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IS SCOLIOSIS A COMMON DEFORMITY IN CHILDREN WITH JUVENILE IDIOPATHIC ARTHRITIS?

ORIGINAL ARTICLE

ABSTRACT

Purpose: Juvenile Idiopathic Arthritis (JIA) is the most common chronic rheumatic disease in childhood. Scoliosis can occur in children with JIA, since it mainly affects joint involvement, and contributes to the asymmetry of body and spine. This study aims to screen scoliosis in JIA, compare it with healthy controls, and evaluate the awareness among parents of children.

Methods: 218 children with JIA (163 girls, 55 boys) and 144 healthy controls (124 girls, 20 boys) aged 4-16 years were involved in this study. Angle of Trunk rotation (ATR) was measured by a scoliometer by applying a forward bending test. Children with more than 5° ATR were referred to take X-ray. To collect demographic data from parents and assess their awareness of scoliosis, forms designed for parents were used.

Results: Scoliosis was seen in 35 of the 218 (16.1%) children with JIA whose joints other than the spine were affected. 183 parents reported that they had never heard of scoliosis before the study. Scoliosis was seen in 25 children of the 183 children whose families had not heard of scoliosis before the examination. In families who had heard of scoliosis before the study, scoliosis was diagnosed in 10 of the 34 children.

Conclusion: Parents of children with JIA should pay attention not only to joints and extremities but also to the spine and asymmetries in the body. However, instead of just assessing the affected joint, professionals should evaluate children with JIA comprehensively, including the spine.

Keywords: Awareness, Deformity, Juvenile idiopathic arthritis, Scoliosis, Spine

JÜVENİL İDİYOPATİK ARTRİTLİ ÇOCUKLARDA SKOLYOZ YAYGIN BİR DEFORMİTE MİDİR?

ARAŞTIRMA MAKALESİ

ÖZ

Amaç: Juvenil İdiyopatik Artrit (JİA), çocukluk çağında en sık görülen kronik romatizmal hastalıktır. JİA eklem tutulumu ile birlikte omurganın asimetrisini etkilediği için JİA'lı çocuklarda skolyoz ortaya çıkabilir. Bu çalışma, JİA'lı çocuklarda omurga taramasını ve sağlıklı kontrollerle karşılaştırmayı ve ebevynlerin bu konudaki farkındalığını değerlendirmeyi amaçlamaktadır.

Yöntem: Bu çalışmaya 4-16 yaş arası 218 JİA'lı çocuk (163 kız, 55 erkek) ve 144 sağlıklı kontrol (124 kız, 20 erkek) dahil edildi. Gövde rotasyon açısı, öne eğilme testi uygulanarak bir skolyometre ile ölçüldü. Gövde rotasyon açısı 5°'den fazla olan çocuklar ortopedist tarafından röntgene yönlendirildi. Ebeveynlerden demografik veri toplamak ve skolyoz farkındalıklarını değerlendirmek için ebeveynler için hazırlanan formlar kullanıldı. Tüm analizlerde Statistical Package for Sciences (SPSS)-24.0 programı kullanıldı. p<0,05 anlamlı olarak kabul edildi.

Bulgular: Omurga dışındaki eklemleri etkilenen JİA'lı 218 çocuğun 35'inde (%16,1) skolyoz görüldü. 183 ebeveyn çalışmadan önce skolyozu hiç duymadığını bildirdi. Taramadan önce aileleri skolyozu duymayan 183 çocuğun 25'inde skolyoz görülürken, skolyozu duyan ailelerde 34 çocuktan 10'ununa skolyoz tanısı konuldu.

Tartışma: JİA'lı çocukların ebeveynleri sadece eklem ve ekstremitelere değil, omurga ve vücuttaki asimetrilere de dikkat etmelidir. Bununla birlikte, sadece etkilenen eklemi değerlendirmek yerine, JİA'lı çocuklar omurga da dahil olmak üzere kapsamlı bir şekilde değerlendirmelidir. Bu konuda ailelelerin skolyoz hakkındaki farkındalığı büyük önem taşımaktadır.

Anahtar Kelimeler: Farkındalık, Deformite, Jüvenil idiyopatik artrit, Skolyoz, Omurga

INTRODUCTION

Juvenile idiopathic arthritis (JIA) is an umbrella term is a chronic autoimmune inflammatory disease characterized by joint inflammation and pain that lasts for at least 6 weeks and affects children who are under 16 years old. The disease involves different clinical pictures rather than being a single disease (1,2). It commonly affects large joints such as the ankle and knee and often causes structural damage (3). The altered load on the joints combined with pain, inflammation, and other mechanical factors contribute to the onset of the injury (1). This pain and inflammation cause postural and muscular imbalance with a limited range of motion in the affected joints (4,5). Persistent inflammation can lead to growth disturbances and deformities such as scoliosis (3). Scoliosis is defined as the increase in the lateral curvature of the spine by 10 degrees to the right or left in the coronal plane, as determined radiologically (6). Moreover, scoliosis is a three-dimensional complex orthopedic deformity that also involves the spine, shoulder, and pelvis (7). Although genetic factors, melatonin disorders, connective tissue disorders, skeletal muscle abnormalities, neurological mechanisms, and biomechanical factors have been attributed to the etiology of scoliosis, the true etiology of this condition is not fully understood, and it is currently defined as a multifactorial disorder (3). Studies show that it is the most common diseased spinal region affected by JIA in 77% of patients. According to the American College of Rheumatology (ACR) and recent research, the true prevalence of spine involvement may be higher due to the subclinical course of the disease (8). Since JIA, which starts with joint involvement and causes body and spine asymmetry, has become an important secondary deformity in these children (9,10). There is no study in the literature investigating the presence of scoliosis in children with JIA. Therefore, in this study, it was aimed to investigate the presence of scoliosis in children with JIA and to investigate the presence of scoliosis compared to their healthy peers.

METHODS

Study design and selection of participants: this observational study was conducted randomly in Istanbul University Cerrahpasa, Division of Pediatric Rheumatology between August-December 2022. The study was approved by the research ethics committee of Istanbul University Cerrahpasa (A-03) and conducted based on the Declaration of Helsinki. Verbal and written information about the study was provided for participants and written informed consent was obtained from all participants. This study was registered with ClinicalTrials.gov under registration number NCT04664231.

The inclusion of 218 participants was above the minimum sample size needed to ensure a power of 95% confidence level and to detect statistical significance at a two-sided significance level of 0.05 (b = 0.20) by considering the effects of size 19,4% in average between the two groups, JIA and controls, using the calculation method for the basis of descriptive studies (11).

218 children diagnosed with JIA clinically according to EULAR criteria were included in the study. 160 healthy children from pre-school to high school were screened and determined as the control group. Children with scoliosis were excluded from the study, and 144 children without spinal problems were included in the study as a healthy control group. Age-matched healthy participants were involved in the study as controls. Participants were examined by a pediatric rheumatologist and an orthopedist and met inclusion criteria.

Eligibility criteria

The following eligibility criteria applied to children with JIA: (1) had been diagnosed with JIA, (2) aged between 4 and 16 years, (3) had been diagnosed within 6 months before the study. The exclusion criteria were as follows: (1) having severe neurological, musculoskeletal, or cardiovascular disease limiting mobility, (2) having conservative scoliosis rehabilitation (exercises and braces) in the past 6 months, and (3) spinal involvement.

Assessments

Each child and parents' knowledge of scoliosis was assessed physically by a specialist physiotherapist (EPK-ET) on the spine.

Assessment of posture

Assessment of the anterior, posterior, and lateral

posture of the children was done through observation. Shoulder height, scapula height, waist-to-arm distance, hip height, genu valgum, and foot deformity were assessed anteriorly and posteriorly. The use of foot orthotics was assessed. Trunk rotation was assessed using the Adam's Bend test. Head position, thoracic kyphosis, and lumbar lordosis were assessed laterally (12).

Assessment of pain

Pain status was evaluated using a numeric rating scale (NRS). The numerical scale is commonly used from 0 to 10, with 10 indicating "the worst pain" and 0 indicating "no pain". The child is asked to draw a circle around which they feel back pain at rest (13).

Scoliometric assessment

To obtain more objective results about the spine, the angle of trunk rotation (ATR) was assessed with a scoliometer (Bunnel scoliometer) during Adam's Bend test, which the physiotherapist measures by moving the scoliometer from the beginning of the thoracic vertebra to the sacrum in the standing forward bending position. The largest ATR measured was recorded (14,15). As a result of scoliometric measurement, children with more than 5° ATR were referred to take radiographic measurement.

Radiographic imaging

The results of the radiographs were compared with the clinical assessment of the physiotherapist at the end of all measurements. Radiographic imaging assesses the degree of scoliosis by measuring the Cobb angle by the orthopedist on the image of a standing anterior-posterior radiograph and recording the largest Cobb angle. The Cobb angle is a gold standard for the evaluation of scoliosis (16,17,18).

Risser classification

To assess the maturity of ossification, the Risser classification scale has been used. The scale provides information about the development stage of the skeleton depending on the degree of ossification and fusion of the iliac crest apophysis. Since stage 0 and stage 5 may look similar, age and long bone growth plates can help distinguish these two. A stage 0 patient will still have open growth plates in most of the long bones, while a stage 5 patient will not have any open growth plates in their long bones (19).

Parent awareness assessment

To assess the awareness of the parents about scoliosis, an assessment questionnaire was prepared with questions about personal and socio-demographical information. Gender, age, weight, height, educational status of children, dominant side, affected side(s), history of the disease, duration of disease, and sports/exercise habits were questioned in detail. To inquire about parents' awareness of their children's posture, questions such as "Have you ever heard of scoliosis? Is there any person among your relatives with scoliosis? When your child bends forward, do you see any asymmetry in his/her back? Does your child have any pain in the back?" were asked from the parents.

Statistical Analysis

Statistical analysis was performed using Statistical Package for Sciences (SPSS) version 24.0 (SPSS inc., Chicago, IL, ABD). Descriptive statistics including frequency, the percentage for nominal variables, and mean and standard deviations were calculated. The Kolmogorov-Smirnov test was used to determine if the data was normally distributed. To compare quantitative variables, the student's t-test was used for normally distributed variables and the Mann-Whitney U-test was used for non-normally distributed variables. All tests were two-sided tests and p-values < 0.05 were considered to indicate statistical significance (20).

RESULTS

A total of 218 children with JIA (age range 4-16 years) whose joints were affected other than the spine was studied (Table 1). Scoliosis was found in 35 of 218 (16.1%) children with JIA. 183 parents reported that they had never heard of scoliosis before the examination. Scoliosis was diagnosed in 25 of the 183 children whose families had not heard of scoliosis before the examination. Scoliosis was diagnosed in 10 of the 34 children whose families had heard about scoliosis before the examination. Only three out of ten children with scoliosis were diagnosed with scoliosis before coming to the clinic (Figure 1).

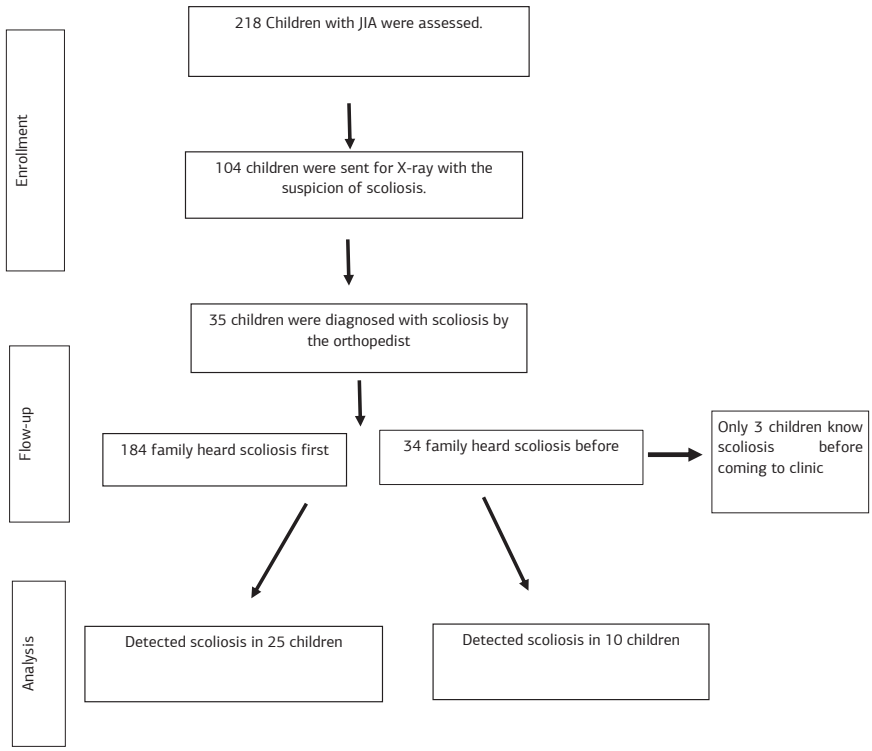

Figure I. Flow chart of parent scoliosis awareness

In the control group, a total of 160 children were examined. Scoliosis was diagnosed in 16 of the 160 (10%) children. Children with scoliosis were excluded from the study, and 144 children without spinal problems were included in the study as a healthy control group (Table 2).

188 of the children with JIA who came to the clinic had no exercise habits (p<0.001). Fifty-seven JIA children were referred to a physician for x-rays because it was suspected in the physical examination that they might have scoliosis. The clinical findings of 35 of the 57 children (%61,40) referred for radi-

Table I. Clinic Features of Children with JIA

	JIA with Scoliosis (n: 35)	JIA without Scoliosis (n: 183)
Affected joint (n, %)		
Hand-wrist	5 (10)	35 (13)
Elbow	7 (13)	41 (16)
Hip	1 (2)	2 (1)
Knee	26 (49)	110 (42)
Ankle	14 (26)	75 (28)
Type of disease		
Oligoarticular	24 (69)	115 (63)
Polyarticular RF+	1 (3)	12 (7)
Polyarticular RF-	8 (23)	51 (28)
Systemic	2 (6)	4 (2)
Disease Duration (month)		
mean±SD (min-max)	32.4±25.3 (2-120)	31.3±25.1 (1-144)
Number of affected joints		
mean±SD (min-max)	2.0±1.1 (1-6)	2.4±1.5 (1-6)

min: minimum, max: maximum, SD: standard deviation, n: number

Table II. Demographic Characteristics of JIA and Healthy Control

	JIA (n: 218)	Healthy Control (n:144)	p
Age (year) mean±SD (min-max)	10.4±2.8 (6-16)	11.1±2.9 (6-16)	0.26
Gender n (%)			
female	168(77)	124(86)	0.33
male	50 (23)	20(14)	
BMI (kg/cm2) mean±SD (min-max)	18.6±2.8 (14.3-29.5)	18.8 ±2.8 (14.2±27.6)	0.50
Children Educational status n (%)			
kindergarten	11 (5)	2 (1)	
primary school	78 (36)	42 (29)	
middle school	90 (41)	35 (25)	**<0.001**
high school	39 (18)	65 (45)	
Economic Status n (%)			
very bad	2 (1)	0 (0)	
bad	30 (14)	3 (2)	
medium	170 (78)	124 (86)	0.10
high	16 (7)	16 (11)	
very high	0 (0)	1 (1)	
Sports Habit			
Yes	31 (14)	70 (49)	**<0.001**
No	187 (86)	74 (51)	

min: minimum, max: maximum, SD: standard deviation, p<0,05, n: number, , Independent t samples t-test; significance was accepted as p<0.05.

ography were consistent with our clinical findings. The mean age of the children with JIA and scoliosis was 10.45±2.27 (p: 0.26), and the scoliometric measures were 6.57±1.89 (5-10) (p<0.001). Mean Cobb angles of 16.14±4.39 (10-27) and Risser degrees of 2.11±1.67 (0-5) were measured on the radiograph (Table 3).

DISCUSSION

Early diagnosis of scoliosis is of critical importance in terms of the negative effects it will have on the lives of adolescents and their parents in the future. This study aimed to investigate the presence of scoliosis in children with JIA and awareness of scoliosis in their parents it was found that children

Table III. Physical assessment and pain level in JIA and Healthy Control

	JIA (n: 218)	Healthy control (n:144)	p
Posture assessment (n,%)			
Shoulder asymmetry	19 (8)	49 (27)	
Scapula asymmetry	5 (2)	7 (4)	
Kyphosis	22 (9)	19 (10)	
Lomber lordosis	60 (24)	9 (5)	
Weiss asymmetry	5 (2)	7 (4)	
Genu valgum	62 (25)	11 (6)	**<0.001**[a]
Pes planus	41 (17)	21 (12)	
No deformity	31 (13)	58 (32)	
NRS mean±SD (min-max)	0.77±1.27 (0-5)	0.67±1.37 (0-6)	0.47
Foot deformity (n, %)			
Have deformity	173 (79)	25 (17)	**<0.001**[b]
Have not deformity	147 (21)	119 (83)	
Using sole (n, %)			
Using	71 (32)	0 (0)	**<0.001**[b]
Not using	125 (68)	144 (100)	
Degree of Scoliometer ( ) mean±SD (min-max)	2.3±2.5 (0-10)	0.9±1.5 (0-6)	**<0.001**[b]

min: minimum, max: maximum, SD: standard deviation, [a] Chi-square test; significance was accepted as p<0.05., [b] Independent t samples t-test; significance was accepted as p<0.05.

with JIA without spinal involvement have a high risk of scoliosis and the families of children with JIA who had scoliosis were not aware of it. Therefore, this study focuses on the importance of spinal screening in children with JIA.

Scoliosis progresses slowly from the beginning. Thus, observation and care are very important to prevent deformities from developing during the acceleration phase. Therefore, school screening for scoliosis during the growth phase is recommended, especially at the age of bone development, which is 13 years for males and 11 years for females (10). Although the American Academy of Orthopedic Surgeons (AAOS), the Scoliosis Research Society (SRS), the North American Pediatric Orthopedic Society and the American Academy of Pediatrics, early detection is low cost and carries a minimal risk of exposure to radiation, supports routine screening, arguing that conservative treatment will increase the chance of effective treatment (21). Children should be screened for scoliosis at any age group, especially in the 10-15 years age group where the growth is the most rapid. Scoliosis screening is recommended by the Scoliosis Research Association (22,23) twice a year for girls aged 10 and 12 years and once a year for boys aged 13 or 14 years.

Scoliosis screening for healthy children is performed regularly in many countries. There is no national scoliosis screening program in our country yet. It is known that regional scoliosis screenings have been carried out from time to time by some university hospitals and with the efforts of district municipalities to reveal scientific data. The current level of evidence in the literature that routine scoliosis screening is necessary is low to moderate. Although various procedures have been developed for the treatment of scoliosis, the most effective treatment still relies on early diagnosis. The lack of routine scoliosis screening program in many regions may delay early diagnosis. In our study, we revealed that the awareness of families about scoliosis is low. Therefore, it is very important to increase the awareness of scoliosis in the society (24,25). However, unfortunately, an additional assessment form for children with rheumatoid arthritis is not available. The fact that we found scoliosis in 16 of the elementary, middle, and high school children whom we considered to be in a healthy control group during our study may indicate that we should increase scoliosis screening in school-aged children. Although scoliosis scans will continue to be performed in schools as a part of the studies, we believe that the comprehensive inclusion of scoliosis scans in routine screening in JIA clinics will be effective in controlling the disease and preventing its progression.

Scoliosis screening will both ensure early diagnosis of scoliosis in children and raise the awareness of this issue among families (26). One of the strengths of our study is that it fills a gap in the literature by looking at the knowledge of scoliosis among parents of children with JIA. This knowledge is crucial for informing families about this condition (27).

Scoliosis occurs in 2-4% of children aged 10-16 years (28). As a result of our study, although the sample size was not large enough to give a prevalence, previously unreported scoliosis was detected in 16 (10%) of the 160 healthy children and 35 (16%) of the 218 children with JIA. These rates suggest that scoliosis rates seen in children with JIA is higher than healthy children. This highlights the importance of examining the spine as well as the affected joints in children who come to the rheumatology clinics even if they have other joint involvement.

Families try to control the symptoms in the affected joint as soon as possible because they are afraid of seeing edema, discomfort, and joint restriction in the affected joints due to the characteristics of the disease. Meanwhile, it neglects other parts of the body. This information was confirmed in our study.

In a study by Weiss et al (29), it is reported that children avoid putting weight on the joints or using the extremities mainly because of pain. The pain is said to cause kinesiophobia in children (30). In our clinical experience, most children with JIA have kinesiophobia with weight transfer due to pain experiments. This causes children to shift their weight to the opposite end of the affected joint or develop a compensatory mechanism in the body to protect the painful extremity. This situation leads to scoliosis as it affects the overall biomechanical alignment. The reason for scoliosis in children with oligoarticular JIA may be the constant shift of the

body's center of gravity to one side due to unilateral joint involvement.

Pain is not expected in children with scoliosis at an early age (31,32). Lonner et al (33) compared two groups of adolescents, including 894 with adolescent idiopathic scoliosis (AIS) and 31 healthy controls. Pain scores on the specific subdomain of the SRS-22 (Scoliosis Research Society Outcomes Questionnaire-22) questionnaire were similar in the scoliosis group and the control groups. Another study that examined a random sample of 310 children concluded that the prevalence of back pain was "moderately high," but the reported data on pain did not appear to be very homogeneous. Severe pain was documented in only 1% of the charts (31). Our study did not aim to ask about rheumatic pain but about the presence of back or low back pain to determine if it was scoliosis-related pain. The results of the study showed similarities in that the level of pain was found to be non-significant in both children with JIA and healthy children.

Norgaad et al (34) examined activity habits in JIA compared to gender- and age-matched healthy controls (age range 10-16 years). They found that children with JIA participated less in sports activities and had more difficulties in physical education than their healthy peers, despite having near-normal functional abilities. However, children with JIA did not differ significantly from their healthy peers. In our study, it was found that both groups had no sports habits. We believe that this is due to the discomfort that children with JIA.

Certain limitations of these previous studies are relevant to the discussion: (i) not all children were sent for X-ray, (ii) there is a need for further studies with larger samples to determine the prevalence, (iii) the study was conducted in a single center and (iv) parent awareness assessment questionnaire is not valid because it was created by us.

In conclusion, these findings suggest that therapists working with JIA should focus on all joints holistically, rather than a single joint. Exercise and assessment programs should involve the whole body, bearing in mind that all biomechanics may be affected rather than just exercising for the affected joint(s). Considering the frequency of scoliosis in children who do not have any disease, it is of great importance to perform routine screening. Families with chronically ill children and families who think they have healthy children should be informed and educated about scoliosis in terms of early diagnosis. We believe that spine scanning in children with JIA should become standard practice in rheumatology clinics and social awareness should be created through the coordinated efforts of families, physiotherapists, and health professionals.

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Ethical Approval: Approval for the study was granted by the Scientific Research Ethics Committee of the Istanbul University Cerrahpaşa (Approval Number: A-03).

Informed Consent: Online informed consent was obtained from participants.

Authors' contributions: ET: Conceptualization, Methodology, Investigation, Writing - Original Draft. EPK: Conceptualization, Methodology, Investigation, Writing - Original Draft. GL: Methodology, Investigation, Writing – Original Draft, Writing - Review & Editing. MAC: Investigation, Writing - Original Draft, Resources. ÖK: Methodology, Writing - Review & Editing, Supervision, Project administration.

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