# Developmental Dysplasia of the Hip in Infants With Congenital Muscular Torticollis

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#### Abstract

Infants with congenital muscular torticollis (CMT) are at increased risk for developmental dysplasia of the hip (DDH), which has led to increased use of diagnostic procedures. Our goal in this study was to establish indications for imaging the hips of infants presenting with CMT. We

reviewed the cases of 292 patients with the diagnosis of CMT, 16 of whom were found to have DDH. Each patient with DDH had an abnormal clinical hip examination.

Our study results demonstrate that, despite the association of these disorders, an infant presenting with CMT does not require routine hip imaging in light of a normal clinical hip examination. The coexistence rate for CMT and DDH requiring treatment is 4.5%, which is lower than the commonly accepted 20%.

he association of congenital muscular torticollis (CMT) and developmental dysplasia of the hip (DDH) has been established and is widely accepted. Since Iwahara and Ikeda<sup>1</sup> reported on the coexistence of these disorders in 1962, it has been confirmed by results from several studies.<sup>2-9</sup> Reported rates have ranged from 0% to the commonly cited 20%, which was established by Hummer and MacEwen in 1972.<sup>10</sup>

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Given this association, identification of one of the disorders initiates an investigation for the other. Most often, the clinically apparent torticollis leads to the hip dysplasia evaluation. In this evaluation, a detailed physical examination is performed, with an emphasis on the hips, and the hips often are imaged. Radiography has long been used in identifying hip dysplasia, but the disorder is more difficult to assess in young infants because of their lack of ossification about the hip. In recent years, ultrasonography has been established as a tool in early diagnosis of hip dysplasia in young infants, but not without expense to the families and the medical system.

The precise etiology of DDH is unknown, but physiologic, mechanical, and genetic factors have been implicated.<sup>11</sup> The higher rate of conditions such as metatarsus adductus, congenital knee dislocation, and CMT in infants with DDH supports the intrauterine molding theory.<sup>12,13</sup> Firstborn infants with breech presentation also have an increased incidence of hip dysplasia. DDH is more common in females and affects the left hip more often—tendencies attributed to hormonal factors and fetal positioning, respectively. Current guidelines<sup>14</sup> call for imaging female infants with either breech presentation or a positive family history because of the significant risk for DDH. There is no consensus regarding imaging infants presenting with CMT, likely because these risk factors have not been thoroughly investigated in this patient population.

For infants with torticollis, the question is still which of them should be imaged in a search for the occult hip disorder. Our goal in this study was to assess the indications for imaging the hips in infants with CMT, to evaluate the risk factors for hip dysplasia, and to determine the coexistence rate of these disorders.

### **MATERIALS AND METHODS**

A patient list was generated by searching computerized outpatient records for the diagnosis of torticollis between 1988 and 2002. Patients selected were younger than 1 year at presentation and were examined by 1 of 5 pediatric orthopedic surgeons at the main hospital or affiliated offices.

Charts were reviewed and data recorded. Data included reason for referral, birth history, physical examination findings, associated conditions, and treatment. Diagnosis of CMT was clinically made on the basis of decreased neck rotation or lateral bending or the characteristic chinocciput positioning with or without palpable sternocleidomastoid (SCM) tumor or tightness. Patients with nonmuscular causes of torticollis were excluded.

Hip examination abnormalities were defined as gross instability (positive Barlow and Ortolani maneuvers), Galleazzi sign, asymmetric abduction or thigh folds, abnormal position of the greater trochanter (Klisic line and Nélaton's lines), and a palpable glide or click (after 2 weeks of age). When hip radiographs were obtained, higher acetabular index, disruptions in the continuity of Shenton's line, and femoral head uncovering were recorded. When ultrasound was performed,  $\alpha$  angle and Graf classification were documented. Instability on dynamic testing was also recorded.

Hip dislocation was defined as gross instability on examination or an irreducible hip. Hip subluxation was defined clinically as the ability to displace but not dislocate the head from the acetabulum or radiographically by femoral head uncovering. Acetabular dysplasia was defined radiographically as a higher acetabular index versus normalized data<sup>15</sup> or ultrasonographically according to Graf classification.<sup>16</sup>

Potential risk factors that were examined included birth position (breech vs vertex), type of delivery (cesarean vs vaginal), gravida (firstborn vs subsequently born), single versus multiple gestations, sex, reason for referral, side of torticollis, and presence of a foot deformity or SCM mass. A history of breech position with successful version was considered breech.

For the statistical analysis, descriptive summaries were obtained for all variables of interest, including birth factors, using frequency tabulations for categorical variables and means and SDs for continuous variables. Predictive and other factors associated with hip dysplasia were determined with the Fisher exact test, and mixed effects logistic regression modeling methods were used depending on the distribution of DDH among levels of the risk factors. The effect of each of the covariates (as fixed effects) on the binary outcome of hip dysplasia was determined with a logistic regression model using attending physician as a random effect. Odds ratios and corresponding 95% confidence intervals were reported. As only 16 of the 292 infants presented with hip dysplasia, single-predictor models were analyzed. For risk factors that did not have any false-positives, the Fisher exact test was used to test the association between the factors and DDH. The t test was used to compare age at imaging between ultrasound and radiograph. All conclusions were made at a .05 level of significance.

#### RESULTS

Searching charts for the diagnosis of CMT, we identified 301 consecutive infants. Nine were excluded: 7 for diagnoses of segmental cervical spine abnormalities, Klippel-Feil syndrome, ocular torticollis, arthrogryposis, and meningomyelocele; 1 for an incomplete chart; and 1 for lack of an ultrasound (examination was normal, and the infant was referred for a hip evaluation but was lost to follow-up before the ultrasound could be obtained).

The remaining 292 infants with CMT were included in the study. The sex distribution was about even: 153 male (52%), 139 female (48%). The torticollis was left-sided in 174 cases (60%), right-sided in 118 (40%). A mass in the SCM was noted in 40 infants, 35 of whom were under 3 months of age. Presenting age was under 3 months for 114 patients (39%), between 3 and 6 months for 86 patients (29%), and after 6 months for 92 patients (32%). Of the 260 patients whose charts included birth history, 31 (12%) had breech position listed, and 47 (18%) cesarean delivery. Patients were firstborn in 58 cases (22%) and part of multiple gestation births in 17 cases (6.5%).

Associated musculoskeletal conditions included talipes equinovarus (5 infants), metatarsus adductus (4), internal tibial torsion (2), clavicle fracture (2), and brachial plexus injury (2). No correlation was found between side of foot deformity and hip dysplasia.

Of the 188 patients with hip imaging, 125 (66%) had ultrasound as their initial evaluation, and 63 (34%) had radiographs. The patients with ultrasounds (mean age, 3.55 months; SD, 2.35 months) were significantly (P<.001) younger than the patients with radiographs (mean, 7.53 months; SD, 4.4 months).

DDH was found in 16 patients, 9 of whom were diagnosed with hip dislocation. Eight of the 9 had gross instability on examination and were successfully treated in a Pavlik harness (they received interval ultrasounds in the harness to ensure reduction and an ultrasound with Graf angles at termination of harness use). The ninth patient had bilateral irreducible hips that required open reduction and femoral shortening. Four other patients, found to have hip subluxation, were also successfully treated in a Pavlik harness. The remaining 3 patients had acetabular dysplasia that was observed with repeat hip imaging; 2 of these patients had Graf IIa hips, and the third had a higher index on radiographs. All patients had radiographic or ultrasonographic resolution on follow-up imaging.

All 16 (11 female, 5 male) patients with DDH had an abnormal clinical hip examination. (Twelve other patients had an abnormal hip examination but no evidence of hip dysplasia.) Fourteen of the 16 were presented within the first week of life, and the other 2 were presented at 3 and 9 weeks, respectively; instability was the most common finding. (Patients with subluxation and acetabular dysplasia were presented later, a mean of 12 weeks after birth, with unilateral limited abduction as the most common finding.) Nine of the 16 had the left hip affected, 4 had the right hip affected, and 3 had both hips affected. All clinical hip abnormalities corresponded to the side on which the dysplasia was diagnosed after imaging.

Of the 292 patients, 27 were referred to the orthopedic surgeon for hip evaluation only (15) or for hip and neck evaluation (12). Fifteen of the 16 patients with hip dysplasia were among this group; the 16th patient, referred for torticollis alone, had hip subluxation noted clinically and radiographically. Of the remaining 265 patients, 4 were initially seen by the orthopedic surgeon for a foot

Table. Pot	ential Risk	Factors	for Hip	Dysplasia
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Risk Factor	Р	OR (95% CI)	
Breech position Cesarean delivery Multiple gestations Female sex Firstborn Side of torticollis Foot deformity <sup>a</sup> Sternocleidomastoid mass <sup>a</sup> Hip evaluation referral <sup>a</sup> Abnormal hip examination <sup>a</sup>	.0033 <.001 .56 .062 >.99 .81 .4 .072 <.001 <.001	4.68 (1.68, 13.03) 5.19 (2.06, 12.04)	

Abbreviations: OR, odds ratio; Cl, confidence interval. <sup>a</sup>Fisher exact test.

evaluation, and their torticollis was identified during follow-up. None of these patients was diagnosed with hip dysplasia.

Analysis showed that breech position and cesarean delivery were significant risk factors (Table). Patients with these factors were 4.68 and 5.16 times more likely to have hip dysplasia, respectively. Birth-history details were insufficient, so the cesarean-delivery group could not be subdivided by reason for delivery (eg, scheduled cesarean delivery, failure to progress). Referral for hip evaluation and an abnormal hip examination were also significant factors associated with hip dysplasia. None of the patients with DDH had a SCM mass.

The hip dysplasia occurred ipsilateral to the torticollis in 12 cases, contralateral in 1 case, and bilaterally in 3 cases.

Five patients under 7 weeks of age had ultrasonographic evidence of physiologically immature hips that were appropriate for age (ie, Graf IIa). Follow-up imaging demonstrated mature hips in all these patients.

#### DISCUSSION

Investigators have reported different rates for the coexistence of these disorders.<sup>2-9</sup> The rate variance derives in part from a lack of precise defining criteria, particularly for hip dysplasia. The criteria for subluxation in the study by Hummer and MacEwen<sup>10</sup> included patients who would be considered normal by current definitions (thus increasing the coexistence rate). Some investigators noted that the severity of limitation in neck rotation correlated with DDH incidence,<sup>2,4,5</sup> but they did not describe their definitions of hip dysplasia. Regardless of the inconsistencies, however, these studies have demonstrated rates lower than the commonly cited 20%.

In their review of 70 infants and children with torticollis, Walsh and Morrissy<sup>8</sup> provided clear defining criteria for DDH and reported a coexistence rate of 8.5%. Hip radiographs were obtained for 77% of these patients. Patients without radiographs were assumed to have normal hips. Of the 6 patients found to have DDH, 4 were referred for treatment of a dislocated hip; the other 2 were referred for torticollis, and the hip dysplasia was detected on clinical examination, suggesting that routine physical examination is sufficient for detecting DDH in children with CMT.

Further supporting the study results of Walsh and Morrissy<sup>8</sup> is the fact that all 16 of our patients with DDH had an abnormal clinical examination and almost all (15) were referred to the pediatric orthopedist for hip evaluation. Of the 265 patients evaluated for torticollis alone, only 1 (0.4%) was diagnosed with hip dysplasia. Among the patients diagnosed with subluxation and acetabular dysplasia, the most common finding was unilateral limited abduction, which has been shown to be an important and specific clinical sign for DDH, particularly in infants older than 3 months.<sup>17</sup>

In our study, 104 infants did not receive hip imaging. If we were to make an assumption similar to the one Walsh and Morrissy<sup>8</sup> made-that these patients did not have hip dysplasia-then the coexistence rate for CMT and DDH requiring treatment would be 4.5%. This assumption is supported by the fact that all these patients had normal hip examinations and were referred for neck evaluations. There were also significantly fewer infants in this group, with its history of breech position and cesarean delivery (Ps = .02 and .002, respectively), than among the infants who received imaging. In addition, although physical examination of infants can be difficult, it is uncommon for experienced examiners to fail to detect a hip dislocation in these patients.<sup>18,19</sup> We included nonimaged patients in our study to obtain a consecutive group of patients; if these infants were excluded, however, our findings would remain valid, and the coexistence rate would change to only 6.9%. In any case, our findings are consistent with recent studies in which the coexistence rate was lower than the commonly accepted 20%.

In evaluations of infants with CMT, an abnormal hip examination should prompt further diagnostic studies. Breech position and cesarean delivery were significant predictors for hip dysplasia in our study. These risk factors should raise the index of suspicion but are not absolute indications for diagnostic imaging. This study provides no evidence that a female infant with CMT is at significantly increased risk for hip dysplasia that would require imaging in the absence of a clinical finding. However, if there is any uncertainty about the quality of the examination (such as might happen with a noncompliant patient), it would be prudent to obtain hip imaging.

In conclusion, this study demonstrates that, despite the association of CMT and hip dysplasia, an infant presenting with CMT does not require routine hip imaging in light of a normal clinical hip examination.

## AUTHORS' DISCLOSURE STATEMENT AND ACKNOWLEDGMENT

The authors report no actual or potential conflict of interest in relation to this article.

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#### COMMENTARY

The paper "Developmental Dysplasia of the Hip in Infants With Congenital Muscular Torticollis" again demonstrates that a proper clinical examination to determine the presence or absence of developmental dysplasia of the hip (DDH) is still appropriate. More sophisticated examinations with sonograms are unnecessary in the vast majority of cases unless the clinical examination cannot be performed by a trained medical practitioner. Factors or conditions associated with DDH such as a breech delivery, metatarsus adductus, congenital muscular torticollis, or a family history of DDH should reinforce in the examiner's mind the need to perform a proper and thorough clinical examination of the hips.

The only reason to perform additional testing such as sonography would be an improperly performed clinical examination of the hips. However, there is no doubt that in our present medicolegal environment, imaging studies may have to be considered in spite of a "good" clinical examination.

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This paper will be judged for the Resident Writer's Award.