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Etiology, Pathogenesis and Possible Prevention of Congenital Dislocation of the Hip

ROBERT B. SALTER, M.D., M.S., F.R.C.S.[C], F.A.C.S.,* *Toronto*

AS a seventh-generation Canadian, I feel particularly honoured in Canada's Centennial Year to have been invited to deliver the Eighth Alexander Gibson Memorial Lecture at the University of Manitoba. Having read the many glowing tributes to Dr. Alexander Gibson—and having been inspired by them—I only regret that, for chronological reasons, it was not my privilege to have had the opportunity to meet and know this outstanding man whom we all honour this evening.

One of the most inspiring tributes to Dr. Gibson came from the pens of the late Dr. R. I. Harris of Toronto and Dr. W. B. MacKinnon of Winnipeg; it reads as follows: "His qualities of integrity and sincerity, his capacity for clear thinking and convincing speech, his great fund of knowledge accumulated by study and experience, and his strong sense of duty made him a great citizen as well as a great surgeon."

As President of the Canadian Orthopaedic Association, Dr. Alexander Gibson designed the official crest of the Association. Of this crest, Dr. Gibson himself wrote as follows: "The significance of the design is that the maple leaf stands for English-speaking Canada, combined with the fleur-de-lys representing French-speaking Canada, the stalk of both emblems being in common."

Of the 77 scientific publications of this truly great scholar, including as they do many original ideas, it has been written that they "presented uncommon clarity of mind and lucidity of language which enabled him to make the complicated simple and the chaotic orderly". It has also been recorded that before permitting publication of any material, Dr. Gibson applied the following rigid formula: "No man has any right to publish unless he has something to say and has done his best to say it aright." Such high standards of academic excellence represent a challenging goal for all of us.

In the First Alexander Gibson Lecture, Sir Walter Mercer[1] referred to Dr. Gibson's lifelong interest in congenital dislocation of the hip among Indian children in northern Canada. In the Fifth Alexander Gibson Lecture, Professor William Boyd,[2] a classmate and close personal friend of Dr. Gibson, discussed what he referred to as "a subject of general interest to every scientist and to every doctor, namely, the relation of *cause* to *effect*". It would seem appropriate, therefore, in the present Alexander Gibson Lecture to include some aspects of both of these subjects in a discussion of the etiology, pathogenesis and possible prevention of congenital dislocation of the hip.

Congenital dislocation of the hip represents one of the most important and most challenging congenital abnormalities of the musculoskeletal system. Since its detection at birth requires a specific method of examination, it is still not being recognized sufficiently early throughout the world, and may even escape detection until after the child has started to walk. Under these circumstances, treatment becomes progressively more difficult and the results become progressively less satisfactory. Furthermore, congenital dislocation of the hip, unless treated early and skilfully, leads inevitably to degenerative arthritis of the hip in adult life (Fig. 1).

In most congenital abnormalities of the musculoskeletal system, such as clubfoot and spina bifida, the anatomical deformity is maximal at birth and it is obvious that the abnormality has been present from an early stage of intrauterine development. In congenital dislocation of the hip, by contrast, the anatomical deformity

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*Surgeon-in-Chief, The Hospital for Sick Children; Research Associate, Research Institute; and Professor of Surgery, University of Toronto, Toronto, Ontario.

Reprint requests to: Dr. R. B. Salter, Surgeon-in-Chief, The Hospital for Sick Children, 555 University Avenue, Toronto 2, Ontario.

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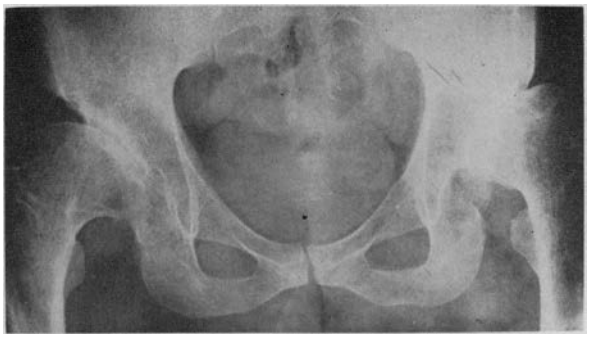

Fig. 1.—Radiograph of the hip joints of a 45-year-old woman who is disabled by painful bilateral degenerative arthritis of the hips as a late result of unsuccessful treatment of congenital dislocation of the hips in early childhood.

is minimal at the time of birth and, if untreated, becomes progressively more marked during postnatal growth. This observation raises the possibility of prevention of at least the postnatal acquired deformities of the hip which are secondary to the original dislocation. It also raises the possibility even of preventing the initial dislocation. Thus, a consideration of the etiological factors and the sequence of events in the pathogenesis of congenital dislocation of the hip is of more than academic interest.

Unfortunately, the controversial question of the etiology of congenital dislocation of the hip has given rise to a plethora of theoretical speculations and a paucity of scientific investigations. Each of the many physical phenomena associated with congenital dislocation, such as acetabular dysplasia, femoral anteversion and elongation of the capsule, has at some time been considered to be the primary cause of the dislocation.

In the organization of this lecture, I have chosen firstly to discuss many of the proven facts about congenital dislocation of the hip as well as my interpretation of these facts; secondly, to develop a hypothesis concerning the etiological factors and the sequence of events in the pathogenesis of this abnormality, and thirdly, on the basis of this hypothesis, to suggest what *might* be done to prevent the initial dislocation. It should be pointed out that the present discussion concerns only the "typical" form of congenital dislocation of the hip to the exclusion of the "atypical" teratologic form associated with arthrogryposis and spina bifida.

What are the known facts about congenital dislocation of the hip? Facts, as opposed to opinions, are of the utmost importance to us; they are the building blocks with which we must build the structure of hypothesis, and the more facts we can gather, the more complete will be our hypothesis. Furthermore, we must make the hypothesis fit the known facts rather than try to make the facts fit a preconceived hypothesis.

FACTS ABOUT CONGENITAL DISLOCATION OF THE HIP

1. *Facts Concerning Embryology*

It has been established by Strayer[3] that the hip joint develops from a single mass of mesodermal tissue in the blastema, or primary limb bud. By about the tenth week, a joint space appears in this mass of mesodermal tissue and joint movement becomes possible. Therefore, it would seem reasonable to assume that the hip joint is not dislocated from the beginning of its formation, but that something must happen to it during its late prenatal or early postnatal development that produces a dislocation.

2. *Facts Concerning Anatomy*

The normal hip joint is one of the most stable of all synovial joints in the body. Three structural factors contribute to this stability: the shape of the opposing bony and cartilaginous joint surfaces of this ball and socket joint, the action of the muscles controlling hip movement, and the integrity of the capsule and ligamentum teres. From a clinical and radiographic study, including arthrography, of congenitally dislocated hips in newborn infants, it is apparent that the shape of the joint surfaces is virtually normal at birth; furthermore, the shape of the joint surfaces becomes progressively abnormal only if the hip remains dislocated during subsequent growth. Therefore, the shape of the joint surface at the time of birth cannot be an etiological factor in the initial dislocation. Clinical studies of paralytic dislocations of the hip secondary to poliomyelitis and spina bifida reveal that while muscle imbalance alone can indeed result in dislocation of the hip, it does so only gradually and over a relatively long period of time. As far as can be determined, in congenital dislocation of the hip the surrounding muscles are perfectly normal at the time of birth. The importance of the fibrous capsule in hip joint stability is emphasized by some studies that we have conducted in the postmortem room on the hip joint of stillborn infants. These anatomical studies reveal that the normal hip can be made to dislocate only after the capsule and ligamentum teres have been divided. I will enlarge upon these studies subsequently.

These clinical, radiographic and anatomical observations suggest that of the three possible structural factors that could account for the instability of the congenitally abnormal hip, the most significant is an abnormality of the joint

capsule and ligamentum teres. Furthermore, the most likely abnormality is an undue laxity of these structures as demonstrated by Andren,[4] as well as by Carter and Wilkinson.[5]

3. *Facts Concerning the Incidence of Congenital Dislocation of the Hip*

(a) *General incidence.*—While the general incidence of clinically detected congenital dislocation of the hip is approximately 1 to 1.5 per 1000 live births, the incidence of this abnormality—unlike the incidence of other equally common congenital musculoskeletal abnormalities, such as clubfoot—varies widely in different parts of the world. This suggests the possibility that postnatal environmental factors may be superimposed upon the original congenital abnormality and have an effect, either for better or for worse, on the natural course of the condition. It also suggests that there may be a racial variation in the incidence of any underlying genetic factor.

(b) *Hereditary and familial incidence.*—In approximately 20% of children with congenital dislocation of the hip, a family history of the abnormality can be obtained. This means that in 80% of the children the abnormality has appeared for the first time in the family. Furthermore, in a clinical study conducted at The Hospital for Sick Children, Toronto, in collaboration with Mrs. D. Wilson Cox[6] of the Department of Genetics, we found that of 226 children—one parent of whom had congenital dislocation—only four had the abnormality. This means that the other 222 children, each of whom had one afflicted parent, were perfectly normal (Table I). Therefore, the genetic factor alone cannot explain the incidence of congenital dislocation of the hip.

TABLE I.—INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP IN OFFSPRING WHEN ONE PARENT IS AFFLICTED

Based on a genetic study of 226 offspring, The Hospital for Sick Children—1963.

Sons	1 in 112	Incidence of 0.9%
Daughters	3 in 114 (1 in 38)	Incidence of 2.6%

Furthermore, studies of identical twins by Idelberger[7] reveal that of 29 pair of identical twins, both twins were afflicted in only 10 of the pairs. Thus, in two out of three pair of identical twins, one twin was afflicted and the other was normal despite an identical genetic composition. All of these observations on the hereditary and familial incidence indicate that while there is definitely a genetic factor in the etiology of congenital dislocation of the hip, there must be other factors as well.

(c) *Sex incidence.*—The repeated observation that 80 to 90% of the children with congenital dislocation of the hip are girls is very significant in that such a predominantly female incidence is not seen in other congenital abnormalities of the musculoskeletal system. Furthermore, there is the impression, which cannot be documented, that the girls with congenital dislocation of the hip tend to be more feminine than the average. They have a very feminine type of body build with wide hips. In addition, they are rather coquettish and know how to use their eyes even as young girls. Furthermore, the boys who do have congenital dislocation of the hip tend to be less masculine (or more feminine) than the average. These observations suggest the possibility of an endocrine or hormonal factor. Recent investigations by Andren and Borglin[8] suggest that an abnormality of estrogen metabolism in afflicted infants may be responsible for the laxity of the hip joint capsule associated with congenital dislocation of the hip.

(d) *Age incidence.*—This may seem to be an unusual point to consider in any condition that is deemed congenital. However, there is growing evidence that the initial dislocation does not always occur at the same chronological age. While the atypical or teratologic type of dislocation almost certainly occurs long before birth, the typical type usually occurs at or shortly after birth and in most infants probably within the first two weeks. Rarely, however, we see a child in whom the initial dislocation does not appear until the child begins to stand. In these children there is no dysplasia of the acetabulum, which is another reason why we believe that dysplasia is secondary to the dislocation; that is, that the dysplasia is the result rather than the cause.

(e) *Geographical and racial incidence.*—The wide variation in the incidence of congenital dislocation of the hip encountered in different geographical areas and among different races is

Fig. 2.—(a) A Nigerian baby being carried on her mother's back with the hips in flexion and abduction. (b) A Chinese baby in Hong Kong being carried in a sling on his mother's back with the hips in flexion and abduction. (c) A northern Canadian Eskimo baby being carried in her mother's parka; the baby is in a sitting position with the hips in flexion and abduction.

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Fig. 3.—(a) A reproduction of one of the figures of Andrea Della Robia (fourteenth century). The infant is swaddled in bandages ("fascia tura"). The original figure is in the Infant's Hospital, Florence, Italy. (b) A contemporary Italian baby living in Toronto, Ontario. The baby is swaddled in a blanket with the hips in extension and adduction. A somewhat similar type of swaddling ("Steckkissen") is used in West Germany. (c) A northern Canadian Indian baby strapped to a cradleboard ("tikonagan") with the hips in extension and adduction. A somewhat similar type of cradleboard ("komse") is used by Laplanders in Northern Scandinavia.

not seen in relation to other congenital abnormalities of the musculoskeletal system. This wide variation is undoubtedly due to a combination of genetic factors and environmental influences. A study of the geographical and racial incidence suggests that one of the environmental influences that may be significant in the etiology of the initial dislocation is the position in which the hips of newborn infants are maintained during the early months of postnatal growth and development. Among those races in which the hips of newborn infants are commonly held in flexion and abduction, the incidence of congenital dislocation of the hip is remarkably low. Low-incidence groups are the Negroes of Central and South Africa,[9, 10] the Chinese of Hong Kong[11] and the Eskimos of Northern Canada (Figs. 2a, 2b and 2c). The position of flexion and abduction is the very position maintained by various types of splints during the treatment of infants with congenitally unstable hips.

By contrast, among those races in whom the hips of newborn infants are commonly held in extension and adduction by various methods of swaddling, the incidence of congenital dislocation of the hip is remarkably high. Examples of high incidence groups are Northern Italians,[12] North American Indians,[13-16] West Germans[17] and Laplanders of Northern Scandinavia[18] (Figs. 3a, 3b and 3c).

These observations suggest that a congenitally unstable and dislocatable hip, which has been maintained in the position of flexion and abduction during intrauterine life, is "protected" by the postnatal position of flexion and abduction and made worse by the postnatal position of extension and adduction. It would seem that the newborn hip joint of the human is not developmentally prepared for a sudden, complete and maintained change from the intrauterine position of flexion to the erect position of extension in the early months of postnatal life, particularly if the hip joint is already congenitally unstable from some cause such as capsular laxity.

While it is difficult to separate the genetic factors from the environmental influences in these observations, a study of Indian children in Northern Canada has proved helpful. We found that whereas the incidence of congenital dislocation of the hip throughout the Indian population

TABLE II.—CORRELATION OF THE CRADLEBOARD (TIKONAGAN) TO THE INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP

Based on a Survey of Canadian Indian Tribes, 1963	
Cradleboard not used	17 cases C.D.H. in 1347 live births (1.2% incidence)
Cradleboard used	250 cases C.D.H. in 2032 live births (12.3% incidence)

Incidence of C.D.H. 10 times greater with use of cradleboard.

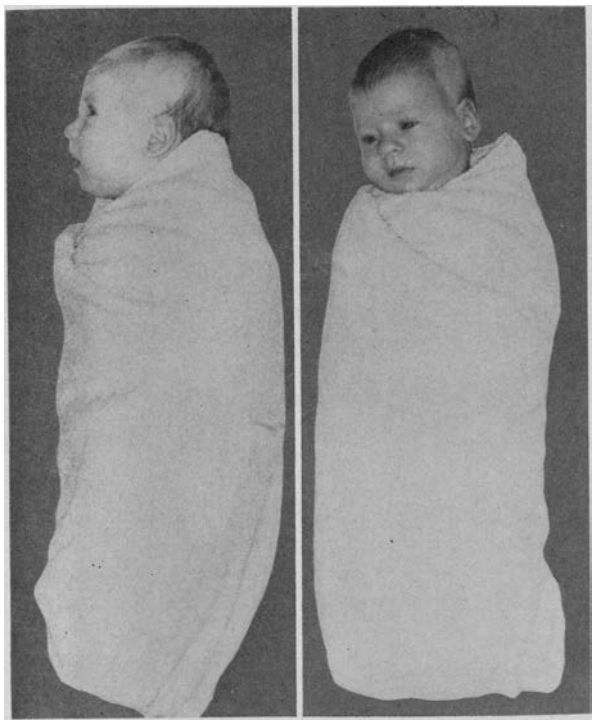

Fig. 4.—Two views of a newborn infant tightly wrapped in a blanket. The hip joints are maintained in extension and adduction.

is higher than in the white population of Canada (possibly due to consanguinity), the incidence in those tribes in which the cradle board (tikonagan) was used for their children was 10 times higher than in those tribes in which it was not used (Table II). Studies by Rabin *et al.*[15] as well as by Houston and Buhr[16] have not, however, shown such a striking relationship between the use of the cradleboard and the incidence of congenital dislocation of the hip. An extremely low incidence, on the other hand, may not be so clearly related to postnatal position of the hips. Edelstein,[9] in an examination of 9000 South African Negro newborn infants, found no congenital instability of the hips—an observation that may be due to the absence of predisposing genetic factors in this particular racial group. Huckstep[10] has made similar observations in Central Africa.

(f) *Seasonal incidence.*—Record and Edwards[19] in England and others in some of the European countries have reported a significantly higher incidence of congenital dislocation of the hip in children who are born during winter months. This has not been noticed in more temperate climates or where central heating is generally used. This interesting seasonal incidence suggests the possibility that, when the weather is cold, the newborn babies and young infants may be more tightly wrapped in blankets which passively extend the hips and limit active movement (Fig. 4). The common habit of wrapping infants tightly in blankets with their hips in extension and adduction should be abandoned.

(g) *Incidence in relation to birth presentation.*—In a combined clinical and genetic study of congenital dislocation of the hip at The Hospital for Sick Children,[20] we found that 23% of all children with congenital dislocation of the hip had been delivered as a breech presentation, whereas in the normal population the incidence of breech presentation is below 5%. Furthermore, the incidence of congenital dislocation of the hip is 10 times higher in children born by breech presentation than in those born by vertex presentation. Another fascinating aspect of this study is that in those children with congenital dislocation of the hip who have been born by breech presentation, the usually predominant female sex incidence is much less striking—only two to one, instead of nine to one. These facts suggest that the initial dislocation in a congenitally unstable hip might be caused (at least in this group of infants) by an iatrogenic factor such as forceful passive extension of the hips associated with traction on the lower limbs during delivery.

4. *Facts About Physical Findings in Children*

The physical findings associated with congenital dislocation of the hip vary with the child's age, and this suggests a progression of changes in the hip after birth. Within the first few days of postnatal life, the most significant

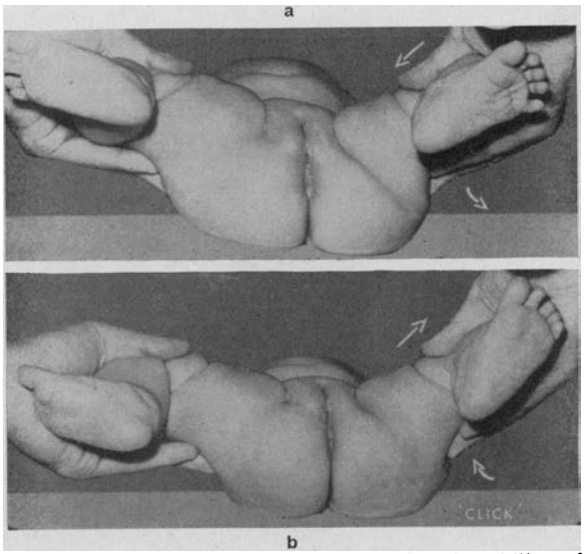

Fig. 5.—The Ortolani sign of congenital instability of the hip joint. (a) With slight adduction and pressure along the long axis of the thigh, the hip can be felt to dislocate or subluxate posteriorly. (b) With slight abduction and traction on the thigh the hip can be felt to relocate.

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