; the orthopedic residents had less success, and the hospitals with no regular screening had the lowest success rates, missing cases with approximately the frequency one would expect, although some, presumably those in which there was a high level of suspicion and vigilance among all the staff dealing with newborn babies, had rates that were better than the average for the 20 hospitals with no regular screening (Tables II and III).

In two of the five cases of congenital dislocation of the hip missed by the orthopedic surgeon the infant, when treated, was found not to exhibit the jump sign; these two infants had presented with limitation of abduction (Michael Bell: personal communication, 1975). In all five cases the diagnosis was made before 6 months of age. Similarly, in two of the six cases missed by the orthopedic residents the infant, when treated, was found not to exhibit the jump sign; they also had presented with limitation of abduction. The diagnosis was made before 6 months of age in four cases and at 1 year in the other two.

Since the completion of this survey almost all the children for whom a late diagnosis was made and hospital admission was required have been treated by pediatric orthopedic surgeons. To the best of their knowledge no child born in a hospital where screening was carried out by an orthopedic surgeon has required an open reduction or an innominate or femoral osteo-

Table II—Results of screening newborns for congenital hip abnormalities, Vancouver, 1967–1971

Screeners; no. of live births	No. of abnormalities identified per 1000 live births	
	In neonatal period	After 1 month of age
One orthopedic surgeon; 16 045	6	0.3
Orthopedic residents at one hospital; 7 189	5	0.8

Table III—Identification of hip abnormalities in hospitals with no regular screening, British Columbia, 1967–1971

Hospital	No. of live births	No. of abnormalities identified per 1000 live births	
		In neonatal period	After 1 month of age
Grace Hospital, Vancouver	13 121	0.7	1.2
Royal Columbian Hospital, New Westminster	8 724	3.6	1.7
Royal Jubilee Hospital, Victoria	8 251	0.7	1.6
Lions Gate Hospital, Vancouver	7 084	0.7	1.1
Prince George Regional Hospital	7 081	0.7	1.1
Burnaby General Hospital	6 969	3.2	1.4
Royal Inland Hospital, Kamloops	4 924	0.6	0.6
St. Vincent's Hospital, Vancouver	4 511	0.7	2.2
Victoria General Hospital	4 486	0.7	0.9
Surrey Memorial Hospital	4 472	0	1.6
Richmond General Hospital	4 191	0.9	2.4
Nanaimo Regional General Hospital	3 377	5.9	1.8
Kelowna General Hospital	3 178	0.3	1.3
Chilliwack General Hospital	2 702	0	0.7
Trail Regional Hospital	1 996	1.0	1.0
Campbell River and District Hospital	1 935	0	1.5
Maple Ridge Hospital	1 897	0	1.0
Powell River General Hospital	1 770	0	1.7
Penticton General Hospital	1 574	0.6	0.6
Matsqui-Sumas-Abbotsford General Hospital, Abbotsford	1 331	0	4.5
Totals and averages	93 574	1.2	1.4

tomy, the three most common surgical procedures performed in late cases (Michael Bell: personal communication, 1978).

Discussion

The figures presented in the tables indicate the benefits of screening when performed by an experienced examiner and the slightly lesser rewards when it is performed by junior staff. They suggest that when the rate of neonatal diagnosis is high the rate of late diagnosis (missing of cases) is reduced and vice versa (Tables II and III). This is understandable, but the relationship is not consistent. In three of the hospitals that had no regular screening (Royal Columbian Hospital, Burnaby General Hospital and Nanaimo Regional General Hospital) a relatively high rate of neonatal diagnosis failed to "protect" the patients from a high rate of late diagnosis. In these institutions we presume that the neonatal diagnosis was frequently mistaken or made only in patients that would have recovered spontaneously. In addition, at two hospitals with no regular screening (Royal Inland Hospital and Victoria General Hospital) there was a low rate of late diagnosis despite a low rate of neonatal diagnosis. For the institutions at the lower end of the list the numbers of births were considered to be too small to permit conclusions.

Table IV—Incidence of late-diagnosed congenital dislocation of the hip as reported in the literature

Authors and year of report	Place of study	No. of late diagnoses per 1000 live births
Barlow[3] 1962	Salford (England)	0
Von Rosen[1,2] 1962	Malmö (Sweden)	0
1968	Malmö (Sweden)	0.07
Hiertonn and James[17] 1968	Uppsala (Sweden)	0.42
Smaill[19] 1968	New Zealand	0.67
McKenzie[14] 1972	Aberdeen (Scotland)	0.91
Mitchell[12] 1972	Edinburgh (Scotland)	0.13
Wilkinson[15] 1972	Southampton (England)	1.3
Williamson[13] 1972	Northern Ireland	1.4
Bjerkreim[21] 1974	Southeast Norway	2.0
Paterson[18] 1976	Australia	0.2
Lehmann 1981	Vancouver; screening	
	By single experienced orthopedic surgeon	0.3
	By orthopedic residents	0.8
	Not done	1.4

The figures for the children born at small outlying hospitals are difficult to interpret. There were approximately 59 000 (a figure arrived at by subtracting the number born in the studied hospitals from the total for the province), among whom only 53 late diagnoses were made, for a rate of 0.91 per 1000. This is below the average for the province and only slightly higher than the rate for St. Paul's Hospital, where orthopedic residents performed screening. It suggests either that neonatal screening and treatment are being carried out effectively in the outlying areas (which seems unlikely) or that late diagnoses are being underrecorded in these areas. Some children in border areas, for example, may have travelled to the neighbouring province for treatment, or they may have been treated as outpatients by their family doctors or by general surgeons rather than by orthopedic surgeons.

Table IV shows the incidence rates tabulated by Place and colleagues[20] for late-diagnosed congenital dislocation of the hip as reported from eight studies, as well as the rates recorded in the earliest reports by von Rosen[1,2] and Barlow,[3] the rate reported by Paterson[18] in 1976 and our results. The rates cannot be taken as precisely comparable because of the varying methods of reporting: the cut-off point for late diagnosis varied from 1 month of age to 1 year. It was impossible to tabulate the incidence of neonatal hip abnormalities from each report without a confusing number of qualifications, but the rates varied from 3 to 20/1000 live births, revealing a consistent pattern of "overdiagnosis".

None of the authors reporting after 1962 was able to duplicate the performance of von Rosen and Barlow, as measured by the number of missed cases (the false-negative rate). The authors who described a low false-negative rate were generally reporting the results of screening in large obstetric units, where the examinations were performed or closely supervised by senior staff.[1,3,12,18]

Overdiagnosis has disadvantages: it makes for unnecessary parental concern and inconvenience for a few weeks, and very occasionally ischemic change may occur in a femoral head due to treatment in an abduction splint. Only one such complication was suspected in the 15 years of experience with newborn treatment at St. Paul's Hospital. There were, however, in the early years a few infants in whom transient slight swelling and tenderness developed over the adductor origin in the groin. A different splint was adopted that required a slightly less abducted posture, and the problem has not recurred.

Criteria are required, but are not yet available, for distinguishing between newborns for whom treatment is essential and those for whom it is unnecessary. Meanwhile, most observers accept overdiagnosis and overtreatment as a reasonable price to pay if they are accompanied by a low rate of missed diagnosis in the nursery. Indeed, there may be virtue in treating some hips that are on the borderline of normality. The relation between osteoarthritis of the hip in middle life and congenital dislocation of the hip treated after late diagnosis is high.[4] Bjerkreim[21] felt that neonatal treat-

ment of some of the lesser degrees of hip deformity will prevent the development of osteoarthritis in middle life in many cases.

We suggest that regional orthopedic surgeons serving peripheral hospitals set up a screening system for the newborn nurseries. If their temperament or location does not lend itself to personal performance of this sometimes tedious exercise, they should find a dedicated person who will do it consistently. Specially trained nurses may prove to be the most effective screeners in some communities. In addition to screening in the newborn nursery, repeated hip examinations should ideally be made routinely during the first year of life to prevent as many late diagnoses as possible. The natural history of the various types of congenital hip disorder requires further study. In addition to the goal of eliminating congenital dislocation of the hip as a crippling disease, there is the prospect of new discoveries about the development of idiopathic osteoarthritis of the hip.

Conclusions

In British Columbia the diagnosis of congenital dislocation of the hip is still frequently delayed. Screening for the condition can be very rewarding, particularly when carried out by experienced examiners. In Vancouver the results have been best when the examiner is an orthopedic surgeon with a special interest in this condition. We recommend the establishment of hip screening programs for all obstetric units.

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The art of diagnosis

Now they say when I come to a patient, I know not immediately what ails him, but I need time to find out. It is true. That they judge immediately is the fault of foolishness; for in the end the first judgement is false and from day to day they know the longer, the less, what it is, and make liars of themselves. Whereas I desire to approach from day to day, the longer, the closer to the truth. For with hidden diseases it is not as with the recognising of colours: in colours one sees well what is black, green, blue, etc. But if there were a curtain before it, thou also wouldst not know. To see through a curtain requires effort where there has been none before. What the eyes see can well be judged hurriedly, but what is hidden from the eyes it is in vain to conceive as though it were visible.

—Paracelsus (1493?–1541)