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Prenatal screening for clubfoot: What factors predict prenatal detection?

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Abstract

Objective—Routine prenatal ultrasound has often resulted in the early detection of musculoskeletal disorders. The purpose of this study was to determine which socioeconomic factors are associated with prenatal detection of clubfoot.

Methods—The Slone Epidemiology Center at Boston University identified infants in three states (MA, NY, NC) who were reported as having a clubfoot. Mothers of these children were contacted, interviewed, and medical records obtained. Data were analyzed by using logistic regression to estimate odds ratios (ORs) and 95% confidence intervals (CIs).

Results—Overall detection of the clubfoot prenatally was 62.3% (421/676) but this varied considerably by state: 81.1% in Massachusetts (154/190), 58.5% in New York (124/212), and 52.2% in North Carolina (143/274). Multivariate analysis revealed the strongest predictors for prenatal detection were maternal age ≥ 35 years (OR: 3.54), non-Hispanic black race (OR: 0.49), the presence of another birth defect (OR: 2.61), residing in Massachusetts (OR: 2.64) and the presence of a bilateral clubfoot (OR: 1.90).

Conclusions—We found a statistically significant increase in higher rate of prenatal detection of clubfoot in Massachusetts and decrease in lower rate in younger mothers (age < 35) and black mothers, even after adjustment for other sociodemographic variables.

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Introduction

Screening prenatal ultrasounds have become routine. This frequent use of ultrasonography, along with improving sonographic technology has often resulted in early detection of musculoskeletal disorders ^{1,2}. Talipes equinovarus, or clubfoot, is typically detected prenatally about 60% of the time ^{3,4}. The detection rate is improving as sonographic technology is improving ⁴, and three-dimensional ultrasound brings a whole new level of ability to detect these malformations ⁵. Other factors, however, play a role in the ability to detect clubfoot prenatally, and those factors are poorly understood.

Prenatal diagnosis of clubfoot can be very beneficial for family education and counseling ^{6,7}. It has been shown that expecting mothers want to know about the possible diagnosis of clubfoot prior to delivery ³ and that bonding with a newborn with clubfoot may be impaired because of the foot deformity ⁸. In order to better understand the deterrents to prenatal diagnosis of clubfoot, we aimed to explore socioeconomic and demographic factors associated with the detection of clubfoot in utero.

Methods

We used data from a large population-based case-control epidemiologic study that has already been described elsewhere in detail ⁹. Cases were infants born with clubfoot between October 2006 and September 2011 and identified through the state birth defects registry in Massachusetts, North Carolina and New York (not including New York City). Infants with clubfoot associated with neural tube defects, Potter syndrome, or genetically inherited syndromes were not eligible for inclusion. The registries provided the study with contact information for cases. Mothers of study subjects were mailed an introductory letter and consent form, and then were called by telephone and invited to be in the study. Mothers who agreed to participate underwent a computer assisted telephone interview by one of the study nurses within 12 months after delivery. Medical records were obtained for those cases whose mothers provided consent. The institutional review boards at Boston University and the state health departments in Massachusetts, North Carolina, and New York approved the study protocol.

The diagnosis of clubfoot was confirmed by medical record, utilizing the orthopaedic record when available. In patients where a medical record was not available, the report by the mother that her child was treated with at least three casts was considered confirmation of the clubfoot diagnosis. Records that were in question were reviewed by a pediatric orthopaedic surgeon (STM) for inclusion in the study. Socioeconomic and demographic data were obtained from the interview.

Questionnaire

Mothers of cases were asked a series of questions on the prenatal diagnosis of their infant's clubfoot. First mothers were asked if the diagnosis of clubfoot was made before birth (i.e. prenatally). Mothers were then asked how the diagnosis of clubfoot first made. Responses included prenatal ultrasound, examination at birth, examination at follow-up pediatrician visit, or other. If a mother responded that the diagnosis occurred by prenatal ultrasound, the

clubfoot was determined to be prenatally diagnosed. All other responses were considered to be postnatal diagnoses.

Data Analysis

For this analysis, data from control mothers were not included. Maternal socioeconomic and demographic factors were compared between for infants with and without a prenatally-diagnosis. Maternal factors that were assessed included race/ethnicity (white, black, Hispanic, other/unknown), age at conception (<20, 20–24, 25–29, 30–34, 35), education (<12 years, 12 years, > 12 years), and body mass index (BMI)(<18.5, 18.5–24.9, 25–29.9, 30+, missing). Household information included was number living in the household (1, 2, 3, 4, 5, missing), income (<\$20,000, \$20,000–\$49,000, \$50,000–\$79,000, \$80,000, missing), whether or not the mother was employed (yes, no), whether or not the father was employed (yes, no, missing) and marital status (married, single living with father, father not present, missing information). Family planning and birth factors included whether or not the pregnancy was intended (yes, no), number of previous liveborn children (none, 1, 2), if there were other birth defects present (no – isolated TEV, yes- multiple birth defects), and laterality of clubfoot (bilateral, unilateral). Finally state of residence (North Carolina, New York, Massachusetts) and urbanicity (rural, urban or missing) were included. Rural compared to urban residence was determined by using maternal zip codes and data from 2010 census data ¹⁰.

Individual factors were first analyzed separately by using logistic regression to produce crude odds ratios (ORs) and 95% confidence intervals (CIs). A multivariate analysis was then conducted to calculate ORs and 95% CI that adjusted for all other maternal factors simultaneously. Analyses were conducted using SAS 9.1 (SAS Institute Inc. Release version 9.1.3. 2000–2004; Cary, NC.:SAS Institute Inc.).

Results

A total of 1,323 cases were eligible and approached for the study. A total of 955 were interviewed (72.2%), 286 could not be contacted due to missing telephone numbers or non-response, and 82 (6.2%) refused participation in the study. Of the 955 interviewed cases, 677 cases had confirmed idiopathic clubfoot. One case mother was unsure about the timing of the diagnosis of clubfoot and was excluded, resulting in a total of 676 cases for this analysis. Complete inclusion and exclusion details have previously been reported ⁹. Overall detection of clubfoot prenatally was 62.3% (421/676) but this varied considerably by state ($p<0.001$ between MA and NC or NY): 81.1% in Massachusetts (154/190), 58.5% in New York (124/212), and 52.2% in North Carolina (143/274).

Several factors were associated with prenatal detection of clubfoot before multivariable adjustment (Table 1). The adjusted odds ratios that were statistically significant predictors of prenatal detection of clubfoot were maternal age at conception ≥ 35 years (OR: 3.54, 95% CI: 1.58, 7.96), study center located in Massachusetts (OR: 2.64, 95% CI: 1.53, 4.54), and bilateral clubfoot in the infant (OR: 1.90, 95% CI: 1.28, 2.80); Maternal race/ethnicity of non-Hispanic black was a negative predictor (OR: 0.49, 95% CI: 0.26, 0.93). The presence

of another birth defect also increased the odds of prenatal detection, though due to small numbers in this group the lower bound of the CI includes 1 (OR: 2.61, 95% CI: 0.85, 8.01).

Discussion

We found a significant increase in prenatal detection of clubfoot in mothers at or over the age of 35 years, for those residing in the state of Massachusetts, and for infants with bilateral clubfoot, as well as a decrease in prenatal detection of clubfoot among non-Hispanic black mothers. The ability to detect clubfoot prenatally is of great importance as it offers the opportunity for families to educate themselves on treatment and prognosis of clubfoot. Families expecting a child known likely to have a clubfoot may pursue formal counseling with someone who specializes in clubfoot treatment^{6,7,11,12} or may pursue education in less formal venues. In fact, one study found that clubfoot informational talks with mothers moderated some of the negative thoughts and behaviors that harmed the mother-child bonding⁸. In addition, early detection of clubfoot can also allow families to establish their social network of support to help them cope with the diagnosis and has been shown to improve the mother's reaction to the child's clubfoot malformation^{8,13}. Prenatal detection also avoids the "surprise" of a congenital birth defect at the time of delivery, which has been shown to impair infant bonding⁸. Furthermore, most mothers have reported that they want to know about the diagnosis of clubfoot prenatally³.

In addition to familial benefits, prenatal detection has other advantages. For example, when a clubfoot is suspected prenatally, a thorough sonographic search for associated anomalies is appropriate, and is typically undertaken once the clubfoot is found^{1,7,14,15}. Prenatal detection of clubfoot can also serve as an indicator of overall prenatal care and access to care and appropriate diagnosis and screening for other more severe birth defects.

A previous study by Radler et al³ found that the rate of prenatal detection of clubfoot was 60% in the Baltimore area, which is similar to the rate we identified in North Carolina (52%) and New York (59%). The higher rate of prenatal detection in Massachusetts (81%) may have been due to the universal health care available in the state. In April 2006, the state of Massachusetts passed legislation that achieved near universal health care coverage resulting in improved access to care for working-age adults and decreased out of pocket costs¹⁶. A study by Long and Stockley¹⁷ compared the impact of recent health care reform in the states of New York and Massachusetts and found the near-universal health care coverage reforms in Massachusetts were much more successful in improving access to care. The current study was conducted after Massachusetts enacted universal health care coverage, and thus most all of the residents of this state had health care coverage; though, we did observe a lower detection rate in 2006 and 2007 (73% for both years), indicating that the reform may have taken some time to show an increased benefit. The universal care in Massachusetts may explain the observed difference in prenatal detection between states, as mothers in Massachusetts would have had improved access to prenatal care.

Older mothers are often screened more closely due to their higher risk of congenital anomalies. This additional attention to older women may explain why prenatal detection in older women was higher. Our findings of greater detection in cases with other major

malformation likely reflects a more thorough fetal survey if one anomaly is identified by ultrasound. Similarly, when two feet are affected with clubfoot, the chance of detection is greater. Our rate of infants with multiple major congenital anomalies was lower than previous studies, due to exclusion of clubfoot considered to be secondary to other anomalies (neural tube defects, Potter syndrome, and known inherited or chromosomal syndromes) in the initial case-control study because its focus was on etiologic risk factors. Prenatal detection of clubfoot can also be an indicator of overall prenatal detection of congenital malformations, most of which are much more serious or even lethal and may indicate for a serious discussion about the viability of the unborn fetus. Thus, the patterns observed here for prenatal detection of clubfoot may apply to prenatal detection of birth defects overall, with greater implications.

The possibility of participation bias in the study is worth considering. Differential participation of various race/ethnicities could certainly be a concern, particularly with the finding of lower prenatal detection in the black non-Hispanic population. However, when the distribution of maternal race/ethnicity in our study was compared to the population from which they came, there were no differences found,⁹ suggesting that such bias was unlikely.

In summary, we found the socioeconomic factors that had the greatest impact on the prenatal detection of clubfoot were mothers at or over the age of 35 years, residence in the state of Massachusetts, the presence of a bilateral clubfoot in the infant, and a negative impact in mother who were non-Hispanic black race.

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References

1. Offerdal K, Jebens N, Blaas HGK, Eik-Nes SH. Prenatal ultrasound detection of talipes equinovarus in a non-selected population of 49 314 deliveries in Norway. *Ultrasound Obstet Gynecol.* 2007 Nov; 30(6):838–44. [PubMed: 17787031]
2. Keret D, Bronshtein M, Weintraub S, Wientraub S. Prenatal diagnosis of musculoskeletal anomalies. *Clin Orthop Relat Res.* 2005 May;(434):8–15. [PubMed: 15864026]
3. Radler C, Myers AK, Burghardt RD, Arrabal PP, Herzenberg JE, Grill F. Maternal attitudes towards prenatal diagnosis of idiopathic clubfoot. *Ultrasound Obstet Gynecol.* 2011 Jun; 37(6):658–62. [PubMed: 21229570]
4. Keret D, Ezra E, Lokiec F, Hayek S, Segev E, Wientraub S. Efficacy of prenatal ultrasonography in confirmed club foot. *The Journal of Bone and Joint Surgery [Internet].* 2002 Sep 1; 84(7):1015–9. Available from: <http://eutils.ncbi.nlm.nih.gov/entrez/eutils/elink.fcgi?dbfrom=pubmed&id=12358364&retmode=ref&cmd=prlinks>.
5. Mercé LT, Barco MJ, Bau S. Three-dimensional volume sonographic study of fetal anatomy: intraobserver reproducibility and effect of examiner experience. *J Ultrasound Med.* 2008 Jul; 27(7):1053–63. [PubMed: 18577669]
6. Mahan ST, Kasser JR. Prenatal Ultrasound for Diagnosis of Orthopaedic Conditions. *Journal of Pediatric Orthopaedics.* 2010 Mar; 30(2 supplement):S35–9.

7. Treadwell MC, Stanitski CL, King M. Prenatal sonographic diagnosis of clubfoot: implications for patient counseling. *Journal of Pediatric Orthopaedics*. 1999 Jan; 19(1):8–10. [PubMed: 9890278]
8. Coppola G, Costantini A, Tedone R, Pasquale S, Elia L, Foschino Barbaro M, et al. The Impact of the Baby's Congenital Malformation on the Mother's Psychological Well-Being: An Empirical Contribution on the Clubfoot. *J Pediatr Orthop*. 2012 Jul; 32(5):521–6. [PubMed: 22706470]
9. Werler MM, Yazdy MM, Mitchell AA, Meyer RE, Druschel CM, Anderka M, et al. Descriptive epidemiology of idiopathic clubfoot. *Am J Med Genet A*. 2013 Jul; 161A(7):1569–78. [PubMed: 23686911]
10. 2010 Urban and Rural Classification - Geography - US Census Bureau [Internet]. [census.gov](http://www.census.gov). [cited 2013 Mar 1]. Available from: <http://www.census.gov/geo/reference/urban-rural-2010.html>
11. Burgan HE, Furness ME, Foster BK. Prenatal ultrasound diagnosis of clubfoot. *Journal of Pediatric Orthopaedics*. 1999 Jan; 19(1):11–3. [PubMed: 9890279]
12. Glotzbecker MP, Estroff JA, Spencer SA, Bosley JC, Parad RB, Kasser JR, et al. Prenatally diagnosed clubfeet: comparing ultrasonographic severity with objective clinical outcomes. *J Pediatr Orthop*. 2010 Sep; 30(6):606–11. [PubMed: 20733428]
13. Crnic KA, Greenberg MT, Ragozin AS, Robinson NM, Basham RB. Effects of stress and social support on mothers and premature and full-term infants. *Child Dev*. 1983 Feb; 54(1):209–17. [PubMed: 6831987]
14. Canto MJ, Cano S, Palau J, Ojeda F. Prenatal diagnosis of clubfoot in low-risk population: associated anomalies and long-term outcome. *Prenat Diagn*. 2008 Apr; 28(4):343–6. [PubMed: 18382991]
15. Sharma R, Stone S, Alzouebi A, Hamoda H, Kumar S. Perinatal outcome of prenatally diagnosed congenital talipes equinovarus. *Prenat Diagn*. 2011 Feb; 31(2):142–5. [PubMed: 21268031]
16. Long SK. On the road to universal coverage: impacts of reform in massachusetts at one year. *Health Aff (Millwood)*. 2008 Jul; 27(4):w270–84. [PubMed: 18522948]
17. Long SK, Stockley K. The impacts of state health reform initiatives on adults in New York and Massachusetts. *Health Serv Res*. 2011 Feb; 46(1 Pt 2):365–87. [PubMed: 21091471]

What's already known about this topic? What does this study add?

- Clubfoot is detected prenatally 60–80% of the time.
- We assessed multiple sociodemographic variables in a large clubfoot population and found several factors influenced prenatal detection of clubfoot:
 - Negative influences
 - ◆ Age<35 years
 - ◆ Black non-Hispanic race
 - Positive influences
 - ◆ Residence in Massachusetts
 - ◆ Another birth defect present
 - ◆ Finding of bilateral clubfoot
- Prenatal screening often detects clubfoot; this may not be available to some groups of women who should be targeted for prenatal care interventions.

Crude and Adjusted Odds Ratios (ORs) for all variables. The adjusted variables are adjusted for all other variables using multivariate analysis. CI denotes confidence interval.

Table 1

	Total	Prenatal Diagnosis		Crude ORs (95% CI)	Adjusted ORs (95% CI)*
		No	Yes		
	N (%)	N (%)	N (%)		
Total	676 (100)	255 (100)	421 (100)		
Maternal race/ethnicity					
White non-Hispanic	491 (72.6)	170 (66.7)	321 (76.2)	Reference	Reference
Black non-Hispanic	82 (12.1)	42 (16.5)	40 (9.5)	0.50 (0.32, 0.81)	0.49 (0.26, 0.93)
Hispanic	76 (11.2)	33 (12.9)	43 (10.2)	0.69 (0.42, 1.13)	0.83 (0.41, 1.71)
Other/Unknown	27 (4.0)	10 (3.9)	17 (4)	0.90 (0.40, 2.01)	0.50 (0.18, 1.37)
Maternal age at conception					
<20	50 (7.4)	24 (9.4)	26 (6.2)	0.94 (0.50, 1.78)	2.21 (0.87, 5.57)
20-24	155 (22.9)	72 (28.2)	83 (19.7)	Reference	Reference
25-29	200 (29.6)	81 (31.8)	119 (28.3)	1.27 (0.83, 1.94)	1.11 (0.62, 1.98)
30-34	165 (24.4)	59 (23.1)	106 (25.2)	1.56 (1.00, 2.44)	1.05 (0.54, 2.03)
35	106 (15.7)	19 (7.5)	87 (20.7)	3.97 (2.21, 7.15)	3.54 (1.58, 7.96)
Maternal education					
< 12 years	87 (12.9)	47 (18.4)	40 (9.5)	Reference	Reference
12 years	171 (25.3)	68 (26.7)	103 (24.5)	1.78 (1.06, 3.00)	1.63 (0.79, 3.37)
> 12 years	418 (61.8)	140 (54.9)	278 (66)	2.33 (1.46, 3.73)	1.41 (0.68, 2.95)
Number in household					
1	18 (2.7)	9 (3.5)	9 (2.1)	Reference	Reference
2	230 (34.0)	82 (32.2)	148 (35.2)	1.81 (0.69, 4.73)	1.98 (0.59, 6.66)
3	213 (31.5)	75 (29.4)	138 (32.8)	1.84 (0.70, 4.83)	1.42 (0.37, 5.50)
4	123 (18.2)	41 (16.1)	82 (19.5)	2.00 (0.74, 5.42)	1.95 (0.49, 7.77)
5	90 (13.3)	48 (18.8)	42 (10.0)	0.88 (0.32, 2.41)	0.94 (0.22, 3.95)
Missing	2 (0.3)	0 (0.0)	2 (0.5)		

	Total	Prenatal Diagnosis		Crude ORs (95% CI)	Adjusted ORs (95% CI)*
		No	Yes		
	N (%)	N (%)	N (%)		
Income					
< \$20,000	126 (18.6)	57 (22.4)	69 (16.4)	Reference	Reference
\$20,000–\$49,999	173 (25.6)	76 (29.8)	97 (23)	1.05 (0.66, 1.67)	1.12 (0.61, 2.03)
\$50,000–\$79,999	97 (14.3)	38 (14.9)	59 (14)	1.28 (0.75, 2.20)	1.13 (0.53, 2.41)
\$80,000	181 (26.8)	40 (15.7)	141 (33.5)	2.91 (1.77, 4.78)	2.04 (0.92, 4.53)
Missing	99 (14.6)	44 (17.3)	55 (13.1)		
Maternal Residence					
Rural	190 (28.1)	87 (34.1)	103 (24.5)	Reference	Reference
Urban	485 (71.7)	168 (65.9)	317 (75.3)	1.59 (1.13, 2.24)	1.29 (0.84, 1.99)
Missing	1 (0.1)	0 (0.0)	1 (0.2)		
Marital Status					
Married	392 (58.0)	128 (50.2)	264 (62.7)	Reference	Reference
Single lives w/Father	184 (27.2)	81 (31.8)	103 (24.5)	0.62 (0.43, 0.88)	1.05 (0.62, 1.80)
Father not present	99 (14.6)	46 (18)	53 (12.6)	0.56 (0.36, 0.87)	1.09 (0.49, 2.41)
Missing	1 (0.1)	0 (0.0)	1 (0.2)		
Pregnancy intention					
No	316 (46.7)	137 (53.7)	179 (42.5)	Reference	Reference
Yes	360 (53.3)	118 (46.3)	242 (57.5)	1.57 (1.15, 2.15)	1.12 (0.71, 1.76)
Number of previous liveborn infants					
None	326 (48.2)	133 (52.2)	193 (45.8)	Reference	Reference
1	201 (29.7)	64 (25.1)	137 (32.5)	1.48 (1.02, 2.14)	1.59 (0.76, 3.32)
2	149 (22.0)	58 (22.7)	91 (21.6)	1.08 (0.73, 1.61)	1.26 (0.57, 2.78)
Mother employed					
Yes	469 (69.4)	163 (63.9)	306 (72.7)	1.50 (1.08, 2.10)	1.11 (0.69, 1.77)
No	207 (30.6)	92 (36.1)	115 (27.3)	Reference	Reference

	Total	Prenatal Diagnosis		Crude ORs (95% CI)	Adjusted ORs (95% CI)*
		No	Yes		
	N (%)	N (%)	N (%)		
Father employed					
Yes	611 (90.4)	231 (90.6)	380 (90.3)	1.01 (0.58, 1.75)	0.6 (0.28, 1.30)
No	58 (8.6)	22 (8.6)	36 (8.6)	Reference	Reference
Missing	7 (1.0)	2 (0.8)	5 (1.2)		
Study center					
NC	274 (40.5)	131 (51.4)	143 (34)	Reference	Reference
NY	212 (31.4)	88 (34.5)	124 (29.5)	1.29 (0.90, 1.85)	1.13 (0.72, 1.77)
MA	190 (28.1)	36 (14.1)	154 (36.6)	3.92 (2.54, 6.05)	2.64 (1.53, 4.54)
Other birth defects present					
No, isolated	645 (95.4)	248 (97.3)	397 (94.3)	Reference	Reference
Yes, multiple BDs	31 (4.6)	7 (2.7)	24 (5.7)	2.14 (0.91, 5.05)	2.61 (0.85, 8.01)
Laterality					
Bilateral	338 (50.0)	99 (38.9)	239 (56.8)	2.05 (1.50, 2.82)	1.90 (1.28, 2.80)
Unilateral	335 (49.6)	154 (60.4)	181 (43.0)	Reference	Reference
Missing	3 (0.4)	2 (0.8)	1 (0.2)		
Body Mass Index					
<18.5	25 (3.7)	7 (2.7)	18 (4.3)	1.41 (0.57, 3.49)	1.36 (0.49, 3.78)
18.5–24.9	310 (45.9)	110 (43.1)	200 (47.5)	Reference	Reference
25–29.9	183 (27.1)	74 (29.0)	109 (25.9)	0.81 (0.56, 1.18)	0.76 (0.48, 1.21)
30+	139 (20.6)	56 (22.0)	83 (19.7)	0.82 (0.54, 1.23)	0.98 (0.58, 1.64)
Missing	19 (2.8)	8 (3.1)	11 (2.6)		

* Adjusted for all other variables