

*Original Research Article***To Swaddle, or Not to Swaddle? Paleoepidemiology of Developmental Dysplasia of the Hip and the Swaddling Dilemma Among the Indigenous Populations of North America**

SAMANTHA H. BLATT*

Department of Anthropology, Boise State University, Boise, Idaho 83725-1950

Objectives: Clinical prevalence of developmental dysplasia of the hip (DDH) is high among modern indigenous populations of North America, yet no systematic study of the paleoepidemiology of this group exists. This study discusses the skeletal criteria, epidemiology, pathophysiology, and risk factors of DDH. A range of cases of DDH from an archaeological Native American population are described and the clinical and prehistoric prevalence of DDH among indigenous populations of North America are calculated and discussed within a biocultural perspective.

Methods: Pelves of 390 adults from the Late Prehistoric (1490 B.P. \pm 70) Buffalo site, West Virginia, were examined for DDH. Morphology of true and false acetabula was classified and other changes of the pelvis, lower limb, and spine were noted along with cranial deformation, providing evidence of infant restriction. Prevalence of DDH among living and archaeological indigenous peoples of North America were calculated and compared. Cranial deformation was assessed as evidence for swaddling.

Results: DDH was identified in 18 adults from Buffalo, resulting in a minimum prevalence of 46.15 per 1,000, within the range reported in modern indigenous groups in North America. Most, but not all, of the DDH cases were associated with cranial deformation, but not all cases of cranial deformation were associated with DDH.

Conclusions: The etiology of DDH suggests that components of both genetic predisposition and swaddling practices have combined to create a high-risk environment for the development of DDH, contributing to high prevalence within archaeological populations, like Buffalo, and modern Indigenous groups of North America. *Am. J. Hum. Biol.* 27:116–128, 2015. © 2014 Wiley Periodicals, Inc.

Developmental dysplasia of the hip (DDH) is an epidemiological conundrum with a controversial etiology. The clinical literature regarding DDH is vast and complicated due to changes to and disagreement over definitions of the disorder, different methods of diagnosis, different age categories of patients, variation in examiner experience, and biological variation in the populations studied (Noordin et al., in press; Roposch and Wright, 2007). Bioarchaeological understanding of DDH is only beginning to catch up with clinical assessments, but biocultural and paleoepidemiological studies can enlighten risk factors and etiology of the disease and even guide potential prevention and treatment programs in high-risk populations. This study will review the most current clinical contributions to the etiology of DDH, compile such knowledge with reports on modern and archaeological prevalence of the disorder within indigenous populations of North America, and integrate these within a cultural context.

DDH is a pathological spectrum that ranges in severity by varying degrees of misalignment of the femoral head from the acetabulum (Aufderheide and Rodriguez-Martin, 1997; Godley, 2013; Ortner, 2003; Weinstein, 2013). In the past, this condition was called congenital dysplasia/dislocation of the hip, referring to complete dislocation at birth. However, a higher frequency of DDH cases begin with only a slight abnormality of the acetabulum and develop over the life of the individual into more severe forms, taking tissue growth and differentiation into consideration, hence the clinical appropriateness of the name change recognizing it as a developing pathology (Gulati et al., 2013; Harcke, 1999; Mitchell and Redfern, 2008, 2011). Misalignment can be the result of trauma or congenital abnormalities in the joint which increase the like-

lihood of further damage (Aufderheide and Rodriguez-Martin, 1997; Godley, 2013; Ortner, 2003; Weinstein, 2013). Trauma can cause dislocation of the hip, but in such cases, it is no longer called DDH, but rather traumatic hip dislocation. Therefore, DDH is a nontraumatic, nonpainful event in babies, and should not be confused as a traumatic pathology.

The mildest form of DDH, acetabular dysplasia, may have no clinical manifestations, but is characterized by the containment of the femoral head within a shallow acetabulum. Though all subluxated hips are by definition anatomically dysplastic, subluxation is a more severe stage and involves partial loss of contact between the joint compartments. Dislocation is the most severe form, with disruption and complete loss of the normal anatomical contact between the joint components. By consequence, the femoral head compensates for the lack of a secure socket by forming and articulating with a false acetabulum, sitting posterolaterally to the true acetabulum (Dunn, 1976; Ortner, 2003). All ranges of DDH have the potential to lead to osteoarthritis and the need for hip replacement after the fourth decade of life, which along with diagnosis, is the main focus of clinical reports. To be detected in the dry bone, however, the disruption of joint activity must be associated with modeling or change in

*Correspondence to: Samantha Blatt, Department of Anthropology, Boise State University, 1910 University Drive, ID 83725-1950, USA. E-mail: samanthablatt@boisestate.edu

Received 24 January 2014; Revision received 28 July 2014; Accepted 20 August 2014

DOI: 10.1002/ajhb.22622

Published online 15 September 2014 in Wiley Online Library (wileyonlinelibrary.com).

the subchondral bone in or adjacent to the joint. This means that dysplasia and subluxation is less likely to be identified and reported than dislocation in skeletal populations (MacFarlane, 1980; Ortner, 2003; Storer and Skaggs, 2006).

Overall, DDH is not uncommon clinically, with a global incidence of 1 to 34 per 1,000 live births and by ultrasonographic detection in 25 to 50 out of 1,000 infants (known as neonate instability). Only in the last decade have sizeable case series been reported in archaeological/historical populations (Marfet et al., 2007; Mitchell and Redfern, 2007, 2008, 2011; Noordn et al., in press; Schoenecker and Flynn, 2007), leading some to infer temporal change in prevalence and to speculate as to why (see Table 4). Consequently, the paleoepidemiology, prevalence, and identifying criteria of DDH amongst populations in antiquity remain relatively unknown and possibly under- or misreported. Furthermore, the few paleopathological studies which have focused on DDH exclusively, report cases mostly from historic cemeteries in Europe that do not necessarily represent natural populations with biological affinity, therefore may not adequately represent heritable etiology of a disease. Recent studies have found lack of significant differences in prevalence of DDH through time among Western European, Mediterranean, and Middle Eastern populations (Blondaux and Millet, 1991; Lodder and Skopelja, 2011; Mafart et al., 2007; Mitchell and Redfern, 2008, 2011). The most notable of these is Mitchell and Redfern (2007, 2008, 2011), who devise a diagnostic classification system for the DDH spectrum in dry bone, including four distinctive types of dislocation anomalies. Further, Mitchell and Redfern (2007) compared the incidence of dislocation between the large Spitalfields cemetery collection and a modern Manchester population (from ultrasounds) and found that there was no significant difference in incidence despite medieval to modern changes in diet, healthcare, and gene flow. These authors go on to state that improvements in healthcare and obstetrics then, may not impact the incidence of dislocation.

Though invaluable in providing a diagnostic range from large collections, such studies cannot provide references for the deep prehistory of DDH, its prevalence among foraging and horticultural communities, and geographically, ethnically, and genetically limit our understanding of the disorder. Weighted mean prevalence of DDH is reported in modern indigenous populations of North America to be as high as 76.1 per 1,000 (Clabeaux, 1977; Lodder and Skopelja, 2011), the highest rate among modern populations, yet there has been no systematic review of DDH in prehistoric North America or any attempt to delve into the local indigenous paleoepidemiology of this anomaly (Kelsey, 1977; Storer and Skaggs, 2006).

The aim of this article is to (1) present the prevalence of DDH among prehistoric and living indigenous populations of North America from available reports, (2) present the largest specific study of DDH in a prehistoric skeletal population from North America, and (3) discuss both the prehistoric and modern epidemiology and pathophysiology of DDH among indigenous North Americans within a cultural-historical perspective with applied clinical knowledge. It is hypothesized that the prevalence of DDH among ancient indigenous groups of North America will not be significantly different from their modern/ living counterparts. Furthermore, this article will explore the

correlation of cultural practices like swaddling and cradleboarding with evidence of cranial deformation in combination with DDH from the target population.

Pathophysiology

About one quarter of the cases of DDH present at birth tend to resolve without treatment as instability diminishes in the first week to first month of life due to increased muscle tone, though some tests become less reliable with increasing age (Dezateux and Godward, 1998; Tegmänder et al., 1999). As diagnostic techniques have become more sensitive, the reported incidence has risen as less severe cases of dysplasia are more easily detected. This does not, of course, mean that the true underlying incidence has changed. In the case of ultrasonographic diagnosis, some studies have reported spontaneous resolution of neonate hip instability in 50% of cases after 5 days (Hadlow, 1988; Wientroub and Gill, 2000), 90% of cases in 2 months (Barlow, 1962), and 97% of cases in 6 months (Abdinejad et al., 1996). Such cases are likely to never be quantifiable from archaeological collections. However, most studies have demonstrated that if dislocation occurs within the first few months of life and is not corrected, then the loss of contact between the acetabulum and femoral head disrupts normal formation of the acetabulum as well as the shape of the femoral head (MacFarlane, 1980).

When the femoral head becomes dislocated, the femoral neck remains both anteverted and in valgus as it is at birth, instead of remodeling with advancing age to the normal anatomical alignment seen in adults. The femoral head and neck are then pulled proximally and laterally by the hip adductors while walking. The repeated motion flattens and misshapes the femoral head to appear mushroom-shaped and the muscles anchoring the hip joint (hamstring, hip adductors, and psoas) become shortened and contracted (Terjersen et al., 1996). Though the capsule of the hip joint expands, the unused and unstimulated acetabulum itself fills with fibrofatty debris (pulvinar), becomes flattened and triangular, and the acetabular labrum becomes enlarged along the superior, posterior, and inferior rim, may become inverted, and blocks reduction of the femoral head (Fig. 1). Similarly, the ligamentum teres becomes hypertrophic and redundant and the transverse acetabular ligament is pulled superiorly, blocking the lower portion of the acetabulum. In bilateral dislocation, the perineal space is widened and therefore the greater trochanter is more prominent than normal (producing the tell-tale DDH waddling gait), the lumbar spine becomes hyperlordotic, and the entire pelvis tilts anteroposteriorly with simultaneous superior elongation of the auricular surface (Roposch and Wright, 2007). Hyperlordosis may result in back pain, but objective functional studies are lacking and it is generally reported that complete bilateral dislocation causes relatively mild functional limitations since leg shortening is similar on both sides, avoiding the development of compensatory scoliosis (Ortner, 2003; Storer and Skaggs, 2006).

Though traumatic hip dislocation is extremely painful, children with DDH do not experience pain and adhere to the same developmental ambulatory schedule as their counterparts, though with a limp. It is only in later childhood and young adulthood, with the onset of secondary

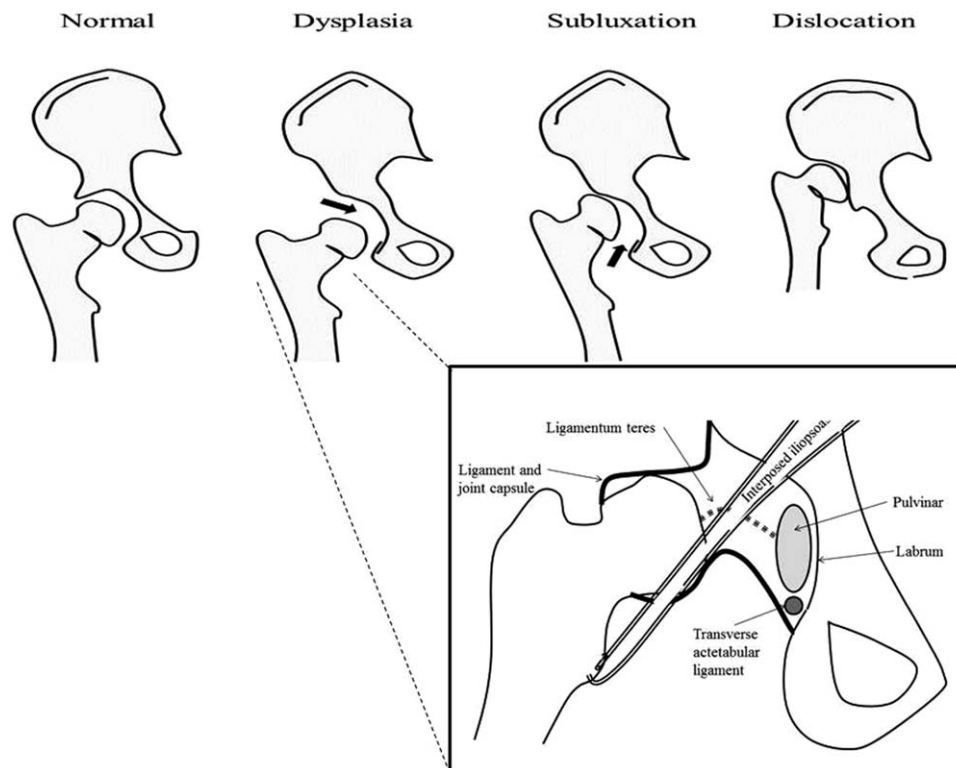


Fig. 1. Redrawn and modified schematic (from Mitchell and Redfern, 2008 and 2011) of the anatomical positioning and modification of the acetabulum, femoral head, and soft tissue of a normal, dysplastic, subluxated, and dislocated hip. Arrows highlight the abnormality.

degenerative changes of the false acetabulum, knee, and spine that pain becomes an issue. In cases of dysplasia, pain and degenerative changes are usually not reported in the first decade of life. Subluxation, similarly, has been associated with sclerosis along with the weight bearing portion of the acetabulum later in life. Early clinical signs include groin pain in both males and females, but females appear to succumb to pain in their mid-30s and suffer degenerative changes in their 40s, while males suffer pain and degeneration one to two decades later (Hartig-Andreasen et al., 2013; Jacobsen, 2007; Weinstein, 2013). In unilateral cases of dislocation, gait disturbance is more noticeable as there is inequality in leg length. In such cases, the contra-lateral unaffected hip is also often found to have acetabular dysplasia and the body adjusts to compensate for the misalignment (Jacobsen et al., 2006). While the adductor muscles of the thigh, in cases of dislocation, can still apply the medial force necessary to adduct the femur, the adductor muscles of the hip cannot initiate abduction without a healthy acetabular joint roof to promote rotation. The hip tilts to the side with the shortened leg (in time this will lead to shortened and contracted muscles), which can further result in compensatory scoliosis in order to keep the femoral head in alignment. Such patients acquire flexion-adduction deformities of the hip which may lead to valgus deformities of the knee with attenuation of the medial collateral ligament and lateral compartment degenerative joint disease (Gulati et al., 2013; Storer and Skaggs, 2006).

Genetic and endocrine factors of etiology

DDH is considered to be a polygenic autosomal dominant disorder with incomplete penetrance and penetrance dependent upon gender (Cylander et al., 2008). Homozygous recessive traits are increased in children with DDH (Cvjeticanin and Marinkovic, 2005). Several loci for DDH have been identified with candidate susceptibility genes (Larchet et al., 1994) involved in bone and joint biology. Feldman et al. (2010) mapped a DDH locus in the region of chromosome 17q21.31-17q22, but no fewer than six DDH susceptibility genes have been identified (GDF5, TBX4, ASPN, IL-6, TGF- β 1, PAPP2). This region contains a cluster of HOX genes that provide specific positional identities to mesenchymal cells in developing joints (Jiang, 2003). Despite this, no unequivocal genes with global significance have been identified. Additionally, no differences in ABO blood types (Dai et al., 2008) or Rh blood types (Gunther et al., 1993) have been found in DDH. Reported frequency of concordance is ~33% in monozygotic twins and ~8% in dizygotic twins (Smith and Aase, 1979). Nevertheless, the majority of monozygotic twins are not concordant, suggesting that an identical genetic background and intrauterine environment does not alone result in DDH.

Preterm infants demonstrate no increase in DDH even after reaching normal development and gestational age (Timmler et al., 2005). Additionally, family history for DDH has been found in 12 to 33% (among the Navajo) of affected patients (Coleman, 1968). A hormonal/endocrine

relationship has long been suspected in DDH since it occurs predominantly in females (80–85%). There has been some evidence that those with DDH have joint laxity caused by altered collagen metabolism (Carr et al., 1993; Jensen et al., 1986) and increased serum relaxin, but with conflicting results. Most studies have demonstrated an increase in the type III/I collagen ratio in the hip capsule and ligamentum teres (Skirving et al., 1984). Increased joint laxity likely explains the early onset and increase in inguinal hernias in DDH children (Uden and Lindhagen, 1988). Abnormal fetoplacental collagen and estrogen metabolism in a mother and child with DDH has been described, but again, with conflicting results (MacLennan et al., 1997). Additionally, women who develop pelvic joint instability and pain in pregnancy have an increased risk of having a child with DDH (7–9 compared with the normal 2–3) (Forst et al., 1997; Saugstad, 1991). These women also have higher serum relaxin levels compared with those without pelvic pain (MacLennan et al., 1997) in the third trimester, but there has been no correlation between serum relaxin in umbilical cord blood and neonatal hip instability (Andersson et al., 2002).

Administration of progesterone in the first trimester for pregnancies at risk of miscarriage (Katz et al., 1985) increases the risk of DDH as does first trimester amniocentesis (Cederholm, et al., 2005), maternal hypothyroidism (Ishikawa, 2008), maternal phenylketonuria (Stevenson and Huntley, 1967) and exposure to radiation. Intrauterine toxoplasmosis (Thiene et al., 1968) or viral infection (Samborska and Lembrych, 1986) does not increase the risk of DDH. Iron deficiency anemia in fetal life may be associated with DDH (Aksoy et al., 1967). Hip dislocation in general (though not DDH proper) is more common in individuals with neuromuscular disorders, such as cerebral palsy, meningomyelocele, and arthrogryposis as well as children with congenital muscular torticollis, metatarsus adductus, spina bifida occulta, and infantile scoliosis (Wynne-Davies, 1982). While neuromuscular disorders can lead to hip dislocation in children, these are distinct from DDH and should be reported in the literature separately from DDH (as they are here). Disuse osteopenia in the long bones of the lower limbs (in non-walkers), signs of flexion contractures on joints surfaces such as the knee and ankles, and congenital structural spinal abnormalities where the condition involves the spine can distinguish DDH from hip dislocation accompanying neuromuscular disorders in skeletal remains.

Mechanical factors of etiology

A number of predisposing obstetric and mechanical factors have been associated with increased risk for DDH. Lack of fetal mobility or mechanical pressure, which hinders abduction and external rotation of the femur and pressure on the greater trochanter appears to be the prime causal factor. Logistic regression analysis demonstrated that breech presentation, in utero postural deformities, oligohydramnios (deficiency in amniotic fluid), female sex, and primiparity were significant positive risk factors for DDH, while low birth weight and prematurity were protective (Helmsted and Asplend, 1983; Hinderaker et al., 1994; Lodder and Skopelja, 2011). Other less reported associations include extrauterine pregnancy, first trimester amniocentesis, paternal age, and maternal stature (Wynne-Davies et al., 1982). There is also much literature documenting the connection

between breech presentation and vertex position and DDH occurrence (Clausen and Nielson, 1988; Lodder and Skopelja, 2011). It is believed that in utero, knee extension of the infant in the breech position results in sustained hamstring forces around the hip and contributes to subsequent hip instability. Though breech delivery occurs in 3 to 4% of births, 25 to 45% of children with DDH were born breech (Hadlow, 1988). A sevenfold increase was also reported for breech babies delivered by elective Caesarean section (Lowry et al., 2005). Similarly, firstborn children are also affected in 6 of 10 cases as compared with subsequent siblings, presumably because of an unstretched uterus and tight abdominal structures in the mother, and DDH occurs less frequently in multiparous women. The left hip is also more frequently involved than the right hip because of frequent left-side fetal positioning near the end of pregnancy (Mahan and Kasser, 2008).

Other mechanical influences on DDH occurrence appear related to postnatal handling of an infant. Many pediatricians today consider swaddling, the tight wrapping of an infant in cloth or bands to restrict limb movement, to be a significant factor in the development of DDH as it maintains the hip and legs in an extended and adducted position. However, radiographs have demonstrated that swaddled infants are allowed 15 to 20° of abduction (Rabin et al., 1965). Swaddling is said to be effective because it mimics the snugness of the womb and limits the Moro reflex (Franco et al., 2005), which can wake and aggravate an infant. Many studies have demonstrated that swaddling decreases crying and increases the duration of uninterrupted sleep (Caiola, 2007). For foragers, swaddling and cradleboarding (cradleboarding involves swaddling) were easy methods of transporting infants, keeping them from harm during the daily activities of the mother, keeping them warm, and diapering them. Wang et al. (2012) used surgical tape to swaddle 112 neonatal rats, divided into a control group and experimental groups. Straight-leg swaddling was demonstrated to increase the prevalence of DDH in this animal model (though a non-bipedal model), especially if the swaddling was early (first 5 days of life) or prolonged (first 10 days of life). The severity of hip impairment varied, but both early and prolonged swaddling resulted in more dislocations, while late swaddling (second five days of life) resulted in more subluxations.

Newborn swaddling has been practiced in many cultures (Lipton et al., 1965) for centuries. Cultures in which swaddling or the use of a cradleboard is especially prevalent have higher rates of DDH. These include Saudis (Moussa and Alomran, 2007; Shaheen, 1989), Japanese, Turkish (Kutlu et al., 1992), Sámi, and Native Americans and Indigenous Peoples of Canada (Lodder and Skopelja, 2011). Infants born in colder winter months demonstrate poorer acetabular development compared with those born in the warmer months (Andren and Palmén, 1963; Siffel et al., 2005) as measured by acetabular depth and acetabular angles; this may explain the increase in DDH in children born in the winter or may represent an effect of increased swaddling or tight clothing to protect the baby from the colder weather (Walker, 1977). In cultures where infant transport involves abduction of the legs around the mother (as in Hong Kong) or where swaddling is absent, DDH is virtually unheard of (e.g. Southern Chinese, African Bantu, Thai, North Korean, Sri Lankan) (Hoaglund et al., 1981; Roper, 1976). For instance, Inuit mothers who

carried their young inside their parkas in a hood, abducting the hips around their backs, have a low incidence of DDH similar to that of Europeans (Lodder and Skopelja, 2011). Although the ethnic/genetic component of the occurrence of DDH is not clear in these cases, several studies have demonstrated dramatic decreases in DDH rates with population-wide educational programs which either decreased swaddling practices, promoted extension swaddling style, or promoted the use of diapers. Kyoto, Japan is an excellent example of this, where the incidence of DDH dropped from 52.9 in 1971 to 5.6 in 1976 after a prevention campaign to avoid prolonged swaddling (Ishida, 1997; Yamada, 1993). The results in Kyoto led to a national Japanese education and prevention campaign resulting in a more than fivefold reduction in the national rate of DDH (Ando, 1993). A similar example can be seen among the nomadic Swedish Sámi, where a DDH prevalence of 24.6–40 has fallen dramatically with both an increasingly sedentary lifestyle and forbiddance of the use of the cradleboard by missionary nurses (Gulati et al., 2013; Holk, 1991; Lehtola, 2004). The current treatment for DDH in infants involves splints or harnesses (e.g. the Pavlik harness), surgical reduction of dislocated hip should harness treatment fail, osteotomies of the pelvis if the acetabulum remains dysplastic with increasing age, and hip replacement surgery if degenerative change occurs in the hip in adulthood (Storer and Skaggs, 2006; Kosuge et al., 2013).

MATERIALS AND METHODS

A systematic review was performed for literature on DDH focusing on etiology, epidemiology, diagnosis, and case studies involving Native Americans (including the Caribbean) and Canadian Aboriginals. Databases used in this study included PubMed Medline, Web of Knowledge, Google Scholar, and various anthropology search engines and journals. Hundreds of manuscripts focused on DDH, but reports were chosen for their breadth of scope in regards to covering a range of etiological factors, as well as those that focused on the target population. The literature search was based on the criteria of covering the spectrum of DDH, but focusing more on the etiology than treatment or clinical diagnostics, which are in most cases not relevant to indigenous or archaeological contexts.

The skeletal sample examined in this study consisted of 679 individuals of all ages from the Buffalo site in Putnam County, West Virginia. During the initial period of study (i.e. Blatt, 2012), the remains were housed at The Ohio State University, Department of Anthropology. The Buffalo remains were subsequently transferred to the state of West Virginia and are currently stored at the Delf Norona Museum and Grave Creek Archaeological Complex in Moundsville in a newly constructed facility dedicated to curation and safe storage of the remains. For the current study, permission to observe and record cases of DDH from the Buffalo collection was obtained from both the museum and the State of West Virginia.

The Buffalo site spans several temporal/cultural components, but the cemetery component of the site is Fort Ancient from the Late Prehistoric period (1490 B.P. \pm 70). The Fort Ancient tradition includes the southeastern edge of Indiana, the southern third of Ohio, the central and eastern portions of Kentucky, and western West Virginia. Fort Ancient is an arbitrary regional grouping of auto-

nous villages, which can be described as transegalitarian horticulturalists, responding in kind to varied circumstances and varying degrees of Mississippian influence (Cook, 2007). Evidence of the subsistence economy at Buffalo is scanty; what is known about their diet conforms to other more thoroughly examined Fort Ancient communities, in which diet overwhelmingly consisted of maize and other domesticated subtropical plants, as well as hickory nuts and a variety of non-domesticated fauna (marine and terrestrial) and domestic dog (Blatt, unpublished data).

Formal excavations took place from 1960 to 1965 (Hanson, 1975) and the site was admitted to the National Register of Historic Places in 1971. Due to time and labor constraints, excavations at the site were primarily conducted with a bulldozer and shovel, exposing 6 m wide trenches in 45 to 60 m intervals. This broad-scale effort did not lend itself to complete and detailed recording of burials and grave goods. A mortuary analysis study by Drooker (2000) reports that during the 1964 to 1965 seasons, burials were avoided, crushed, disturbed, and displaced by the bulldozer, and feature locations were recorded inaccurately. Looting and vandalism of burials was an ongoing problem as well. Thus, burial number most likely represents a minimum. This sample was chosen for a study focused on DDH because (1) a number of DDH cases had been reported previously (Blatt, 2012), (2) the remains are well preserved and are considered representative of the Buffalo population, and (3) this size of this sample makes it the second largest assessment of DDH published to date and the largest, most comprehensive assessment of DDH from the prehistoric New World.

All remains were analyzed according to *Standards for Data Collection from Human Skeletal Remains* (Buikstra and Ubelaker, 1994). Sex was assigned to each individual from pelvic and (Phenice, 1969) cranial morphology (Krogman and Iscan, 1986). Age was assigned to each individual from auricular surface morphology (Lovejoy et al., 1985) and when possible combined with pubic symphysis morphology (Meindl et al., 1985). The prevalence of DDH out of 1,000 was calculated from the Buffalo site, other archaeological populations, and modern clinical cases involving Indigenous North Americans. Only cases in which the number of total cases examined was reported (rather than nonpopulation-based case studies) were included in prevalence calculation, though they are listed in Tables 3 and 4 as reference. Individuals with neuromuscular disorders and immature individuals where DDH was not clearly observable from skeletal elements were excluded from this study.

Cases of identified DDH from Buffalo were seriated in order to identify a pattern of criteria and characteristics associated with the increasing severity of DDH stages to segregate criteria of dysplasia from subluxation from dislocation within this population. In accordance with the guidelines and classifications presented by Mitchell and Redfern (2008, 2011) the entire pelvis and sacrum of each individual with DDH was examined, as were the vertebrae, femur, and upper extremities in order to gauge any full-body patterns of skeletal modification in conjunction with DDH. Individuals identified as having DDH were also examined for artificial cranial deformation, including cranial depressions produced by binding. The diagnostic criterion for DDH (based upon the works of Mafart et al., 2007; Mitchell and Redfern, 2008, 2011) were shallow, irregular, or obliterated true acetabulum that were not

TABLE 1. Summary of paleopathological changes observed in DDH cases from the Buffalo sample (based upon the diagnostic classification of Mitchell and Redfern, 2008, 2011)

Structure	Dysplasia	Subluxation	Dislocation
True acetabulum	Comparatively shallow and broad with slight superior and lateral osteophytic rim. Smooth acetabular floor and roof.	Oval-shaped. Shallow and broad with moderate superior and lateral osteophytic rims (indicating labral tears). Degenerative changes and porosity to the superolateral rim.	Triangular with irregular and porous floor. Cysts along margin of true acetabulum, which may indicate labral tearing.
False acetabulum	None.	None.	Type 1-narrow or broad, shallow depression with or without patchy bone layer on the posterior and superolateral ilium. Eburnation may be present. Type 3-raised "bony plaque" on the posteolateral ilium. Eburnation occasionally present on segments of the plaque.
Other regions of Ossa Coxa	None.	None.	Wider greater sciatic notch, more triangular obturator foramen, shorter and broader ilium. Posterior tilting of ilium with anteroinferior alteration of sacroiliac joint.
Femoral head	Normal or minimal subchondral cysts.	Medial degenerative change or mushroom-shaped. May show eburnation.	Mushroom-shaped and flattened. May show eburnation and porosity.
Femoral neck	Normal to minimal anteversion.	Intermediate anteversion.	Short with thin circumference. Valgus, varus, or normal in anteroposterior plane.
Femoral trochanters	None.	May show porosity.	Lesser trochanter may show subtle lateralization. Greater trochanter is comparatively smaller than normal and may be mushroom-shaped and/or elongated. May be normal.
Femoral shaft	None.	The leg on the side with the abnormal hip may be shorter than the contralateral leg.	Smaller circumference and may be shorter in length.
Vertebrae	None.	None.	May have asymmetric inferior facets, and transverse processes. Osteophytes on bodies of the opposite side of affected hip. Sacrum has lipping on the affected side. May show evidence of fusion and spina bifida.
Shoulder	None.	None.	May show degeneration and lipping at glenoid fossa of scapula and humeral head on affected sides.

capable of articulation with the femoral head, bony changes (see Table 1 for stages) on the lateral aspect of the ilium suggestive of the formation of a false acetabulum for the displaced femoral head. As described in Table 2, in conjunction with the pelvic abnormalities of DDH, cases were diagnosed by the presence of supplementary postcranial abnormalities such as mushroom-shaped femoral head, short femoral neck, reduced femoral circumference, and asymmetrical vertebral lipping.

RESULTS

Of the 679 individuals in the skeletal series, 390 adults were suitable for observation of DDH due to reasonable preservation of at least one hemi-pelvis. Eighteen of the 390 individuals were identified as having some form of DDH, giving an overall minimum prevalence of 46.15 per 1,000 of the entire spectrum of the disorder in this population. A minimum prevalence is given due the absence and/or fragmentary nature of both halves of the pelvis for some individuals. Four other cases of hip dislocation were observed in individuals who also had spina bifida and therefore they were not included in prevalence calculations.

Three individuals had unilateral dislocation; six displayed bilateral dislocation; five were identified as unilateral subluxation; one bilateral subluxation; and three

TABLE 2. Buffalo individuals with DDH

Indiv.	Sex	Age	DDH type	Cranium
C7-2	F	—	Bilateral dislocation	D ^a
C10-16	M	20–30	Dislocation (left)	D
C10-17	M	40–50	Subluxation (left)	D
D10-13	F	30–40	Subluxation (left)	—
D11-4	F	—	Bilateral dislocation (left)	D
D11-20	F	30–40	Bilateral subluxation	D
D11-30	M	40–50	Subluxation (right)	D
E8-24	F	20–30	Dislocation (right)	D
E10-14	M	18–25	Bilateral dislocation	ND ^b
E10-74	F	20–30	Subluxation (left)	D
E10-82	F	20–30	Bilateral dislocation	D
E11-10	F	30–40	Bilateral dislocation	D
E11-10A	F	30–40	Bilateral dislocation	D
E11-13	M	18–25	Dysplasia (left)	D
E11-15	F	40–50	Dislocation (left)	—
E11-23	F	45–55	Subluxation (left)	D
E11-37	F	20–30	Subluxation (left)	D
E11-39	F	—	Dysplasia (right)	D

^aDeformed crania.
^bNot deformed crania.

appeared to have unilateral dysplasia. Consequently, the frequency of dysplasia, subluxation, and dislocation within this population was 0.8%, 1.5%, and 2.3%, respectively. Of the unilateral cases in this study, seven

TABLE 3. DDH prevalence per 1,000 among Indigenous North Americans

Authors	Location	Culture/period	Sample	DDH	Prev.
Corrigan and Segal, 1950	Island Lake, Manitoba	Cree-Ojibwa	1,253	107	35.9
Kraus and Schwartzmann, 1957	Fort Apache, AZ	Apache	3,500	31	30.6
Rabin, 1965	Many Farms District, AZ	Navajo	818	59	37.9
Houston and Buhr, 1966; Houston, 1988	Northern Saskatchewan	Cree	4,453	250	13.2
Salter, 1968	Ontario	Navajo	2,032 ^a	17	123
			1347 ^b	77	12.6
Coleman, 1968	Fort Defiance, Ship Rock, Gallup, NM	Navajo	155	420	66.7
Walker, 1977	Island Lake, Manitoba	Cree-Ojibwa	1,248	18	336.5
Pratt, 1982	Many Farms District, AZ	Navajo	548 ^c	89	32.8
			270 ^d		330
Clinical prevalence			15,624	1,113	71.2
Hooton, 1920 ^e	Madisonville, OH	Fort Ancient	—	1	—
Hooton, 1930 ^e	Pecos Pueblo	Pre-contact	—	1	—
Wakefield, 1937	Eastern Arkansas	Pre-contact	100	1	10
Snow, 1948 ^e	Indian Knoll, KY	Late Archaic	—	2	—
Goldstein, 1957	Texas	AD 800–1700	146	0	0
Morse, 1963 ^e	Stueben Village and Mounds, IL	Hopewell	—	1	—
McPherron, 1967 ^e	Juntunen, MI	Late Woodland	—	1	—
Morse, 1969 ^e	Morse, IL	Pre-contact	—	1	—
Miles, 1975	Mesa Verde, CO	AD 750–1300	179	0	0
Clabeaux, 1977	Orchard, Ontario; Trenton, NJ	Pre-contact	286	2	7
Gregg, 1981	Crow Creek, SD	14th C.	486	2	1
Pfeiffer, 1984	Uxbridge, Ontario	1490	312	1	3.2
Loveland, 1985	Red River County, TX	AD 1100–1800	75	1	—
Merbs and Vestergaard, 1985	Sundown, Prescott, AZ	AD 1100–1200	26	0	0
Wheeler, 1985	El Morro Valley, NM	13th C.	26	0	0
Drusini, 1987	Maguana, Santo Domingo	Late 15th C.	108	5	46.3
Lahr and Bowan, 1992	Kechipawn, NM	1300–1600	54	0	0
Orton, 2003 ^e	Kwasteryerkiva (NMNH 271828)	Pueblo	—	1	—
Orton, 2003 ^e	Proctorville, OH (NMNH 368989)	Fort Ancient	—	1	—
Orton, 2003 ^e	Jones Point, AL (NMNH 372897)	Pre-contact	—	1	—
Present Study	Buffalo, WV	Fort Ancient	390	18	43.6
Archaeol. prevalence			2,198	40	17.6
Total weighted avg.			17,822	1,153	64.7

^aUsed cradleboard.^bDid not use cradleboard.^cChildren.^dAdults.^ePrevalence not given or included in average because total sample number is unknown.TABLE 4. DDH prevalence comparison^a

Population	Clinical prev.	Archaeol. prev.
Western Europe	3.6	4.9
Eastern Europe	35.8	6
Middle East	10.6	6.2
Africa	0.06	—

^aBased on the accounts of Lodder and Skopelja (2011).

presented on the left side and three on the right. Of the 18 individuals, 12 of the cases are identified in females, giving a female: male ratio of 2.4:1.

Two of the types in Table 1 of false acetabula described by Mitchell and Redfern (2008) were identified from the Buffalo sample (Figs. 2–4). Type 1 as a narrow, shallow depression superolateral to the true acetabulum with or without eburnation and Type 3 as a raised bony plaque with or without evidence of eburnation. When placing the pelvis and sacrum in anatomical position, it appears to lean posteriorly so that the true acetabula face anteriorly and the iliac crest falls several centimeters posterior to the sacrum, which in turn elongates the auricular surface. Dislocation was also associated with a host of pathological features found at other joints (Figs. 5 and 6). For instance, femoral heads of affected sides were always mushroom-shaped or flattened and the shaft and neck circumference

and length and trochanter size of the affected femur were reduced. Additionally, several individuals showed signs of periostitis of the lesser trochanter as result of abnormal pull on the iliopsoas muscle. Several individuals displayed asymmetrical inferior processes of the lower thoracic and lumbar vertebrae and exaggerated lipping on the opposite affected side and sacral lipping on the affected side. In one individual, the glenoid fossa of the scapula showed significant degeneration on the affected side, possibly suggesting over compensation of upper arm strength. Additionally, 15 of the 18 individuals had cranial deformation (Table 2), one individual did not have cranial deformation, and the cranial preservation of two individuals was too fragmentary to determine presence or absence of deformation.

All the clinically observed cases of DDH among indigenous peoples of North America have been reported from communities that have traditionally and continually swaddled their infants in one manner or another. These cases are presented in Table 3. Based on ethnographic data from northern Saskatchewan, multiple methods of swaddling were probably used by prehistoric cultures. In northern Saskatchewan, the Cree had no less than four methods of swaddling, all of which bind the legs tightly together (i.e. cradleboard, mossbag, hammock, and tight wrapping in blankets) (Houston, 1988). Among Fort Apache inhabitants in Arizona, incidence was 31. Among



Fig. 2. Example of a raised bony plaque false acetabulum (Type 3 from criterion of Mitchell and Redfern, 2008, 2011) and triangular, deformed true acetabulum. This individual had bilateral dislocation of the hip. Individual E10-14.

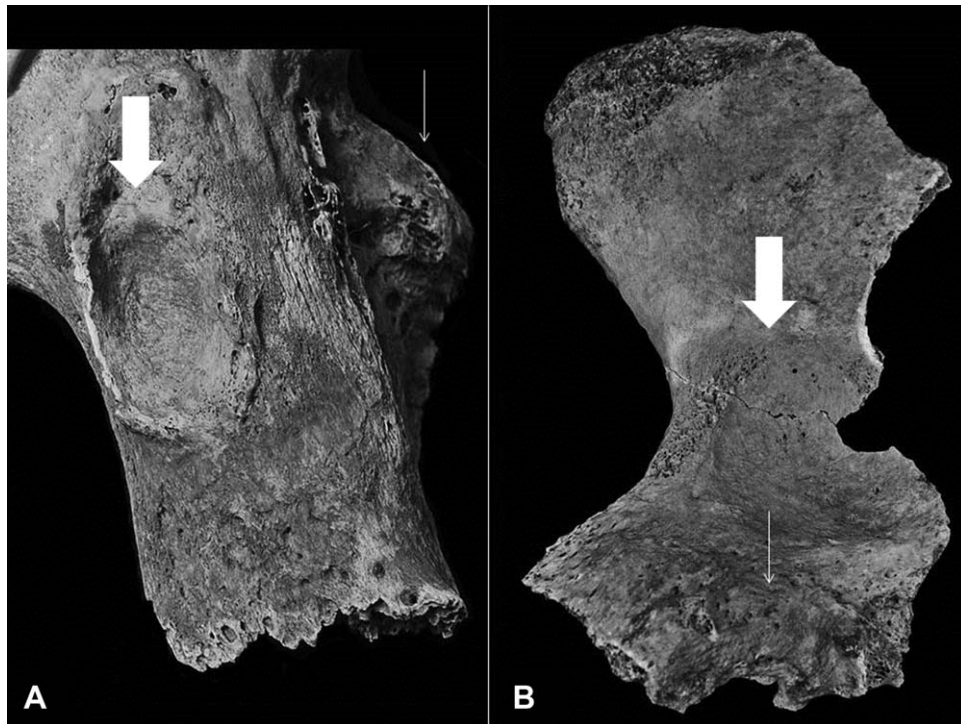


Fig. 3. Examples of Type 1 false (based on the criterion of Mitchell and Redfern, 2008, 2011) acetabulum from Buffalo. A) small, oval depression with active porosity, lipping, and centrally located eburnation. Note the degenerated and triangular true acetabulum to the right with signs of labral tearing. From individual E11-10. B) Large, rounded depression with porosity set superior-posteriorly to the true acetabulum. From individual E8-B24. Narrow arrows indicate true acetabula and broad arrows indicate false acetabula.



Fig. 4. Example of the malformation and degeneration of true acetabulum from Buffalo. A) Shallow acetabulum suggestive of acetabular dysplasia. From individual E11-23. B) Fragmented (postmortem) triangular acetabulum consistent with complete dislocation. Abnormal shape and cross-hatching appearance of the lunata surface, reminiscent of ossification of connective tissue bundles. From individual E10-82.



Fig. 5. Mushroom-shaped femoral heads. This individual had bilateral dislocation of the hip. From individual D11-4.

Navajo children from Fort Defiance and Gallup, it was 67 (Coleman, 1968; Kraus and Schwartzmann, 1957). Navajo from the Many Farms District had a prevalence of 40.1 in children and 33.3 in adults with dysplasia to dislocation ratios of 4.5:1 and 0.3:1.10 respectively (Pratt et al., 1992). In another Navajo sample (Rabin et al., 1965), the fact that dislocation was present in more adults than children was believed to be a consequence of the recent transition from exclusive use of the cradleboard to that of diapers. The results were not completely straightforward, as not all cases with dislocation were cradleboarded (two cases) and not all cradleboarded infants had DDH (a few even showed spontaneous improvement of DDH while on the cradleboard). Among Aboriginal groups in Ontario (Salter, 1968) and Quebec (Ghibely, 1990) there is a 10-fold increase in DDH (123 vs. 12.6) among cradleboard users and the Cree-Ojibwa of Northern Saskatchewan and Manitoba, who have a near universal practice of cra-

dleboard swaddling, have a DDH prevalence of 336 and 13.2, respectively for all ages (Houston and Burh, 1988). Among the Cree-Ojibwa, however, 113 of the 427 untreated cases of DDH, including 21 cases of complete dislocation, improved within the first year of life (Walker, 1977), which appears to support clinical findings of the commonness of spontaneous correction after birth. Using weighted averages of the reports from clinical cases, the prevalence of DDH among all cases of living Native American and Aboriginal Canadian populations is 71.2. Further, a positive family history increases the risk of DDH (Coleman, 1968; Dogruel et al., 2008; Heikkila, 1984; Houston and Buhr, 1988, Mamouri et al., 2004; Kremli et al., 2003; Mirdad, 2002; Romero et al., 1989; Stein-Zamir et al., 2008; Wynne-Davies, 1970) and among indigenous groups, it was 33% in the Navajo (Coleman, 1968; Kraus and Schwartzmann, 1957), in northern Saskatchewan Cree it was 16% in sisters and 14% in mothers (Houston and Buhr, 1988), but there was no correlation in the Manitoba Cree-Ojibwa.

If sampling of archeological data was adequate, then statistical analysis should reveal if differences between prehistoric and modern prevalence were real. The data (DDH n /sample n) were subjected to a χ^2 test to analyze temporal differences (archeological vs. living) (Table 3). With the incomplete nature of sampling archeological materials in mind, prevalence of DDH among indigenous North Americans is significantly different ($\chi^2 = 81.78$; $P < 0.00$) in prehistoric (40 of 2,198) and living (1,113 of 15,624) populations. However, it is likely that other archaeological studies overlooked or underreported DDH. Therefore, as the largest and only systematic archaeological case study of DDH among Native Americans or Canadian Aboriginals, the prevalence of DDH at Buffalo (46.15) could be more realistic. If this is the case, there could be no significant temporal differences in prevalence ($\chi^2 = 3.24$; $P < 0.07$).

DISCUSSION

Seriation by severity of DDH resulted in clear distinction among the three stages of the disorder. As a developmental anomaly, criteria overlap with development from one stage of severity to another, but the common characteristics of each stage observed in this sample are listed in Table 1 (based on Mitchell and Redfern, 2008, 2011). Cases of dysplasia and subluxation were identified from comparatively shallow and wide true acetabula. While both forms had labral lipping and superolateral porosity, subluxed hips also had evidence of labral tearing as well as moderate alterations to the shape of the femoral head. As predicted, cases of dislocation were immediately obvious due to the formation of a false acetabulum and degeneration or obliteration of the true acetabulum with or without ossified ligament damage.

Causal factors due to intrauterine environment may have been unimportant among indigenous North Americans as compared with other populations. For instance, among the Saskatchewan Cree, the unassisted birth of large infants is frequent (which presumably means the infant was crowded), but breech-positioned births are extremely infrequent (Coodin et al., 1975). In cases of a breech birth, unlike other populations, among Native Americans and Canadian Aboriginals there is no

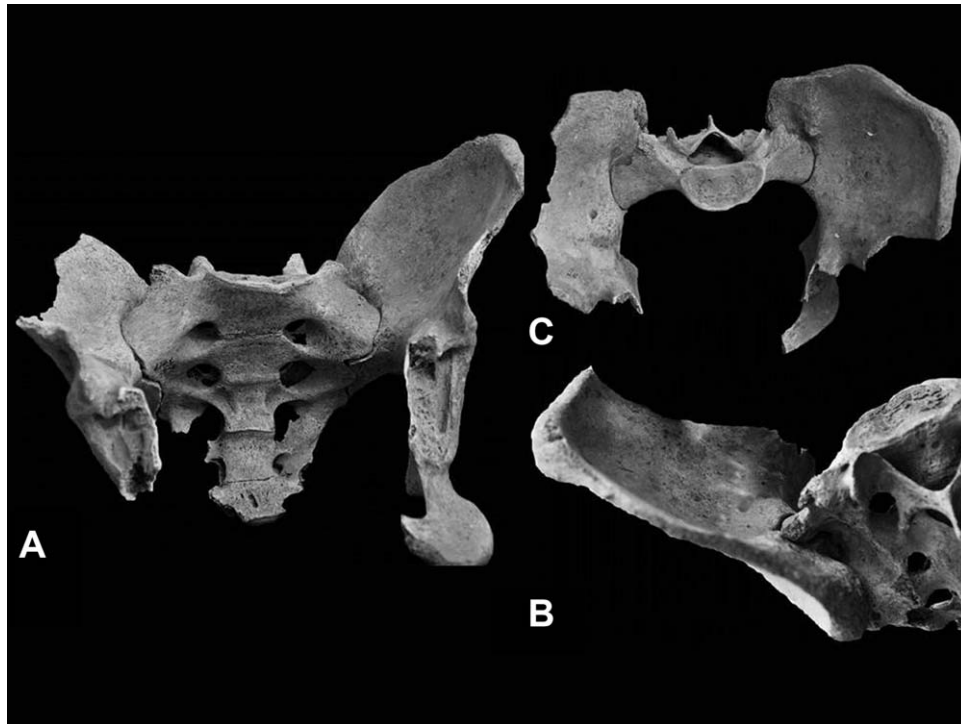


Fig. 6. Full pelvic view of the modification of the position of the ilium in relation to the sacrum and the anterior position of the true acetabula (A). Note the posterior placement of the iliac crest and anterior-superior tilt of the ilium (B, C). From individual E10-82.

correlation between breech presentation and DDH incidence (Bower et al., 1987; Walker, 1977). It is possible that selection for easier births of large infants and women with lax pelvic ligaments had a trade-off of increased frequency of DDH. While remaining primarily closed systems in regards to gene flow, temporal change among these indigenous groups has been found in specific cases where modern diapers replaced the use of traditional swaddling methods. In such cases, DDH cases have declined from earlier records. Therefore, while the indigenous populations of North America may have a genetic predisposition to DDH, and swaddling practices alone may not cause the hip dysplasia, such practices probably have an unfavorable effect on the future course of a dysplastic hip.

The increase in frequency with increase in severity is likely due to difficulty of identifying milder forms of DDH and the correction of mild forms before adulthood, rather than the absolute higher frequency of dislocation. Furthermore, as a developmental disorder, most cases of dysplasia and subluxation would likely be found in infants and young children, which in the case of skeletal remains has the added difficulty of identifying criteria of mild forms of DDH in immature and unfused skeletal elements. Dislocation and subluxation are not clinically associated with reactions in the skeleton needed by paleopathologists to identify the disorder in skeletonized remains. These less severe stages on the DDH spectrum have only been clinically identified in infants with the aid of radiography, ultrasonography, or physical examination using the Galeazzi sign and the Ortolani or Barlow maneuvers (Storer and Skaggs, 2006) which assesses the

relationship of the femoral head to the acetabulum within its soft tissue context. Therefore, determining prevalence and range of DDH in a skeletal series is presented with multiple challenges, which may have both resulted in the infrequent reporting of DDH archaeologically and also influenced reported differences in the epidemiology of DDH in modern versus prehistoric and historic populations.

Many archaeological reports of DDH are single case studies without reference to overall population size and most likely overlook all but severe and obvious cases of dislocation. Therefore, though these reports used the criteria of mushroom-shaped femoral head, triangular acetabulum, and false acetabulum, the archaeological prevalence of DDH in North America has most likely been under-reported. Nevertheless, in those studies reporting DDH where the population data are known (see Table 3), 74% were female of whom 35% demonstrated bilateral involvement, similar to the figures of 76% female and 27% bilateral in this review. As indicated in Table 3, including the current sample, the total archaeological prevalence of DDH among the prehistoric indigenous peoples of North America was 17.6 and the weighted mean prevalence of clinical and archaeological cases was 64.6 of 1,000. As mentioned, these figures must be taken with caution given the changes in criteria for identifying DDH in the skeleton and the fragmentary nature of the archaeological record. As such, a clinical understanding of the prevalence of DDH among the target population are confounded.

Delays in the onset of walking have not been correlated to the restraint from being cradleboarded/swaddled

(Kamath and Bennet, 2004), but societal considerations later in life influence the perceived functional ability in cases of DDH. In a study of 420 cases of DDH in the Cree–Ojibwa Island Lake Community population of Manitoba, Walker (1977: 503) reported that, “functional disability in everyday life was neither admitted nor observed . . . People viewed DDH in the same way as urban societies view left-handedness. . . The sole disability admitted by an Island Lake individual (unaffected) was that it was difficult to portage a canoe with a man who limps.”

The only reasonable means by which to infer the practice of cradleboarding and swaddling in a prehistoric community from skeletal remains is through ethnohistoric comparison and evidence of artificial cranial deformation. Cranial deformation is performed on infants while the cranial bones are unfused. Ethnographic accounts conclude that the process requires hours of daily manipulative pressure, in the form of head wrappings, bands, and/or boards, which in turn requires swaddling constraints (Blatt, 2010). Despite the fact that nearly all individuals identified as exhibiting DDH had cranial deformation, many more individuals within the entire series had deformed crania without DDH. This suggests that the swaddling and restraint practices involved in creating cranial deformation cannot be said to always result in hip adduction or extension severe enough to lead to successive stages (therefore more easily identifiable in the skeleton) of DDH in adults. This is compounded by the fact that, as mentioned above, early stages of DDH have been shown to frequently self-correct in infancy or childhood.

CONCLUSIONS

Past reports of DDH prevalence among prehistoric Native Americans and Canadian Aboriginals may be underestimated by lack of focus on the pathology and identification of all its criteria, as well as small sample sizes. Based on the holistic evidence provided here, it is possible to begin discussing risk factors of DDH among prehistoric populations of modern descendant at-risk groups. Paleoepidemiology of disease in antiquity can further provide insight into possible relationships between modern behavior changes and disease frequency. Despite the incomplete nature of the archaeological record, native North Americans have the highest reported global prevalence of DDH now and in prehistory (Table 4). The etiology of DDH suggests that components of both genetic predisposition and swaddling practices have combined to create a high-risk environment for the development of DDH, contributing to its high prevalence within archaeological populations, like Buffalo, and modern indigenous peoples of North America. This suggests that either modern healthcare or obstetric practices among North American indigenous people does not directly impact DDH incidence (e.g. it is more genetically influenced) or that modern innovations have not served to influence factors related to incidence in these communities in particular. Swaddling is still highly popular in North America and Europe today and it is important to note that when done properly, most modern swaddling methods and specialized swaddling clothes, which allow infants to move and bend their legs, are not associated with health risks. That being said, in 2013 several states and municipalities legally banned swaddling practices by U.S. day care workers, citing numerous harmful effects of the practice, despite

opposition from the American Academy of Pediatrics (Karp, 2008; Pearson, 2013). So, it is clear that the cultural and biological views of swaddling in regards to infant health merit further study.

It is possible, with future advances in molecular paleopathology and more specific designation of the chromosomes and genes involved in DDH, that the etiology of the underlying risk of the disease in indigenous peoples of North America will become clearer and the processes at play be revealed more distinctly. Nevertheless, this study attests to the importance of investigating various lines of evidence, both pathological and ethnohistoric, to understand health from a paleoepidemiological and biocultural perspective.

ACKNOWLEDGMENTS

The author is grateful to Heather Cline and Charles Morris of the Delf Norona Museum and Grave Creek Archaeological Complex in Moundsville, West Virginia, who worked tirelessly and enthusiastically to grant the author, the permission to examine the Buffalo collection and make his stay comfortable. Appreciation is also extended to Paul Sculli, Gabriela Jakubowska, Ana Casado, and most sincerely to the Editor-in-Chief and the anonymous reviewers for their suggestions and aid. This work is dedicated to Penelope.

LITERATURE CITED

- Abdinejad F, Takapouy J, Eskandari N. 1996. Incidence of congenital dislocation of the hip in Shiraz. *Med J Islamic Repub Iran* 9:275–280.
- Aksoy M, Erdem S, Dincol K. 1967. Iron deficiency anemia as a possible contributory factor in the development of dislocation of the hip. *Blut* 15: 153–156.
- Andersson JE, Vogel I, Uldbjerg N. 2002. Serum 17 β -estradiol in newborn and neonatal hip instability. *J Ped Orthop* 22: 88–91.
- Ando M. 1993. Prevention of congenital dislocation of the hip in infants: experience and results in Japan. Asahikawa, Japan: Yamada Company.
- Andrén L, Palmén K. 1963. Seasonal variation of birth dates of infants with congenital dislocation of the hip. *Acta Orthop Scand* 33:127–131.
- Aufderheide AC, Rodriguez-Martin C. 1997. Cambridge encyclopedia of human paleopathology. Cambridge: Cambridge University Press.
- Barlow TG. 1962. Early diagnosis and treatment of congenital dislocation of the hip. *J Bone Joint Surg* 44:292–301.
- Blatt SH, Sculli PW. 2010. Deformed or not deformed, that is the question: quantifying artificial cranial deformation. *Am J Phys Anthropol* 141:69.
- Blatt, SH. 2012. Epidemiology of developmental dysplasia of the hip in Amerindians: cases from the Late Prehistoric Buffalo Site, West Virginia. *Am J Phys Anthropol* 147:101.
- Blondiaux J, Millot F. 1991. Dislocation of the hip: discussion of eleven cases from mediaeval France. *Int J Osteoarch* 1: 203–207.
- Bower C, Stanley FJ, Krickler A. 1987. Congenital dislocation of the hip in Western Australia. *Clin Orthop* 224:37–44.
- Buikstra JE, Ubelaker DH, editors. 1994. Standards for data collection from human skeletal remains. In: Proceedings of a seminar at The Field Museum of Natural History organized by Jonathan Haas. Arkansas: Arkansas Archaeological Survey Research Series.
- Caiola E. 2007. Swaddling young infants can decrease crying time. *J Pediatr* 150:320–321.
- Carr AJ, Jefferson RJ, Benson MKDA. 1993. Joint laxity and hip rotation in normal children and in those with congenital dislocation of the hip. *J Bone Joint Surg* 75:76–78.
- Cederholm M, Haglund B, Axelsson O. 2005. Infant morbidity following amniocentesis and chorionic villus sampling for prenatal karyotyping. *BJOG Int J Ob Gyn* 112:394–402.
- Ceylaner G, Ceylaner S, Ustunkan F, Inan M. 2008. Autosomal dominant inheritance of congenital dislocation of the hip in 16 members of a family. *Acta Orthop Traumat Turcica* 42:289–291.
- Chan A, McCaul KA, Cundy PJ, Haan EA, Byron-Scott R. 1997. Perinatal risk factors for developmental dysplasia of the hip. *Arch Dis Child* 76: F94–F100.
- Chan A, Cundy PJ, Foster BK, Earne RJ, Scott RB. 1999. Late diagnosis of congenital dislocation of the hip and presence of a screening

- programe: South Australian population-based study. *Lancet* 354:1514–1517.
- Clabeaux MS. 1977. Congenital dislocation of the hip in the prehistoric Northeast. *Bull NY Acad Med* 53:338–346.
- Clausen H, Nielsen, KT. 1988. Breech position, delivery route and congenital hip dislocation. *Acta Obst Gyn Scand* 67: 595–597.
- Coleman SS. 1968. Congenital dysplasia of the hip in the Navajo infant. *Clin Orthop Relat Res* 56:179–193.
- Coodin FJ, Dilling LI, Haworth JC. 1975. Birthweights of Manitoba Indians. *Ann R Coll Phys Surg Can* 8:69.
- Cook R. 2007. *SunWatch: Fort Ancient Development in the Mississippian World*. Tuscaloosa: University of Alabama Press.
- Corrigan C, Segal S. 1950. The incidence of congenital dislocation of the hip at Island Lake, Manitoba. *Can Med Assoc J* 62:535–540.
- Cvjeticanin S, Marinkovic D. 2005. Genetic variability in the group of patients with congenital hip dislocation. *Genetika* 41:1142–1146.
- Dai J, Shi D, Zhu P, Qin J, Ni H, Xu Y, Yao C, Zhu L, Zhu H, Zhao B, Wei J, Liu B, Ikegawa S, Jiang Q, Ding Y. 2008. Association of a single nucleotide polymorphism in growth differentiate factor 5 with congenital dysplasia of the hip: a case control study. *Arth Res Ther* 10:R126.
- Dezateux C, Rosendahl K. 2007. Developmental dysplasia of the hip. *Lancet* 369:1541–1552.
- Doğruel H, Atalar H, Yavuz OY, Sayli U. 2008. Clinical examination versus ultrasonography in detecting developmental dysplasia of the hip. *Int Orthop* 32:415–419.
- Drucker, P. 2000. *Eastern Fort Ancient Mortuary Patterns: Preliminary Results from Buffalo, WV*. Philadelphia: SAA Poster.
- Drusini, A, Businaro F, Calderón, FL. 1987. Skeletal biology of the Taino: a preliminary report. *Int J Anthropol* 2:247–253.
- Dunn PM. 1976. The anatomy and pathology of developmental dysplasia. *Clin Orthop Rel Res* 119:23–27.
- Feldman G, Dalsey C, Fertala K. 2010. The otto aufranc award: identification of a 4Mb region on chromosome 17q21 linked to developmental dysplasia of the hip in one 18-member, multigeneration family. *Clin Orthop Rel Res* 468:337–344.
- Forst J, Forst C, Forst R, Heller K-D. 1997. Pathogenetic relevance of the pregnancy hormone relaxin to inborn hip instability. *Arch Orthop Trauma Surg* 116:209–212.
- Franco P, Seret N, Van Hees JN, Scaillet S, Groswasser J, Kahn A. 2005. Influence of swaddling on sleep and arousal characteristics of healthy infants. *Pediatrics* 115:1307–1311.
- Ghibely A. 1990. Congenital dislocation of the hip in the Cree Indian population of Quebec, Canada. *Acta Orthop Belgica* 56:37–42.
- Godley DR. 2013. Assessment, diagnosis, and treatment of developmental dysplasia of the hip. *J Am Acad Phys Ass* 26:54–58.
- Goldstein MS. 1957. Skeletal pathology of early Indians in Texas. *Am J Phys Anthropol* 15:299–310.
- Gregg JB, Zimmerman LJ, Steele JP, Ferwerda H, Gregg PS. 1981. Antemortem osteopathology at Crow Creek. *Plains Anthropol* 26:287–300.
- Gulati V, Eseonu K, Sayani J, Ismail N, Uzoigwe C, Choudhury MZ, Gulati P, Aquil A, Tibrewal S. 2013. Developmental dysplasia of the hip I the newborn: a systematic review. *World J Orthop* 4:32–41.
- Gunther A, Smith SJ, Maynard PV, Beaver MW, Chilvers CED. 1993. A case-control study of congenital hip dislocation. *Public Health* 107:9–18.
- Hadlow V. 1988. Neonatal screening for congenital dislocation of the hip. A prospective 21-year survey. *J Bone Joint Surg* 70:740–743.
- Hanson L. 1975. The Buffalo site: a late 17th century Indian Village site in Putnam County, West Virginia, report of Archeological Investigations, no. 5. Morgantown: West Virginia Geological & Economic Survey.
- Harcke HT. 1999. Developmental dysplasia of the hip: a spectrum of abnormality. *Pediatrics* 103:152.
- Hartig-Andreasen C, Soballe K, Troelson A. 2013. The role of the acetabular labrum in hip dysplasia. *Acta Orthopaedica* 84: 60–64.
- Heikkilä E. 1984. Congenital dislocation of the hip in Finland. An epidemiologic analysis of 1035 cases. *Acta Orthop Scand* 55:125–129.
- Helmsted A, Asplend S. 1983. Developmental dysplasia joint: a biomechanical study in autopsy specimens. *J Ped Orthop* 3:491–497.
- Hinderaker T, Daltveit AK, Ingens LM, Udén A, Reikerås O. 1994. The impact of intra-uterine factors on neonatal hip instability. *Acta Orthop Scand* 65:239–242.
- Hoaglund FT, Kalamichi A, Poon R, Chow S, Yau A. 1981. Congenital hip dislocation and dysplasia in Southern Chinese. *Int Orthop* 4:243–246.
- Holck P. 1991. The occurrence of hip joint dislocation in early Lappic populations of Norway. *Int J Osteoarch* 1:199–202.
- Hooton EA. 1920. Indian Village Site and Cemetery near Madisonville, Ohio. In: *Papers of the Peabody Museum of American Archaeology and Ethnology*, Harvard University 8(1):1–134.
- Hooton, EA. 1930. The Indians of Pecos Pueblo: a study of their skeletal remains. New Haven: Yale University Press, p 530.
- Houston CS, Weiler RL, McKay RW. 1979. Native Children's lung. *J Can Ass Rad* 30:218–222.
- Houston CS, Buhr RH. 1988. Swaddling of Indian infants in northern Saskatchewan. *Musk-ox* 36:5–14.
- Ishida K. 1977. Prevention of the development of the typical dislocation of the hip. *Clin Orthop Relat Res* 126:167–169.
- Ishikawa N. 2008. The relationship between neonatal developmental dysplasia of the hip and maternal hypothyroidism. *J Pediatr Orthop* 28: 432–434.
- Jacobsen S, Rømer L, Soballe K. 2006. The other hip in unilateral hip dysplasia. *Clin Orthop* 446:239–246.
- Jacobsen S. 2007. Adult hip dysplasia and osteoarthritis. *Acta Orthop Suppl* 77:2–37.
- Jensen BA, Relmann I, Fredensborg N. 1986. Collagen type III predominance in newborns with congenital dislocation of the hip. *Acta Orthop Scand* 57:362–365.
- Jiang J, Ma H, Lu Y, Wang Y, Wang Y, Li Q, Ji S. 2003. Transmission disequilibrium test for congenital dislocation of the hip and HOXB9 gene or COL1A1 gene. *Chin J Med Genet* 20:193–195.
- Kamath SU, Bennet GC. 2004. Does developmental dysplasia of the hip cause a delay in walking? *J Pediatr Orthop* 24:26.
- Karp HN. 2008. Safe swaddling and healthy hips: don't toss the baby out with the bathwater. *Pediatrics* 121:1075–1076.
- Katz Z, Lancet M, Skornik J, Chemke J, Mogilner BM, Klinberg M. 1985. Teratogenicity of progestogens given during the first trimester of pregnancy. *Obstet Gynecol* 65:775–780.
- Kelsey JL. 1977. The epidemiology of diseases of the hip: a review of the literature. *Int J Epidemiol* 6:269–280.
- Kosuge D, Yamada N, Azegami S, Achan P, Ramachandran. 2013. Management of developmental dysplasia of the hip in young adults. *Bone Joint J* 95:732–737.
- Kraus BS, Schwartzman JR. 1957. Congenital dislocation of the hip among the Fort Apache Indians. *J Bone Joint Surg* 39:448–449.
- Kremlí MK, Aishahid AH, Khoshhal KI, Zamzam MM. 2003. The pattern of developmental dysplasia of the hip. *Saudi Med J* 24:1118–1120.
- Krogman WM, Iscan MY. 1986. *Human skeleton in forensic medicine*, 2nd ed. Springfield, IL: Charles C. Thomas.
- Kutlu A, Memik R, Mutlu M, Kutlu R, Arslan A. 1992. Congenital dislocation of the hip and its relation to swaddling used in Turkey. *J Pediatr Orthop* 12:598–602.
- Lahr, MM, Bowan, JE. 1992. Paleopathology of the Kechipawan site: health and disease in a south-western pueblo. *J Archeol Sci* 19:639–654.
- Larchet M, Bourgeois JM, Billon P, Chillard C, Simon J, Aldebert B, Amram D, Touati R, Vely P, Chevalier L, Hartmann JM, Bonnet P, Mares P, Lesbros D. 1994. Évaluation comparée du dépistage clinique et échographique de la luxation de la hanche dans une population bretonne et languedocienne. *Arch Pédiatr* 1:1093–1099.
- Lehtola V-P. 2004. *The Sámi people: traditions in transition*, 2nd ed. Fairbanks, AK: University of Alaska Press.
- Lipton EL, Steinschneider A, Richmond JB. 1965. Swaddling, a child care practice: historical, cultural, and experimental observations. *Pediatrics* 35: 521–567.
- Loder RT, Skopelja E. 2011. The epidemiology and demographics of hip dysplasia. *ISRN Orthop* 2011:1–46.
- Lovejoy CO, Meindl RS, Pryzbeck TR, Mensforth RP. 1985. Chronological metamorphosis of the auricular surface of the ilium: a new method for the determination of adult skeletal age at death. *Am J Phys Anthropol* 68:15–28.
- Loveland CJ, Gregg JB, Bass WM. 1985. Ancient osteopathology from the Caddoan burials at the Kaufman-Williams site, Texas. *Plains Anthropol* 30:29–43.
- Lowry CA, Donoghue VB, O'Herlihy C, Murphy JF. 2005. Elective Caesarean section is associated with a reduction in developmental dysplasia of the hip in term breech infants. *J Bone Joint Surg* 87:984–985.
- MacFarlane A. 1980. Congenital dislocation of the hip - an epidemiological conundrum. *J Mater Child Health* 13–15.
- MacLennan AH, MacLennan SC. 1997. Symptom-giving pelvic girdle relaxation of pregnancy, postnatal pelvic joint syndrome and developmental dysplasia of the hip. *Acta Obstet Gynecol Scand* 76:760–764.
- Mafart B, Kefi R, Beraud-Colomb E. 2007. Palaeopathological and palaeogenetic study of 13 cases of developmental dysplasia of the hip with dislocation in a historical population from southern France. *Int J Osteoarch* 17:26–38.
- Mahan ST, Kasser JR. 2008. Does swaddling influence developmental dysplasia of the hip? *Pediatrics* 121: 177–178.
- Mamouri GH, Khatami F, Hamed AB. 2004. Congenital dislocation of the hip in newborns of Mashhad City. *Intern J Ped Neonat* 4(1).
- McPherron A. 1967. The Juntunen site and the Late Woodland Prehistory of the Upper Great Lakes Area. *Anthropological papers. Museum Anthropol* 30: 214.

- Meindl, RS, Lovejoy, CO, Mensforth RP, Walker RA. 1985a. A revised method of age determination using the os pubis, with a review and tests of accuracy of other current methods of pubic symphyseal aging. *Am J Phys Anthropol* 68:29–45.
- Merbs CF, Vestergaard EM. 1985. The paleopathology of Sundown, a prehistoric site near Prescott, Arizona. In: Merbs CF, Miller RJ, eds. *Health and disease in the prehistoric Southwest*. Tempe, AZ: Arizona State University. *Anthropol Res Pap* 34:115–127.
- Miles JS. 1975. Orthopedic problems of the Wetherill Mesa populations. Washington, DC: US Department of the Interior National Park Service.
- Mirdad T. 2002. Incidence and pattern of congenital dislocation of the hip in the Aseer region of Saudi Arabia. *West Afr J Med* 21:281–222.
- Mitchell PD, Redfern RC. 2007. The prevalence of dislocation in developmental dysplasia of the hip in Britain over the past thousand years. *J Pediatr Orthop* 27:890–892.
- Mitchell PD, Redfern RC. 2008. Diagnostic criteria for developmental dislocation of the hip in human skeletal remains. *Int J Osteoarch* 18:61–71.
- Mitchell PD, Redfern, RC. 2011. Brief communication: developmental dysplasia of the hip in Medieval London. *Am J Phys Anthropol* 144:479–484.
- Morse DF. 1963. The Steuben Village and Mounds: a multicomponent late Hopewell Site in Illinois. *Anthropological Papers*. Ann Arbor, MI: University of Michigan. *Museum Anthropol* 21:93.
- Morse D. 1969. *Ancient Disease in the Midwest*. Springfield, IL: Illinois State Museum Pub No. 15.
- Moussa M, Alomran A. 2007. Acetabular dysplasia in adult hips of a Saudi population - a possible relation to coxarthrosis. *Saudi Med J* 28:1059–1061.
- Noordin S, Umer M, Hafeez K, Nawaz H. 2010. Developmental dysplasia of the hip. *Orthop Rev* 2(2):19.
- Ortner DJ. 2003. *Identification of pathological conditions in human skeletal remains*. Washington, DC: Academic Press.
- Pearson C. 2013. Swaddling ban: why are day cares banning baby burritos? *The Huffington Post*. Retrieved from http://www.huffingtonpost.com/2013/04/03/swaddlingban_n_2885662.html. Accessed on December 10, 2013.
- Pfeiffer S. 1984. Paleopathology in an Iroquoian ossuary, with special reference to tuberculosis. *Am J Phys Anthropol* 65:181–189.
- Phenice TW. 1969. A newly developed visual method of sexing the os pubis. *Am J Phys Anthropol* 30:297–301.
- Pratt WB, Freiburger RH, Arnold WD. 1982. Untreated congenital hip dysplasia in the Navajo. *Clin Orthop* 162:69–77.
- Rabin DL, Barnett CR, Arnold WD, Freiburger RH, Brooks G. 1965. Untreated congenital hip disease: a study of the epidemiology, natural history, and social aspects of the disease in a Navajo population. *Am J Pub Health Nat Health* 55:1–44.
- Romero MI, Julián M, Gaete V, Bedregal P, Pinto JI, Castiglione C. 1989. Epidemiologic characteristics of congenital hip dysplasia in a Chilean population. *Rev Chilena Ped* 60:268–271.
- Roper A. 1976. Hip dysplasia in the African Bantu. *J Bone Joint Surg* 58:155–158.
- Roposch A, Wright JG. 2007. diagnostic information and understanding disease: uncertainty in the diagnosis of developmental hip dysplasia. *Radiology* 242:355–359.
- Samborska B, Lembrych, S. 1986. Congenital dysplasia and congenital luxation of the hip joint in newborns. Assessment of the course of pregnancy and labor. *Gin Pol* 57:102–107.
- Saugstad, LF. 1991. Persistent pelvic pain and pelvic joint instability. *Eur J Obstet Gyn Rep Biol* 41:197–201.
- Shaheen, AE-KM. 1989. Mehad: the Saudi tradition of infant wrapping as a possible aetiological factor in congenital dislocation of the hip. *JR Coll Surg Edinb* 34:85–87.
- Schoenecker PL, Flynn JM. 2007. Screening for developmental dysplasia of the hip. *Pediatrics* 119:652–653.
- Siffel C, Alverson CJ, Correa A. 2005. Analysis of seasonal variation of birth defects in Atlanta. *Birth Def Res* 73:655–662.
- Skirving AP, Sims TJ, Bailey AJ. 1984. Congenital dislocation of the hip: a possible inborn error of collagen metabolism. *J Inher Metab Dis* 7:27–31.
- Smith DW, Aase JM. 1979. Polygenic inheritance of certain common malformations. Evidence and empiric recurrence risk data. *J Pediatr* 76:653–659.
- Snow CE. 1948. *Indian Knoll Skeleton*. University of Kentucky Reports in Anthropology, Vol. 4. Lexington, Kentucky: University of Kentucky. p 514–517.
- Slater BCS, Watson GI, McDonald JC. 1964. Seasonal variation in congenital abnormalities. Preliminary report of a survey conducted by the Research Committee of Council of the College of General Practitioners. *Br J Prev Soc Med* 18:1–7.
- Stein-Zamir C, Volovik I, Rishpon S, Sabi R. 2008. Developmental dysplasia of the hip: risk markers, clinical screening and outcome. *Pediatr Int* 20:341–345.
- Stevenson RE, Huntley CC. 1967. Congenital malformations in offspring of phenylketonuric mothers. *Pediatrics* 40:33–45.
- Storer SK, Skaggs, DL. 2006. Developmental dysplasia of the hip. *Am Fam Phys* 74: 1310–1316.
- Taboridze II, Aladashvili LT, Lordkipanidze EF. 1991. Association of the ABO blood group system and hip joint dysplasia. *Ortop Travmatol Protez*. 8:23–26.
- Tegnander A, Holen KJ, Terjesen T. 1999. The natural history of hip abnormalities detected by ultrasound in clinically normal newborns. A 6–8 year radiographic follow-up study of 93 children. *Acta Orthop Scand* 70:335–337.
- Terjesen T, Holen KJ, Tegnander A. 1996. Hip abnormalities detected by ultrasound in clinically normal newborn infants. *J Bone Joint Surg* 78:636–640.
- Thieme WT, Wynne-Davies R, Blair HAF, Bell ET, Loraine JA. 1968. Clinical examination and urinary estrogen assays in newborn children with congenital dislocation of the hip. *J Bone Joint Surg* 50:546–550.
- Timmler T, Wierusz-Kozłowska M, Markuszewski J, Woźniak W. 2005. The hip joints of preterm neonates in sonographic evaluation. *Chir Narz Ruchu Ortop Pol* 70:301–305.
- Üden A, Lindhagen T. 1988. Inguinal hernia in patients with congenital dislocation of the hip. A sign of general connective tissue disorder. *Acta Orthop Scand* 59:667–668.
- Wakefield EG, Dellinger SC, Camp JD. 1937. A study of the osseous remains of the "mound builders" of eastern Arkansas. *Am J Med Sci* 193:488–495.
- Walker JM. 1977. Congenital hip disease in a Cree- Ojibwa population: a retrospective study. *Can Med Ass J* 116:501–504.
- Wang, E, Liu, T, Li, J, Edmonds, EW, Zhao, Q, Zhang, L, Zhao, X, Wang, K. 2012. Does swaddling influence developmental dysplasia of the hip? An experimental study of the traditional straight-leg swaddling model in neonatal rats. *J Bone Joint Surg Am* 94:1071–1077.
- Weinstein SL. 2013. Lovell and Winter's pediatric orthopedics. Philadelphia: Lippincott Williams and Wilkins.
- Wheeler RL. 1985. Pathology in late thirteenth century Zuni from the El Morro Valley, New Mexico. In: Merbs CF, Miller RJ, editors. *Health and disease in the prehistoric Southwest*. Tempe, AZ: Arizona State University. *Anthropol Res Pap* 34:79–84.
- Wientroub S, Grill F. 2000. Ultrasonography in developmental dysplasia of the hip. *J Bone Joint Surg* 82:1004–1018.
- Wynne-Davies R. 1970. Family study of neonatal and late diagnosis congenital dislocation of the hip. *J Med Genet* 7:315–333.
- Wynne-Davies R, Littlejohn A, Gormley J. 1982. Aetiology and interrelationship of some common skeletal deformities. *J Med Genet* 19:321–328.
- Yamada Y. 1993. Prophylaxis of congenital dislocation of the hip at Tokoname city. In: Ando M, editor. *Prevention of congenital dislocation of the hip in infants: experience and results in Japan*. Asahikawa, Japan: Yamada Co. Ltd. p 27–30.