Incidence, trends, and associated risks of developmental hip dysplasia in patients with Early Onset and Adolescent Idiopathic Scoliosis

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ARTICLE INFO

Keywords:

Early onset scoliosis
Adolescent idiopathic scoliosis
Degenerative dysplasia of the hip
Hip arthroplasty
Recurrent hip dislocations

ABSTRACT

Introduction: Early Onset and Adolescent Idiopathic Scoliosis, relatively common diagnoses (~3% general population), have been associated with developmental dysplasia of the hip (DDH); a more rare spectrum of anomalies related to the abnormal development of acetabulum, proximal femur, and hip joint. To the best of our knowledge, no high powered investigations have been performed in an attempt to assess incidence and associated risks of DDH in scoliosis patients.

Methods: The KID database was queried for ICD-9 codes from 2003 to 2012 pertaining to EOS (Congenital and Idiopathic < 10y/o) and AIS patients. Descriptive analysis assessed patient demographics and yearly trends in hip dysplasia rates. EOS and AIS patients with hip dysplasia were isolated, and incidence of hospital admissions for associated anomalies (osteonecrosis, osteoarthritis, recurrent hip dislocation, hip ankylosis) and hip arthroplasty (total + partial) were investigated. Univariate analysis of hip pathology determined significant predictors of hip arthroplasty. Binary logistic regression analysis was used to determine the relationship between these predictors.

Results: 111,827 scoliosis patients (EOS: 25,747; AIS: 77,183) were included. AIS patients were older (15.2 vs 4.3), more female (64.2% vs 52.1%), had a higher CCI (0.84 vs 0.64), and less racially diverse (all $p < 0.001$). The incidence of hip dysplasia was 1.4% for AIS patients and 3.9% for EOS patients ($p < 0.001$). Of the AIS ($n = 1073$) and EOS ($n = 1005$) patients with hip dysplasia, 0.3% ($p > 0.05$ between groups) developed hip osteonecrosis, 0% of patients were coded as having a hip labral tear, hip ankylosis, and 0.6% (EOS: 0.2%; AIS: 0.9%, $p = 0.025$) developed hip osteoarthritis. AIS patients were more likely to have recurrent hip dislocations (35.4% vs 17.0%, $p < 0.001$), and both groups had similar primary hip arthroplasty rates (6.7% vs 5.4%, $p = 0.118$) and revision hip arthroplasty rates (0% vs 0.4%, $p = 0.053$). Hip osteoarthritis (OR: 13.43[5.21–34.66], $p = < 0.001$) and older age (OR: 1.039[1.007–1.073], $p = 0.017$) were the only significant predictors of hip arthroplasty ($p = < .001$).

Conclusions: The incidence of hip dysplasia in EOS and AIS populations is higher than that of the general population. The rate of DDH was 3.9% and 1.8% for EOS and AIS, respectively. While the incidence of DDH is higher, associated anomalies of osteoarthritis, osteonecrosis, labral tears, and ankylosis appear to be a minimal risk for AIS and EOS patients with Hip Dysplasia.

1. Introduction

Early onset and adolescent scoliosis are three dimensional deformities of spinal curvature that, combined, occur at a relatively common

rate in the general population (~3%, respectively).^{1–3} Early onset scoliosis (EOS) presents before age 10 and encompasses Idiopathic, congenital, neuromuscular, and syndromic scoliosis.⁴ Adolescent Idiopathic Scoliosis (AIS) is the most common form of scoliosis, and

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<https://doi.org/10.1016/j.jor.2018.08.015>

Received 24 June 2018; Accepted 5 August 2018

Available online 15 August 2018

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presents between the age of 10 and skeletal maturity.^{3,5}

EOS, and more rarely AIS, have been associated with significant comorbidities such as neurologic, musculoskeletal, cardiopulmonary, and urogenital anomalies.^{2,6,7} Included under the umbrella of musculoskeletal anomalies is Developmental Dysplasia of the Hip (DDH), which is the range of deformities involving the growing hip. These deformities can include frank dislocation, subluxation and instability, or dysplasia of the femoral head and acetabulum.⁸ It has been previously established that EOS and AIS patients have an increased rate of DDH compared to the general population (6.4 per 100 live births vs. 5.1 per 1000 live births), however, there have not been any recent high powered investigations into the current rate of DDH in the EOS and AIS population.^{1,9,10}

Spine and hip pathology are related due to the pelvic attachment of the lumbosacral junction. Sagittal spine balance affects pelvic tilt at this point of attachment and thus pelvic tilt is able to function as a compensatory mechanism for sagittal malalignment.¹¹ Due to the nature of the pelvis, this change in tilt can lead to hip comorbidities existing with spinal deformities.^{11–13,17} Hip morbidities in the EOS and AIS populations, such as hip osteonecrosis, hip labral tear, hip ankylosis, hip osteoarthritis, etc, have been understudied to date.^{9,10,14,15} Therefore, this study aimed to determine the true rate of DDH in patients diagnosed with EOS and AIS and the proportion of those patients who had concomitant diseases of the hip.

2. Methods

2.1. Study design and data source

This study was a retrospective review of all inpatient entries in the Kids Inpatient Database (KID), a subset of The Agency for Healthcare Research and Quality's Healthcare Cost and Utilization Project (HCUP). The KID database is the largest all-payer pediatric (< 21 years at admission) inpatient database in the United States containing data for approximately 3 million discharges a year. Released every 3rd year, it can be used to identify, track, and analyze national trends in health care utilization, access, charges, quality, and outcomes. Discharge level weighted variables for the years 2003–2012 in our study.

2.2. Study population

Hospitalized patients with EOS and AIS were identified using International Classification of Disease, Ninth Revision, Clinical Modification (ICD-9CM) codes. The diagnosis codes used were congenital and idiopathic scoliosis (7370 73710 73720 73730 73739 7378 7385 75610 73731 7542 73732) from 2003, 2006, 2009, 2012. Patients with an age of < 21 y/o were included for analysis. Patients less than 10 years old were stratified into the EOS group, while those greater than or equal to 10 years old were stratified into the AIS group.

2.3. Variables of interest

EOS and AIS patient demographics were assessed for each year. Demographics consisted of age, gender (male or female), and race/ethnicity (black, Hispanic, white, Asian and Pacific Islanders, or others). We then assessed the overall and annual incidences of DDH with respect to EOS and AIS patients. The incidence of hospital admissions of various hip anomalies within our populations were also assessed using specified ICD-9 codes, which included osteonecrosis, osteoarthritis, recurrent hip dislocation, hip ankylosis and hip arthroplasty (71885, 71895, 7265, 73343, 73342, 7151, 7150 7152, 71515, 71525, 71535, 71595, 71509, 71995, 7159, 71835, 71855, 71875, 71825, 8151, 8140, 8152, 8153, 0070, 0071, 0072, 0073, 0074, 0075, 0076, 0077, 0085, 0086, 0087).

2.4. Statistical analysis

National estimates for annual EOS and AIS hospitalization incidence were quantified using KID weighted discharges. Descriptive, chi-square, and independent sample *t*-test analyses assessed frequencies and means of demographic and hip diagnosis variables amongst EOS and AIS patients. Univariate analysis of all coded hip pathologies in the KID database was performed. Binary logistic regression analysis of significant univariate hip pathologies was used to determine significant predictors of hip arthroplasty. All tests were two sided, and significance was set to a *p*-value of less than 0.05. All statistical analyses were performed utilizing IBM Statistical Package for the Social Sciences (SPSS) version 23.0132 (Armonk, NY: IBM).

3. Results

3.1. Demographics

111,827 scoliosis patients were included in the study. There were 25,747 EOS and 77,183 AIS patients. The average age of AIS patients was 15.2 years vs. 4.3 for EOS. 64.2% of AIS patients were female compared to 52.1% for EOS and had a higher Charlson Comorbidity Index (CCI) (0.84 vs 0.64). Racial demographics were 49.9% White, 13.9% Hispanic, 11.9% Black, 4.2% Other, 2.0% Asian or Pacific Islander, 0.5% Native American (Table 1).

3.2. Incidence of DDH and hip anomalies in EOS and AIS

1073 AIS (1.4%) and 1005 EOS (3.9%) patients presented with DDH. The annual incidence of AIS was 186 (2003), 206 (2006), 309 (2009), 372 (2012). The annual incidence of EOS was 153 (2003), 195 (2006), 299 (2009), and 357 (2012). The incidence of hospital admissions for patients diagnosed with osteonecrosis, osteoarthritis, recurrent hip dislocation, hip ankylosis and hip arthroplasty (total + partial) in the EOS and AIS populations was then calculated. We found 0.3% ($p > 0.05$ between groups) developed hip osteonecrosis, and 0.6% (EOS: 0.2%; AIS: 0.9%, $p = 0.025$) developed hip osteoarthritis. There were 0% of patients coded as having a hip labral tear or hip ankylosis, AIS patients were more likely to have recurrent hip dislocations (35.4% vs 17.0%, OR: 2.67, $p < 0.001$), and both groups had similar primary hip arthroplasty rates (6.7% vs 5.4%, $p = 0.118$) and revision hip arthroplasty rates (0% vs 0.4%, $p = 0.053$) (Table 2).

3.3. Predictors of hip arthroplasty

Univariate regression analysis was performed to find the significant predictors of hip arthroplasty for all hip pathologies coded in the KID database. Significant predictors were identified as having $P < 0.05$. We identified age ($p = .003$, 1.048[1.016–1.081]), and osteoarthritis

Table 1
Demographics of EOS and AIS patients isolated from KID database.

Demographic Variable	Incidence (%) or Mean
Sample Size (N)	111,827
EOS Size	25,747
AIS Size	77,183
Age	15.2 (AIS), 4.3 (EOS)
Sex (female)	64.2 (AIS), 52.1 (EOS)
Race	
White	49.9
Hispanic	13.9
Black	11.9
Asian or Pacific Islander	4.2
Native American	2.0

Racial demographics were % White, % Hispanic, % Black, % Other, % Asian or Pacific Islander, .5% Native American. (Table 1).

Table 2

Incidence of degenerate dysplasia of the hip and hip pathology in EOS and AIS patients.

Degenerative Dysplasia of the Hip	Percent (n)
EOS	3.9 (1005)
AIS	1.4 (1073)
Hip Pathology	
Osteonecrosis	0.3 (4 EOS, 3 AIS)
Osteoarthritis	0.6 (2 EOS, 10 AIS)
Recurrent Hip Dislocation	35.4 (133 EOS, 281 AIS)
Hip Ankylosis	0.0 (0)
Hip Labral Tear	0.0 (0 EOS, 1 AIS)
Hip Arthroplasty (total + partial)	6.7 (54 EOS, 86 AIS)

Table 3

Binary logistic regression model predicting risk of arthroplasty.

Independent Predictors	Adjusted Beta [95% Confidence Interval]	Significance
Age	1.039[1.007–1.073]	p = .017
Gender (Female)	.913[.642–1.297]	p = .610
Hip Osteoarthritis	13.43[5.21–34.66]	p = < .001

($p = < .001$ 16.11[6.32–41.06]) as being significant predictors of hip arthroplasty. A multiple binary logistic regression analysis was then used to determine the relationship between the significant univariate predictors of hip arthroplasty. Both of the significant univariate predictors, age ($p = .017$, 1.039[1.007–1.073]) and hip osteoarthritis ($p = < .001$, 13.43[5.21–34.66]) remained significant (Table 3).

4. Discussion

EOS and AIS patients have been shown to have increased rates of comorbidities when compared to the general population. Insults to the immature spine can have lasting effects on the development of multiple organ systems. However, diseases of the hip, as a consequence of spinal deformity, have been relatively understudied in the context of EOS and AIS. This study investigated the rate of dysplasia of the hip as well as the incidence of hospital admissions for hip disease in the EOS and AIS populations.

We evaluated 111,827 KID scoliosis patients from the years 2003–2012. This was the first high powered investigation into the EOS and AIS rate in the North American population. Previously, Wynne-Davies⁹ conducted a survey of Infantile Idiopathic scoliosis in 134 infants in Edinburgh. They found that DDH occurred in 3.5% of cases. This was later investigated by Hooper,¹⁵ who reviewed the radiographs of 156 scoliosis patients in Edinburgh. They reported a much higher rate of 6.4% of patients having dysplasia. We found a DDH rate of 1.4% and 3.9% in the AIS and EOS populations, respectively. These rates are much higher than those of the general population, which range from 3 to 6 per 1000 (0.3–0.6%) live births in European countries and 0.7–7.6 per 1000 (0.07–0.76%) live births in the United States.¹⁰ The higher incidence of DDH in the EOS population versus the AIS population can be explained by the fact that EOS patients may have pathologies such as Spinal Muscular Atrophy (SMA), Osteogenesis Imperfecta, Marfan's Syndrome, vertebral anomalies, etc., that cause scoliosis and concurrently affect the developing hip (e.g. SMA can cause contractures of the lower extremity that leads to hip dysplasia).^{5,7,14,16}

It has been shown that scoliosis patients experience increased rates of acetabular over coverage, which can predispose patients to hip

pathology.¹¹ We found that hip osteoarthritis occurred in 0.6% (EOS: 0.2%; AIS: 0.9%) of patients and 0.3% of patients had developed hip osteonecrosis. AIS patients were also more likely to have recurrent hip dislocations (35.4% vs 17.0%, OR: 2.67, $p < 0.001$), which was likely a product of the older AIS population having more time to experience a recurrence. These findings argue that, while EOS and AIS patients with DDH are at a significant risk for recurrent hip dislocations, they are experiencing minimal rates of additional hip pathology (osteonecrosis, osteoarthritis, hip ankylosis and hip arthroplasty). Our regression analyses did find that hip osteoarthritis and older age were significant predictors of patients having to undergo hip arthroplasty. While there wasn't a significant proportion of patients undergoing arthroplasty in this database, this investigation may have been identifying future arthroplasty patients early in their disease progression. These results are reflective of the pathogenesis and epidemiology of hip arthroplasty in the adult population, where older patients with significant arthritis make up the majority of the cohort undergoing arthroplasty.^{17,18}

Potential follow up investigations could look to compare the rates of hip pathology in adult patients who were diagnosed with EOS/AIS and compare the age at which they experienced those pathologies to controls. Other studies could look at bone mineral density in early hip pathology patients with EOS/AIS. It has been previously shown that 20–38% of adolescent patients with scoliosis may have osteoporosis, but it was unclear in our investigation what percentage of the patients who experienced hip pathology were due to bone mineral defects or mechanical defects.¹⁹

This study was limited by the inherent constraints of the KID database. The reliance on ICD-9 diagnostic and procedural codes is a potential source of error as there could have been mistakes at the data entry level. The KID also only provides inpatient data, lacking aspects of temporality that could be utilized when studying the onset of varying diagnoses and subsequent outcomes. The KID also is lacking longitudinal data, limiting our ability to capture outpatient claims and readmissions. Despite these limitations, the breadth of patients studied within this sample, which included extensive demographic, comorbidity, and inpatient outcomes data, maintains significant merit. Furthermore, the national design of the KID dataset increases the generalizability of our results.

5. Conclusion

This study identified the rate of degenerate dysplasia of the hip (DDH) in the Early Onset Scoliosis (EOS) and Adult Idiopathic Scoliosis (AIS) populations using the KID database. The rate of DDH was found to be 3.9% and 1.8% for EOS and AIS, respectively. There was an overall increase in the incidence AIS and EOS patients with DDH from 2003 to 2012 (339 cases vs. 729 cases). This investigation also found the incidence of hospital admissions of osteoarthritis, osteonecrosis, labral tears, and ankylosis to be minimal for AIS and EOS patients with DDH. Older age and hip osteoarthritis were the only significant predictors of hip arthroplasty in AIS and EOS patients. These findings may benefit risk management and disease prevention in EOS and AIS patients.

Source of funding

None.

Conflicts of interest

None.

Supplemental data. Univariate analysis of predictors of hip arthroplasty

Independent Predictors	Significance
Age	p = .003 1.048[1.016–1.081]
Gender	p = .644 1.085[.768–1.533]
Hip Osteonecrosis	p = .99 [0]
Hip Osteoarthritis	p = < .001 16.11[6.32–41.06]
Recurrent Hip Dislocation	p = .853 1.042[.674–1.611]

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