THE UNIVERSITY OF MANITOBA

A PRELIMINARY INVESTIGATION OF CONGENITAL HIP DISEASE IN THE ISLAND LAKE RESERVE POPULATION, MANITOBA

by

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A THESIS

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Chapter 1

INTRODUCTION

The study of populations is basic to anthropology, epidemiology, and genetics. Population studies focus attention on groups rather than the individual, and permit study of statics and dynamics relative to evolutionary processes.[101] Epidemiology requires accurate enumeration of the population at risk of the disease. An advantage of population studies to biological and medical science lies not only in understanding the genetic basis of variability, but also in elucidating the environmental determinants of diseases.[151,4] Definition of population differences in disease susceptibility is permitted. Differences in disease incidence between populations can be described in demographic and biological terms, making the study of differences in environmental determinants more effective.[151]

Congenital malformations are, increasingly, important foci for epidemiologic and genetic research. Their relative incidence has increased, due to decrease in mortality from other conditions and partly due to improved diagnosis. Defects of bone and joint systems are the most frequent in

many studies.[108,222] The hip joint is more often involved when dislocation occurs, in isolation or as part of a 'complex' of congenital abnormalities.

The majority of common malformations are considered to be of multifactorial causation.[34,123] Smithells[195] noted that an epidemiological approach is therefore likely to be the most fruitful in the long run. Neel[145] suggested integration of epidemiologic-genetic strategies in research of congenital malformations and chronic diseases.

Island Lake Research Project

Corrigan and Segal[47] reported an estimated prevalence of 60 cases per thousand for congenital dislocation of the hip (CDH) in the Cree-Ojibwa Island Lake population. Gray[76] verified the high frequency of CDH. In addition, these authors reported frequent consanguinity in the Island Lake population, and associated CDH with consanguineous marriage. CDH incidence of one[232] to four (neonates included[171]), in 1,000 live births can be given for Caucasian populations.

Research commenced in 1968 by Dr. D. Rokala, Department of Anthropology, University of Manitoba, has generated a genetic-demographic data base for the Island Lake reserve population which spans six generations. Relevant demographic data has been integrated into individual records.

Records have in turn been integrated into biological sibships, with individuals referenced into a single sibship sharing a common progenitor and progenitrix. Sibships have been linked into biological lineages. The data base has been compiled from research in archives, vital records registers, church and administrative records, and interviews. Rokala[170] has stated the specific objectives of the research as:

1. to ascertain the segregation frequency, mode of inheritance or heritability, and recurrence risk for congenital hip disease (CHD) and bifid uvula, a reported microform of cleft palate.

2. to investigate the population structure of Indian reserves characterized by elevated prevalence of one or more diseases of purported genetic etiology.

The target population was characterized by high fertility, high mortality, and filter immigration rates following the establishment of the Island Lake reserve with subsequent familial emmigration.[170] In small isolated populations consanguinity may result in elevated prevalence of diseases which involve, at least in part, genetic or familial mechanisms.[184,220,221,218,85,170] Rokala[170] suggested that the Island Lake population might be considered "an isolate with elevated random or non-random consanguinity between mating types as contributory to the population pathology." Schull[185] reported Japanese studies on consanguinity that revealed a picture of small but pervasive effects detectable

only with large numbers of observations.

Proposed Study

This study is a preliminary stage in the proposed investigation of congenital hip disease (CHD)*. The target population includes the four administrative bands of Treaty Indians resident at Island and Red Sucker Lakes. Treaty Indians with residence outside this area who have been traced to the Island Lake population due to prior residence and/or genealogical connections are included. Objectives of this study are:

1. Description of basic epidemiological features of CHD. These include sex ratio, unilateral/bilateral hip involvement, left/right hip involvement, seasonality, birth order, birth presentation, birth weight, parental age, prevalence and variability within the atypical/ teratological cohort.

2. Calculation of annual prevalence rates. Consideration of the trends in specific subtypes of CHD should enable more definitive statements to be made concerning the future health care needs within the target population.

3. Description of familial clustering. Families 'at risk' may be identified should familial clustering be revealed.

4. Consideration of the attitude of members in the target population, affected and unaffected, to CHD, and the functional disability experienced by probands.

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* The term congenital hip disease will be employed to encompass the variability in pathological changes, designated specific terms (dysplasia, subluxation, frank dislocation) by medical practitioners.

Relationship to Total Project

A CHD data base has been created which has been integrated into the genetic-demographic data base, and will permit investigation of the genetic processes involved. Data are based on attempted complete ascertainment of all newborns in the target population since 1955. Representation of all families, not just those exhibiting the disease is therefore possible. Investigation of associated anomalies may detect genetic markers of utility in intra- and inter-population analyses.

Outline of Methods

Radiological examination of all members in the target population is necessary to elicit complete ascertainment of CHD. This has not been undertaken. Probands have been identified through retrospective study attempting truncate ascertainment, locating probands from medical records.[28,217] Personal interviews and observations were employed to obtain data on attitude and functional disability, and detection of social factors which may have bearing on the incidence of CHD in the target population. Data description has been conducted with the aid of the SPSS (Statistical Package for Social Sciences) system of computer programs.

Sources of Data

Data on the target population was derived from the following sources:

1. Winnipeg Office, Northern Medical Services, Department of National Health and Welfare (DNHW).

2. Records of medical personnel (Segal, Gray, Singh) who, in the employment of DNHW, conducted annual CDH surveys in the target population, commencing in 1949.

3. Medical records of six Winnipeg hospitals (Children's, General, Shriner's, St. Boniface, Rehabilitation, and the D.A. Steward Centre), and the Charles Camsell Hospital (DNHW), Edmonton.

4. Health station records at the Garden Hill, St. Therese Point, Wasagamach, and Red Sucker Lake communities.

5. Field work of approximately six weeks duration in the four communities of the Island Lake reserve.

6. The genetic-demographic data base was utilized to complete data from all sources and to locate probands in the genealogical matrix.

7. Provincial Atmospheric Environment Canada records were obtained for utilization in the analysis of seasonality.

Organization of Data

The data was numerically coded and placed on two computer cards per individual. Each individual has a unique identity number, identical to that used in the main data base. Record linkage is based on the individual identity numbers, randomly coded surnames, and birthdates. All information on individuals and families, derived from all sources, is thereby treated in a confidential manner. CHD cards are

sequenced in the main data base.

Theoretical Support

Edwards[59] noted that the knowledge of pathologic aspects of human population genetics was severely limited by the paucity of data relevant to the genetic structure of human populations. The need for extensive data, to permit investigation of the transmission of hereditary traits and detection of environmental factors is well recorded.[217,218,185,151,25] The data presented herein, are based on annual surveys with attempted complete ascertainment of all newborns. These data are therefore considered representative of the population of individuals born in and from 1955 to the present. The problem of bias inherent in hospital samples is minimized by the majority of probands located from hospital records having been previously ascertained in the annual surveys. Additional sources of bias in ascertainment are reduced as the population is composed of Treaty Indians (with an, as yet undefined, degree of white admixture), with similar environment and socio-economic status.

The epidemiological parameters to be investigated have been delineated in numerous reports and studies on CHD.[218,217,161,232,176,233,50] Definition of these parameters permits inter-population comparison. Detection of similarities or differences between populations may contribute to

the understanding of CHD etiology. Increased incidence amongst relatives is not proof, by itself, that inheritance is involved. It may be due to environmental factors shared by family groups.[158] The continued custom of swaddling infants in cradleboards is one of the environmental factors existent at Island Lake. This factor is implicated in prevention of natural remission of the neonatal dysplastic hip.[176,98]

In the majority of modern industrialized populations acceptance of infant prophylactic and/or remedial treatment precludes study of the natural progression of the disease and functional disability attendant on advancing age. Non-availability of treatment prior to 1949, long standing reluctance and limited acceptance of treatment by members in the target population since 1949, permits investigation of the natural progression of the disease. Comparison is therefore possible with the Navajo, Many Farms study[161] in which similar features were present.

The data base may enable delineation of association between major defects and CHD, a factor Neel[145] noted as frequently absent in studies. Atypical and teratological cases arising from developmental defects early in foetal life are reported but excluded from the analysis proper. These cases are usually clinically distinct and are considered

also to be separate etiologically.[67,213,176]

Thesis Format

A review of the extensive literature on CHD is presented in Chapter 2. Definition of the population of Island Lake reserve within a temporal and spatial context is presented in Chapter 3. Historical aspects of medical services in the area, and the CDH project (DNHW) are included to provide background information from which the CHD data base originated. The fourth chapter contains data presentation, epidemiological features, functional disability assessment, and description of cradleboard usage.

Discussion of data, hypotheses, and indications for future health care needs form the major portion of the fifth and final chapter.

Chapter 2

REVIEW OF THE LITERATURE

Congenital dislocation of the hip has been known to man since Hippocrates (460-357 B.C.) who first described and named the condition characterized by 'a waddling gait'. Roser (1879) recognised that diagnosis could be made soon after birth, and Ortolani (1937) described a test to detect dislocation in children under one year of age. Routine screening of neonatal hips was not commenced until the early 1950's.

Prior to routine neonatal examinations, the incidence rates reported for CDH were approximately 0.65 per 1,000 live births[163] to 1.0 per 1,000.[35] With the introduction of routine screening in neonates, and inclusion of neonatal 'unstable' hips, incidence rates have shown a fourfold increase.[227,19,191,91,137]

Definitions

It is doubtful that more than two percent of cases diagnosed CDH are in fact complete frank dislocations.[213] Salter (1968:933) commented that in most congenital

abnormalities:

> The anatomical deformity is maximal at birth and it is obvious that the deformity has been present from an early stage of intra-uterine development. In congenital dislocation of the hip, by contrast, the anatomical deformity is minimal at birth and, if untreated, becomes progressively more marked during postnatal growth.

McKeown[129] considered that a general definition of congenital malformations should include 'any microscopic or macroscopic structural abnormality'. This definition is applicable to CDH. Howorth (1963:172) commented, ". . . the word congenital is useful as a date line but it is not inclusive". Synonyms in common usage are 'congenital luxation', 'congenital hip disease', 'pre-luxation', 'dislocatable hip', 'congenital dysplasia', 'acetabular dysplasia', 'instability of the hip', and 'unstable hip'. In Italy, 'pre-dislocation' is preferred to dysplasia.[30]

Tuell (1966: 225f.) has defined the three conditions, dysplasia, subluxation (pre-luxation), and dislocation (luxation) as follows:

> Congenital dysplasia of the hip is a condition in which the head of the femur is in full contact with a formed acetabulum that is commonly more shallow than normal, and whose roof is more sloping than normal.
>
> By subluxation is meant a hip with the femoral head riding in the rim of the dysplastic acetabulum.
>
> In CDH no part of the head of femur is in contact with the acetabulum.

The head of femur is always dislocated upwards, the direction

may be anterior, lateral, or posterior. Dunn (1969:1037) considered it rational to define CDH as: "an anomaly of the hip joint, present at birth, in which the head of femur is, or may be, partially or completely dislocated from the acetabulum."

Classification of Types

There is no universal agreement on the classification of CDH. Hass's 1951 classification is frequently used. Summarized this is:

1. Typical
   a. dysplasia
   b. subluxation
   c. dislocation
2. Atypical
3. Isolated cases of specific known etiology

Tachdjian[205] distinguished two main groups:

1. Teralogic - develops in utero
   a. part of a generalized congenital abnormality
   b. independent
2. Typical - characterized by its postnatal appearance
   a. the 'unstable hip'
   b. the 'subluxated hip'
   c. the 'dislocated hip'

In 1967 Finlay, et al., proposed a classification based on the clinical state of the hip with expected findings for each level. There has been little attempt to adopt this classification. Mitchell (1972:4) used:

1. confirmed positive 'clunk' test, condition classified as luxation
2. unconfirmed 'clunk' test but laxity found, classified as 'unstable'
3. no abnormality found, classified as normal*

Terms in current usage are those defined by Wynne-Davies (1970:315):

1. Primary
   a. neonatal CDH: diagnosed in the first four weeks of life, usually in the first week, with a 'clunking hip'
   b. late-diagnosis CDH: diagnosed after the first four weeks, usually months or years later. A well defined group in which there is invariably deformity and no doubt as to diagnosis.

Atypical CDH

'Atypical', 'true', or 'teratological' CDH cases are clinically distinct, and the majority are likely to be separate etiologically. A high incidence of CDH in children with multiple congenital abnormalities has been noted. "In many of these, CDH represents a minor anomaly that is not emphasized" (Warkany, 1971:994). These constitute approximately 2 percent of all cases of CDH.[97] Prevalence rates quoted range between 4.3 percent to sixteen percent.[140,163,158,174,50] Variability exists in reported studies whether teratological cases are excluded from CDH studies.[138,67,44,152] The common associated abnormalities and anomalies in

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* Italics in the original

in 'atypical' CDH recorded in the literature, are presented in the Appendix, Table 1, p.134. Similarities exist between typical CDH and several of the associated anomalies (joint laxity, plagiocephaly) which are present in index cases and their relatives in greater frequency than would be expected from a random sample of a population.[234] Wynne-Davies (1970:715) considered it possible, ". . . there is some change in the type of collagen or in its rate of maturation."

Normal Development

Strayer[203] in a major study of the embryology of the hip joint noted that all the elements differentiate from one mass of the blastema. Rotation of limb buds occurs prior to separation of the component parts of the hip joint.[16] The joint cavity opening commences in the 23mm. specimen. The joint space is visible in the 33mm. specimen, and is completed in the 37mm. specimen, approximately at eight to nine weeks of fertilization age.[202,123,135] Gardner and Gray[70] concluded that processes up to this stage are genetically determined, and that intra-articular structures arise in situ. CDH can not occur before the opening of the joint cavity.[202,135] The role of movement in joint development is emphasized, with functional modelling of the hip joint considered especially marked in the first year of life.[111,

79,55

Diagnosis*

Neel[144] commented that CDH is more diagnosable at one year of age. Researchers concur in the diagnostic signs of CDH in late-diagnoses but variability is evident in the diagnostic tests employed and considered valid for the neonate. Two tests commonly used in the neonate or early postnatal infant are Ortolani's sign and Barlow's sign (Salford test). Ortolani's sign was devised for hips over six months of age. Barlow found it unreliable in the newborn and modified it slightly.[191,201] MacKenzie (1972:20) commented:

> . . . that the "jerk" or "jolt" described by Ortolani was mistranslated as "click" in English. Fascial clicks in the region of the hip, and vacuum clicks arising in the hip and knee are common in infants and should not be confused with the jerk of an unstable hip.

The 'clicking' hip test is more accurately described as the 'clunking' hip test as the positive sign is a 'heavy sound'. Many of the positive high pitched clicks present in the newborn disappear in a few days.[137]

Hart's test for limitation of abduction in flexion is considered reliable by the majority of examiners. With

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* Diagnostic determinants are given in Appendix B.

dysplasia and dislocation, abduction is usually limited to between 45 and 50 degrees (normal 90°), and if less than 60 degrees an x-ray is warranted. The rule in the neonate does not preclude CDH later.[83] Lowrie (1970:73) considered that ". . . limitation of abduction alone does not indicate a diagnosis of CDH, reflecting only adductor tone." The test was considered of no value by Barlow in 1962. In series of newborn and older children, unequal abduction between sides was found in a small percentage, at all ages.[175,29]

Babb and Sundberg (1970:15) considered telescoping, the 'push-pull'sign to be ". . . probably the most dependable sign." This is present in roughly four percent of newborns and is rare at twelve months.[175] The sign of asymmetrical skin creases is regarded as unreliable. Ryder and Mellbin[175] noted that only fifty to sixty babies in 1,501 had symmetrical skin folds about the hip. In a series of 139 cases Barlow (1962:295) found:

> . . . far less than half the infants with dislocated hips had asymmetrical skin folds, and the great majority of children with asymmetrical skin folds were found to be normal.

There is general agreement on the value of radiographs at three to four months of age and thereafter. They are, however, considered of limited value in the neonate.[62, 211,118] In the newborn the relationship of the femur to the

acetabulum does not show clearly, as the major portion of both these elements are formed in cartilage. Andren and Von Rosen[10] described a technic which is generally employed.[126] Roentgenograms are considered generally useful when a 'clunk' is not detected clinically in the neonate, but joint laxity is present.[137] The features of a newborn hip roentgenogram and markers are shown in Figure 1.1. Wynne-Davies[234] determined the age at which the ossific nucleus of the femoral head appeared. However, Barlow[18] found bilateral size variation in 10 percent of capital epiphyses x-rayed at birth and one year. Barlow, and MacKenzie[126] reasoned that this was not a true indication of persistence.

In the mature hip, the 'CE' angle and Sharp's acetabular angle are used (Fig. 1.2, 1.3). "The CE angle is a measure of the covering of the femoral head and the depth of the acetabulum" (Wynne-Davies, 1970:320). Wiberg[222] considered a CE angle of 20 to 25 degrees borderline; less than a sign of defective development, greater than a normal acetabulum. Estave[64] detailed Severin's six categories of the CE angle.

The acetabular angle (Sharp's) has been described by Wittenborg (1964:240) as:

> A morphological maturation index of the acetabulum . . . it shows a normal variation within the individual and among individuals reflected in a frequency distribution curve similar to many biological parameters.

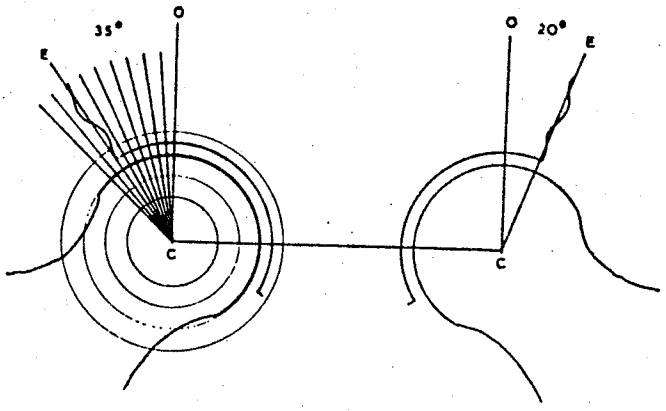

Figure 1.2 *The " C E " angle.*
At left : 35°. Normal angle. At right: 20°.
This angle or less indicates acetabular hypoplasia.
(Strange,1965:15)

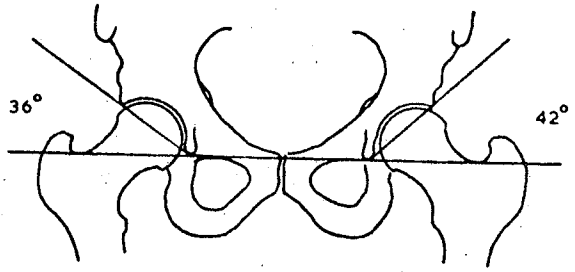

Figure 1.3 *Sharp's angle.*
At left: 36°. Lying within the normal range of 32 - 38°. At right: 42°. The very outside of normality: slight acetabular hypoplasia.
(Strange,1965:15)

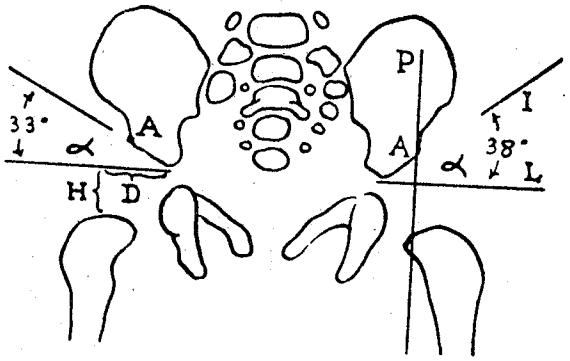

Figure 1.1 *Drawing from roentgenogram of pelvis of newborn infant. (This infant's hips were normal on physical examination.) (A) Antero-inferior iliac spine. (L) Horizontal line of Hilgenreiner, drawn through comparable points on triradiate cartilage. (I) Line drawn parallel with the acetabular roof. The angle formed between "I" and "L" is the acetabular index. (P) Perkins' line, a perpendicular dropped through the antero-inferior iliac spine at right angles to "L". (H) The distance between "L" and the highest point on the femoral neck. (D) The distance between the triradiate cartilage and the intersection of "H" and "L".*
(Coleman,1968:180)

Ryder and Mellbin[175] found the acetabular angle to be larger on the left, with angles in females greater than males in all ages, in a small but statistically significant group. Caffey, et al.[29] concurred with the greater left side finding, and commented that if the [then] criteria had been employed, prophylactic treatment would have been given to 2,850 normal hips. The authors found 44 right and 54 left hips in white females with angles greater than 29 degrees, and observed that this angle was slightly larger in Negroes at six and twelve months in both sexes, and on both sides.

Ethnic variability has been demonstrated in femoral anteversion and acetabular depth. Getz[71] found the anteversion angle to be larger in children than in adults, and in normal Lapps compared with normal Norwegians. Values of 40 degrees for white, and nine degrees for black (Senegal) foetuses at full term were reported by Cheynel and Huet.[40] Reported values for this angle at birth range between 25 and 35 degrees. Getz[71] found the acetabular depth to be larger in normal Lapp men than females, and larger in Norwegian than Lapps. A difference of 3.5mm. was found between full term white and black foetuses by Cheynel and Huet.

MacKenzie[126] found that 37 percent of neonates with positive radiographs had 'stiff hips' without instability

on clinical examination. Seventy percent with negative radiographs had definite clinical instability. He concluded ". . . a negative radiograph in a child with stiff hips does not exclude dislocation" (1970:20).

Arthrography may be used to confirm the diagnosis, the accurateness of reduction,[115] and obstacles to reduction, at any age.[64,103,143,201]

Additional well recognised clinical signs in the late-diagnosis cases are; an externally rotated leg posture, unequal rotation of the feet in prone, the appearance of excessive pelvic width, prominence of the greater trochanter, atrophy of the gluteal muscles, increased lumbar lordosis, Trendelenberg sign and gait.

Geographic and Ethnic Incidence

There is considerable geographic and ethnic variability in reported incidence rates (Appendix C, p.139). High incidence is reported in Scandinavian countries, Scotland, northern Italy, Hungary, Poland, and Japan. "Nests" of high prevalence have been noted in Germany, southwest Brittany and Holland.[88,154] Stevenson, et al.[200] reported a survey of 14 world centres, in which 73 percent of the cases of 'luxation' or 'subluxation' were from Bogota, Columbia and Ljubljana, Yugoslavia. The authors considered

the data invalid as differing criteria must have been employed.

Low incidence (or prevalence) rates are given for South American and African continents, Hong Kong, India[204] and China.[229] The incidence in Africans is considered lower than would be expected to occur randomly in the population.[233] Le Damany's Sudan study (n=40,000) revealed no cases.[88]

There is ethnic variation in incidence rates within geographic regions. The low incidence in African, and African-derived populations[43,107,26] may reflect genetically determined structural differences between Negroid and Caucasian populations.[40,29] CDH has been reported in Negroes.[125,137,23] Maoris, and possibly Polynesians, may be low incidence groups.[158] CDH in a Eskimo has not been reported in the literature. High prevalence rates given for Laplanders have been attributed to anatomical differences,[71] and to postnatal swaddling practices.[110]

The North American Indian groups of the Navajo, Apache, and Cree-Saulteaux Chipewyan peoples have elevated prevalence rates. Crowe[49] described CDH as "widespread" in a survey of orthopaedic problems among the Navajo in 1948. A report on congenital malformation rates for Indians and all races in the first year of life recorded no cases of CDH.[210] Adams and Niswander[2] reported a study of 18,811 American Indian and Alaskan infant births in predominantly

reserve populations. They stated, ". . . the frequency in American Indian infants is apparently not remarkable" (1968: 227f.). Caution must be observed when reports are based on the assumption that all births in reserve populations occur in Indian hospitals, or that those that do are representative of the total population.[170] Research has established the Navajo and Apache as high prevalence groups.[44,21,109,128,161]

Many reports are based on the impressions of orthopaedic surgeons, and are influenced by the location of services, the interest and reputation of the observers, and variables as diagnostic criteria and time duration of study.[213]

Sex Ratio

A male/female sex ratio of 0.15 per 1,000 is cited for Britain.[34] In all surveys a female predilection is evident with no study located reporting a male preponderance. A female/male ratio range of 2.5 to 20.0 to 1 was collated from 29 studies.[212] In a small series (n=342) Medalie, et al.[133] found an indication that males with CDH tended to be associated with a higher incidence of complicated deliveries. There is an impression that the incidence in females is higher in the 'luxation-subluxation' group than in the 'dysplasia' group. The same authors reported a female

incidence of 8.7 percent in the 'luxation' group, and 7.6 per cent in an 'all types' group. Wynne-Davies[233] reported 3.2 percent females in the neonatal group, and 6.6 percent females in the late-diagnosis group. Czeizel, et al.[50] noted fewer males in the 'luxation-subluxation' group (18.6%) compared with the dysplasia group (28.5%). In MacKenzie's study[126] the rate of abnormality at birth was more common in girls (6:4), as was the spontaneous correction rate, and at six months the ratio of "still abnormal" hips was about 5:4 (♀:0). Further, girls required operations six times more than boys. The female sex disparity was significantly less in those born by breech presentation, 2:1, compared with the noted overall sex ratio of 9:1 in Salter's 1968 study.[176]

The higher incidence in females was considered by Le Damany (1912) to be due to sex difference in the structure of the pelvic bones, particularly in the inclination of the acetabulum. Sex differences in anatomical structure specific to the acetabulum are reported.[52,71,29,134,130] MacKenzie[126] observed that the hip develops more slowly in females than males. In more girls than boys the epiphyseal centres are radiologically visible at three and six months. A common observation is that more girls are walking independently at the age of one year.

Salter (1968:935) stated that ". . . boys who do have

CDH tend to be less masculine (or more feminine). . .", and with Andren[7] observed the subjective impression that girls with dislocation of the hip are a very feminine type of body build, with wide hips.

Unilateral/Bilateral Hip Involvement

Unilateral hip involvement is more common than bilateral hip involvement. Bilateral hip preponderance is reported in three studies.[111,158,138] In one study, bilateral hip involvement was nearly five times higher in infants with a family history of CDH.[45] Wilkinson and Carter[226] reported that the sex ratio was more equal in bilateral hip involvement (38 ♀:40 ♂) than in unilateral hip involvement (94 ♀:13 ♂). A higher frequency in the 'luxation-subluxation' group (38.3%), than in the 'dysplasia' group (28.5%) has been reported.[50]

The left hip was involved four times more than the right hip.[120] Right hip preponderance was reported in four studies (n=17).[212] The left/right ratio ranges from 1.08 to 5.5. MacKenzie[126] observed a higher spontaneous cure rate in left hips. In his study more left hips required operative treatment (40:19). There was a marked association between female sex and left hip.

Hasse observed ". . . that the right side of the

pelvis exceeded the left side in volume and extent. . ."[88] Side inequality is recorded in several anatomical studies. Lowrie (1970:72) questioned that the left hip predilection may ". . . suggest that detection of abnormalities in the left hip is easier for right handed examiners." This comment, if applicable, is relevant only in neonatal findings. The greater involvement of the left hip is recorded in many late-diagnosis studies, and remains unexplained.

ETIOLOGY

"Etiologic heterogeneity exists for congenital hip disease" (Woolf, 1971:16). In addition to the structural differences noted, seasonality, birth presentation, birth order, parental age, postnatal posture, and genetic factors are considered to be involved in the etiology and expression of CDH.

Seasonality

A higher prevalence of CDH births in the winter months has been reported, with four of 23 studies in which this variable is noted, finding no seasonal variation.[98,50, 158,161] Wilkinson[223] found no seasonal variation in the prevalence of breech births, but a prevalence of summer births in his 'reducible hip' group, the reverse of the normal ratio reported.

Record and Edwards[163] theorized that the winter excess was related to heavier clothing and crib coverings in the winter months, their hypothesis strengthened by the reported higher incidence of CDH in populations who swaddle their babies in cradles. No seasonal variation was reported by Rabin, et al.[161] in a study of the Navajo who use the cradleboard. While Wynne-Davies (1970:322) considered this relationship possible she commented:

> . . . this explanation, however, cannot account for the increase of winter CDH noted by direct examination of the neonate during the first few days of life, and some additional factor must be acting.

Chen, et al. (1970:291) hypothesized that, ". . . the foetus in Tel Aviv is vulnerable to the seasonal factor which tends to increase the numbers of CDH cases in its fourth and fifth months of development." This relates to Gordon's[74] hypothesis that the hip joint is most vulnerable at the time the lower limbs have enough space to extend, between the fourteenth and sixteenth to twentieth to twenty-fourth week. Elwood[61] suggested the prevalence of winter births associated with congenital malformations is partly due to teratogenic factors operating on summer conceptions. These factors remain unknown.

Prenatal Posture and Breech Births

In several studies (Appendix D, p. 141) a strong

association with breech births is demonstrated. Wilkinson[223] has reported the sole prospective study of breech births in CDH analysis. In 23 cases of 'reducible hip' displacement, 19 had breech presentations. In 14 cases of 'irreducible hip' displacements, 6 had breech presentations, and only these 6 were diagnosed at birth. The prevalence of this deformity in single breech presentations confirmed Wilkinson's opinion that single breech born babies are 'at risk' of CDH. Evidence of delayed leg folding was present in 80 percent of newborn infants with hip displacement, compared with 65 percent of normal babies born by breech.

The breech posture with extended knees is commonly adopted by the foetus in the second trimester. There is a greater tendency for this posture to persist in the primigravida, in whom version may be delayed up to the 34th week of pregnancy. In 10 percent of foetuses normal version fails to occur, and babies are born as frank breech presentations.[225] In experiments with young rabbits, Wilkinson found that only the combined effect of induced hormonal laxity and breech malposition produced changes in females. The results were similar to hip displacement in man. Wilkinson[223] considered the higher female sex incidence in breech-born 'reducible hip' displacement cases (18 of 19) may have some etiological significance, or may be related to a higher

prenatal male mortality. He found support for his theoretical sequence of development (Fig. 2) in Vartan's (1945) survey of nearly 4,000 pregnancy radiographs.[223]

Foetal posture is dependent upon consecutive development of muscle function and dominance. Development proceeds from proximal to distal. At the end of the 8th week the legs lie in abduction and extension. The 'primary' foetal posture is adopted when innervation of psoas, the adductors, and the quadriceps occurs, their activity producing hip flexion, internal rotation, and knee extension. "The extended breech posture is probably normal and universal at approximately the twelfth week of gestation" (Wilkinson, 1966:1106). Folding of the foetal legs occurs between 12 and 36 weeks with the innervation of the hamstrings. Leg folding permits spinal flexion which precedes cephalic version, the vertex posture at 26 to 40 weeks. If the primary breech posture persists beyond 30 weeks breech presentation is probable.

Wilkinson[223] recorded four newborn babies with locked breech birth posture, in whom the "snapping test" was negative but radiological evidence of posterior dislocation was present. The foetal period in which Wilkinson theorizes leg folding should occur coincides with Gordon's[74] "most vulnerable period" for the hip joint. Wynne-Davies (1970: 712) stated that whatever the genetic predisposition a

Figure 2.1
Birth posture. External rotation breech malposition.

Figure 2.2
Locked external rotation breech malposition 'position of dislocation'.

Figure 2.3
Birth posture. Locked internal rotation breech malposition.

(Wilkinson,1966:1108)

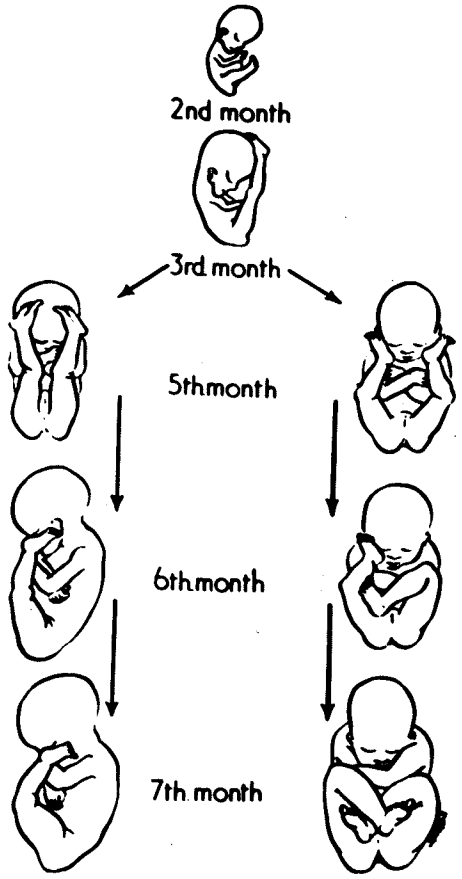

Figure 2.4 *The development of breech malposition in utero. The left side shows the persistent medial rotation breech posture. The right side shows the pre-version folding of the more common lateral rotation breech posture.*

(Wilkinson,1963:282)

"trigger" is needed to establish dislocation of the hip, ". . . the most obvious one is the breech position of the foetus." Woolf (1971:15) reported that his data:

> . . . are compatible with the hypothesis that close relatives of breech cases have a lower frequency of CDH than the close relatives of non breech cases, in support of the existence of a breech malposition effect.

The same author observed the effect was more noticeable in male than female prospositi.

Factors which interfere with foetal leg folding play a part in the etiology of congenital hip dislocation. These factors may be physiological or pathological.[224]

Birth Weight*

Birth weight is considered a sensitive indicator of foetal development.[50] McIntosh, et al.,[132] from a study of infants with congenital malformations, reported a higher proportion of infants weighing less, than over 2.5 kilograms. Record and Edwards[163] considered the difference between the means of prospositi and controls implied a very considerable risk to small babies. This was not correlated with breech deliveries, nor was there anything unusual in the gestation period. Lower than expected incidence in children with a birth weight under 1.2 kilograms was reported.[126,19] This

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* Data converted to a single unit of measurement

was theorized to be related to birth prior to development of joint laxity. Phillips[158] recorded eight (5%) premature infants $\leq$ 2.5 kilograms, with an incidence of one in 701 abnormal hips compared with one in 209 normal hips for full term infants. Robinson[166] found controls weighed .45 kilograms less at birth ($p < .05$). When cases delivered by breech were compared with matched controls, the difference in mean birth weights was even greater (1.88 kgs.) and significant ($p < .005$). He inferred that low birth weight was a characteristic of breech delivery rather than direct association with CDH. Palmen[152] recorded fourteen premature cases (n=97), three times the normal proportion of prematurity, but noted the proportion of prematurity in the neonatal preluxation group was not increased. This may be due to premature babies escaping examination because they are placed in incubators, and these may account for "failure to detect in newborn."

The significance of birth weight is unclear. Several authors consider the premature is not more liable.[168,18,50 191,232,233,227]

Parental Age and Birth Order

Data of Record and Edwards[163] and Czeizel, et al.[50] suggested the incidence of CDH is high in children born to

women aged thirty-five to forty years and over. While Bick[23] suggested the rate of occurrence appeared to increase with increase in maternal age, Woolf, et al.[232] and Robinson[166] observed the incidence was higher in children born to younger mothers. Robinson concluded this implied a greater risk to children born of mothers who bear children in the early adult life.

A preponderance of first born affected children is evident.[212] This was considered independent of maternal age effect, and remained following exclusion of breech births.[50, 233] Miyazaki (1961:144) commented that ". . . birth order and maternal age had nothing to do with the occurrence of this anomaly." Woolf, et al.[232] concluded the primary affect was birth order, and that the preponderance of cases born to young mothers resulted from the fact that first born tend to be born to young mothers. Considered significant, is the finding by Wynne-Davies[233] of a higher excess of first born in the group of parents with acetabular dysplasia.

Record and Edwards[163] concluded maternal fertility showed some impairment both before and after the birth of an affected child. No additional data on this was located.

Postnatal Posture

High incidence rates are given for Laplanders, Navajo,

Apache, and Cree-Saulteaux-Chipewyan populations who swaddle their babies in cradleboards, and for Central European populations who bind the infant's lower limbs.[134] While the incidence rates range from 3.5 percent (min.) at Fort Apache[109] to 38.6 per 1,000 in Poland[52]; extremely low prevalence rates are given for populations that carry their infants on the back, for example, 0.01 per 1,000 in Hong Kong,[92] and 0.11 per 1,000 for South African Bantu.[58]

Salter[176] reported an incidence of 1.2 percent in Canadian tribes not using the cradleboard; 12.3 percent in those tribes using the cradleboard.

When babies are carried on their mother's backs, the child's legs are maintained in a similar position to that used in treatment of 'unstable' hips. The use of the gietka or komsen (Lapps), or tihkinākan* (Cree), hammocks, and any form of tight wrapping maintains the infant's legs in extension, adduction and possibly internal rotation. "Thus a hip which would reduce itself if the child were allowed to kick is maintained out of joint and secondary acetabular dysplasia develops" (Wynne-Davies, 1970:322). Use of the cradleboard is decreasing. Coleman[44] considered usage was inconsequential as nearly all Navajo infants were tightly bound throughout

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* tihkinākan : singular, Plains and Woodland Cree[230]

the year, irrespective of cradleboard usage. While the spontaneous improvement in spite of cradleboard usage has been observed,[161] results of animal studies support the hypothesis that cradling is a predisposing factor.[186,177,225]

Familial Incidence and Genetic Theories

Genetic mechanisms have long been assumed to play a part in CDH because of the predilection of certain families for the disorder.

Idelberger[96] examined 138 of 236 pairs of twins. Concordance was higher in monozygotic (42.7%) than dizygotic (3%) twin pairs. In a smaller neonatal series, Kambara and Sasakawa[102] found 68 percent of 'monochorionic' twins and 33 percent of 'bi-ovular' twins concordant. That monozygotic twins are not invariably concordant, but they are to a significantly higher frequency than dizygotic twins suggests polygenic/multifactorial etiology. Idelberger's data demonstrated that the twin of an affected dizygotic member was no more likely to have the defect than any other sibling of the same sex.[163]

Family prevalence rates reported range from 6 percent[126] to 33 percent.[154] Muller and Seddon[140] found that the incidence in offspring (3.4%) was higher than the incidence in sibs (2.2%). The average risk to a subsequent child

has been estimated to be about 5 percent for girls.[163] In a population noted as 'markedly inbred', the CDH incidence in first degree relatives was 15 times the population incidence, with increased risk for relatives of affected males.[98]

Le Damany is credited for applying concepts of continuous variation and polymorphism to man.[163] Le Damany distinguished cases developing as a consequence of gross abnormalities, from the majority which he regarded as merely one end of a continuously variable distribution of hip stability. Hart[87] supported Faber's theory that dysplasia of the hip is inherited as a Mendelian dominant of incomplete penetrance. Mizizaki[138] concluded that the type of inheritance was not sex-linked but sex-limited, females having a higher frequency, with the mode of inheritance having both the tendencies of dominant and recessive inheritance.

Wynne-Davies[233] considered her data supported hypotheses proposed by Carter and Wilkinson.[36] The rapid fall in incidence from about 12 percent in affected sibs and children, to 0.3 percent in affected cousins, indicated a polygenic rather than dominant inheritance. Employing Falconer's methods, 'heritability' was estimated at nearly 80 percent for a "probably polygenic mechanism" concerned with the development of the acetabulum in which a high proportion of late-diagnosis cases occurred. The second

mechanism, was concerned with joint laxity and thought to be a single gene of dominant effect (heritability greater than 100%). This contributed to the preponderance of neonatal cases. The majority of neonatal cases, which contained a preponderance of the joint laxity type, exhibited spontaneous improvement.

Woolf[231] considered his data tended to negate a polygenic mode of inheritance. He thought it to be more compatible with dominant inheritance only when assuming gross under-reporting occurred for parents and second degree relatives. It was considered plausible that the two types proposed by Carter and Wilkinson did occur. Sasaki[178] and Woolf theorized that exogenous facts may be more important for females than males. Sasaki found that pelvic traits of twins with normal hips, as neck-shaft angle, anteversion angle, and femur neck length were shown to have a lower "hereditary environmental ratio" in women. His study revealed that:

> . . . traits to demonstrate the transverse extension of the acetabulum were found to be more influenced by hereditary and the traits to demonstrate the vertical extension of the acetabulum were found to be more influenced by the environment (1960/1:1530).

Orthopaedic surgeons have commented that the risk of treatment failure increases somewhat exponentially when treatment is delayed.[124,176,127] This indicates a strong

influence by environmental factors whatever the genetic liability present.

Hormonal Mechanisms

Sex-limited joint laxity was demonstrated radiographically by Andren[7] who attributed this to increased reaction to maternal hormones; females excreting oestrogen in a larger amount, over a longer period, in the urine than males. This hypothesis was supported by the results of animal studies.[236,159,225] Andren and Borglin[8] suggested that laxity may be intensified when the foetal liver has a reduced capacity to conjugate oestrogen. Experiments by Aarskoog, et al.,[1] and Thieme, et al.,[206] have not confirmed an anomaly in oestrogen metabolism. The hypothesis that some hormonal imbalance is present remains attractive to several researchers to explain the female predilection.

Maternal Diet

Aksoy, et al.[3] reported two cases, female, in whom CDH appeared as part of a syndrome with severe iron deficiency anaemia. The authors cited Reinmann who had noted that the onset of iron deficiency anaemia in early childhood, or during foetal life due to inadequate maternal iron diet, may lead to foetal insufficiency with consequent fundamental and far-reaching effects. A special relationship was

speculated between the development of CDH and iron deficiency due to nutrition factors.

While Robinson[166] had suggested seasonal changes in maternal diet may be one reason for the seasonal variation in CDH births, Bailar and Curian[17] commented that this had been investigated "with indifferent success."

Joint Laxity

A relationship has been demonstrated between CDH and generalized joint laxity.[119,36,234,98] Carter and Wilkinson[36] concluded that two types of generalized joint laxity were present. A physiological generalized joint laxity, present in females, temporary, due to hormonal imbalance, and developing prenatally to disappear in the neonatal period. The second type, a familial generalized joint laxity is persistent in both sexes, acts as if due to a dominant mutant gene, and is more important etiologically in males. Wilkinson[223] found 23 cases of 'reducible hip' displacement (n=3368) and eleven cases of abnormal joint laxity. Three remaining abnormal at one year of age were "with extreme degrees of persistent joint laxity." (Ibid., p. 43). Wynne-Davies[234] confirmed Carter and Wilkinson's results in establishing the presence of "excessive joint laxity" both in probands and their first degree relatives.

This effect was more noticeable in the neonatal group. She considered that up to the age of two years factors other than age were significant, namely female sex and social class.

Ives (1969:51) found:

> generalized ligamentous laxity was more than twice as common in CDH cases than in controls and was not of simple familial type since it predominates in females and diminished with age like neonatal hormone-influenced laxity.

The author concluded that prolonged laxity in Indian females with the custom of swaddling was the most significant etiological factor so far identified in the Cree and Chipewyan population.

Key[105] reported familial incidence of hypermobility of joints. Kirk, et al.[106] considered their data gave support to the opinion that "generalized familial ligamentous laxity" represents the extreme of a wide normal variation in joint mobility.

> Another view is that isolated ligamentous laxity is a mild mesenchymal developmental disorder which lies at one end of a spectrum of heredofamilial connective tissue disorders with the fully developed picture of Marfan's syndrome and the Ehler-Danlos syndrome at the other. (Ibid., p.243)

The association of a distinct familial generalized laxity with genetically determined connective tissue disorders was noted by Carter and Wilkinson.[36,37] Beighton and Horan (1970:147) questioned if the etiology of "generalized arti-

cular hypermobility", involved, ". . . inborn abnormality of connective tissue, probably collagen . . ."

A characteristic feature of the literature on CDH reappears again in relation to joint laxity. Authors refer to 'joint laxity', 'persistent joint laxity', 'excessive joint laxity', 'generalized joint laxity', 'generalized articular hypermobility', and 'abnormal joint laxity'. Is the 'set of signs and symptoms' given these terms one and the same condition, with variation expressed along a continuum, or is there some significant difference between each, or some of the terms? Are children of parents who demonstrate joint laxity without acetabular dysplasia "at risk" as Wynne-Davies's data suggests? Should these children be subjected to more intensive examinations as has been proposed?[36,37,223,126] If this is valid it may be possible to detect which cases will persist from the large numbers of neonates with 'unstable hips' presently being treated. In addition, there is a need to be able to detect the "missed cases" which present as late-diagnosis cases. Mitchell[137] estimated these to be one in 8,000, while these cases formed 5 percent of MacKenzie's study.[126] Williamson (1972:17) noted that despite the institution of neonatal examinations and early splintage in Northern Ireland, ". . . this has failed to prevent established dislocation in about 2.4% of hips so

treated."

Pathological Changes

Murk (1929) proposed that because of structural differences, animals did not develop CDH, and gave rise to the concept of 'true', 'typical' and 'anthropological' CDH. Numerous animal studies have shown that a condition very similar to that seen in man does occur in animals. Animal studies give support to the following hypotheses:

1. that hormone-induced joint laxity is a predisposing factor in the disease,[236,225,159]
2. that there is possible variability in metabolic capacity,[7]
3. that postnatal extension of the hips is important in the pathogenesis of CDH in man,[177,186]
4. that acetabular dysplasia appears to be the consequence or result, and not the cause of dislocation.[176,86]

These studies confirm operative findings that increased anteversion of the femoral neck is not alone a significant factor, and that hypertrophy and lengthening of the ligamentum teres and the capsule seem to be necessary for dislocation to occur.[11,194]

Elongation and laxity of the capsule, with or without absence or enlargement of the ligamentum teres and psoas

tendon have been noted.[199,94,181,123,6,67] Capsule laxity may be the only abnormality seen in the "non-dysplastic clinical CDH" hip.[124] Badgley[16] considered dysplasia was primary, leading to a flat socket and dislocation, a view supported by other researchers.[97,10] Arnold, et al.[13] described dysplasia as a healing phase of the temporally dislocated or dislocatable hip in the early neonatal period. Hart found primary dysplasia and considered congenital subluxation should be recognised as an entity in itself.

Sommerville,[196,197] contributed greatly to the management of resistant cases by reporting his operative findings of inverted limbus formation (Fig. 3). From this he considered the sequence of events to be that of lateral rotation with anteversion---extension of the hips---subluxation---dislocation---then inverted limbus formation which prevents re-entry of the head of femur into the acetabulum. Limbus formation may be present at birth.[127,207,143,137,223] The presence of inverted limbus formation is noted in neonatal cases with a negative 'clunk' test.

Several authors have noted a defect or 'notch' in the acetabular labrum at operations.[157,177,66,126] Fellander, et al.[66] considered the femoral head slipping over this ridge gave rise to the 'clunk' detected on clinical tests. These authors reasoned that subluxation was

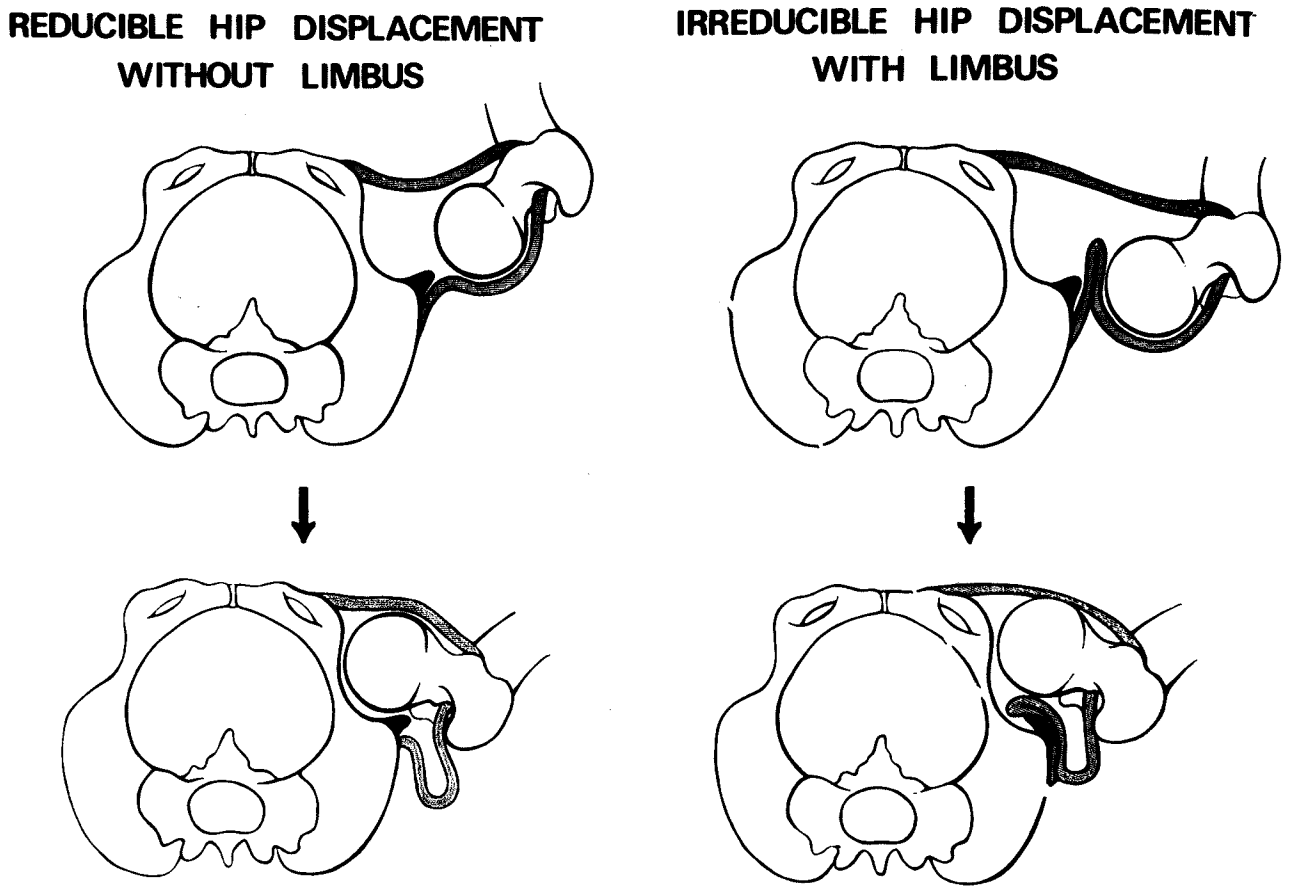

Figure 3. *Diagrams to illustrate concentric and eccentric reductions.* (Wilkinson,1972:48)

caused by insufficient firmness of the chondral part of the roof resulting in instability of the head in the acetabulum. Salter (1968:938) observed:

> The radiographic dysplasia of the acetabulum is minimal and indeed often undetectable, on the first day of life, but it becomes progressively more severe as long as an abnormal relationship exists between the head of the femur and the acetabulum.

He considered that, at least in the first year of life, the acetabular dysplasia was to a large extent reversible, provided a normal relationship between the femoral head and the acetabulum had been restored. Salter supported the concept that dysplasia is not a primary genetic defect, but is secondary to the displacement. ". . . the most impressive feature of the dysplasia was the abnormal direction the entire acetabulum faced" (Ibid. p.939). The hip that dislocated at, or shortly after birth lacked corrective force applied to the acetabulum, whose direction then continued to face "relatively forward and laterally, instead of facing downwards." When the hip is extended and adducted, the head of femur is inadequately covered by the acetabulum both anteriorly and laterally. This hip will then redislocate or resubluxate in the position of extension and adduction, whereas the reduced hip is stable in flexion and abduction. This effect is most marked in the late-diagnosis cases in which maldirection of the acetabulum is more pronounced.

Successful results of innominate and femoral osteotomies in persistent and late-diagnosis cases support this hypothesis. Early recognition of inverted limbus formation, via arthrography, and its excision, reduces the number of persistent neonatal cases. Splintage of the neonatal hip in a Von Rosen, Stracathro, or Barlow type of splint maintains the hip in the stable position, and has not been shown to do a hip any harm. The latter factor is important as a large number of 'unstable' hips will and do recover spontaneously.[161,171] Many babies are splinted unnecessarily. Hiertonn and James[90] estimate probably 15 to 20 times the incidence of dislocation. Consequently, many surgeons delay application of the splint in neonatal 'unstable' hips for as much as three weeks. As treatment is more difficult the older the child, it is considered preferable to splint too many, thus overtreating, than to delay and assess the hip beyond the immediate postnatal period. Matles[124] found no normal hips, on clinical or roentgenographic examinations, when treatment had commenced after the age of six months. A risk of failure, 1 in 3 was cited by MacKenzie, et al.[127] if treatment commenced after the age of five years.

SOCIAL EFFECTS

Social Position

Consideration of "social position"[50] and "socio-economic status"[233] has revealed the previously unrecorded observation of a social class effect in CDH.

Wynne-Davies[233] employed the father's occupation, while Czeizel, et al.[50] utilized the mother's occupation, as an index of social circumstances. The findings in both studies were significant at the 1 percent level. There was a higher proportion of the 'higher income groups' in the CDH groups compared with controls. Wynne-Davies commented the social class effect was most obvious when neonatal cases were included. This finding corresponded ". . . to the findings of a higher proportion of joint laxity in the same income group to the joint laxity control survey" (1970:712). She questioned a possible genetic basis rather than better medical care in the higher income group, while Czeizel, et al. related their findings to better socio-economic conditions since CDH was more common in infants of high birth weight.

Attitude

McDermott, et al.[128] aptly noted that health is a relative matter, and what is considered a disease to one population may not be regarded as such by another. The

Navajo apparently do not view CDH as a real handicap even when there is bilateral involvement. A Navajo with CDH displays little functional impairment in his/her participation in everyday life, in potential for marriage, or in fertility. Treatment of the late-diagnosed cases by fusion procedures converted what was regarded as "almost a blessing" into a very real handicap. The Navajo with a fused hip is unable to sit on the ground and thus join the family a mealtimes, or do weaving. He cannot ride horseback, and life round the hogan became very complicated.[128,54,161,49,21] CDH may be regarded as a crippling disease with adverse socio-economic effects in 'western' populations. It may not, however, be so regarded in other populations and disease attitude may adversely effect acceptance of prescribed treatment.

SUMMARY OF THE LITERATURE

CDH is an important disease with considerable ethnic and geographic variability in prevalence rates. Etiological heterogenity is apparent. The 'child at risk' appears to be female, first born, and a single breech presentation in the autumn or winter months. This child belongs to parents who have abnormal joint laxity, and/or acetabular dysplasia, and a family history of CDH is present. If the condition is not recognised, or not treated in the early postnatal period, the

risk of treatment failure and the cost of treatment is greatly increased.

While excellent results are being achieved in certain countries through mass screening and prophylactic methods, a significant number of cases still present as 'missed late-diagnosis' cases. The absence of well-established criteria for diagnosis, and the lack of agreement among researchers on what constitutes dysplasia, dislocation, and the neonatal 'hip at risk' creates a major deficit in the literature, and in validity of data.

Chapter 3

ENVIRONMENTAL AND HISTORICAL ASPECTS

Location

Island Lake is located approximately 290 miles northeast of Winnipeg, 135 miles east of Norway House, and 60 miles south of God's Lake. Red Sucker Lake is located 40 miles northeast of Island Lake. The administrative territory of the Island Lake Band is bounded by latitudes 53°45' and 54° 00' north, and longitudes 94°00' (second Meridan) and 95°00' west.[72]

Geography

Island Lake is one of the several major lakes in the region. It forms part of the Hayes River Basin, drains northwards into God's Lake and eventually into Hudson's Bay (Fig. 4). The main body of the lake is 45 miles in length and varies from 9 to 13 miles in width.[72] The whole area lies within the Precambrian Shield, and is characterized by relatively low relief, averaging 50' to 75', with well rounded knobs of rock exposure and the occurrence of muskeg in the intervening low areas. The lake, with some 3,000 islands

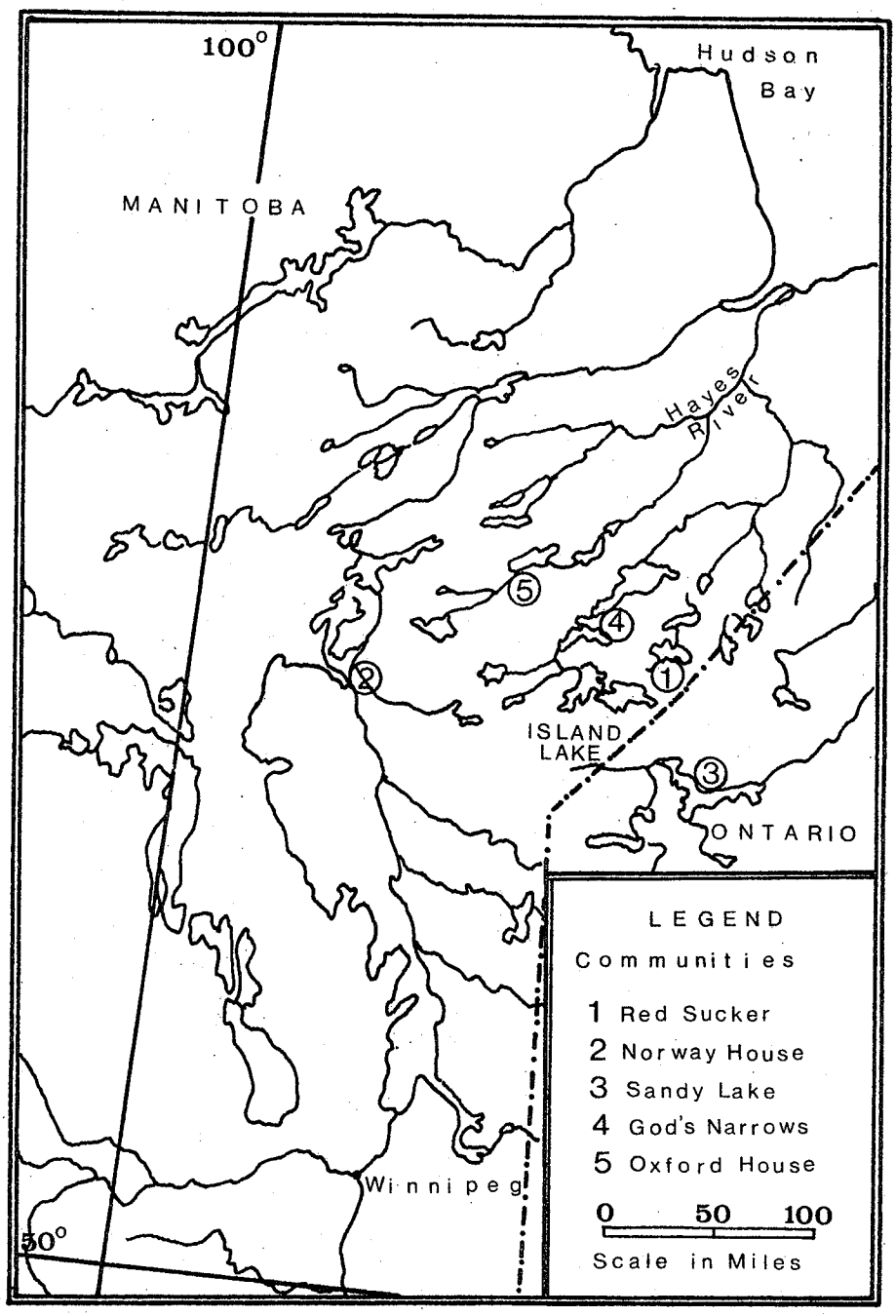

Figure 4

Location of Island Lake and Other Communities

distributed through its length, has a deeply indented shoreline. The nature of the terrain makes land communications between the four communities difficult. Movement is confined to boats in the summer months, tracked transport across the ice in the winter, with increasing usage of small ski and float planes.

Flora and Fauna

The area is in the Northern Coniferous Forest Belt, and all four community sites are wooded. The natural vegetation is spruce and aspen, with jackpine and white birch dominating the secondary growth.[209]

Game is not abundant in the area. While trapping as an occupation has declined absolutely and relatively as a source of income, moose, bear, beaver, muskrat, mink, martin, fisher, lynx, wolf, deer, fox and rabbit are reported by residents operating traplines in the winter.[5,72,209] The spruce partridge is the most common game bird, but the ruffed grouse is also found. Other species frequently seen are Canada goose, mallard, teal, wood duck, loon, gulls and terns.[72] Fish are plentiful and constitute the only dependable local source of food.[72,95,149,209] Northern pike (Jackfish), yellow perch, and pickerel (Walleyed Pike) are widely distributed, with whitefish and lake trout forming

the principal species for commercial fishing.

Climate

Situated north of the 54th parallel the area is subjected to extremes of temperature with long cold winters and short, warm summers. The mean daily temperature (in degrees F. from the nearest official weather station, Norway House) is 29.6° with a January daily mean of -9.7°, and a July daily mean of 65.1°. Daily temperatures less than -35° may occur during six months of the year.[63] There are only 90 frost-free days per annum. The mean annual precipitation is about 19.4 inches spread uniformly throughout the year.[179] The prevailing winds are from the west.

Ethnohistory

Ethnohistorical data pertaining to Island Lake are meagre. The model given for the structural arrangement of pre-reserve populations as Cree and/or Ojibwa units is that of small winter hunting-trapping groups dispersed throughout the subarctic forest region. Each group was quite small, composed of two to five closely related families, and numbering in all 7 to 25 individuals.[68,169,189] During the short summer period several hunting-trapping groups congregated to form an aggregate group of 50 to 200 individuals who concentrated on communal subsistence and ritual activities.[68,169,

168,80 The summer aggregate local band was agamous. Historical and ethnographic evidence suggests that the preferred pattern of marriage was that of first cousin or classifactory cousin, of the cross cousin type.[68,75] The practice of polygamy decreased sharply with the arrival of missionaries and government agencies at the turn of the century.[75,81]

With the growth of the fur trade and the establishment of trading posts in and adjacent to the Island Lake area from around 1818, these posts provided an additional focal point for gathering. Fluctuations in fur trade economy resulted in widespread population mobility in and adjacent to the Island Lake region throughout the 19th and early 20th centuries. Geographical features served as one factor separating the residents of Island Lake from adjacent populations to the north and west, for example, God's Lake and Oxford House.[75] Grant (1929:4) noted a certain aloofness which the Island Lake people demonstrated towards fellow countrymen: "even when they meet them on the trail they neither camp with them nor do they eat together."

The Hudson Bay Company posts in and adjacent to Island Lake fluctuated in occupation and location depending on the status of the local biotic communities.[69,149,179] Following the Winnipeg treaty (No. 5) concluded in 1875 with the Saulteaux and Swampy Cree tribes at Norway House

and Berens River, an adhesion was sought and accepted on July 29, 1909. The formerly independent local bands occupying the Island Lake area and the surrounding district were joined together to form the government band of Island Lake. The initial aggregate of 449 individuals present at the first treaty payments was derived of Cree and Ojibwa population aggregates from Red Sucker Lake to the north, Stevenson Lake to the west, the Cobham and Severn River District system to the south, and Sandy Lake to the east.[47,33]

The settlements which spread outwards from Wasagamach at the westerly end of the lake grew round Hudson Bay Company trading posts and missions to create the present day established communities of Wasagamach, St. Therese Point, and Garden Hill. In the 1950's, due to the influx of individuals to Island Lake and a desire to be closer to traditional trapping and fishing grounds, a group, predominantly derived from Red Sucker Lake, returned to Red Sucker Lake on off-reserve land.[146,170] In 1969 the Island Lake reserve was subdivided into four administrative bands, Garden Hill, St. Therese Point, Wasagamach, and Red Sucker Lake.[31] From their inception the bands have been under the supervision of the Canadian Government.

Missionary activity which commenced in the latter half of the 19th century, mainly by travelling lay preachers,

was intensified in 1903 with the assignment of a resident missionary to the band by the United Church.[80] Father Marius Dutil commenced Roman Catholic missionary activity in 1921, founding a mission at Garden Hill in 1944. In 1952 the Oblate priests and sisters of the Order of Grey Nuns of Ste. Hyacinth opened a mission and school at the site of the first Catholic mission, Mascinicap, or St. Therese Point.[57, 156] A second United Church mission was established at Red Sucker Lake in 1951 under a licenced Indian lay reader. The four communities still today display distinct religious affiliation, Garden Hill and Red Sucker Lake are predominantly Protestant, St. Therese Point and Wasagamach Roman Catholic.

The early emphasis of the Department of Indian Affairs was given to education with the policy of sending students south to residential schools.[179] Later, emphasis was directed towards improvement of health and provision of medical services.[31]

Medical Services

Development of local medical services was slow, and initially supplied by missionaries.[46,32,156] In 1945 medical services for both Indians and Eskimos became a branch of the Department of National Health and Welfare (DNHW). Initially known as Indian Health Services, it became Indian

Indian and Northern Health Services after 1954.[76] Today, it is known as Northern Medical Services. Medical contact was rare, commencing with the attendance of a physician at the annual treaty payments.[46] In the period 1940 to 1947 a tuberculosis program was introduced by Indian Affairs Branch, the continuing program managed by the Manitoba Sanitorium Board. A chest x-ray survey was conducted with the annual treaty party.[47] The Garden Hill health station was opened in 1948. A second opened at St. Therese Point in 1952, with a third established at Wasagamach in 1970. Hospital services have been provided by the Norway House Indian Hospital, and hospitals in the south.

Congenital Dislocation of the Hip

Correspondence and reports of early contact travellers, traders and missionaries does not reveal comments concerning a remarkable number of lame people. Grant, the anatomist, conducted an anthropometric study in the Island Lake area in 1927, noting only, "A few were lame . . ."[75] No identification of the disease process, or indication that the affected number was significant was given. Dr. Corrigan first visited Island Lake in the summer of 1940 as government doctor at the annual treaty payments. He stated, "he had never seen so many cripples all gathered together in one

place outside of a hospital" (Corrigan and Segal, 1950:535). The older residents at Island Lake, interviewed in 1972, consistently stated that there were fewer people limping when they were young.

In 1949 the opportunity arose to conduct a CDH survey in conjunction with the chest x-ray survey at treaty time. Forty-five positive cases were located, an estimated prevalence of 6 percent (included reported cases), with genealogical relationship shown in 100 percent.[47] As a result of the implied hereditary mechanism and noted consanguinity, a genetic study (unpublished) was conducted in 1955 by Segal and Steinberg.[33]

Annual CDH surveys were conducted at Island Lake from 1950 in conjunction with the treaty x-ray surveys. Gray, Orthopaedic Surgeon, Charles Camsell Hospital, Edmonton, conducted the surveys between 1955 and 1963. His successor, Singh, Orthopaedic Surgeon, Charles Camsell Hospital, continued the surveys through to 1970. The majority of cases requiring hospitalization were directed to Edmonton until 1971 when the Children's Hospital (Health Sciences Centre), Winnipeg, assumed this responsibility.[33]

The annual x-ray surveys encountered considerable difficulties with equipment and quality of films. Gray noted:

> . . .The 1959 films on the whole were of very poor quality by comparison with previous surveys and it was extremely difficult and in some cases impossible to do a satisfactory analysis or compare them with previous films with any degree of accuracy (DNHW, 1959).

In his 1960 report Gray noted that of 153 films "49 unreadable, and only 18 of remaining 104 useful for diagnostic purposes."[33] ". . .Films very poor quality, approximately 50 percent could not be interpreted."[33]

The 1949 survey examined only individuals with obvious limps. The 1955 survey concentrated on children aged two years and under. Subsequent annual surveys attempted to examine, either by x-ray or/and clinical examination, all newborns of that year. In 1965 the policy was instituted of x-raying all newborns born at the Norway House Indian Hospital for preliminary evaluation, then to Charles Camsell Hospital for final evaluation and treatment recommendations.

Today the Island Lake population is recognised as a high incidence CHD population. Detection methods are performed on newborns. X-rays are taken of babies born at Norway House Indian Hospital and are read by specialists in Winnipeg. Treatment is initiated from medical services in Winnipeg. The total program is currently under review as it is recognised that a number of children escape detection.[24]

Tihkināākan (Cradleboard)

The tihkināākan* has been reported in use at Island Lake since the first white persons travelled through the region.[80,95,149] Hallowell[80] described the tihkināākan used in the 1930's as having a geometric pattern on the top similar to present day design. A 1962 Medical Services report[33] stated that the tihkināākan was in almost continuous usage from birth to approximately eighteen months of age at Garden Hill, Red Sucker Lake, and St. Therese Point. Corrigan and Segal reported that children may be kept in a mossbag "sometimes as late as three years" (1950:535). The 1962 report stated that cradleboards were used extensively in adjacent areas of Northern Ontario (Sandy Lake, Trout Lake, Sioux Lookout region); at God's Lake to the north of Island Lake. Only the waspison (inner bag minus the board) was used by a few at Oxford House to the west. Use of the cradleboard was not reported for Norway House. Sister Grenier[78] noted that Island Lake people were the only ones using cradleboards** at Norway House in the 1930's and 1940's. They tended to

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* The term tihkināākan applies to the whole 'complex' board and bags. tihki- : 'place child's body in just perfectly'; -nakan : container. [198,230]

** tihkināākans: small, toy tihkināākan, singular. Plural form expressed as / number greater than one / tihkināākana. Use of tihkināākans is incorrect. [198,230]

cease tihkinākan usage when they remained in that community.

The tihkinākan was incriminated from the beinning of CHD investigations at Island Lake as placing the infant's limbs "in the worst possible position" (Gray, 1960:49). Gray gave one objective of his program as "to modify or eliminate the tik-in-agan" (Ibid.). The Regional Superintendent of Medical Services, Manitoba, in a communication concerning the CDH project noted the importance of "proceeding with caution when we suggest changes in long standing customs, otherwise the people may react adversely to the project as a whole" (DNHW, 1965). With the aid of local residents Gray designed a new tihkinākan which permitted greater range of abduction. He was unsuccessful in his attempts to obtain acceptance of the new design. The efforts of medical personnel through the 1950's and 1960's to bring about a decrease in tihkinākan usage were not fruitful. Reasons for this lack of success will be discussed in Chapter 5.

Summary

Construction of all weather runways, and increasing usage of air travel is breaking down the isolation by distance, of the Island Lake reserve. The period between 1949 and 1973 has seen a vast improvement in medical services. Expert medical attention for the treatment of CHD is given

in Winnipeg, and infants requiring treatment are not sent to Edmonton. Despite the continuous investigations since 1949 into CHD with treatment recommendations, the Island Lake people exhibit a certain apathy to change in tihkinākan shape and usage, and to acceptance of treatment.

Chapter 4

DATA PRESENTATION

Hospital records were searched first, followed by annual survey records and field work, respectively. Data collection resulted in multiple ascertainments of 27 cases initially seen at annual surveys and later hospitalized. When detailed survey records were available, the data source was recorded as that of the first ascertainment and CHD diagnosis. Field work was undertaken for three weeks in September and one week in December, 1972. Vital records were checked against Indian Affairs Band Records. Official birthdates were recorded where discrepancy existed between survey and/or hospital data and Band records.

Objectives of Field Work

1. Examination of children in attendance at the Garden Hill school for clinical evidence of CHD and the presence of generalized joint laxity.

2. Examination of 101 cases diagnosed by survey personnel as 'probable' or 'suspected' cases (negative and positive), or cases given diagnoses other than frank dislocation or subluxation (n=47).

3. To identify affected adults, living and deceased.

4. To ascertain the prevalence and variability in tihkinākan (cradleboard) usage.

5. To ascertain the degree of functional disability experienced by affected individuals in the local environment.

Field Methods

1. Clinical Tests

a. Five joint tests were used to assess generalized joint laxity. Finger, thumb, elbow and ankle tests were given with the subject sitting. The knee joint was held at ninety degrees during assessment of ankle dorsiflexion and eversion. All tests were given bilaterally. Elbow and knee extension was graded for zero degrees (full range) and greater than ten degrees (hyperextension). Individuals who had received obvious limb trauma, e.g. fractures, burns, were excluded. Description of the tests is given in the Appendix B, p. 135. The five tests are illustrated in the Fig. 5.

b. Limb length was evaluated by palpation of the anterior superior iliac spines with the subject in the standing position, feet together. No attempt was made, due to the time factor, to measure the actual limb length.

c. Trendelenberg sign test: Individuals were requested to stand on one leg, preferably without hand support. Pelvic raise or lowering on the unsupported side was noted. Difficulty was encountered obtaining co-operation of children

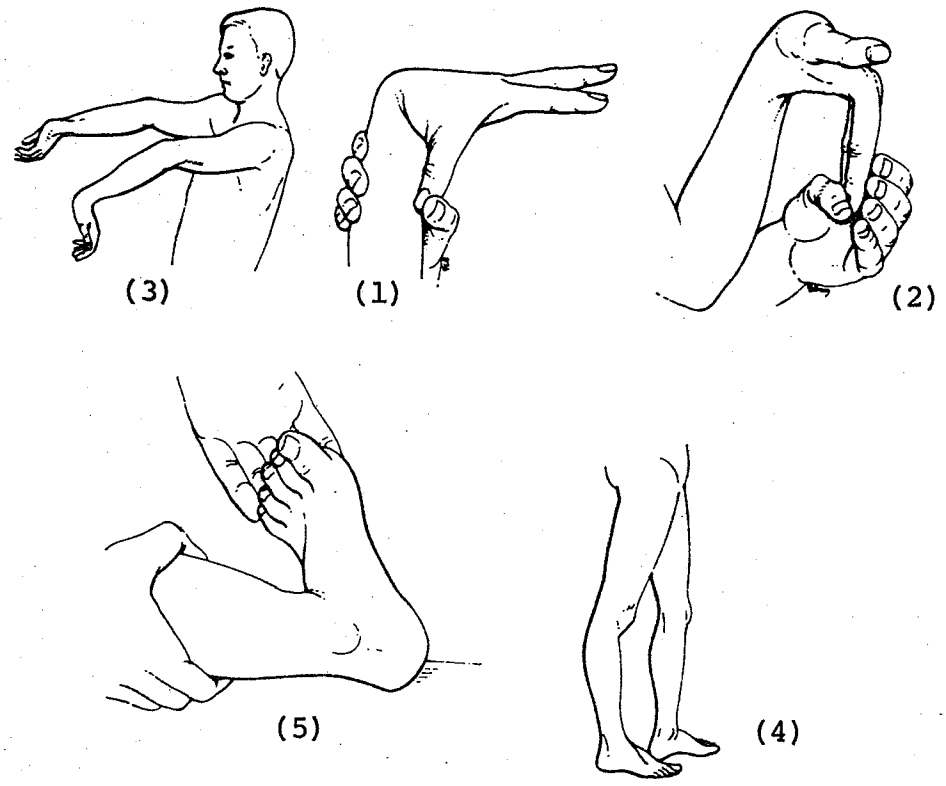


Figure 5

Joint Laxity Tests, Range Regarded as Excessive
(Test Detail Given in Appendix B)
(Wynne-Davies,1970:706)

under four years of age.

d. Trendelenberg gait examination: Individuals were requested to walk a specific distance away from, and towards the examiner. The examination was repeated when doubt existed on the gait status. The presence of trunk sway and pelvic drop was noted. The distance covered was determined by the site of examination. The distance in the school was approximately eighteen feet. The distance varied when examination was carried out in homes or at health stations. In- or out-toeing in the gait pattern was noted.

e. In the standing position the presence or absence, and degree of lordosis was observed but not measured.

2. Data collection

Names of reported 'limping' adults were collected from survey lists since 1966, health workers, and adults in the four communities. Children seen in the schools were questioned whether any member of their family limped. Reported presence of a limp of any kind was noted and investigated. Individuals present were examined and the personal history recorded. In the case of deceased individuals attempts were made to interview relatives to elicit several descriptions of the gait pattern and time of onset. Individuals reported but not seen were recorded as 'reported' cases (n=31). Individuals positive for Trendelenberg sign and gait

with prominence of the Greater Trochanter(s), lordosis, limb inequality in unilateral cases, and a history of a limp since commencement of walking were recorded as confirmed frank dislocations subsequent to a review of the medical records to exclude other pathology. Schoolchildren who presented with mild or moderate Trendelenberg gait or sign tests were, in the absence of radiological confirmation recorded as 'probable' cases.

Factors noted by observation and questioning in relation to the tihkinākan were:

a. age of the child in a tihkinākan;

b. situations in which babies were carried in the tihkinākan;

c. position of the tihkinākan when not being carried by the mother:

d. variation in size and design;

e. age at which a baby is first placed in a tihkinākan;

f. amount of time daily a baby would spend in the tihkinākan;

g. whether the infant slept in the tihkinākan;

h. age at which the tihkinākan is no longer used;

i. customs related to placement of the infant in the tihkinākan;

j. additional methods of infant positioning, and when used;

k. number of rooms and beds available in each family residence.

Functional disability was evaluated subjectively.

The evaluation was based on response to questioning, and observations of the performance of activities of daily living in the community. For example, the following activities were noted:

a. the ability to rise and lower to a chair, or the floor;

b. walking on level ground and traversing snowy, icy and muddy surfaces;

c. ascending or descending slopes, with or without a load;

d. ability to chop, saw and carry firewood, and haul water;

e. embarking and disembarking from small boats and canoes, either directly onto the rocky shoreline or on to the docks;

f. boat and skidoo handling, performance in manual work in the community as building, road construction, and freight handling was observed in males. An attempt was made to determine the type of employment held by affected males, whether they were welfare cases and if they were active trappers.

g. children's participation in playground activities.

These factors were observed during the ten days spent at the Garden Hill school, home visits, attendance at well baby clinics and at health stations.

Individuals absent from Red Sucker Lake community during the September visit were examined at the December visit as were a small number in the other three communities.

Classification

1. Typical and Atypical groups were created on the basis of presence or absence of one or more congenital abnormalities, ascertained by medical personnel. In addition, where doubt existed on the etiology of CHD because the individual had poliomyelitis prior to CHD diagnosis, cases (n=5) were placed in the Atypical group. Detected congenital abnormalities are listed in Table 4. Description of these cases is given in the Appendix E, p.142. Unless explicitly stated, the following data description and analysis is based upon the Typical Group (no additional abnormalities reported). Description of the Atypical group is given in the Appendix F, p. 144.

2. Three types of diagnoses are distinguished:

a. confirmed: no doubt expressed by the medical examiner.

b. probable: doubt expressed by the medical examiner on the basis of poor quality radiographs, clinical or radiological examination only, child not of walking age when examined, and borderline signs.

c. reported: case never seen, or no records of examination located; deceased or living off the reserve; known present but not located.

3. Cases were subdivided into positive and negative groups

Table 4

Detected Congenital Abnormalities (Atypical Cases)

Type	Frequency
Calcaneo-valgus*	28
Umbilical Hernia	3
(with Calcaneo-valgus	2)
Sacralization of Lumbar 5**	2
Axenfeld's Syndrome	1
Cleft Palate# & Calcaneo-valgus (mild)	1
'Congenital Encephalitis'	1
Congenital Heart Disease	1
Crouzon's Disease (craniofacial dysostosis)	1
Synostosis Radio-Ulnar Joints (bilateral)##	1
'Talipes'	1
(Poliomyelitis###	5)
(with Talipes Equino-varus	1)

* Mild Calcaneo-valgus diagnosed under two years of age considered within normal limits and classified as Typical (n=22)

** Detected incidentially on radiographs; may be under-reported

\# One other case located, not included in sample

Reported present in other members of the family, not investigated

Placed in Atypical Group when CHD diagnosis was subsequent to Poliomyelitis infection

on the basis of the initial diagnosis made by a medical practitioner. Typical negative cases proved not to have CHD nor were other congenital abnormalities reported.

4. Positive cases were subdivided into three diagnostic categories:

a. True: given the diagnostic label of frank dislocation or subluxation by the medical examiner at the first examination, without consideration by this investigator of the radiological and clinical findings.

b. Dysplasia : abnormal radiological findings designated dysplasia by the medical examiner at the first examination.

c. Other: a residual category employed to encompass variability in diagnoses. This category includes cases given the diagnoses of 'stiff hip', 'unstable hip' (includes "tendency to subluxate"[33]),and 'CDH' abbreviation in the absence of other details such as radiological and/or clinical findings.

5. Cases examined for generalized joint laxity were graded either positive or negative. Positive grading was given when an individual was bilaterally positive for three or more joint tests. The elbow and knee tests were taken as positive when hyperextension of ten degrees or more was present.

6. Trendelenberg sign test: this was graded a) negative, no

pelvic drop on the unsupported side, b) mild positive, neither a pelvic raise or obvious drop occurred on the unsupported side, and c) positive, a definite pelvic drop occurred on the unsupported side.

7. Trendelenberg gait examination: individuals were graded a) negative, no trunk sway or pelvic drop on the unsupported side, b) mild positive, some trunk sway and pelvic drop suspected, and c) positive, an obvious trunk sway towards the weight-bearing limb with noticeable pelvic drop on the unsupported side.

The coding form is presented in Appendix G, p. 151.

RESULTS

DIAGNOSIS AND ASCERTAINMENT

Congenital Hip Disease status is known for 1248 individuals. The Typical group (n=1204) contains 783 negative cases, and 420 positive cases. The positive group includes two hundred and forty-three (243) cases classified True, one hundred and twenty-three (123) Dysplasia, and fifty-four (54) Other (Fig. 6).

Type of Diagnosis

Sixty-six percent of typical cases have confirmed diagnoses. Probable cases form 30.32 percent, and reported cases 3.2 percent. Reported cases were ascertained from

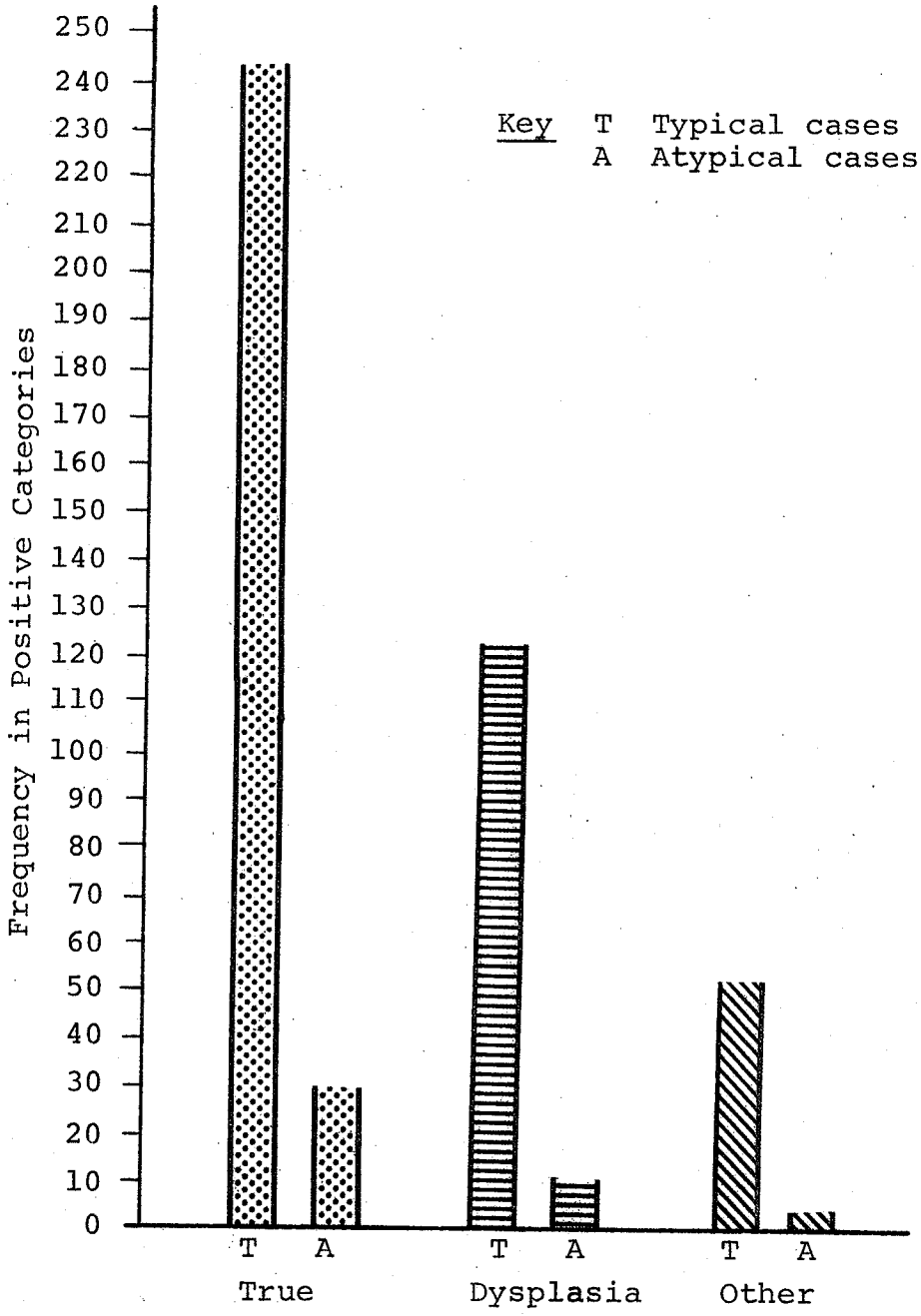


Figure 6

Typical and Atypical Cases in Three Diagnostic Categories

survey data from 1966 onwards or during the 1972 field work. Probable cases form 16.73 percent (n=209) of 984 cases ascertained from survey data. In 380 positive cases, seventy-two (5.76%) retain a probable diagnosis. Probable diagnostic status was given to a higher percentage of negative than positive cases (22.27% compared with 6.97%) (Table 5). Probable cases occur irregularly through the twenty-two years of data collection as indicated by the following:

Year Diagnosed	Frequency & Percentage of Total	Frequency of Diagnoses	
		Positive	Negative
1956-1961	184 (50.4)	62	122
1964-1971	25 (6.85)	10	15
1972	156 (42.74)	15	141
	365		

The greater number of probable cases between 1956 and 1961 reflects difficulties encountered with radiological equipment in that period.

Case Location

Nine hundred and seventy-four cases were located from survey records, 233 were located on reserve in 1972, twenty-one from health station records, and forty-two cases from hospital medical records (Fig. 7). Cases located from hospital records were all positive for CHD, and in the Typical group had been examined initially at annual surveys. In forty-two cases located from hospital records, twenty-two

Table 5

Positive and Negative Cases by Diagnostic Type for Typical Group, and Less Cases Diagnosed in 1972

	Diagnostic Type *			
Diagnosis	Confirmed	Suspected	Reported	Total
Positive	323	87	9	419
	(25.88)	(6.97)	(0.72)	(37.09)
Negative	475	278	31	784
	(38.0)	(22.27)	(2.48)	(62.9)
Total	798	365	40	1203**
	(66.28)	(30.32)	(3.2)	(96.32)
Frequencies less cases diagnosed in 1972 (n=219)				
Diagnosis	Confirmed	Suspected	Reported	Total
Positive	308	72		380
	(24.66)	(5.76)		(30.42)
Negative	467	137		608
	(37.4)	(10.96)		(48.67)
Total	775	209		984
	(62.04)	(16.73)		(78.78)

* Percentage of total sample (n=1249) in brackets
** Diagnostic type not reported for one case

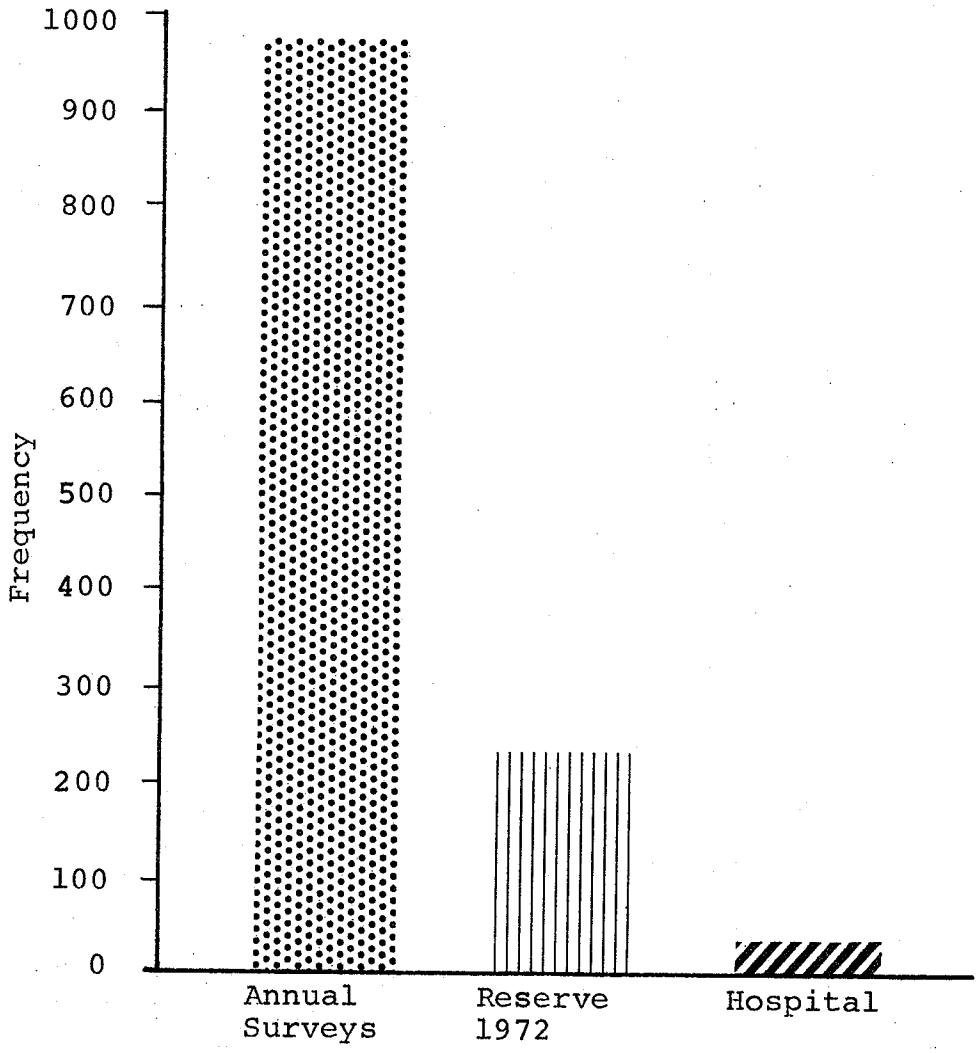


Figure 7

Case Location in the Three Major Data Sources

were located from Winnipeg hospitals and twenty from the DNHW hospital in Edmonton.

Preliminary analysis of case ascertainment by community revealed that 77 percent (n=329) of positive cases were located at Garden Hill and St. Therese Point. In 423 cases for which community residence is known, 183 cases were from Garden Hill, 146 from St. Therese, 45 from Red Sucker Lake, and 49 from Wasagamach. Frank dislocation cases form a larger percentage of cases located at Red Sucker Lake (48.8%) and Wasagamach (46.9%), with approximately 33 percent of cases at Garden Hill and St. Therese Point.

New Case Ascertainment

New case ascertainments vary considerably over the twenty-two years of data collection. Frequencies in individual years are presented in Table 6 for Typical and Atypical cases, by negative and positive diagnosis. The total number ascertained and the number positive, for individual years, 1949 to 1972, is presented in Figure 8. The greatest number were ascertained in 1972 (n=220), with the least (n=1) in 1962. Omitting 1972 data, the greatest number of cases were ascertained in 1964 when children born in the years 1962 and 1963 were first reported in survey lists. Data for survey years 1951 to 1953 were lost in a fire, and/or unobtainable.

Table 6

Case Ascertainment by Year of Diagnosis (n=1249)

	YEAR OF DIAGNOSIS 19-																			
	49	50	54	55	56	57	58	59	60	61	62	64	65	66	67	68	69	70	71	72	TOTAL
TYPICAL CASES																					
Negative				1	48	2	29	116	1	103		146		95	1	54		2	5	179	782
Positive	20	5	2	48	47	12	11	29	49	39		23	11	23	9	28	6	11	8	40	421
Unknown										1											1
Totals	20	5	2	49	95	14	40	145	50	143		169	11	118	10	82	6	13	13	219	1204
ATYPICAL CASES																					
Negative										1											1
Positive	1	2		2			2	2			1	4	7	1	14		6	1		1	44
Totals	1	2		2			2	2		1	1	4	7	1	14		6	1		1	45
TOTALS	21	7	2	51	95	14	42	147	50	144	1	173	18	119	24	82	12	14	13	220	1249

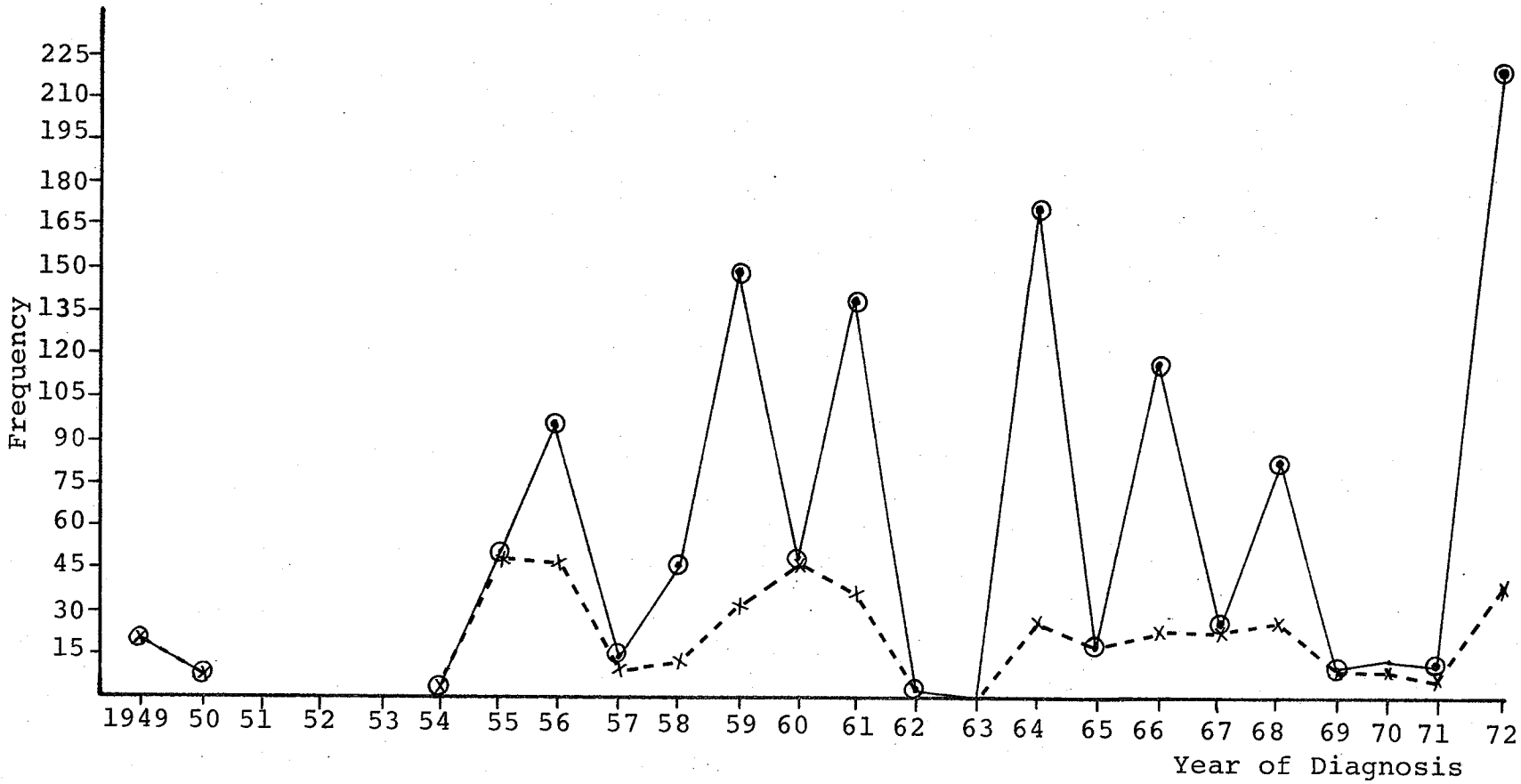


Figure 8

Case Ascertainment, Total and Frequency Positive by Year of Diagnosis

Over all years, the greatest number of positive cases were diagnosed in 1960, 1955 and 1956 (n=49, 48, 47). More cases were seen at annual surveys than the frequencies presented indicate. In most years, a number of children ascertained the previous year were re-examined.

The twenty cases located for 1949 are less than half (n=45) the number of new cases reported by Corrigan and Segal (45). A complete list was not located. It is therefore possible that some of the adult cases ascertained in 1972 may have been seen initially in 1949. Data were not located for cases examined and proven negative for the years 1949, 1950 to 1955, 1962 and 1965. Complete lists of negative cases seen in 1967, 1969 and 1970 have not yet been located.

Ascertainment Rates

Considerable fluctuation is evident in annual ascertainment rates, with the range from 35 cases per 1,000 live births (undetection) to 600 cases per 1,000 live births. Rates for five year periods exhibit steady decline from 345 positive cases per 1,000 in 1955-59, to 223 per 1,000 in 1960-64, and 127 per 1,000 in 1965-69. Rates for True cases only (dislocation and subluxation) over the same three five year periods are 108 per 1,000; 129 per 1,000, and 92 per 1,000 live births.

Positive cases form 18.85 percent of all live births between 1950 and 1969 (n=1,814)[170]; 21.1 percent with atypical cases included.

Age at Diagnosis

Neonatal cases (< 28 days) form 3.57 percent, while Late-Diagnosis cases (< 28 days) form 95.93 percent (n=49, 1155). Fifteen positive cases were neonatal diagnoses. Cases were grouped into six age categories.

1. Neonatal: 28 days and less.
2. Under four months and greater than 28 days.
3. Under one year but greater than four months.
4. Under three years but greater than one year.
5. Under eighteen years but greater than three years.
6. Over eighteen years (in several cases day and month of birth was unknown).

The percentage of total (n=1204), with percentage positive and negative at each of the six age categories is presented in Figure 9. A total of 521 cases (43.27%) were diagnosed under one year of age, in which 233 (55.48%) were positive cases. Children in this age group are not walking well, if at all, and the clinical signs of Trendelenberg gait and sign, etc., can not be fully utilized. More than three-quarters of positive cases were diagnosed under the age of

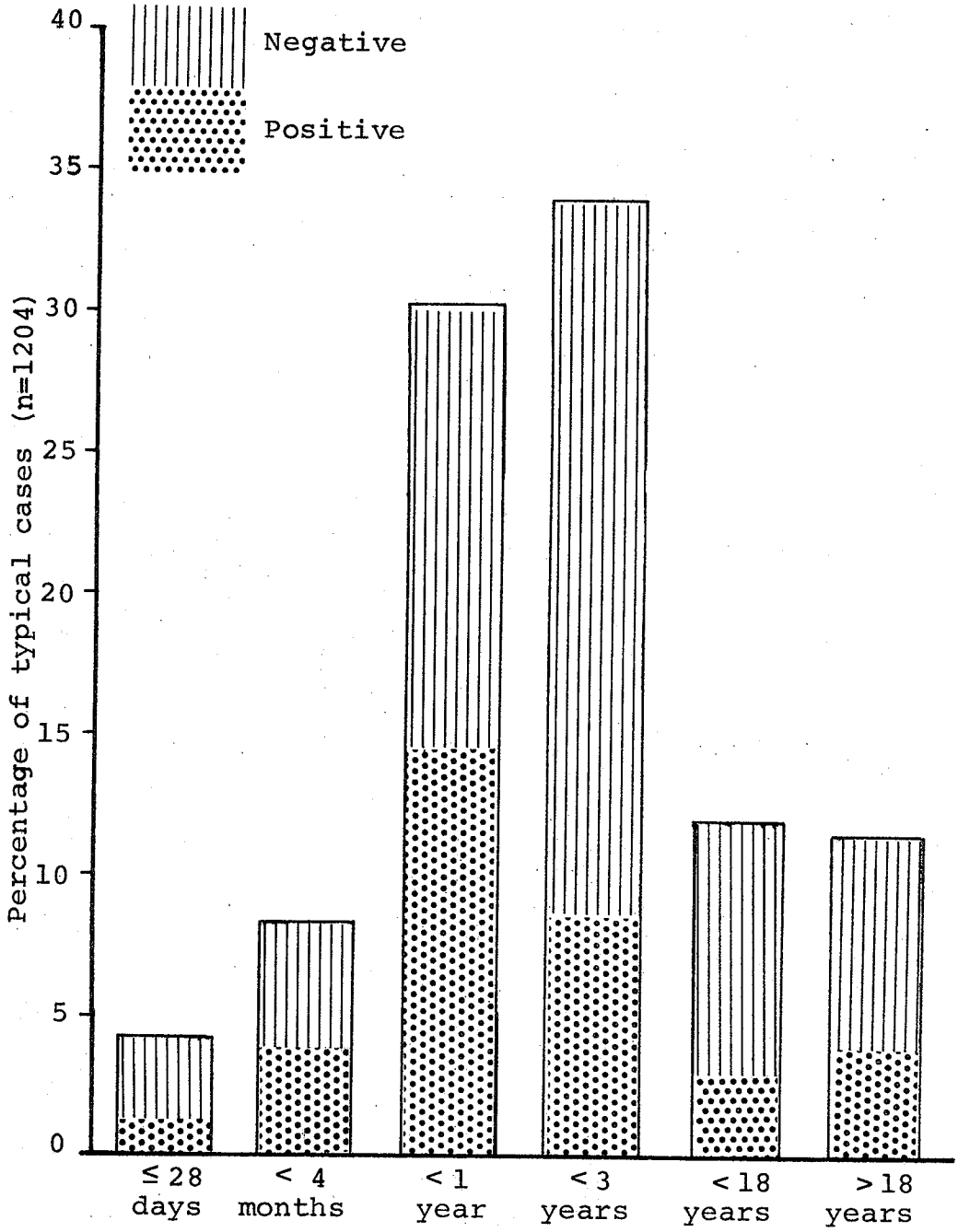


Figure 9

Age at Diagnosis in Six Age Categories
for Positive and Negative Typical Cases

three when walking is usually an established practice (n=343, 80.71%). One hundred and thirty-seven cases were diagnosed between ages 3 and eighteen, and 129 cases were ascertained at an age greater than eighteen years. Of 129, forty-seven (11.19%) were positive cases, many of them adults who have not been seen by a medical practitioner for their hips. Four reported cases, for whom chronological age was unknown, are included in the over eighteen years positive group.

CHARACTERISTICS

Sex

In 1204 Typical cases there are 587 males and 617 females. One hundred and forty-five males and two hundred and seventy-six females are positive, while one male is diagnosis unknown. The sex ratio is 1:1.05 (male to female). Fewer males with positive diagnosis were observed (145) than expected (205.1), and more females positive were observed (276) than expected (215.9) X^2_1 51.91, $p < .001$). Within the negative cases the male to female ratio is 1:0.77 compared with 1:1.03 in positive cases. The sex ratio is highest in the True diagnostic category, 1:2.48, while it is 1:1.51 in the Dysplasia category, and is least in the Other category, 1:1.07. For all positive categories a female preponderance is evident (Table 7).

Table 7

Frequencies and Sex Ratios by Diagnosis and Side Affected

Diagnosis	Frequency		Sex Ratio
	Male	Female	Male:Female
Negative	441	341	1 : 0.77
Positive[1]	145	276	1 : 1.903
Total	587	617	
Diagnostic Category			
True	70	174	1 : 2.48
Dysplasia	49	74	1 : 1.51
Other	26	28	1 : 1.07

Side Affected	Sex Ratio			
	True	Dysplasia	Other	Positive
Unilateral				
Left	1:1.9	1:1.22	1:1.3	1:1.55
Right	1:2.16	1:1.87	1:1.28	1:1.85
Combined	1:2	1:1.35	1:1.3	1:1.66
Bilateral	1:2.86	1:1.6	1:0.85	1:2.12

[1] χ^2_1 51.91 $p < .001$

Side Affected

In 420 Typical cases 651 hips were abnormal. The frequency of affected hips in the three diagnostic categories is presented in Table 8. Bilateral hip involvement exceeds unilateral hip involvement, a ratio of 1.22:1 (calculated on final initial diagnosis).

Within unilateral hip involvement the left hip is affected more than the right (1:0.68). Laterality findings are not significant at the .05 level. The sex ratio has greater disparity in the group with right hip involvement (1:1.85) than in the left hip involved group (1:1.55). Frequencies of side involvement with type of categories and type of diagnosis are presented in Table 9. Table 7 details the sex ratio for each diagnostic category by the side affected.

In ninety-six unilateral True cases, thirty-eight were right side affected and fifty-eight were left. The side affected was equal (26) for frank dislocation cases, but unequal in forty-four subluxation cases (12 right, 32 left). In 147 True cases with bilateral hip involvement, 117 were either frank dislocation, subluxations or a combination of the two. Radiological findings were reported for fifty-nine percent of True cases (n=144). In this group unilateral cases exceeded bilateral cases (subcategory frank dislocation and subluxations, ratio 1.43:1). Four percent of cases with

Table 8

Frequency of Affected Hips in Three Diagnostic Categories

Diagnostic Category	Frequency		
	Cases	Hips	Affected Hips
True	243	486	390
Dysplasia	123	246	183
Other	54	108	78
Total	420	840	651

Table 10

Side Affected : Reported Clinical Findings for Trendelenberg Gait and Sign Tests

Trendelenberg Gait		Trendelenberg Sign	
	Freguency		Result
Bilateral 'waddle'	34	25	Positive (all True)
		1	Negative (Other)
		8	Unknown
Positive Left	27	20	+ Left (16 True)
		3	Negative (Other)
		4	Unknown
Positive Right	32	24	+ Right (22 True)
		4	Negative (1 Dysplasia, 3 Other)
		4	Unknown

Table 9

Side Affected by Diagnostic Category and Type

Diagnostic Category	Unilateral		Bilateral	Total
	Right	Left		
True	38	58	147	243
Dysplasia	23	40	60	123
Other	16	14	24	54
Total	77	112	231	420
Percentage	18.28	26.6	54.87	
Diagnostic Type	**Unilateral**		**Bilateral**	**Percentage**
	Right	Left		
Confirmed	52	85	186	76.9
Suspected	24	24	39	20.7
Reported	1	3	5	2.14

χ^2 tests not significant

radiological findings were reported to be negative. Frank dislocations form 47.3 percent, with forty-two unilateral and twenty-nine bilateral. Subluxations form 46.6 percent with forty-one unilateral and twenty-nine bilateral. Dysplasia was reported for one unilateral case and two bilateral cases were frank dislocation plus subluxation, and seven were frank or subluxation with a contralateral shallow acetabulum.

In an individual of walking age, mature enough to co-operate, side involvement may be easily detected by the Trendelenberg sign and gait test. This data was available for 93 Typical cases (Table 10).

In twelve cases, only gait findings were located. Cases positive for Trendelenberg gait were not all positive for Trendelenberg sign test. Eight were negative, none in True category.

Right dysplasia with left 'CDH' was reported for three of 60 bilaterally affected cases in the Dysplasia category. In the Other category thirty-one hips were 'CDH', of which twelve cases (24 hips) were bilateral, and seven cases (7 hips) were unilateral.

Hospitalization and Treatment

Sixty-six (5.28%) of the total cases were hospitalized in connection with CHD. Positive, Typical cases number 55, with two negative Typical cases and nine positive Atypical

cases (0.72%). In the fifty-five positive Typical cases 47 (3.76%) were True, three (0.24%) Dysplasia and five were Other (0.4%). CHD was the primary diagnosis for 75.7 percent (n=50), with sixteen cases (24.24%) in which CHD was a secondary diagnosis.

Thirty-three cases were hospitalized and treated for CHD; twenty-nine were Typical, with CHD the primary diagnosis in twenty-four. Nineteen cases had bilateral hip involvement. At initial diagnosis 24.1 percent were over three years; 75.9 percent were under three years, with 48.1 percent under one year of age. Treatment given is summarized in Table 11. Plaster of Paris splintage was given to 27 cases, fifteen cases received innominate osteotomies, and fourteen cases received manipulation under anaesthetic. All cases received more than one form of treatment with the most frequent combinations being simple splintage, plaster of paris and manipulation under anaesthetic (n=5), and plaster of paris and innominate osteotomy (n=5). Treatment is only known for cases hospitalized. The figures presented on splintage omit an unknown number of infants who received pillow splints on the reserve.

Data on findings at a later medical examination are available for 64 of sixty-six hospitalized cases. Ten cases were seen once, thirty-seven had their diagnosis repeated,

Table 11

Treatment Modalities : Frequencies in Typical and Atypical Hospitalized Cases

Typical Cases		Treatment Type	Atypical Cases	
Yes	No		Yes	No
10	17	Splintage	2	2
27	1	Plaster of Paris cast	4	
9	18	Traction	1	3
14	13	Manipulation under Anaesthetic	3	1
7	15	Adductor Tenotomy		4
15	12	Innominate Osteotomy	2	2
7		Other Reconstructive Surgery	1	
(n=29)			(n=4)	

six were graded worse , six improved, one improved and subsequently negative, with four cases negative. Five of the 25 cases who received reconstructive surgery were seen by the investigator in 1972, and examined clinically without the knowledge that the individual had received surgery. Two of the five proved negative, one a right frank. In two cases seen once in hospital, one was graded a left frank and one a bilateral frank. Three of the five remained positive clinically.

Birth Characteristics

There were 1182 single births (417 positive); six twins, three males and three females, with one female a positive case. In fifteen cases this aspect was unknown (1.25%).

Data on birth presentation was available for 4.88 percent of the total sample, 3.32 percent (n=40) Typical cases of which thirty-four were positive. In forty Typical cases thirty-six had a vertex presentation with breech presentation reported for four positive cases. One breech presentation was delivered by Caesarean section.

Birth weight was reported for 4.8 percent of the total sample in which 4.4 percent (n=55) were positive CHD cases. The mean birth weight, Typical cases, (n=46), was 3.35 kilograms, with the mean for positive cases (n=41) being

3.39 kilograms. Birth weight was reported for one breech case (3.20 kgs.). The mean birth weight for the five Typical negative cases was 3.03 kilograms.

Familial Clustering

In sibships with proband(s) the sibsize has a range from one to 16. One quarter of siblings in 249 sibships were affected.[170] The range of affected siblings has a maximum value of seven in 9 (all diagnoses included). A preponderance of first borns is not evident.

Seasonality

In 1199 cases, the three months with the highest number of births over all years, are March (n=114), January (n=112) and November (n=111). The least number of births occurred in the months of August (n=74), February (n=86), and December (n=87). The three months with the highest number of affected cases are January (n=53), March (n=49) and November (n=46). Analysis of individual birth months, for homogeneity of positive and negative cases was not statistically significant. Males only were slightly significant at $p. < .025$ (X^2_{11} 21.92).

Birth months were grouped into quarters with the first, January to March; the second, April to June; the third, July to September; and the fourth, October to December.

Testing for homogeneity in births of affected and non-affected cases by birth months, in quarters was significant at $p < .005$ (X^2_3 15.23). In separate analyses for sexes, males retained a level of significance ($p < .01, X^2_3$ 12.21). Births expressed in quarter-year for positive and negative cases are presented in Fig. 10, and by sex, in Table 12. Birth months in quarter-year were grouped into winter (first and fourth) and 'summer' (second and third) half year. Testing for homogeneity of positive and negative cases was highly significant at $p < .001$ (X^2_3 13.36). The grouping of the quarters first and second, third and fourth, was not significant.

Consecutive temperature readings were available for the period 1955 to 1964. Variation over this period was not significant. Two five year periods 1955-1959, and 1960-1964, were selected and tested for homogeneity in births of positive and negative cases in quarter-year groups (First - January to March, etc.). This was slightly significant for the years 1955 to 1959 ($p < .025$ X^2_9 19.84). Separate analyses for each sex demonstrated that only males were slightly significant for years 1955 to 1959 (X^2_3 8.50, $p. < .05$). These results, for both sexes, are presented in Figure 11.

Diagnostic Criteria

Information related to diagnostic findings was

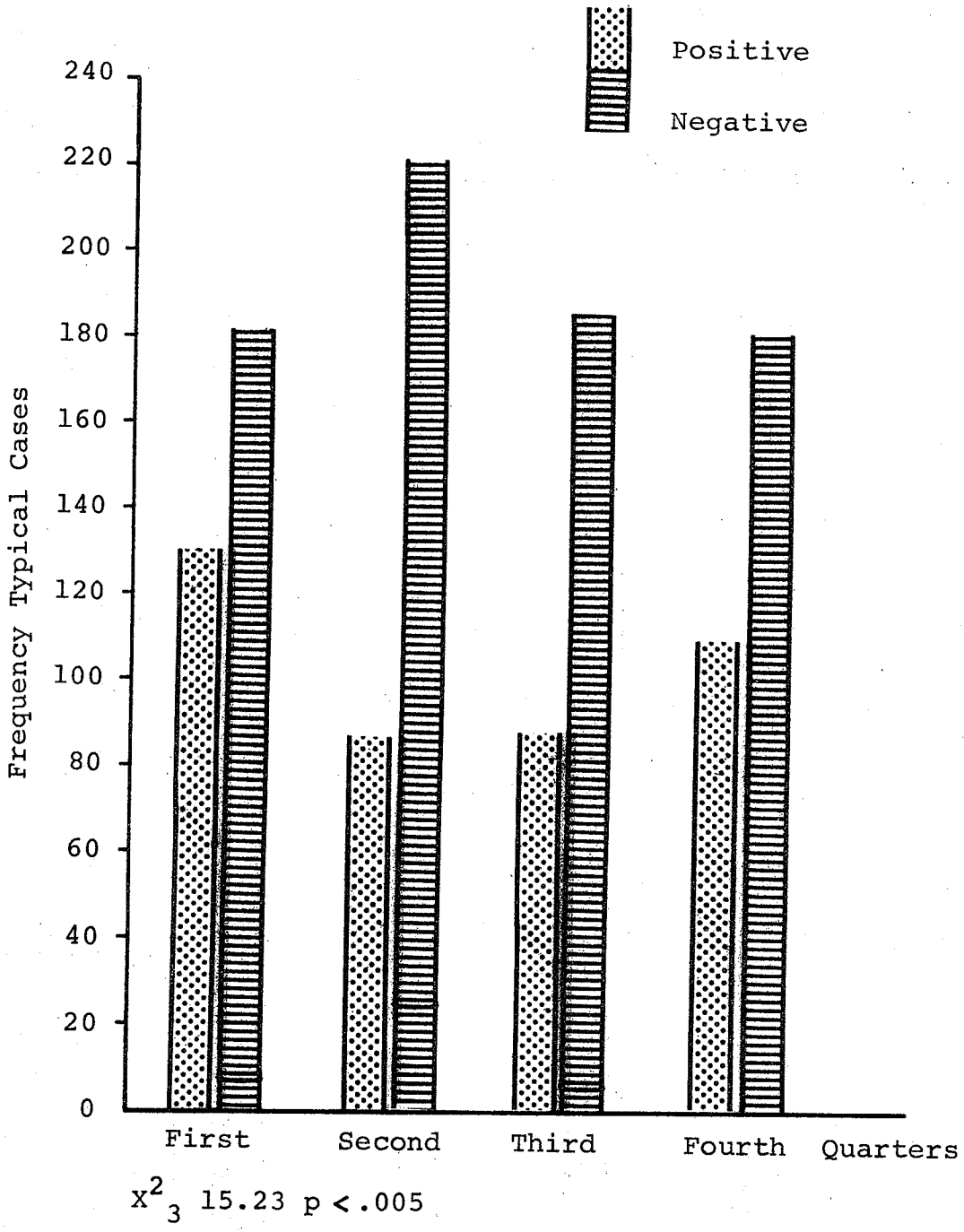


Figure 10

Frequency of Births in Quarter-Years
for Positive and Negative Cases

Table 12

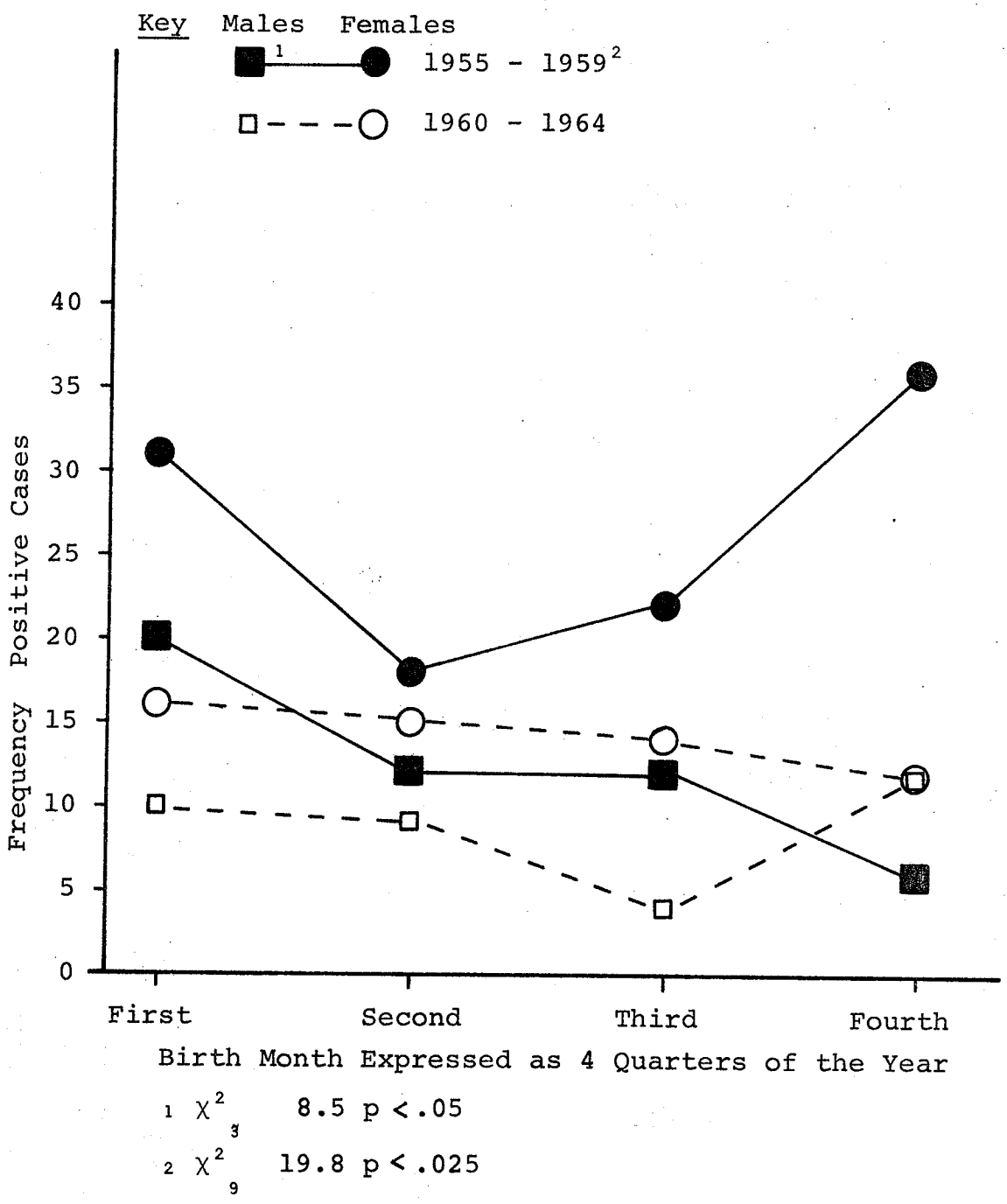
Frequency of Births By Sex
By Quarter Year

Sex	Diagnosis	Quarters				Total
		1	11	111	1V	
Males*	Positive	52	31	25	34	142
	Negative	98	125	106	105	434
Females**	Positive	78	55	62	77	272
	Negative	84	97	79	75	335
	Total	312	308	272	291	1183#

* χ^2_3 12.21 $p < .01$

** χ^2_3 7.44 $p < .1$

\# 21 cases with unknown birth month (1.74 per cent)



1 χ^2_3 8.5 $p < .05$

2 χ^2_9 19.8 $p < .025$

Figure 11

Frequencies, by Sex, for Births in Quarters of the Year, for 1955-1959, and 1960-1964

scanty. Data, with frequencies of cases for which data is known, for each test, and also expressed as percentage of total Typical cases, is presented in Table 13. Tests are listed in order of the frequency for which each was reported. This order does not reflect the clinical importance attached to each test.

Arthrography is the only test which is estimated to be accurately reported (excluding the possibility of non-located cases treated in hospitals not included in this investigation). The higher frequencies for tests of limb length, Trendelenberg gait, Trendelenberg sign, lordosis, and prominence of the Greater Trochanters reflect data collected during field work. These are all easily recognized, late-diagnosis signs. Presence or absence of pain was one facet of field work investigation. Except in two (hospitalized) cases, this is reported only for adults (n=8), all of whom have never been seen by a physician concerning their hip(s) problem or who were seen once, in 1949. Acetabular angle values were reported for 46 cases. The mean of the right hips (n=18) was 27.6^{o}, with a range from 13 to 44 degrees. The left hip (n=19) mean was 31.6^{o} with a range from 25 to 40 degrees. In 20 Dysplasia cases two hips had angles greater than 38 degrees, while in nine Other cases the value exceeded 38 degrees in four hips.

Percentages of the total positive for each clinical test, with frequencies, are presented in Figures 12 and 13.

Table 13

Clinical Tests: Frequencies and Percentages

Test	Percentage of Total (n=1204)	Number	Percentage Positive
Arthrography	96.42	1162	100.00*
Radiology	71.51	861	27.87
Limb Length	25.4	306	74.18
Trendelenberg sign	19.7	237	35.02
Prominence of Greater Trochanter	16.2	195	25.13
Trendelenberg gait	12.87	155**	65.8
Pain	12.3	148	5.4
Lordosis	11.9	144	8.33
Crude Abduction range	9.22	111	36.02
Skin Folds	9.05	109	24.7
Clunk test	8.63	104	11.54
Abduction range in degrees	7.3	88	22.73
Telescoping	6.89	83	25.3
Palpation Head of Femur	5.2	63	11.1
Acetabular size	3.99	48	97.9
Additional signs and symptons	2.57	31	100.0
Wide Perineum	0.58	7	71.43

* Cases given test (n=8)
** Figure omits children not at walking age at date of examination

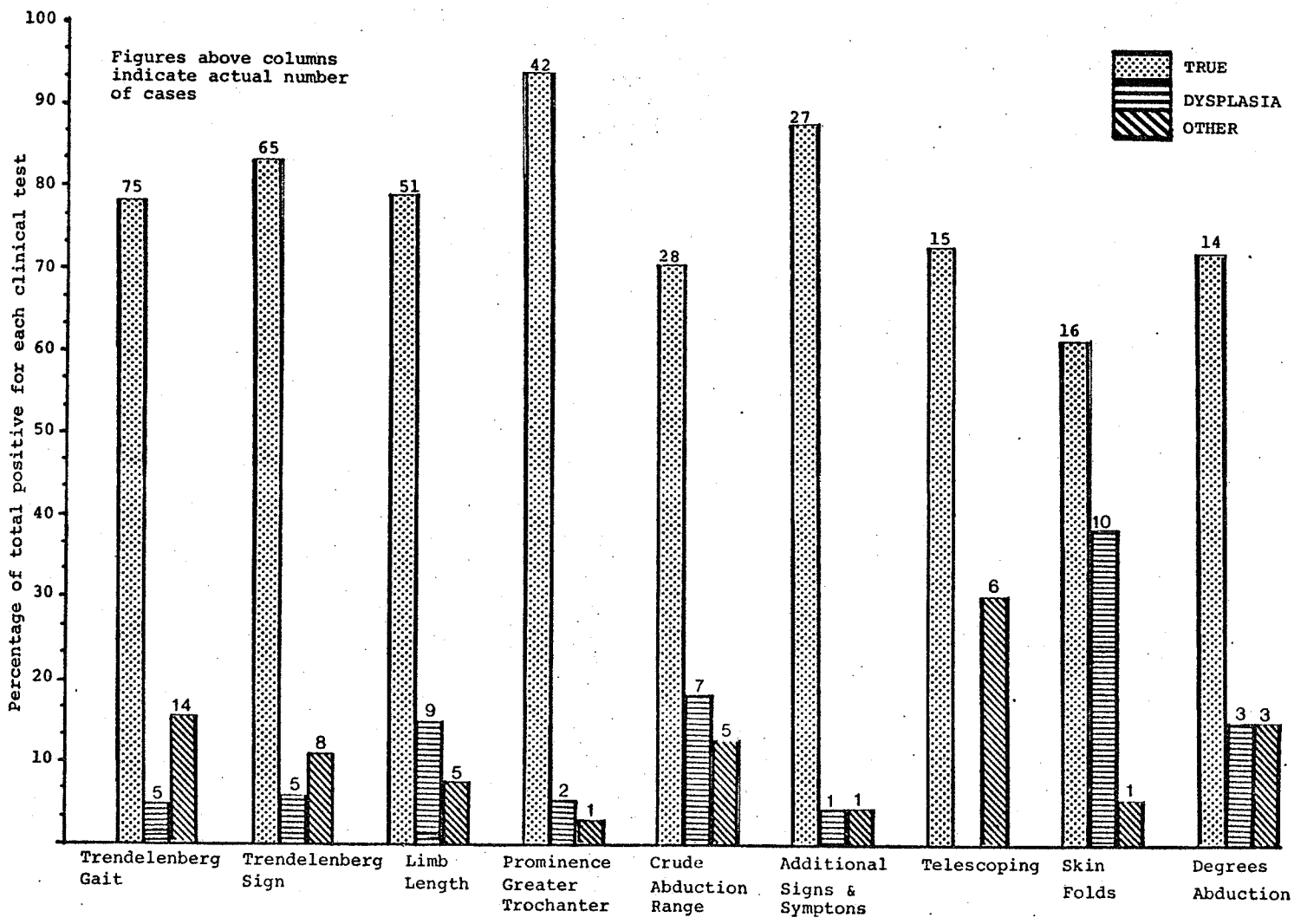


Figure 12

Clinical Tests Reported by Diagnostic Category (A)

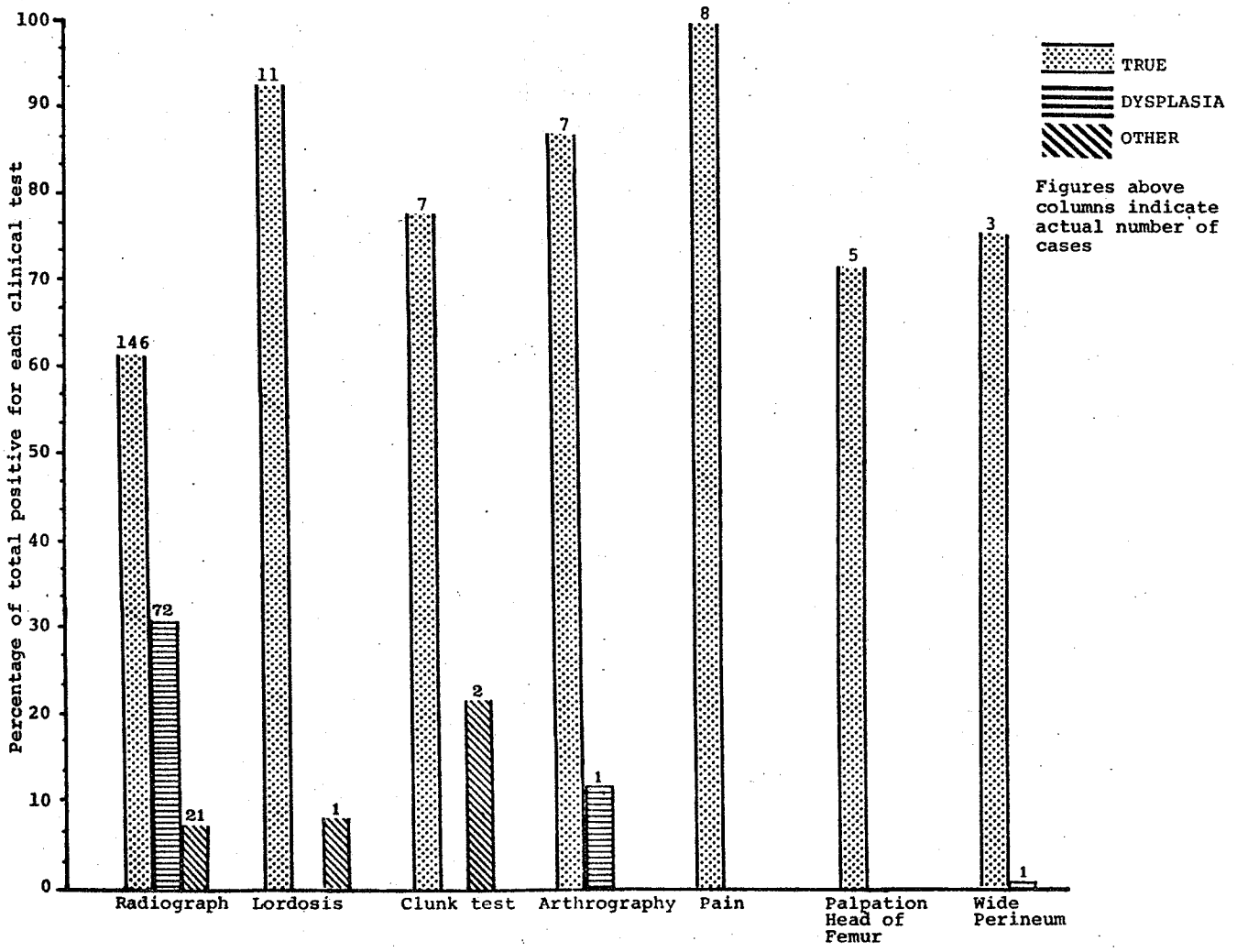


Figure 13

Clinical Tests Reported by Diagnostic Categories (B)

These demonstrate that the best reporting of clinical findings occurred for True cases (frank dislocations and subluxations). Due to the scanty data only a brief description has been presented.

Generalized Joint Laxity

Four hundred individuals, predominantly between the ages of five and sixteen years, were examined for generalized joint laxity. Fifty-five were positive for three or more paired joint tests. In the 55, thirty-four (13 males, 21 females), were negative CHD cases, and twenty-one (9 males, 12 females), were positive. Chi square tests for each sex, by CHD and laxity, were not significant.

Two hundred and fifty-three individuals tested had been seen and diagnosed negative for CHD. Fourteen of this group (5.53%) had positive clinical CHD status in 1972. All fourteen were negative for the joint laxity tests. Two of the fourteen were classified as left frank dislocations while the remaining twelve exhibited mildly abnormal findings for Trendelenberg gait, and/or sign.

Results of Subsequent Medical Examination

The majority of cases seen more than once by medical personnel were ascertained initially, and subsequently, at the annual surveys. A total of 627 (79.97%) negative cases,

and 168 (40.09%) positive cases were only seen once. Diagnosis was repeated in fifty-six (7.14%) negative, and seventy-six (18.14%) positive cases. Two negative cases (0.25%) and fourteen positive (3.34%) had deteriorated, while thirty-six (8.59%) were improved, but not normal. One hundred and thirteen positive cases (26.9%) were stated to be normal. This data is presented in Table 14.

Analysis was conducted to investigate the association between initial diagnosis, by the type of initial examination, and the field work findings. Cases diagnosed initially in 1972, and cases who had received treatment were excluded in order to detect cases exhibiting remission or spontaneous improvement.

One hundred and thirty negative cases, who had received x-ray and clinical examination at the initial examination, were seen in 1972. In the 130, ninety-nine had only been seen once by medical personnel. Positive clinical grading was given to fifteen, three of which were frank dislocations and had only been seen once by a physician.

Eighty positive cases, who had received clinical and x-ray examination at initial diagnosis, were seen. Forty-one had one medical, and twenty-five were graded negative (11 True, 14 Dysplasia). Twenty-six cases remained positive at a second medical examination and eleven of these were graded

Table 14

Results of Subsequent Medical Examinations

Initial Diagnosis	Frequency	Percent*	Subsequent Med. Exam Diagnosis	Frequency
Negative	627	79.97	Diagnosis	56 (7.14%)
Positive	168	40.09	repeated	76 (18.14%)
Negative	2	0.25	Deteriorated	
Positive	14	3.34		
Positive	36	8.59	Improved but not normal	
Positive	113	26.9	Normal	

* Percentages expressed as percent of diagnostic category

negative (7 True, 2 Dysplasia and 2 Other). Thirteen cases, on subsequent medical examination, were noted as improved and subsequently normal. In these 13, eight were negative, one was graded a left frank dislocation, and four were mildly abnormal for Trendelenberg gait and sign tests.

Ten cases seen in 1972 were diagnosed initially on clinical findings alone. All had received additional medical examinations at which 4 were graded normal, with three still positive. These three were clinically negative (Table 15). In all, 41 True, 27 Dysplasia and eleven Other cases were graded clinically negative in 1972.

FUNCTIONAL DISABILITY

Affected individuals (frank dislocations, uni- or bilateral) carry out identical routine daily activities around the home and in the community, as unaffected individuals. Twenty-five adults over thirty were questioned in relation to the presence of pain. One admitted to pain derived from the hip which interferred with normal activities. One male, (Case 2589), aged twenty-nine years was on welfare, and had been treated (surgery) at age twenty-six for a painful hip which prevented manual labor. This individual was observed to be active in construction of his own house. When the presence of pain was acknowledged, the site indicated

Table 15 Initial Diagnosis by Subsequent Medical Examination(s) Diagnosis by 1972 Clinical Status*

Initial Diagnosis	Frequency	Type of Initial Examination	Subsequent Med. Exam. Diagnosis	Frequency	1972 Status Type	Frequency
Negative	130	x-ray & clinical	seen once	99	positive (3 frank)	15
Positive	80	x-ray & clinical	seen once	41	negative (7 True)	25
			improved & later normal	13	negative (5 True)	8
Positive (True)	10	clinical only	still positive	6	negative	3
			improved & later normal	4	negative	4
Positive	14	x-ray only	seen once	9	negative	6
			still positive	2		
			improved & later normal	5	negative	4
Positive	6	clinical, x-ray not in same year			positive	3
					negative	3
Positive	45	x-ray, clinical not in same year			positive	26
					negative (15 frank)	19

* Each section is exclusive, cases selected on type of initial examination

by older individuals was the knee joint region. Frequently older people spend a considerable portion of the day sitting on the floor, with their feet tucked under their buttocks, a position which must surely contribute to the presence of pain.

Affected males are known to have been employed on freight transport between Norway House and Island Lake. Between 200 and 400 pounds were carried by each individual at the portages which averaged three-quarters of a mile, with the longest portage being one and three-quarter miles. The load carried by affected men was stated by these individuals, and supported by others at independent interviews, to be identical to that carried by unaffected individuals. One male, (Case 0205), a bilateral frank dislocation, was observed hauling freight while employed by a local airline company. No disability was obvious apart from the presence of a marked limp. Case 0348, a confirmed left frank dislocation complicated by poliomyelitis, has never walked, yet he was reported to have been the "best trapper" in his community. Case 1244, a left frank dislocation, was reported to be "the fastest man in his community on snow shoes" in his youth, and at age 58 is still actively employed.

Speed in walking over the relatively rough terrain - there are no level walkways in the entire Island Lake area - appears unaffected by the presence of a dislocated hip.

Several individuals reported that, with age they tire more easily over distances greater than five miles. Walking on the slippery ice characteristic of 'break-up', was reported to be more difficult by one female unilateral case. The sole disability admitted by an Island Lake individual (Case 0719, unaffected) was that it is difficult to portage a canoe with a man who limps. "He dips and you don't".

TIHKINĀKAN (CRADLEBOARD)

A present day tihkinākan has the approximate dimensions of length 29½ inches, a bottom width of 15 inches, a bar width of 14 inches, and a top width of 16 inches (Fig. 14). The inner birch rim has a height of two and three-eights of an inch and an overall length of 50 inches. The internal rim width increases gradually from approximately six inches at the bottom to nine and three-eights of an inch at the rim top. Adults interviewed gave the impression that formerly (time not established) the rim was less rounded and was slightly flatter at the bottom.

The rim is attached to the board by leather thongs (formerly Sinew or string[74]). Traditionally there are three pieces to a tihkinākan. The cradleboard with a half bag cover attached to the rim by leather thongs, the inner 'bundle bag' (or pouch bag, waspison, sometimes referred to incorrectly as

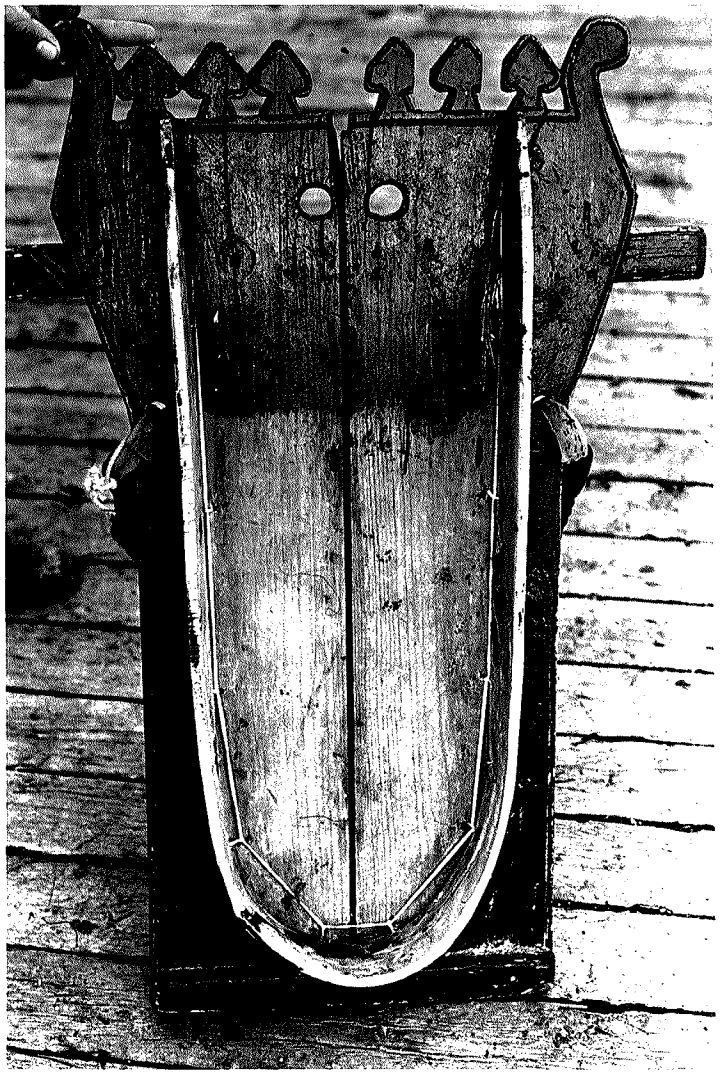


Figure 14.1 Cradleboard

Figure 14.2 Infant in Tihkinākan

the 'mossbag'), and the flannelette moss bag. Usage of the moss bag has steadily decreased since the 1960's and is rarely used today. Occasionally a grandmother will use a moss bag on a baby with a diaper rash. The moss used belongs to Genus Sphagnum, plants which apparently produce antiseptic substances and were used in surgical dressings through World War One.[100] Analysis of mosses used at Island Lake is given in the Appendix H, p. 152. The moss bag was like a joined double envelope, and moss was inserted into the two 'pockets'. The baby was placed on the opened bag, the two 'pockets' wrapped round each leg, then with the other wrappings placed in the waspison which was laced firmly. The waspison was then placed in the half-bag of the cradleboard and that was laced. Today diapers have replaced the moss bag, and many mothers have abandoned the use of the waspison, simply placing the infant, in all the required wrappings, directly onto the cradleboard. The infant's knees are held in extension with the lower limbs in approximately the neutral position, blankets are folded firmly over the legs and the outer bag is laced to the hip region. The arms are then placed at the side of the trunk, blankets folded over, and the outer bag is completed. Not all mothers are conscientious in ensuring that the feet are against the bottom rim and are not turned in. The babies are wrapped and laced securely enough so that

no 'slumping' of the infants in the cradleboard appears to occur.

Three sizes of cradleboards may be used depending on the size of the infant. A mother with a large family may require two cradleboards, and new bags may be made for each baby. Normally two bags are taken to fishing camps, and when the mother accompanies her husband to the traplines. Cradleboards may be borrowed, and they can be given to anyone when no longer required.

It is stated that cradleboards are used chiefly for travelling purposes outside the home; to go to the church, health stations, the Bay store, et cetera; that in the home babies are taken out, and do not sleep in them. Home visits revealed that several mothers do in fact keep their babies in the cradleboards in the home. Several mothers stated that their babies spend the day in, and sleep in their cradleboards. Only on two occasions was an infant observed out of the tihkinākan in the home. One was lying on a bed, another in a hammock strung between two walls. A greater number of mothers stated that the child, wrapped in a blanket, sleeps with the parents. Size of the home, number of rooms, and family size greatly influences whether an infant will sleep with the parents.

Mothers of probands interviewed stated, without

exception, they had used a tihkināākan (n ⩾ 30). Use of the bundle bag alone was preferred by one young mother. In this instance the baby was under 4 weeks of age. While several people reported usage of hammocks only one was observed. When the tihkināākan is not being carried on the mother's back it is commonly propped up in a vertical or semi-vertical position; on a bed, resting against a wall, or chair, or the mother's legs. Occasionally when the weather permits the tihkināākan may be strung up outside, from a tree branch. The tihkināākan is often rocked gently from side to side when an infant is restless. Breast feeding is performed without removing the infant from the cradleboard, or undoing any of the lacing.

Time did not permit evaluation of the general statement that the tihkināākan is not used once a child achieves walking age. It was observed that infants known to be walking (toddling) were brought to well baby clinics, to the Bay store, and to church in cradleboards. Further, when an infant was ill, he or she was brought to the health station in a tihkināākan.

Firm customs appear to exist on the correct placement of an infant in the tihkināākan. Older women commented that "young mothers today do not take enough care putting the baby in the tihkināākan". It was stated that the feet should

sit "square on the bottom", resting on the rim. Blankets (formerly moss) should be placed between the legs, particularly at the knees, "so that the legs are not pushed out at the ankles, and between the feet so that the feet do not turn in". "When you see people who walk like a bear you know that they did not pack the moss in right". A repeated statement was that the "baby must be put in tihkinākan properly or the legs will not be right".

Tihkinākan usage is still near 100 percent on the Island Lake reserve, in all four communities. Infants are placed in the tihkinākan from day one (when not delivered in hospital), and usage is often continued past commencement of walking. Except in illness, it is doubtful that infants are kept in cradleboards until eighteen months of age, or more, as occurred in the past. Island Lake mothers will use the tihkinākan when visiting Norway House or Winnipeg for the first time; however on subsequent visits, or should they remain in either community, they tend to stop usage. Cradleboards are not used by residents of those communities.

A small number of infant walkers and 'Jolly Jumpers' were observed. Apparently enthusiasm exists for their use. Both can be obtained from local Bay stores though the presence of only one doorway in one-roomed houses limits the use of 'Jolly Jumpers'.

SUMMARY

Description of data pertaining to Congenital Hip Disease at Island Lake over a 22 year period has been presented. Ascertainment rates in five year periods indicate declining incidence which is minimal when True cases only are considered.

Chapter 5

DISCUSSION AND SUMMARY

High incidence of CHD has been reported for the Island Lake reserve population. Description of the epidemiological features was presented in Chapter 4. Analyses which will permit discussion of the findings relative to other studies is yet to be completed. The following discussion will focus on case ascertainment, the factors of attitude, treatment, tihkināākan usage effects, and will conclude with recommendations on health care needs.

The present study was mainly based on data obtained at annual surveys. Two main types of sampling were involved; case controls or probands located from hospital records, and community studies. Kessler and Levin[104] considered the retrospective approach unsuitable for conditions which did not invariably lead to hospital or clinic visits. This is not the case for CHD at Island Lake, however, as the annual surveys are analagous and compensate for this defect. Initially, it was hypothesized that with attempted ascertainment of all newborns in each year since 1955, the data represented the population born in and after 1955. There were 1,525 live births

at Island Lake between 1955 to 1969[170] of which the data forms 94.13 percent.

Several biases exist in case ascertainment. The initial survey in 1949 specifically examined individuals with abnormal gaits, of any age. Apart from inquiry into affected relatives of probands in the late sixties, no attempt has been made to determine the number of affected adults. Field work was undertaken in an attempt to reduce this bias by locating, either in person or by name, affected adults. Since subluxation and dysplasia may not exhibit marked clinical signs, if any, under-detection of adults and adolescents is suspected. A potential bias in truncate ascertainment is omission of families with no affected sibs. Smith (1958:192) considered it was ". . . generally true that the more affected there are in a family, the more chance the family has of being included . . ." Singh[187] noted that certain families did not attend surveys, and that these were often reported to have a child with frank dislocation. He suspected the surveys under-reported the frequency of frank dislocations. High frequency families are documented. It is apparent from data that certain families were consistently present at treaty/survey time, and presented their children for hip examinations year after year, while other families appear never to have been examined. Increased probability of complete ascertainment,

of newborns, existed when surveys were conducted concurrently with treaty payments, since nearly everyone returned for that event. When separate hip surveys were conducted and were limited to the main communities by bulky radiological equipment, families at summer fishing camps were omitted. In some years surveys were restricted to Garden Hill and St. Therese Point. The greater concentration of reserve population in the latter communities is reflected in the 77 percent positive case ascertainment from those communities.

Anderson[5] noted the danger of over-representation of interesting families. This exists in the target population due to the high frequency of one surname, whose holders have gained a reputation for high prevalence of CHD. This led to the disease being casually termed 'X' disease at one Winnipeg hospital. At another, individuals with this surname and an Island Lake address have a definite elevated probability of hip examination and detection. Ten of eleven cases located at the latter hospital were given on admission CHD as a secondary diagnosis, despite previous survey ascertainment.

The initial hypothesis that search of personal files at the local health stations would result in complete ascertainment of examined affected cases was refuted. In the majority of examined cases, at routine medical examinations, or surveys, the CHD status positive or negative, was not

noted. Children sent south for treatment of other conditions therefore did not have CHD status recorded on their files. This is then detected, seemingly for the first time. Local nurses commented that the presence of a limp is so common that it fails to elicit special mention. A potential cause of labour difficulties due to malformed pelves,[21] it is surprising that frank dislocations are not noted at ante-natal examinations, or at delivery. Data was not located to indicate probands experienced greater difficulties in labour, nor were they significantly characterized by small families.

Surveys were conducted at specific times of the year. It can be expected that, in a number of cases, abnormality present at birth may have proceeded to normality prior to examination. Further, the data omitted infants who failed to survive to the survey date.

Failure to attend surveys is considered to be directly related to the local attitude towards CHD. Island Lake people view affected individuals more as a left-handed person may be viewed in urban societies. Adults questioned as to presence of affected individuals in their community may reply 'no one', yet the community will have several with severely abnormal gaits. Two of 26 reported affected adults were clinically negative, demonstrating local residents can distinguish a CHD limp from other gait abnormalities. CHD is not visibly

manifest at the age of preferred examination, neonatal, and many adults consider 'there is nothing the matter' with their infants at this age. Native midwives stated they can detect which babies will limp. It is not known if their knowledge of recognised clinical signs arises from native lore, or from observations of medical examinations with explanation of findings. Emphasis on the correct placement of a child in the tihkinākan indicates concern that the legs will be 'normal'. Failure to recognise CHD as a significant disability at any age, requiring treatment, lessened and lessens participation in surveys, and in carrying out treatment recommendations, without conscientious efforts by local health personnel.

Standardization of diagnostic measures with the importance of 'minimal observer error' was noted by Anderson[5] as important in investigations of this type. There have been three major medical examiners in the 22 years of data collection. Clinical signs considered reliable in the 1950's are either no longer valid since they occur with reasonable frequency in normal children[19,175], or their values have been upgraded. Acetabular angle values greater than 40 degrees are today considered diagnostically significant. The means for right and left positive hips fall well below this value. Normally, diagnosis is made on the constellation of signs present, however the data contains a percentage of infants

who received only one type of examination. Lack of detail in clinical findings does not permit further comment on possible standardization of clinical signs utilized.

An unexpected number of children examined during field work exhibited a mildly abnormal Trendelenberg sign, description of which has not been located in the literature. In unilateral stance, the pelvis is raised on the non-weightbearing side. Dislocation interferes with efficiency of hip abductor stabilization function, and in unilateral stance on the abnormal hip, the pelvis 'drops' on the unsupported side. Twenty-eight individuals (8 negative cases) showed abnormal findings in that the pelvis was neither raised, nor was there appreciable 'drop' on the unsupported side. In addition, 16 demonstrated mild abnormality in the gait pattern, towards a Trendelenberg type. This finding is difficult to interpret in the absence of radiographs which, it is suspected, may reveal mild subluxation or dysplasia which has altered the efficiency of abductor muscle action. Justification of the clinical import of this finding awaits further investigation which would include simultaneous radiography.

Inadequate data on birth presentation prevents comparison with findings of other investigators that a breech presentation baby is at greater risk. In the past, and occasionally today, birth occurs on reserve, or at fishing

camps in the absence of trained personnel, and details are not recorded. Native midwives reported that most births were 'easy'; few presenting buttocks first. Singh[187] considered the number of breech births located to indicate under-detection, and estimated a frequency of 12 annually since 1966. Not all were CHD cases. Personal files were searched for birth data, however; this search was incomplete with regard to negative cases. Houston[93] commented that medical practitioners in the north have reported a low frequency of difficult births in northern native populations. Increased risk to small babies suggested by Record and Edwards in 1958 is not apparent in the data (n=60).

Seasonal variation in CHD births, significant for males, is more noticeable in the late fifties than the early sixties. The number of affected births in the winter half of the year was highly significant (sexes combined, $p < .001$). Division of the year into winter and 'summer' halves may be more valid than the artificial division into calendar quarters. Other investigators have theorized that seasonality is an effect of the month of birth, or an indication of some factor operating on summer conceptions. Analyses for the total population were not significant. Small sample size and changing patterns in residence mobility, in this time period, probably account for the observed seasonality effect.

A preponderance of first borns is not evident in the data.

Treatment recommendations evolve from detection of abnormality. The effect of well motivated, but possibly poorly executed treatment in the initial stages of investigations into CHD at Island Lake is difficult to assess. Immobilization of affected children in plaster of paris spicas on the reserve met with a highly unfavorable reaction among the local residents. Infants were then taken to Norway House[33] and treated there. Unfortunately, one child died of gastroenteritis. This case is still recalled, with death attributed to the hip treatment. In an early survey outdoor photographs were taken of unclothed affected females, of varying ages. These individuals are mothers of present day children. The potential psychological trauma of this event may have contributed to local attitudes towards surveys and treatment.

Early efforts to promote wearing of pillow splints on all newborns as a prophylactic measure met with little success. Nurses have reported[33] that these were hung as an ornament, thrown away, and that few were worn, often only when visiting the health station. For treatment, other than simple splintage, infants were sent to Winnipeg, or for a period, to Edmonton. These children were frequently absent for a minimum of six months, often longer than a year. While

parents readily assent to children being 'sent out' for treatment of other conditions, which usually involves a shorter absence, there is considerable opposition to children leaving the reserve for hip treatment. The hospitalized child may spend a further period in a foster home before final return. During this highly formative period of development, children are exposed to another language (English), to different and more easily attained standards of hygiene, different foods, and often a higher standard of material welfare in terms of toys, television and personal attention. Considerable re-adjustment is required, both of the child and the parents, on return to the reserve.

Efforts to return children to the reserve in spicas, for the period of immobilization, instead of foster homes, have not proven successful. The child frequently returns in a few weeks with a sodden, filthy, ineffective plaster. In a number of cases the home conditions were not deemed suitable. The difficulties of transporting a child whose legs are fixed in a 'frog-legged' position, by boat and skidoo can not be discounted. Occasional cases of poor hygiene have reinforced the attitude held by some foster parents and social workers that it is preferable for children to remain in the south, until treatment is completed. Insufficient knowledge concerning the local conditions may contribute to these

attitudes.

Consent for treatment of a child is influenced by visible results of treatment. The child that left with a limp, yet still returns with a limp, albeit improved by medical evaluation, is, in the eyes of the local people 'no better'. Parents question the value of treatment from these results. As the majority of cases detected and treated early, can achieve normal status, medical personnel recommend treatment of all affected cases. It is reasoned that affected individuals may not always reside at Island Lake. With increasing exposure to values of modern urbanized societies it is likely that CHD will be eventually viewed as a significant disability. It is probable that an affected (frank dislocation) individual residing in the south would gradually become aware of their unique status, and be exposed to social and economic discrimination. Should parents have the right to refuse treatment in cases of frank dislocation, detected under one year of age? The degree of personal motivation, and the lack of recognition of disability, by affected and unaffected Island Lake individuals alike, play major roles in determining whether an individual leads a normal or restricted life. Functional disability in everyday activities was neither admitted nor observed, and only one application for welfare was based on inability to perform manual labor due to hip

abnormality.

Efforts to reduce tihkināk̄an usage have met with failure, not due to opposition to medical recommendations, but because of the practicality of the tihkināk̄an in the Island Lake environment. Transport is by boat and skidoo, and there are no level walkways which would permit use of a perambulator. The infant in the tihkināk̄an is kept warm despite a rigorous winter. Securely bound, the infant is in no danger of burns from unprotected iron stoves, falling into the water, or crawling/toddling into trouble. No change in tihkināk̄an use is likely to occur for as long as present travelling methods and terrain remain.

The tihkināk̄an is considered potentially harmful to the 'dislocatable' or dislocated hip as it maintains the joint in an abnormal postnatal posture of extension and adduction. It seems unlikely, while not verified, that the 'swaddled' hip is in true extension and adduction, but that the hips are adducted and extended relative to the normal postnatal posture of flexion and abduction. Rabin, et al. (1965:11) commented that, "the straight-legged position tends to pull the head of femur away from its resting place in the acetabulum." He observed on a radiograph of one Navajo child that the legs are not maintained in firm abduction, and that "the child can partially abduct his lower limbs."

Houston[93] considered a maximum of 15 to 20 degrees of abduction possible in a cradleboard. Diaper usage by both Navajo and Island Lake people ensures that the hips are in slight abduction. Further, there is the custom of placing sufficient wrapping between the legs "so that the legs will be straight." Greater flexor tone limits extension in the neonatal hip, and it is likely that lordotic motion in the flexible lumbar spine contributes to the extended position in the cradleboard.[199] Radiographic investigations are required to establish the exact hip and spinal posture.

Island Lake and Navajo Indians frequently prop the cradleboard in a vertical position. The infant hip may be weight bearing at an early age.[161] In bilateral stance, each hip would transmit half the body weight. Considerable weight will be taken throughout the entire body due to the firm lacing extending to the upper chest, in a tihkināk̄an. This produces a reduction in load forces across the developing hip. The leg length disparity present in unilateral dislocation will produce adduction of the pelvis on the longer side, and increase the pressure forces across this, the normal hip. The resultant forces through the normal hip would be directed towards the lateral margin while the load on the shorter, dislocated hip, will be reduced. Currently dysplasia is considered to be a healing phase of the dislocated

or dislocatable hip.[176,161] When the hip is flexed and abducted the head of femur is 'well seated' in the acetabulum, and will provide normal stimulus for development of a deep, concave structure.[176] The combined factors of neonatal ligamentous laxity, a semi-extended and minimally abducted hip posture, with lack of pressure stimulus, may combine to retard the normal development of the acetabulum and produce dysplasia, with or without dislocation. Spontaneous improvement, noted in cases diagnosed during the neonatal period, may be prevented.

Neonatal cases constitute less than 5 percent, and some of the late-diagnosed dysplasia cases may have been dislocated or 'dislocatable' hips at birth. The suggested harmful effect of cradling is reduced by reports of spontaneous improvement despite cradleboard usage. Rabin, et al.[161] found spontaneous improvement in 9 of 13 cases while on cradleboards. Two children with dislocations were never on cradleboards. At Island Lake where cradling is estimated to approach 100 percent, 113 cases underwent spontaneous improvement. At field work, 41 frank dislocation or subluxation cases were clinically negative. Genetic analysis, to be performed, may enhance understanding in this area should significant heritability estimates be found in cases which fail to improve, compared with those which exhibit spontaneous

improvement. Diagnosis of CHD in neonates further mitigates against cradling as a prime etiological factor.

There is a second potentially harmful effect of tihkinākan usage. The most prevalent illness of young children in northern communities is chest conditions. Mothers tend to keep the tihkinākan fully laced up. The child's arms are held firmly down against the sides of the thorax, free arm movement and thorax mobility is thus restricted. The absence of arm mobility which has a secondary effect on the thorax, and the restriction of normal thoracic motion through tihkinākan usage, in infants up to walking age, may impede normal development of thoracic mobility, and strength of thoracic musculature. This practice may contribute to the multifactorial etiology of chest conditions in this population. Further, mothers rock the tihkinākan as soon as the baby starts to cry, thus limiting the ventilatory effect of crying. Support for this hypothesis was derived from observations that many children with 'chests' demonstrated shallow, ineffective coughs.

Rabin, et al.[161] hypothesized that primary abnormality in pelvic and acetabular shape could be responsible for the high incidence in the Navajo population. The Navajo range of acetabular values fell well within the limits of normality for each age group, and mean values were practically identical

to those reported by Caffey, et al.[29] in a Caucasian population. No difference was detected between adult Navajo and non-Navajo pelves. The acetabular angle measures the slope of the acetabulum relative to a horizontal. (Fig. 2, p.29). It does not measure acetabular depth shown by Cheynel and Huet,[40] and Getz[71] to vary significantly between ethnic groups. A wider range of variability, within normal limits, may be present at certain ages. It is hypothesized that the Cree-Ojibwa acetabular depth dimension may exhibit disparity with Caucasian hips, whose values are utilized in detection of the abnormality. This has been demonstrated in the Lapps, a 'swaddling-high incidence' population.

Woolf[231] and Sasaki[178] concluded exogenous factors may be more important in females than males. Sasaki suggested that traits demonstrating the vertical extensions of the acetabulum (i.e. depth) were more influenced by the environment. A preponderance of females with a significant seasonality effect may indicate a greater environmental component in CHD causation, while a sex ratio closer to unity and absence of a seasonality effect may indicate a higher genetic component. Sex disparity is most marked in the True category (1:2.58), and is considerably less than the 1:6 (♂:♀) observed in 1950. The female excess is low compared with other reported findings.

Comparison with Coleman's[45] finding of higher bilateral hip involvement when a family history was present will be permitted when analysis is completed. Few studies have reported the bilateral hip preponderance observed at Island Lake. Coleman's findings may be repeated. The sex ratio in bilateral hip involvement was (True, 109 ♀:38 ♂) not more equal than in unilateral hip involvement, as reported by Carter and Wilkinson.[37] The finding of greater frequency of bilateral hip involvement in the True category (dislocations and subluxations, 52.25%) than in the Dysplasia category (34.95%)* is similar to that reported by Czeizel, et al.[50] It is probable that a polygenic mechanism regulates or controls the development of the acetabulum.[233] In a population characterized by physical isolation, with observed frequent consanguinity and high inbreeding coefficients,[170] it is hypothesized that genetic mechanisms may play a greater role, and may also account for the bilateral hip preponderance.

RECOMMENDATION

Establishment of a community hospital at Island Lake. Population size, and utilization of in-patient beds in the south by Island Lake people, with factors detailed below,

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* Percentages calculated on confirmed diagnosis cases.

justify this recommendation. Minor surgery (adductor tenotomies), traction, and application of plaster spicas could be performed by a visiting surgeon; the hospital itself staffed by nurses. Under the present conditions, antagonism to CHD treatment is greatly influenced by the treated child's long period of absence from the community. Such a facility would enable parents to visit their offspring, would lessen socio-cultural 'dislocation' and problems of re-integration into reserve life. Further, infants would be exposed to their own language during the period of language acquisition.

Considerable reduction in health care costs may be derived with in-patient facilities at Island Lake. Cost of infant hip treatment includes airfares to and from Winnipeg, to which escort fare and accommodation in Winnipeg must be added. Cost of treatment is considerable. One case, in whom treatment commenced at eighteen months, incurred expenses of approximately $4286 over six months, with treatment incomplete. A second case, diagnosed at 20 months, completed the treatment program in 18 months save for reviews, and incurred costs of $11,769.00.[33] MacKenzie[119] noted the exponential increase in treatment costs, for late-diagnosis cases which require more than 'simple splintage'. Acceptance of treatment, with an understanding of the benefit derived from early treatment must be gained if institution of rigorous

neonatal detection is to be worthwhile, for reasons other than prevalence rates.

CHD education programs would most profitably be directed at the younger section of the population, and should be incorporated into school courses as Human Biology, Health, and Physical Education. It is suggested that information should be increased in detail over successive grades, to ensure that tomorrow's parents will seek and accept medical treatment of affected infants. Mean school leaving age, for both sexes, should be determined in the planning of the suggested education program, to ensure exposure to CHD information prior to leaving school. The practice of loosening the top lacing of the tihkināk̄an when inside a building, is considered worth emphasis in community and school health programs.

Investigation of plastic materials currently utilized in, for example, scoliosis braces, with the aim of providing a lighter, easier to clean and more durable spica.

It is suggested that information related to type of hip examination, hip status (negative and positive), treatment recommendations, and parental response to suggested treatment, should be accorded the same status on personal health files as the immunization record. Family history of CHD clearly noted on health files may increase the probability

of hip examination in offspring.

The local attitude to CHD should be taken into greater account when planning treatment for children diagnosed well after commencement of walking. Realistic time periods for treatment should be given to parents, even when a period greater than six months is indicated. Part of the antagonism to treatment is surely derived from provisional statements that the child will return in 'a few weeks, or months', yet the child is absent for a considerably longer period. Such instances weaken the relationship between Island Lake people and health personnel.

SUMMARY

Ascertainment rates for True cases (dislocation or subluxation) are the highest reported for any population, and are considered reliable indicators of congenital hip disease in the Island Lake Cree-Ojibwa population. There is a small excess of female affected cases, and preponderance of bilateral hip involvement in all diagnostic categories. Genetic factors are highly suspected to play major roles in disease causation. Description of disease characteristics with discussion of social factors has been given. Analyses to be conducted by Dr. D. Rokala, will interpret observed incidences in terms of heritability, and enable prediction

of 'risk', of utility in genetic counselling.

Despite the high incidence of this 'disabling' disease, function in everyday activities exhibits little impairment, emphasizing that health is a relative matter. What is considered a crippling disease in one population, may not be so regarded in another. Knowledge of local attitudes towards specific disease processes is important to ensure acceptance of treatment programs, and that treatment will, by local rather than medical evaluation, improve an individual's ability to function normally within his environment.

APPENDIX

Appendixes

APPENDIX A

Table 1. Associated Abnormalities Reported in Congenital Dislocation of the Hip Studies

Type	Reference
Spina Bifida	50,163,51
Arthrogryposis Multiplex Congenita	7,91,15,23,191,88
Talipes	50,119,158,163,174,233
Torticollis	50,119,145,163,174
Hernia	50,163,234
Cleft Palate	50,158,163
Genu Recurvatum	126,234
Plagiocephaly	214,223,234
Marfan's Syndrome	36,89,162,188
Aplasia	158,234
Chromosomal Deletions	148,174
Amyotonia Congenita	158,163
Congenital Cutis Laxa	164
Ehlers-Danlos Syndrome	131
Cerebral Palsy	20
Central Core Disease	12
Scoliosis	234
Congenital Heart Disease	158
Syndactyly, Polydactyly	50
Strabismus	50 (28.6%)
Congenital Dislocation of the Knee	234
Congenital Ocular Pathology	47

APPENDIX B

Diagnostic Tests

Test Position

Unless otherwise specified, all tests are performed with the subject lying on his back (supine), legs towards the examiner.

Tests

1. Ortolani's sign (1973): With flexed knees, the hips are flexed to a right angle and then abducted. The head of femur can be felt, when the test is positive, to slip across the posterior lip of the acetabulum.

2. Barlow's sign (1962): Barlow has found this test [Ortolani's sign] unreliable in the newborn and has modified it slightly. He carries the similarly flexed leg into mid-abduction and presses forward on the great trochanter with the fingers while holding the thumb in the femoral triangle. . . . A slip can then more easily be felt and often seen. Thumb pressure over the lesser trochanter can redislocate the head. If the head reduces as the thumb pressure is released, Barlow calls the hip 'unstable - no dislocated but dislocatable'. Von Rosen independently also reached this method of examination. (Strange, 1965:69)

Barlow's test, also termed the Salford test, has more control over rotation than Von Rosen's test but the results are equal.[18]

3. Hart's Test: for limitation of abduction in flexion.

. . . the lower limbs are first adducted and extended. The thighs - still adducted are then flexed to a

> right angle with the trunk. With the tips of the fore and middle fingers round the knee the thighs are abducted . . . . Interpretation : In the early weeks of life decreased abduction, whether unilateral or bilateral, is highly suggestive of subluxation of the hip joint. (Egan, et al. 1969:7f.)

The 90° of abduction present in the newborn hip decreases to between 60 and 70° of range by the ninth month.

4. Telescoping : The femur is pressed rostally, then pulled caudally, to detect excessive hip joint motion.

5. Galeazzi's sign : 'Apparent femoral shortening' is applicable for unilateral dislocations at all ages. "The knees flexed, feet flat, and the limb is examined for shortening (apparent) of the femur" (Tachdjian, 1967: 328).

6. The Birth Posture is considered by Wilkinson (1966:1108), ". . . an accurate record of prenatal posture, but only represents its final stage." The 'position of dislocation', locked external rotation breech malposition is an easily recognisable sign (Fig. 2.2, p. 29).

7. Trendelenberg sign : In the erect position, when a limb is raised from the ground, the pelvis drops to the unsupported side, and the trunk sways over the stance limb. Normally this is prevented by the action of the hip abductors, gluteus medius and minimus, acting on the stance hip, to maintain coronal balance. In hip dislocation a stable fulcrum is absent. The Trendelenberg sign appears in the gait every-

time weight is taken on the affected limb (Trendelenberg gait), and is most marked when the condition is bilateral.

8. Roentgenographic Determinants : Several roentgenographic determinants which can assist the diagnosis of neonatal hip dysplasia are given by Coleman (1968:185, Fig. 1.1, p.18).

1. an acetabular index above 40°
2. lateral disposition of femoral 'beak' with relation to the vertical line of Perkins
3. unilateral* shortening of "H" line of Hilgenreiner
4. failure of the acetabular roof to reduce its slope during the early months of growth; and
5. failure of the roof to form a graceful curve over the developing femoral head.

The acetabular index (Hilgenreiner's angle) is not applicable after the disappearance of the "Y" cartilage.[201] Used up to 36 months of age, an angle which exceeds 25° indicates dysplasia is present.

9. Joint Laxity : Carter and Wilkinson (1964:1) devised a series of tests to determine generalized joint laxity. This was diagnosed when three of the following tests were positive.

1. passive opposition of the thumb to the flexor surface of the forearm
2. passive hyperextension of the fingers so that they lie parallel with the extensor suface of the forearm
3. ability to hyperextend the elbow greater than 10°
4. ability to hyperextend the knee greater than 10°

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* Italics in the original

5. an excessive range of passive dorsiflexion of the ankle and inversion of the foot.

The tests, numbered as presented above, are shown in Fig. 5, p. 64. Wynne-Davies[234] accepted 0^{o} for the knee and elbow tests, and dorsiflexion of the ankle greater than 45^{o}.

APPENDIX C

Table 2: Geographic and Ethnic Incidence of Congenital Dislocation of the Hip

Locale/Ethnic Group		Time Duration of Study	per 1,000	Reference	
Sweden	Malmo	8	1.7	171	#
		7	2.1	153	#
		2	3.0	153	#
		10	5.7	153	*
	Goteburg	2	19.5	91	#
	Uppsala	3	24.4	90	#
	Lapps		24.6	134	*
	Uppsala	3	33.1	99	#
Finland		1	0.6	111	
	Lapps		50.2	142	
Norway	Lapps(pop.)		0.12	71	
	Lapps		50.2	216	*
Hungary		11	10.5	155	*
	Budapest	5	27.5	50	*#
Poland			38.6	52	#
Italy,North			4.3	180	*
Yugoslavia			405.0+	27	#
Israel	Rehovet	c.4	2.5	108	#
	Tel Aviv	12	2.7	215	#
	West Jerusalem	3	5.8	82	#
	Jerusalem	7	9.8	133	#
Great Britain					
	Birmingham	10	0.65	163	*
	Salford	3	1.55	19	
	Glasgow	1	3.2	233	##
	Middlesex	4	4.1	67	#
	Southampton	1	5.9	224	#
	London		6.1	228	
	Edinburgh	1	6.2	233	##
	Edinburgh	7	7.1	137	#
	Dublin	1	7.7	147	#
	Salford		18.2	18	#
	Aberdeen & N.E.Scotland	10	21.8	126	#

: includes neonatals; ## : only neonatals;
* : orthopaedic patients
+ : data probably invalid

Table 2 (continued)

Locale/Ethnic Group	Time Duration of Study	per 1,000	Reference
New Zealand			
Auckland	14	3.4	158 #
Wellington	3	8.5	191 #
Australia			
Brisbane & Adelaide	14	1.2	38 *
Japan	6	0.24	145 #
Niigata-Shi		1.01	102 *
	7	44.0	103 #
Hong Kong	10	0.01	92 *
South America			
Santiago		c.0.37	200 #
Medellin		c.0.24	200 #
Mexico City		c.0.08	200 #
Panama City		c.0.18	200 #
Africa			
Johannesburg Bantu		0.11	58 #
Kampala		2.0	167 *
North America			
Winnipeg	1	0.225	114 #
British Columbia	11	c.1.0	113 #
New York	2	0.4	182 #
Salt Lake City	2	6.1	232 *
Utah	8	0.67	232 *
Minnesota	6	1.03	84
Minnesota	10	1.08	83
New York	7	1.44	132
North Saskatchewan Cree-Chipwyan		min.7.0	98
Many Farms,Arizona Navajo	3	10.9	128 *
Fort Defiance Navajo	3	c.11.7	21 #
New York		17.1	23
Fort Apache Apache		min.30.5	109
Many Farms, Arizona Navajo	6	33.5	161
New Mexico	4	35.7	73 *
Manitoba Cree-Saulteaux	1	36.0 (est,60.0)	47
Fort Defiance,Shiprock, Gallop -Navajo		66.6	44

: includes neonatals,
* : orthopaedic patients

APPENDIX D

Table 3 Prevalence of Breech Presentations in Index Cases (23 studies)

CDH patients Percentage	Comment	Reference
3		126
6	mode of delivery known for 216 of 342 cases	133
9		214
11	0.1% of survey population	191
11.4	1.9% of normals	158
11.4	frequency in all births 3.1% ($p < 0.001$)	50
14		147
15	known for 95 of 264, 15% despite 169 unknown, usual incidence 1.25%	140
16	plus 9% with therapeutic version late in pregnancy	37
17.3	4.4% in 8,814 normals	19
17.7	Neonatals	233
17.9	Late-diagnosis	233
19		125
20	higher than general population	215
21	4 times that of controls at all birth ranks, correlated with low birth weight	166
21	delivery with history of version	67
22	hospital incidence, live births 4.6%	158
23	incidence in normal population less than 5%	175
25	figure excludes all that may have undergone spontaneous version	224
72	2.6% of all births in 1968-69	223
0	Ugandans	167
0	Maryland,U.S.A.	139
0	897 in 16,678 births, South African Bantu	58
0	Cree-Chipewyan,Canada	98

APPENDIX E

Detected Congenital Abnormalities

The most frequent type of congenital abnormality detected in addition to congenital hip disease, was Talipes Calcaneo-valgus (n=28). Thirteen were graded severe at an age of one year or greater. Additionally, twenty-two cases were diagnosed as mild Calcaneo-valgus, under the age of two years. These cases are considered within normal limits (Singh, personal communication 1972) and were placed in the Typical Group. There were three cases of umbilical hernia, two of which also had calcaneo-valgus (one severe). There were two cases of sacralization of Lumbar five vertebra, both detected incidentally on radiographs. Under-reporting of this anomaly in the total sample is possible.

The Atypical group includes five poliomyelitis cases. All five had poliomyelitis prior to CHD diagnosis and one additionally had Talipes Equino-varus. These five were placed in the Atypical group due to doubt as to the etiology of the hip dislocation. The Typical group includes one case with established poliomyelitis, post CHD diagnosis.

Congenital eye abnormalities (e.g. congenital

corneal opacity) was observed in a number of individuals (mainly adults) in 1949. No attempt was made to detect this anomaly which was not otherwise reported in the data available.

APPENDIX F

Atypical Group

Atypical cases form 3.6 percent of the total sample, with forty-four cases positive and one negative. In the 44 positive cases, thirty (30) are classified True, ten (10) 'Dysplasia' and four Other. Forty-two positive cases have a confirmed diagnosis; two positive and one negative have a probable diagnosis. There are no reported cases in this group. The three probable cases were ascertained prior to 1972 and form 6.6 percent of all probable cases.

Neonatal cases constitute 2.2 percent and late-diagnoses cases 97.7 percent. Ten cases were diagnosed between the years 1949 to 1959, with thirty-two cases diagnosed between 1964 to 1969 when a more detailed examination was given at annual surveys. Thirty-five cases (77.7%) were diagnosed under one year of age; three (6.6%) under three years, five less than eighteen years (11.1%), with one case over eighteen (2.2%).

The sex ratio, male to female, is 1:1.58. Seventeen males and twenty-seven females have a positive diagnosis. One male was negative. Sixty-eight hips are affected in a

total of 90 hips. Bilateral cases number 24. Twenty were unilateral; fourteen left and six right. The right to left ratio is 1:2.33. Frequencies for side affected, in the three diagnostic categories, and by type of diagnosis are presented in Table 16.

All cases were single births. Type of birth presentation was known for twenty-one positive cases. There was one breech presentation and twenty vertex presentations. Birth weight data was available for fourteen (14) cases. The mean weight is 3.23 kilograms, the range 2.38 to 4.19 kilograms, with the mode 3.18 kilograms (n=3). Seven True cases, seven Dysplasia and two Other cases were normal deliveries. Three cases were first borns, one sibsize two and two sibsize five. Seven cases were sibsize eight; three birth order eight.

Thirty-seven cases were located at annual surveys. Eight cases located from hospital medical records proved to have been initially seen at annual surveys. Radiological examination was given to forty-one cases. No data for this was located for five. In thirty True cases radiological findings were present for twenty-six. There were twelve frank dislocations, nine subluxations and four negative cases. The diagnostic tests reported, frequencies for cases with data, and cases positive for each test in the total number

Table 16

Atypical Group : Side Affected by Diagnostic Category and Diagnostic Type

Side Affected	Positive	Diagnostic Category True Dysplasia	Other
Unilateral			
Left	14	8 5	1
Right	6	3 1	2
Bilateral	24	19 4	1
Total	44	30 10	4

Diagnosis	Confirmed	Diagnostic Type Probable	Suspected	
Negative			1	
Positive				
Unilateral				
Left	14			
Right	5	1		
Bilateral	21	3		
Total	40	4	1	45

positive, is presented in Table 17, and Figures 15 and 16.

Nine positive cases were hospitalized (0.72% of all hospital cases). In five CHD was the primary diagnosis, with secondary diagnosis made in four cases. Four cases received treatment in hospital for CHD (12.12% of all hospitalized and treated cases). One case was a bilateral frank dislocation, and three cases had the left hip affected, (two frank dislocations and one a subluxation). CHD was the primary diagnosis in the four treated cases. A resume of treatment received is given in Table 11, p. 89.

Twenty-one cases (47.73%) were seen once by medical personnel, and twenty-three were seen more than once. The diagnosis was repeated in nine cases (20.45%), four were 'improved but not normal' (9.09%), three 'improved and subsequently normal' (6.8%) with seven cases normal (15.9%).

Seventeen cases were examined in 1972 for generalized joint laxity and nine (7 females, 2 males) were graded positive. Five of these nine were True cases, three Dysplasia and one Other. In the 17 cases, fifteen were graded clinically negative (negative Trendelenberg sign and gait test, no limb inequality or marked lordosis). Two cases remained right frank dislocations. Two non-ambulatory cases were not examined.

Table 17

Atypical Group; Clinical Tests, Frequencies and Percentages

Test	Percentage of total (n=45)	Number	Percentage positive
Arthrography	100.00	45	100.00*
Radiology	91.1	41	75.6
Limb length	73.3	33	69.7
Clunk test	68.86	31	9.7
Abduction range in degrees	66.6	30	3.33
Skin folds	62.3	28	37.7
Crude abduction range	60.0	27	40.74
Palpation Head of Femur	46.66	21	4.76
Telescoping	44.44	20	30.0
Prominence of Greater Trochanter .	20.00	9	44.4
Additional signs & symptons	17.7	8	100.00
Trendelenberg gait	15.55	7**	85.7
Trendelenberg sign	13.3	6	83.3
Acetabular size . .	4.4	2	0.0
Lordosis	2.2	1	100.0
Wide Perineum . . .	2.2	1	100.0

* Cases given test (n=2)
** Figure excludes children not walking at date of examination

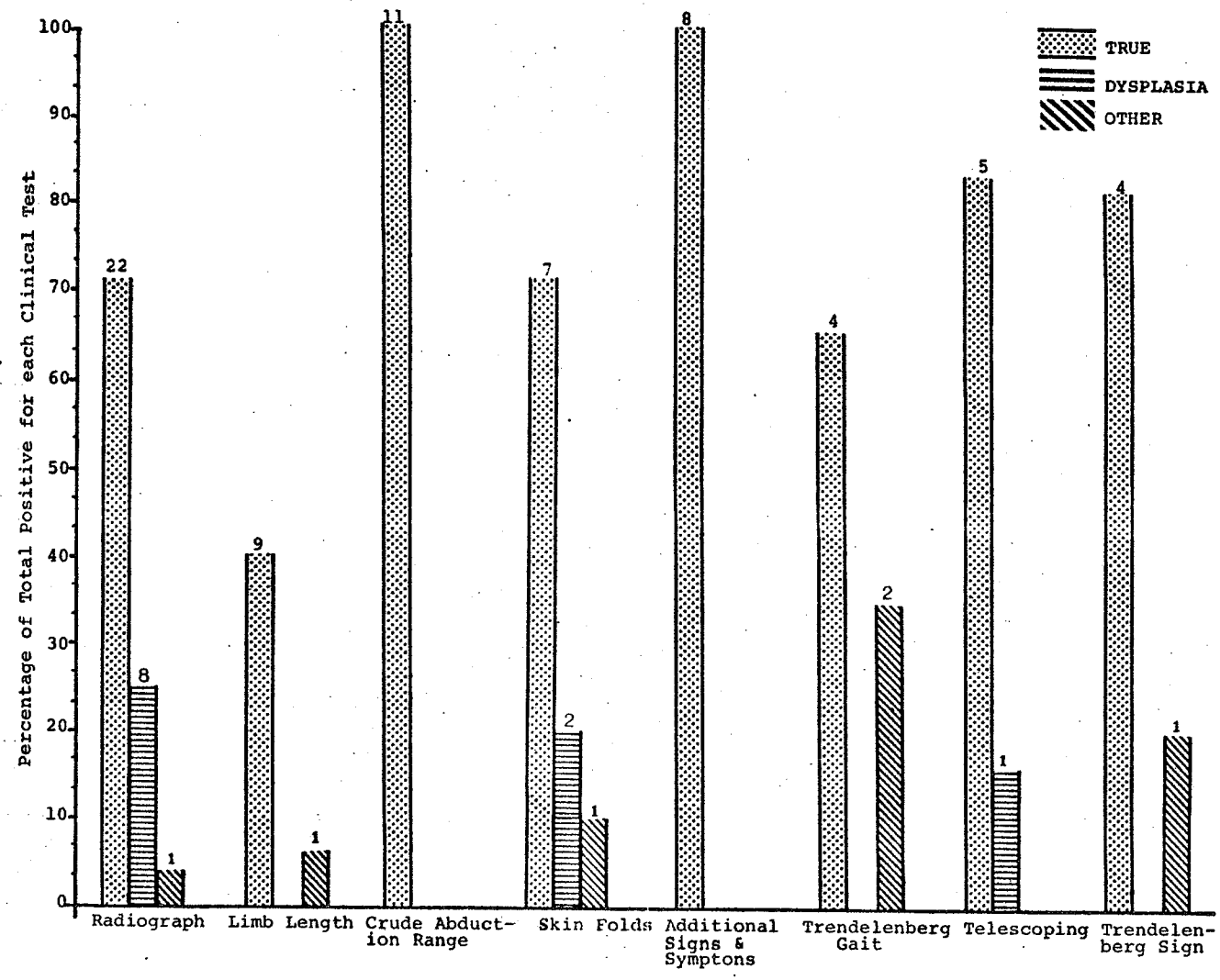

Figure 15

Clinical Tests Reported by Diagnostic Categories (A) Atypical Cases

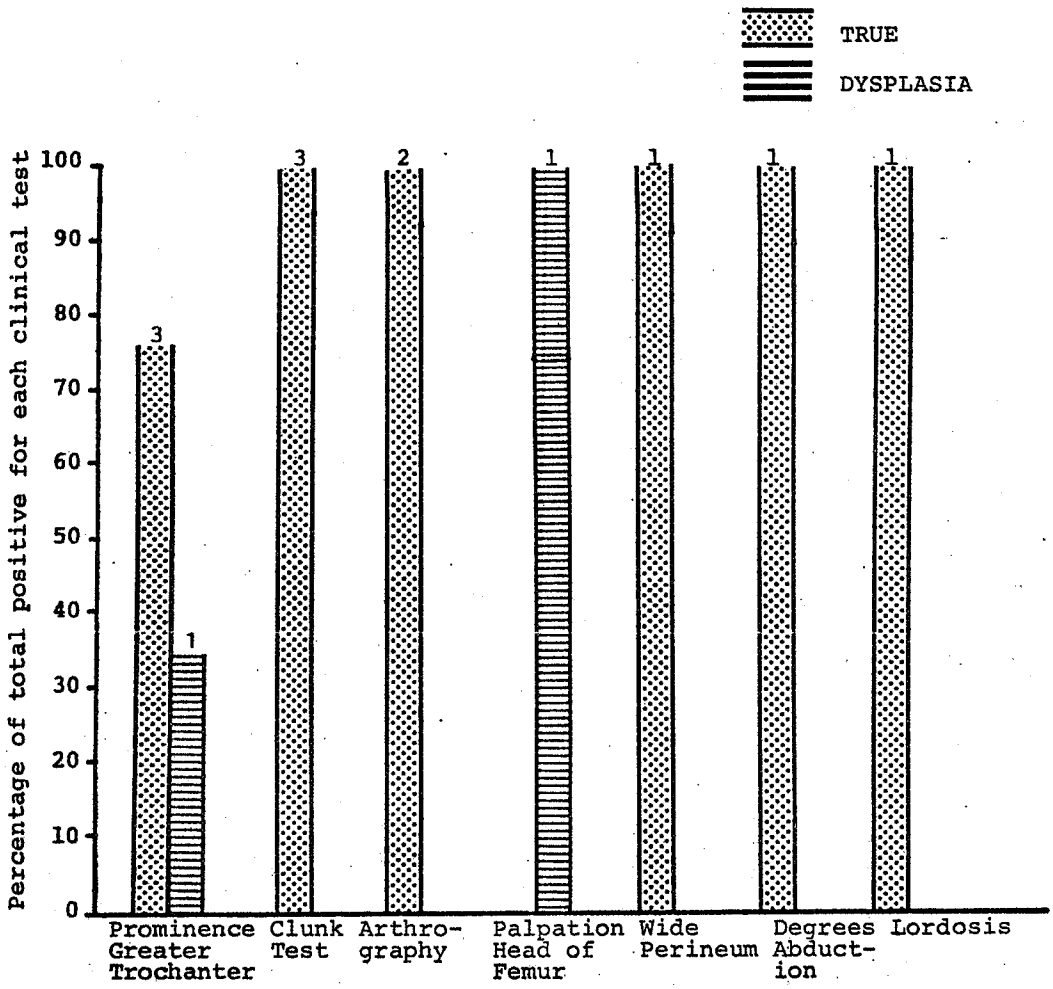

Figure 16

Clinical Tests Reported by Diagnostic Categories (B) Atypical Cases

CHD COMPUTER

1. NAME CODE — 1

2. CARD NUMBER — 5

3. DATA SOURCE — 7

01 = Annual Survey
02 = Child.,Hos. Med.
03 = Gen.Hos. Med.
04 = Rehab.Hosp.
05 = D.A.Steward
06 = St.Bon.Hos.
07 = Shriner's Hos.
08 = Charles Cam.Hos.
09 = Nor.Hse.Ind.Hos.
10 = Reserve Nurs.Stat.
11 = On Reserve,Walker
99 = Other

4. SEX — 9

1 = male
2 = female
9 = unknown

5. BIRTHDATE — 10 — Day Mth Year — 16

7 digit
ie 7th May,972 =0705972
9999999 = Unknown

6. BIRTH — 17

1 = single
2 = twin
9 = unknown

7. SIBSHIP SIZE — 18

8. BIRTH ORDER — 20

99 = unknown

9. BIRTH WEIGHT — 22

in ozs.
8lbs 12ozs = 140
999 = unknown

10. BIRTH PRESENTATION — 25

1 = vertex
2 = breech
9 = unknown

11. BREECH PRESENTATION — 26

1 = frank
2 = version
3 = history version
4 = not breech
9 = Unknown

12. DELIVERY — 27

1 = normal
2 = low forceps
3 = high forceps
4 = Caesarian
5 = other
9 = unknown

13. PREGNANCY LENGTH — 28

1 = full term
2 = premature
3 = postmature (>40wks)
9 = unknown

14. DATE OF DIAGNOSIS — 29 — (6) — Day Mth Yr

15. TYPE OF DIAGNOSIS — R. L. — 35

01 = frank dis.
02 = frank + dysp.
03 = sublux.
04 = subl.+ dysp.
05 = dysplasia
06 ='Stiff'
07 = unstable
08 = 'CDH'
09 = other
10 = normal
99 = unknown — 39

16. DIAGNOSIS KIND

1 = MD confirmed
2 = suspect,probable
3 = reported
9 = unknown

17. STATUS 1972 — 40

1 = + R.Frank
2 = + L.frank
3 = + bilat.fr.
4 = + mild gait & Trend.
5 = + gait only,mild
6 = + Trend. only,mild
7 = negative
8 = deceased
9 = unknown

18. ASSOC. ANOMALIES — 41

01 = Cer. Palsy
02 = Spina Bifida
03 = Men.Ret. + Microc.
04 = Arthrogry.
05 = Tal.Eqvarus
06 = Tal. Cal.valgus
07 = Talipes
08 = Torticollis
09 = Cleft Palate
10 = Other
11 = NOT REPORTED
12 = POLIO.
13 = Umbilical hernia
14 = C.Heart Dis.
15 = Synostosis R/U jts
99 = unknown

19. HOSPITALIZATION FOR — 43

1 = CHD
2 = Other
3 = NOT
9 = unknown

20. DATE FIRST X-RAY — 44 — Mth Yr

9999 = not taken

21. CLINICAL EXAM. — 48

1 = complete
2 = gait only
9 = not given/ unknown

22. DATE FIRST CLINICAL EXAMINATION — 49 — Mth Yr

9999 = not given

23. RADIOLOGY — 53 — R. L.

01 = frank
02 = fr. + dysp.
03 = fr. dysp. fal.acet.
04 = fr.dysp.f.ac.,DUFE
05 = fr.dysp.f.ac.def.H.F.
06 = lat.pos.H. Femur'
07 = lat.pos.+ dysp.
08 = lat.pos.,dysp. DUFE
09 = lat.pos.dysp.def.H.F.
10 = shallow acet.
11 = sh.acet. + antever.
12 = sh.acet.+ DUFE
13 = fal.acet.(dev. or est.)
14 = NEGATIVE
15 = NO DETAILS GIVEN
16 = other
17 = DUFE
99 = unknown

CODE FORM

24. X Ray ACET.MEASUREMENTS

TYPE: 1 = 'CE' — 57
2 = Sharps
9 = unknown

MEASURE — 58 — R. L.
R:32 L:39 =3239
9999 =unknown
7777 = increased
8888 = normal

62

25. ARTHROGRAPHY

1 = yes
3 = NOT
9 = unknown

26. ' CLUNK ' TEST — 63 — R. L.

1 = positive
2 = negative
9 = unknown

27. ABD.TEST TYPE — 65

1 = in flexion
2 = in extension
9 = unknown

28. ABDUCTION RANGE — 66 — R. L.

1 = limitation
2 = Grt. than 50°
9 = unknown

29. DEGREES ABDUCTION — R. L.

9999 = unknown

30. TELESCOPING — 68 — NC 7 — R. L.

1 = positive
2 = negative
9 = unknown

31. PALPATION HEAD FEMUR — 9 — R. L.

1 = yes
2 = no
9 = unknown

32. PROMINENCE GRT.TROCH. — 11 — R. L.

1 = yes
2 = no
9 = unknown

33. WIDE PERINEUM — 13

1 = yes
2 = no
9 = unknown

34. LIMB LENGTH — 14 — R. L.

1 = equal
2 = real shortening
3 = apparent "
4 = no shortening
9 = unknown

35. TRENDELENBEERG SIGN — 16 — R. L.

1 = positive
2 = negative
9 = unknown

36.TRENDELENBERG GAIT — 18

1 = positive right
2 = positive left
3 = bilat./waddle
4 = not walk.date exam.
5 = negative
9 = unknown

37. PAIN — 19

1 = in hip
2 = in back
3 = down leg
4 = not confin.to hip
5 = NOT STATED
6 = NOT PRESENT
9 = unknown

38. LORDOSIS — 20

1 = present
2 = negative
9 = unknown

39. SKIN FOLDS — 21

1 = symmetrical
2 = asymmetrical
9 = unknown

40. OTHER SIGNS & SYMPTONS — 22

1 = ↗ ROM Add.
2 = ↙ long dist.walk.
3 = inab.do heavy man.work
4 = muscle wasting
5 = abn.jt.lax.lst clin.
8 = other
9 = unknown

41. YEAR JOINT LAX.TEST — 23

99 = not

42. GEN.JOINT LAXITY — 25

1 = positive (3+)
2 = negative (≤ 2)
9 = unknown

43. SPLINTAGE — 26

1 = yes
2 = no
9 = unknown

44. P.O.P. CAST — 27

1 = yes
2 = no
9 = unknown

45. TRACTION — 28

1 = yes
2 = no
9 = unknown

46. M.U.A. — 29

1 = right
2 = left
3 = bilateral
4 = NOT
9 = unknown

47. ADDUCTOR TENOTOMY — 30

1 = right
2 = left
3 = bilateral
4 = NOT performed

48. INNOMINATE OSTEOTOMY — 31

1 = right
2 = left
3 = bilateral
4 = NOT performed

49. OTHER HIP TREATMENT — 32

1 = surgery
2 = applicances
3 = other
9 = unknown

50. SUBSEQ. MD EXAMIN. — 33

1 = seen only once
2 = diagnosis repeated
3 = given more sev.status
4 = given less sev.status
5 = " & subseq.normal
6 = NORMAL STATUS given
7 = reported case never seen
8 = seen 1, no details

APPENDIX H

Island Lake Moss

Analysis conducted by Professor R. E. Longton, Department of Botany, University of Manitoba, revealed that moss used in the mossbag was largely composed of *Sphagnum fuscum* Klingrr., with smaller amounts of *Sphagnum capillaceum* (Weiss) Schrank, *Sphagnum magellanicum* Brid., and a variety of other mosses.

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