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Is it worthwhile to routinely ultrasound screen children with idiopathic clubfoot for hip dysplasia?

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Abstract

Purpose—Patients with idiopathic clubfoot are considered at increased risk for having developmental dysplasia of the hips (DDH). However, the studies showing this association have been relatively small. Many clinicians who treat idiopathic clubfoot routinely screen the hips of these patients with ultrasound or radiograph due to the concerns of increased risk of DDH. We evaluated a large clubfoot population to determine the risk of DDH and compare this to a population of children without clubfoot. We also evaluated if the clubfoot patients found to have DDH would have been discovered by standard DDH screening.

Methods—We identified infants in three states (MA, NY, NC) who were reported to each state's birth defects registry as having a clubfoot. A second cohort of infants without clubfoot was also identified as a control group. Mothers of these children were contacted to be included in the study, and a computer-assisted telephone interview was administered by one of the study nurses, including questions about treatment of DDH. The child's median age at interview was 7 months. Mothers of clubfoot cases were also contacted for follow-up at mean age of 3.3 years.

Results—Families of 677 patients with clubfoot and 2037 controls were interviewed. 5/677 (0.74%) patients with clubfoot and 5/2037 (0.25%) controls reported having their infant treated with a brace or harness for hip problems ($p=0.134$). Of the patients with clubfoot, two of them did not need treatment for their DDH and two would have been discovered by standard hip screening. Follow-up study at 3.3 years of age found no serious late hip dysplasia.

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Conclusions—Treatment of DDH was uncommon in all children; the higher proportion in infants with clubfoot was not statistically different than controls. Of the patients with clubfoot and DDH, standard hip screening would have been appropriate and others did not need treatment. These data suggest that routine hip ultrasound or radiographic screening of idiopathic clubfoot patients is not necessary unless indicated by the standard infant hip screening.

Background

The American Academy of Pediatrics (AAP) recommends screening for developmental dysplasia of the hips (DDH) by physical exam of all infants and by ultrasonography of infants considered at high risk for hip dysplasia (breech presentation at time of delivery, family history of DDH, or physical exam concerning for DDH^{1,2}. The prevalence of DDH in the general population varies from 0.14% to 3.5% live births, depending on the study and the method of detection^{1,3}. Among children with talipes equinovarus (TEV or clubfoot), the prevalence of DDH was thought to be higher due to the presumption that both result from intrauterine compression^{4–6}. As understanding of the etiology of both of these conditions improved, the link became less clear. Ponseti showed that the development of the clubfoot occurred long before intrauterine compression was significant⁷. Studies showing an association between clubfoot and DDH are small, clinical series of patients^{8–10}. However considerable concern remains that there is an increased rate of hip dysplasia in patients with idiopathic clubfoot^{8–10}. Many orthopaedic practitioners who routinely treat clubfoot often still utilize ultrasound or radiographic screening of the hips in all their patients with clubfoot because of concerns for this association. However, it is not clear if the cost and time burden to the care of these patients is warranted.

The primary purpose of this study is to evaluate a large idiopathic clubfoot population to determine their risk of having DDH and compare this to a similar population of children without idiopathic clubfoot. The secondary purpose is to determine standard infant DDH screening would have discovered the cases of hip dysplasia in patients with clubfoot or if additional screening is necessary.

Methods

This was part of a large population-based case-control epidemiologic study that has already been described elsewhere in detail¹¹. Between 2007 and 2011 infants who were reported to the birth defects registry as having a clubfoot in the states of Massachusetts, New York (not including New York City), and North Carolina were identified. New York City was not included because it conducts birth defects surveillance separately from the rest of NY State. Additionally, infants without a malformation were identified in the same three states from birth certificates (MA and NC) or birth hospital (NY) to serve as controls. Mothers of cases and controls were mailed an introductory letter and consent, and then had a follow-up phone call. They then underwent a computer-assisted interview by a study nurse. Medical records of the cases, when approved by the mother, were obtained. The mothers of both the cases and controls were contacted when the infant was on average 7 months old. For cases whom were 18 months of age, mothers were re-contacted for a follow-up interview when the child was on average 3.3 years of age with an additional set of questions. The institutional

review boards at Boston University and the state health departments in Massachusetts, North Carolina, and New York approved the study protocol.

Case classification

The mother of each infant who was identified through the birth defects registry as having a clubfoot was queried about their child's foot and its treatment to confirm the diagnosis of idiopathic clubfoot. Medical records were reviewed for the validity of the clubfoot, as well as other birth defects or associated disorders. Any treatment or involvement of the hips was noted. Orthopaedic notes, when available, were used for confirmation of the diagnosis of clubfoot; in the absence of medical records, the mother's report of at least 3 casts applied for the foot disorder defined a clubfoot case. Questionable records were reviewed by a pediatric orthopaedist (STM) for review who was blinded to the results of the questionnaire during this review. Patients with known syndromes or neurogenic clubfoot were excluded.

Control group selection

Controls were selected from the birth certificates (MA and NC) or birth hospitals (NY) by each state, selecting approximately 4 controls per case. Mothers of controls were sent an introductory letter invited to participate; mothers who agreed underwent computer assisted telephone query by one of the study nurses. Telephone query confirmed that each control subject did not have any malformations or birth defects.

Data collection

In the initial telephone interview, questions regarding hip dysplasia and its treatment were asked of both the cases and controls. These questions were as follows: "Was your baby treated with a harness or brace for his/her hips?"; if yes, "was it for a dislocated hip?"; and if yes, "what age was your child at the time of treatment?" The follow-up telephone interview of mothers of idiopathic clubfoot asked the following questions: "Has your child been diagnosed with hip problems?"; "did they receive an ultrasound or xray?"; "do/did they wear a brace?"; and "has your child been treated for hip dysplasia?"

Data Analysis

Idiopathic clubfoot cases were compared to controls for maternal report of treated hip dysplasia, based on the initial interview questions. Proportions were compared using Fisher exact test with statistical significance set at $p < 0.05$, two tailed. Analysis was done both on the aggregate as well as the individual state (MA, NC, NY) level. Descriptive analysis was done on individual patients.

Results

The mothers of 677 patients with confirmed idiopathic clubfoot and 2037 controls participated in the study. Distributions of demographic factors for cases and controls have previously been reported¹¹. Briefly, cases were more likely to be male and first born than controls; both groups were generally similar with regard to maternal age, race/ethnicity, and education. Approximately 40% of case subjects resided in North Carolina and 28% and 31% resided in Massachusetts and New York, respectively. Treatment for hip dysplasia was

reported by only five mothers of patients with a clubfoot (0.74%) and five mothers of control subjects (0.25%). Although the proportion of treated hip dysplasia was three-times higher among cases, the difference was not statistically significant different ($p=0.134$). See Table 1. These proportions were evaluated according to maternal residence. In Massachusetts, treated hip dysplasia was reported for one case and one control (0.52% versus 0.22%, $p=0.504$). In New York, one case and three controls were reported to have hip dysplasia (0.47% versus 0.53%, $p=1.0$). In North Carolina, three cases and one control were reported to have hip dysplasia (1.10% versus 0.10% $p=0.032$).

Medical records were available for review in 77% of the patients with clubfoot and 4 of the 5 cases with maternal report of treated hip dysplasia. In the single patient from Massachusetts, the medical record was not available for review, but the family reported that the diagnosis of acetabular dysplasia was made at 6 months of age, which was treated with a Pavlik harness. In the single patient from New York with clubfoot and DDH, the patient was noted to be breech presentation at time of delivery and treated in a Pavlik harness for hip dysplasia. There were three patients from North Carolina with clubfoot and DDH. One infant was diagnosed at birth with DDH, and the mother noted that the hip relocated within the first 4 days, and no further brace or treatment was done. One infant was treated for “mild” acetabular dysplasia that resolved after six weeks in a Pavlik harness. The family noted that there was family history of hip dysplasia and “loose” hips were found on the physical exam by the orthopaedic surgeon. The last patient had a screening hip ultrasound at 4 weeks of age that showed the right hip had 45% coverage and an alpha angle of 57 degrees; the left hip was noted to be normal. The patient was treated in a Pavlik harness and the hips resolved. See Table 2.

Among the 226 patients with clubfoot (43% of eligible) whose mothers were re-contacted and who had initial reports of hip dysplasia confirmed the diagnosis and treatment. Another 12 families who initially reported “no” to treatment for hip dysplasia responded “yes” when asked if their child had been *diagnosed* with hip problems. Of those, 8/12 reported that imaging of the hips had been done (radiograph or ultrasound). Two patients reported that they had “treatment” for hip dysplasia, one patient noted treatment with nighttime brace for hip immaturity, and another with “stretchy pants”. One child was diagnosed late with mild cerebral palsy and underwent hip screening without treatment. Six families noted that their child’s hips were described as “out of alignment” or “positional” or “loose” and had no treatment. No child was treated for severe hip dysplasia. See Table 3.

Although no follow-up was conducted on control subjects, information was obtained during the initial interview on breech presentation at delivery and family history of hip dysplasia, covering two of the AAP criteria for ultrasound screening for DDH. Treated hip dysplasia was reported in two of 158 (1.3%) control subjects with either of those AAP criterion compared to 3 of 1879 (0.16%) without either criterion.

Discussion

Historically, talipes equinovarus and hip dysplasia have been considered to be related^{4,5}. However, as the etiologies of the two conditions have been better understood, the link has

become less certain^{6,7,12}. Nonetheless, there has been ongoing concern that patients with idiopathic clubfoot have an increased risk for hip dysplasia^{8–10}. Most of the studies that have investigated this link have been small in numbers, and typically an individual surgeon's case series. We assessed the prevalence of treatment for hip dysplasia based on maternal report in a large idiopathic clubfoot population and a similar control group of patients without clubfoot. We found the prevalence of hip dysplasia was low, both in infants with clubfoot (5/677, 0.74%) and controls (5/2037, 0.25%). Although not significantly different ($p=0.134$), the three-fold higher proportion for clubfoot subjects is apparent. We also assessed the medical record documentation, as available, of the patients with clubfoot treated for hip dysplasia to determine if the hip treatment was indicated and if standard infant hip screening would have discovered the hip dysplasia. Two of the five patients with clubfoot who report treatment for hip dysplasia did not have treatable hip dysplasia, and an additional two patients' hip dysplasia would have been found by standard infant hip screening. Follow-up on average at 3.3 years of age was also done for many of the clubfoot population and this found no severe case of late presenting hip dysplasia, leaving just one of 677 patients with treatable DDH identified by image screening alone. While longer follow-up of the control group was not done, if this had been done and discovered any late-presenting hip dysplasia, it would only lessen the slight difference in hip dysplasia in the clubfoot group compared to the controls, rather than increase it.

Several authors have argued for increased screening of hip dysplasia in patients with clubfoot. Canavese et al⁸ reported two patients who were treated for clubfoot and later found to have hip dysplasia. Perry et al⁹ presented an observational cohort study of 119 infants with clubfoot that found 7 of them had hip dysplasia. Only four of them had dysplasia of Graf III or IV severity¹³. Carney et al¹⁰ reported a case series of 51 patients treated for idiopathic clubfoot who had a plain hip radiograph at 4 or more months of age to screen for DDH. They found the acetabular index measured more than 28 degrees in 8 infants; however, no child had treatment for their hip dysplasia and all resolved untreated.

There have been several authors who have advocated against increased screening for hip dysplasia in patients with idiopathic clubfoot. Paton et al¹⁴ reported on a prospective study over 11 years of neonatal foot deformities and hip dysplasia. Of 614 infants with foot deformities, 60 had true clubfoot. Of those with clubfoot, 7/60 had acetabular dysplasia of Graf type II severity and all resolved without treatment. Westberry et al¹⁵ reported on the largest prior study of hip dysplasia in idiopathic clubfoot. They followed 349 patients with clubfoot followed to average of 8.4 years; 127 patients had radiographs of the hips and the remaining 222 were followed clinically. Of those who had a radiograph, only 1 (0.8%) was found to have DDH and she was known to have hip dysplasia on presentation to the orthopaedic clinic. No further patients were found to have hip dysplasia radiographically or clinically¹⁵.

Results from our series of 677 infants with clubfoot do not support the need for increased screening of hip dysplasia in this patient population. Of the five patients in our study reported to be treated for hip dysplasia, two did not warrant treatment and two had hip dysplasia that would have been found by standard infant hip screening. On follow-up at average 3.3 years of age, no child was found to have late presenting severe hip dysplasia.

We don't know what percentage of the clubfoot population in our study had a screening hip ultrasound or radiograph because of the clubfoot, but all had increased scrutiny of their hips because of the routine contact with a pediatric orthopaedic surgeon. Thus, it is reasonable to assume that all clubfoot cases with treatable hip dysplasia in this study would have become clinically apparent and that routine ultrasound or radiographic screening of newborns would have conferred no benefit. The overall prevalence of hip dysplasia was low in both the patients with clubfoot (0.75%) and the control population (0.25%), representing the low end of the previously reported prevalence of hip dysplasia in the general population of 0.14% to 3.5%³.

The number of clubfoot patients and controls was different in each state due to correspondingly different numbers of births in the catchment areas of each state.; This was a population-based study and the distribution of demographics for our participants are similar to the populations from where we ascertained cases and controls¹¹. It is unlikely that the association of clubfoot and hip dysplasia is differential by location; hence, state of residence would not be a confounder in this study.

This study was part of a larger study investigating the etiology of idiopathic clubfoot¹¹ and therefore other congenital foot anomalies were not included. While it would be interesting to investigate the association between other congenital foot anomalies (such as Calcaneovalgus foot and positional clubfoot) with hip dysplasia, this is outside the scope of this study.

There has been some recent controversy about whether or not to screen for hip dysplasia in the general population^{16–18}, however the American Academy of Pediatrics still recommends physical exam screening for all infants with utilization of ultrasound in infants considered at high risk for hip dysplasia (breech presentation at time of delivery, family history of DDH, or physical exam concerning for DDH)^{1,2} and more recent studies have confirmed this recommendation^{16,19}. Based on maternal reports of treated hip dysplasia in our control population, routine screening of the general newborn population would identify DDH in only 0.16%. Each year there are, on average, around 4 million births in the United States²⁰; clubfoot typically occurs in 1 per 1000 live births⁶, so roughly 4,000 infants are born with a clubfoot each year in the US. The cost of an infant bilateral hip ultrasound is around \$625. Some of those infants with clubfoot are not idiopathic and some will warrant a hip ultrasound for other risk factors. Assuming 50% of the 4,000 annual births with clubfoot are idiopathic and they were not to receive routine hip ultrasound screening, the healthcare system would save 1.25 million dollars each year. This study did not address the syndromic or neurogenic clubfoot; as hip dysplasia shows increased prevalence in many of these infants, routine hip screening in this group is still recommended.

In summary, data from this largest study to date on the relationship between idiopathic clubfoot and hip dysplasia suggest little or no benefit from specialized hip screening of newborn infants with idiopathic clubfoot. These patients will benefit from a careful physical exam of the hips by their treating orthopaedic surgeon. For those infants with positive family history of hip dysplasia, breech presentation at time of delivery, or concerning physical exam, a hip ultrasound appears to be indicated based on our observed two-fold greater prevalence of DDH in such infants.

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Answers to the question: Was your child treated with a brace or harness for hip problems?

Table 1

Hip dysplasia treatment	Case			Controls			p-value from Fisher's exact test
	Hip Problems	N	%	Hip problems	N	%	
Overall	5	677	1%	5*	2037	0%	0.134
MA	1	191	0.52%	1	455	0.22%	0.504
NY	1	212	0.47%	3	568	0.53%	1.0
NC	3	274	1.10%	1	1014	0.10%	0.032

* Mother of one control was unsure.

Table 2

Patients with clubfoot who report treatment for DDH. Patient and treatment characteristics.

Patient	State	Age DDH Dx	Treatment	Details
1	MA	6 months	Pavlik	Medical record not available
2	NY	6 weeks	Pavlik	Breech presentation at delivery, hip located but shallow acetabulum
3	NC	Birth	None	Hip relocated on own, no treatment
4	NC	5 weeks	Pavlik	Positive family history of DDH and “loose” hips on exam; acetabular dysplasia on ultrasound
5	NC	4 weeks	Pavlik	Ultrasound at 4 weeks showed right hip alpha angle of 57° and 45% coverage; left hip normal

Table 3

Patients with clubfoot who, when asked on follow-up contact, “has your child been diagnosed with hip problems” responded “yes” when on initial contact had reported “no” to being treated for hip dysplasia

Patient	Details of hip issue	Any imaging done?	Any treatment done?
1	Doctor said child had hip problem, mom not sure of details	No	No
2	Pain in hip, no diagnosis	No	No
3	“Positional” hip problem, will start PT for intoeing	No	No
4	Hips “out of alignment” according to chiropractor	No	No
5	Hips “out of alignment”	Yes	Yes, “stretchy pants”
6	Leg length inequality, hips are out of alignment	Yes	No
7	Muscle weakness due to mild cerebral palsy	Yes	No
8	Intoeing due to “hip problem”	Yes	No
9	“Loose hips”, not dysplasia	Yes	No
10	Hips are “premature”	Yes	Yes, nighttime brace age 6–18 months
11	Slight hip dysplasia, thought positional due to breech positioning at birth	Yes	No
12	Doctor told mom hips are “hairline away from being normal” but not sure what he meant	Yes	No