

Patient factors associated with delayed diagnosis of developmental dysplasia of the hip

A. W. Lindberg^{1,2}
V. Bompadre¹
E. K. Satchell³
A. C. R. Larson⁴
K. K. White^{1,2}

Abstract

Purpose Early detection and intervention for developmental dysplasia of the hip (DDH) is important for normal hip development. Previous studies have shown disparities in access to paediatric specialty care among different racial and socioeconomic backgrounds. This study aims to identify whether these factors are related to timely referral for infants with DDH.

Methods A retrospective cohort study of patients seen and treated for DDH between July 2006 and June 2011 at a single institution were reviewed. The patients were divided into early-presenting (seen before six months of age) and late-presenting patients (seen at six months of age or later).

Results A total of 457 patients met the eligibility criteria. There were 378 early and 79 late presentations. Late presentations were significantly more likely to be vertex at birth (85% vs 41%, $p < 0.001$). Bivariate analysis also demonstrated that late presentations were more likely to be non-white (65% vs 45%, $p = 0.004$), non-English speaking (20% vs 8%, $p = 0.003$), from lower income areas (\$70 769 vs \$61 591, $p < 0.001$) and hold public insurance (25%, $p = 0.001$). However, a logistic multiple regression analysis showed that only vertex birth presentation ($p = 0.000$), absent family history

of DDH ($p = 0.047$) and affected right side ($p = 0.001$) were significantly associated with late presentation.

Conclusion Despite screening algorithms to facilitate early diagnosis of infants with DDH, better research is needed to understand how different demographic and socioeconomic factors play into the delayed access to paediatric orthopaedic care for DDH so that we may ultimately improve rates of early treatment.

Cite this article: Lindberg AW, Bompadre V, Satchell EK, Larson ACR, White KK. Patient factors associated with delayed diagnosis of developmental dysplasia of the hip. *J Child Orthop* 2017;11:223-228. DOI 10.1302/1863-2548.11.160228

Keywords: DDH; access to care; late presentation

Introduction

The incidence of hip dislocation at birth has been reported as one in 1000 births, and the incidence of hip subluxation or dysplasia reported as ten in 1000 births.¹ Proper geometric development of the hip joint in childhood is dependent on the presence of a spherical femoral head positioned within the acetabulum. Natural history studies have shown that untreated subluxation leads to early degenerative joint disease in the hip and untreated bilateral dislocations can lead to excessive lumbar lordosis and chronic low-back symptoms.² Early detection and treatment of developmental dysplasia of the hip (DDH) is critical for the best chances of obtaining a well-functioning, pain-free hip joint well into adulthood, and research has shown that a child's age at initial reduction is correlated to radiographic outcomes.³

Previous studies have shown that many disparities exist in paediatric medical and dental care among different racial/ethnic backgrounds. In a study looking at the National Survey of Children's Health data, Latinos, Asian/Pacific Islanders and Native Americans were found to have significantly higher odds of encountering difficulty obtaining specialty care.^{4,5} The same authors also found discrepancies in medical care between children in households that were primarily English-speaking and those which were non-English-speaking. Children from non-English-speaking households were reported to have increased problems obtaining specialty care (40% vs 23% in English-speaking households).^{6,7}

¹ Seattle Children's Hospital – Department of Orthopaedics and Sports Medicine, 4800 Sandpoint Way NE M/S MB.10.620, Seattle, WA 98105, USA

² University of Washington – Department of Orthopaedics and Sports Medicine, Seattle, Washington, USA

³ University of Pennsylvania Perelman School of Medicine – Department of Psychiatry, Center for Neuromodulation in Depression and Stress, 3400 Civic Center Blvd, Philadelphia, Pennsylvania, USA

⁴ Cincinnati Children's Hospital Medical Center – Division of Pediatric Orthopaedics, Department of Orthopaedic Surgery, 3333 Burnet Ave, Cincinnati, Ohio 45229, USA

Correspondence should be sent to: Dr A. W. Lindberg, Seattle Children's Hospital, 4800 Sandpoint Way NE M/S MB.10.620, Seattle, WA 98105, USA.

E-mail: antoinette.lindberg@seattlechildrens.org

Medicaid health insurance coverage has also been shown to affect access to paediatric specialty care. A study conducted in Chicago, United States, evaluating eight different paediatric subspecialties, including orthopaedics, found disparities across the board for private *versus* public insurance. Specifically for orthopaedics, of 40 offices contacted, 98% would see a patient with private insurance, but only 20% would see the same condition if the patient had public insurance.⁸ Similar studies have uncovered similar disparities in other cities.^{9,10} However, these studies primarily focus on paediatric fracture care, and to our best knowledge, there are no similar studies addressing insurance status and care of orthopaedic issues such as DDH.

DDH is a fairly common paediatric orthopaedic disorder, and early detection and treatment will continue to be a key factor in reducing both lifetime healthcare costs as well as long-term patient disability. The goal of our current study was to evaluate the relative proportions of different racial, ethnic and language backgrounds between early-presenting and late-presenting DDH patients and assess differences in socioeconomic status using median income and insurance indicators. We suspected that the late-presenting patients would be disproportionately non-white, Hispanic, non-English speakers and come from poorer communities and be more likely to be covered by public insurance, such as Medicaid. As we continue to see and treat a multitude of cases of late-presenting DDH, this study aims to identify more clearly some factors associated with late diagnosis so that these areas can be targeted for better educational and diagnostic outreach.

Patients and methods

We conducted a retrospective chart review to evaluate all patients presenting for the first time to the orthopaedics clinic at a tertiary paediatric hospital with the diagnosis of DDH during a five-year period from July 2006 to June 2011. Patients were identified using ICD-9 codes from billing records after Institutional Review Board (IRB) approval. For this type of study, formal consent is not required. Charts of patients identified were individually screened to ensure they met the inclusion criteria of a diagnosis of hip dysplasia and/or dislocation and had their initial clinic visit during our study timeframe. Exclusion criteria disqualified patients whose treatment (orthotic or surgical) was initiated at an outside facility, patients who had abnormal exam or ultrasound findings in the early neonatal period but normalised without additional treatment within the first 12 weeks of life, and hip dysplasia/dislocation associated with other disorders (i.e. myelomeningocele, cerebral palsy, arthrogryposis, skeletal dysplasias and other teratologic causes). Patients were divided into two groups

based on age at initial presentation to the clinic. Early presentation DDH was defined as children presenting prior to age six months, while late-presenting DDH was defined as presentation at six months of age and older. The choice of the six-month age cutoff was selected as the failure rate of Pavlik harness treatment exceeds 50% after that age.¹¹

Study variables were collected from medical charts as well as hospital demographics data. In our institution, language spoken at home is consistently documented. Race was categorised as 'white', 'non-white' and 'refused to indicate' based on information provided by the families. Ethnicity was similarly recorded as 'non-Hispanic', 'Hispanic' and 'refused to indicate'. Insurance was categorised as 'public' for Medicaid and its affiliated programs and the remainder of insurance plans were considered 'private'. Patient's home zip codes were used to calculate distance from our institution using readily accessible online distance calculators. Zip codes were also used in conjunction with publicly available United States Census data through the American FactFinder website (factfinder2.census.gov) to search mean household income. Other data points with known association with DDH were collected from the medical record, such as gender, family history and birth presentation (vertex vs breech).

Statistical analysis

For categorical variables, we give the relative frequencies (percentages). For the quantitative variables, we provide the estimated means and standard deviations. We also provide bivariate tests of associations between each key characteristic and late presentation. We used *t*-tests for unequal variances for the quantitative characteristics and Chi-square tests for the categorical characteristics. In the first model, we ran a list-wise logistic regression analysis. However, with 32% of missing data among the predictors, it seemed reasonable to use multiple imputation in a second model. Statistical significance was set at $p \leq 0.05$ and all confidence intervals at 95%.

Results

A total of 457 (82.9% female, 17.1% male) patients were included in the study. There were 378 (83%) early presentations and 79 (17%) late presentations. Among the early presentations, 59% were white, 20% were non-white and 21% refused to indicate. Bivariate analysis determined the average income for early presentations was significantly higher than the average income for late presentations (\$70 769 vs \$61 591, $p < 0.001$). Non-whites were more likely to present late than whites (65% vs 45%, respectively, $p = 0.004$). Non-English speakers were significantly more likely to present late than early (20% vs 8%, $p = 0.003$). Late presentations were significantly more likely to

be vertex than early presenters (85% vs 41%, $p < 0.001$). Late presenters were also more likely to have public insurance than private (41% vs 21%, $p < 0.001$). Patients with bilateral DDH (57%) were less likely to present late than those with unilateral DDH ($p < 0.001$). Ethnicity and family history were not significant predictors. Table 1 shows these results.

We used these key characteristics in a list-wise logistic regression model to determine the effects of these variables on late or early presentation. The list-wise analysis identified birth presentation ($p < 0.001$) and family history ($p = 0.005$) as being significantly associated (Table 2). However, because there was a high percentage of data missing (32%), we used multiple imputation to minimise the bias. In the second model, after accounting for missing data, birth presentation, side of presentation and family history had a significant effect on late presentation. The odds of presenting late with breech birth was 0.16 times the odds of those with vertex presentation after accounting for race, ethnicity, insurance type, income, state of residency, family history and side of presentation (odds

ratio (OR) = 0.16, 95% confidence interval (CI) = 0.07-0.36; $p = 0.000$). The odds of late presentation for children with a family history of DDH was 0.48 times the odds for those without history (OR = 0.48, 95% CI = 0.23-0.99; $p = 0.005$). The odds of late presentation for children with left affected side was 0.26 times the odds of those with right side (OR = 0.26, 95% CI = 0.12-0.58; $p = 0.001$) (Table 3).

Discussion

The current guidelines from the American Academy of Pediatrics outline their recommendations for screening of all newborns' hips with physical examination and an algorithm for repeat examination, orthopaedic referral and radiographic evaluation.¹² Recently, the American Academy of Orthopedic Surgeons recommended performing an imaging study before six months of age in infants with one or more of the following risk factors: breech presentation, family history or history of clinical instability. They also highlighted the lack of evidence to support universal ultrasound screening of newborns.¹³

Table 1. Bivariate analysis of key characteristics.

Characteristic	Early (%) n = 378	Late (%) n = 79	p value
Income (median)	\$70 769	\$61 591	< 0.001*
Race			0.004*
White	59	48	
Other	20	37	
Refused	21	15	
Ethnicity			0.147
Non-Hispanic	68	50	
Hispanic	12	13	
Not indicated	19	8	
English language			0.003*
No	8	20	
Yes	92	80	
Birth presentation			< 0.001*
Vertex	41	85	
Breech	59	15	
Side			< 0.001*
Right	10	24	
Left	28	41	
Bilateral	61	35	
Insurance type			< 0.001*
Private	79	59	
Public	21	41	
Family history			0.236
No	77	83	
Yes	23	17	

* significant at $p < 0.05$

Table 2. List-wise logistic regression.

Characteristic	OR	SE	95% Confidence interval	p value	
Income	0.85	0.17	0.58	1.27	0.433
Race					0.400
White	(ref.)	1.00			
Other	1.33	0.65	0.51	3.49	
Refused	0.31	0.35	0.03	2.84	
Ethnicity					0.690
Non-Hispanic	(ref.)	1.00			
Hispanic	0.65	0.41	0.19	2.24	
Not indicated	1.72	1.97	0.18	16.1	
English language					0.685
No	(ref.)	1.00			
Yes	1.31	0.88	0.35	4.87	
Birth presentation					0.001*
Vertex	(ref.)	1.00			
Breech	0.06	0.03	0.02	0.16	
Side presentation					0.159
Right	(ref.)	1.00			
Left	0.64	0.35	0.22	1.86	
Bilateral	0.39	0.20	0.14	1.09	
Insurance type					0.153
Private	(ref.)	1.00			
Public	1.94	0.90	0.78	4.83	
Family history					0.005*
No	(ref.)	1.00			
Yes	0.27	0.13	0.11	0.67	

OR, odds ratio; SE, standard error

* significant at $p < 0.05$

Table 3. Logistic regression using multiple imputations technique.

Characteristic	OR	SE	95% CI	p value
Birth presentation				0.000*
Vertex	(ref.)	1.00		
Breech		0.16	0.07 0.36	
Side				0.001*
Right	(ref.)	1.00		
Left		0.26	0.12 0.58	
Bilateral		0.12	0.14 1.09	
Family history				0.005*
No	(ref.)	1.00		
Yes		0.48	0.23 0.99	

OR, odds ratio; SE, standard error

* significant at $p < 0.05$

In other countries, particularly in Europe and Australia, universal ultrasound screening is done to try to capture all cases of DDH in infancy, but the debate between universal *versus* selective ultrasound screening is ongoing.^{14,15} A recent Cochrane review found that neither of the ultrasound strategies have been demonstrated to improve clinical outcomes including late diagnosed DDH and surgery.¹⁶ Further conflicting information regarding the optimal screening procedure for DDH has arisen after the United States Preventive Services Task Force found insufficient evidence to recommend routine DDH screening, including physical examination.¹⁷

More recently, authors using decision-analysis modeling concluded that for the goal of a non-arthritis hip at age 60 years, the optimal screening strategy was to screen all newborns with physical examination and use ultrasonography selectively for patients with positive physical exam, breech presentation or family history.¹⁸ It is difficult to ascertain the 'normal' rate of missed DDH diagnoses in neonates, but a large randomised controlled trial in Norway looking at ultrasonographic screening found that the prevalence of late subluxation/dislocation was 0.3 in 1000 for universal ultrasound screening, 0.7 in 1000 for selective ultrasound screening and 1.3 in 1000 for no ultrasound screening.¹⁹ In our study, we found that breech infants and patients with known positive family history of DDH were significantly less likely to present late with DDH even after factoring in other variables. These findings can be interpreted as a sign that our current screening algorithms are working for these known high-risk populations since these children are over-represented in the early presentation group. Similarly, Azzopardi et al reported that breech presentation and delivery by Caesarean section were protective for late diagnosed DDH in Australia.²⁰

Race and language differences were detected in our study group between early and late DDH groups in bivariate analyses, but these effects were no longer significant with multivariate analysis, suggesting that the

relationships are more complex. Non-white race and foreign language-speaking household are often correlated with each other, likely decreasing the chances that either factor would be an independent predictive factor in multivariate analysis. Additionally, reports of relationships between family income and access to paediatric orthopaedic care are scarce, most likely due to the difficulty in obtaining reliable data through chart review. In our study, we used publicly available census data based on home zip codes as a proxy for income since we do not routinely collect household income data directly. Public health insurance and family income are inextricably linked factors. Although our method of gauging family income by using census data is imprecise, the associations between public insurance and income to late DDH presentation seen in our current study suggest that these patient factors are relevant and need further investigation.

Our inclusion of five years of new DDH patients in this study design did allow for the capture of a relative large number of late-presenting cases (79 separate patients) which accounted for one in every six new cases. In this study, we found, by bivariate analysis, that there are associations between demographic characteristics and late presentation. However, they should be interpreted with caution as they do not control for confounding factors. More compelling results come from the logistic regression model using multiple imputations, which considers the key characteristics simultaneously (Table 3). Vertex birth presentation, side of presentation (right) and absence of family history seem to be the predictors of late presentation. One limitation of our study is the reliance on a retrospective design which limited our available data, especially with demographic information. In multiple instances information on patients' race, ethnicity and home language were not reported by the families or not recorded at the point of registration. Almost all of the 32% missing data elements were related to one of these demographic characteristics, but we were able to use the statistical technique of multiple imputation in order to perform multivariate analysis.

Although ethnicity, race, median income and language are not independent risk factors for late presentation once factored into multivariate analysis, our data suggest they are still relevant factors to consider for DDH screening and diagnosis. Further studies may help support promotion of screening outreach to these populations that may not be physiologically at risk of DDH like breech patients or patients with a family history of DDH, but may be demographically at risk of having their DDH missed during routine screening. Effective screening, however, is highly dependent on the ability and confidence of the primary care provider (PCP) in conducting the hip examination and following the proposed algorithms. Multiple studies have demonstrated that education in musculoskeletal issues is

suboptimal in both medical school and primary care residency curricula.²¹⁻²⁴ Furthermore, non-physicians have been found to have a greater propensity for providing primary care to underserved populations.²⁵ Increased educational outreach to cover all types of paediatric primary care providers, including advanced practice providers such as physician assistants (PAs) and nurse practitioners (NPs), may be a straightforward target for improving early diagnosis of DDH.

More research is needed to evaluate the relationships of different factors affecting access to care so that better outreach programs can be deployed. Once we can better understand how different patient factors play into the reliability of access to care for DDH, we will be able to target our outreach efforts more effectively and can also reconsider our algorithms for physical examination and ultrasound screening programs.

Received 11 August 2016; accepted after revision 1 March 2017.

COMPLIANCE WITH ETHICAL STANDARDS

FUNDING STATEMENT

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

OA LICENCE TEXT

This article is distributed under the terms of the Creative Commons Attribution-Non Commercial 4.0 International (CC BY-NC 4.0) licence (<https://creativecommons.org/licenses/by-nc/4.0/>) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed.

ETHICAL STATEMENT

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent was not required to be obtained as this study was a retrospective review of medical records.

Source of Funding: Pediatric Pilot Grant. Center for Clinical and Translational Research. Seattle Children's Hospital

ICMJE CONFLICT OF INTEREST STATEMENT

Dr White receives royalties for UpToDate and honoraria for consultancy in BioMarin Pharmaceuticals and reports grants from Seattle Children's Hospital during the conduct of the study. The other authors declare no conflicts of interest.

REFERENCES

- Guille JT, Pizzutillo PD, MacEwen GD. Development dysplasia of the hip from birth to six months. *J Am Acad Orthop Surg* 2000;8:232-242.
- Weinstein SL, Mubarak SJ, Wenger DR. Developmental hip dysplasia and dislocation: part I. *Instr Course Lect* 2004;53:523-530.
- Lindstrom JR, Ponseti IV, Wenger DR. Acetabular development after reduction in congenital dislocation of the hip. *J Bone Joint Surg [Am]* 1979;61-A:112-118.
- Flores G, Tomany-Korman SC. Racial and ethnic disparities in medical and dental health, access to care, and use of services in US children. *Pediatrics* 2008;121:e286-e298.
- Flores G, Tomany-Korman SC. Erratum: Racial and ethnic disparities in medical and dental health, access to care, and use of services in US children. *Pediatrics* 2009;124:999-1000.
- Flores G, Tomany-Korman SC. The language spoken at home and disparities in medical and dental health, access to care, and use of services in US children. *Pediatrics* 2008;121:e1703-e1714.
- Flores G, Tomany-Korman SC. Erratum: The language spoken at home and disparities in medical and dental health, access to care, and use of services in US children. *Pediatrics* 2009;124:1265.
- Bisgaier J, Rhodes KV. Auditing access to specialty care for children with public insurance. *N Engl J Med* 2011;364:2324-2333.
- Iobst C, King W, Baitner A, et al. Access to care for children with fractures. *J Pediatr Orthop* 2010;30:244-247.
- Skaggs DL, Clemens SM, Vitale MG, Femino JD, Kay RM. Access to orthopedic care for children with medicaid versus private insurance in California. *Pediatrics* 2001;107:1405-1408.
- Weinstein SL. Developmental hip dysplasia and dislocation. In: Morrissy RT, Weinstein SL, ed. *Lovell and Winter's Pediatric Orthopaedics*. Philadelphia: Lippincott Williams and Wilkins, 2006:1007.
- No authors listed. Clinical practice guideline: early detection of developmental dysplasia of the hip. Committee on Quality Improvement, Subcommittee on Developmental Dysplasia of the Hip. American Academy of Pediatrics. *Pediatrics* 2000;105:896-905.
- Kishore M, Song K. AAOS clinical practice guideline: detection and nonoperative management of pediatric developmental dysplasia of the hip in infants up to six months of age. *J Am Acad Orthop* 2015;23:206-207.
- Portinaro NM, Pelillo F, Cerutti P. The role of ultrasonography in the diagnosis of developmental dysplasia of the hip. *J Pediatr Orthop* 2007;27:247-250.
- Laborie LB, Markestad TJ, Davidsen H, et al. Selective ultrasound screening for developmental hip dysplasia: effect on management and late detected cases. A prospective survey during 1991-2006. *Pediatr Radiol* 2014;44:410-424.
- Shorter D, Hong T, Osborn DA. Cochrane Review: screening programmes for developmental dysplasia of the hip in newborn infants. *Evid Based Child Health* 2013;8:11-54.
- Shipman SA, Helfand M, Moyer VA, Yawn BP. Screening for developmental dysplasia of the hip: a systematic literature review for the US Preventive Services Task Force. *Pediatrics* 2006;117:e557-e576.
- Mahan ST, Katz JN, Kim YJ. To screen or not to screen? A decision analysis of the utility of screening for developmental dysplasia of the hip. *J Bone Joint Surg [Am]* 2009;91-A:1705-1719.
- Rosendahl K, Markestad T, Lie RT. Ultrasound screening for developmental dysplasia of the hip in the neonate: the effect on treatment rate and prevalence of late cases. *Pediatrics* 1994;94:47-52.
- Azzopardi T, Van Essen P, Cundy PJ, Tucker G, Chan A. Late diagnosis of developmental dysplasia of the hip: an analysis of risk factors. *J Pediatr Orthop B* 2011;20:1-7.

21. **Lynch JR, Schmale GA, Schaad DC, Leopold SS.** Important demographic variables impact the musculoskeletal knowledge and confidence of academic primary care physicians. *J Bone Joint Surg [Am]* 2006;88-A:1589-1595.
22. **Haywood BL, Porter SL, Grana WA.** Assessment of musculoskeletal knowledge in primary care residents. *Am J Orthop (Belle Mead NJ)* 2006;35:273-275.
23. **Matzkin E, Smith EL, Freccero D, Richardson AB.** Adequacy of education in musculoskeletal medicine. *J Bone Joint Surg [Am]* 2005;87-A:310-314.
24. **Freedman KB, Bernstein J.** Educational deficiencies in musculoskeletal medicine. *J Bone Joint Surg [Am]* 2002;84-A:604-608.
25. **Grumbach K, Hart LG, Mertz E, Coffman J, Palazzo L.** Who is caring for the underserved? A comparison of primary care physicians and nonphysician clinicians in California and Washington. *Ann Fam Med* 2003;1:97-104.