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Assessment of Scoliosis and Factors Associated with Severity in Individuals with Cerebral Palsy

Serebral Palsili Bireylerde Skolyoz Şiddeti ile İlişkili Faktörlerin İncelenmesi

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ABSTRACT Objective: The aim of this study was to determine the prevalence of scoliosis in individuals with cerebral palsy (CP) and to investigate the factors associated with its development. **Material and Methods:** In this retrospective, descriptive cross-sectional study, children diagnosed with CP were examined. Demographic data, Risser grade, triradiate cartilage, scoliosis type, Cobb angle, previous surgeries, and developmental hip dysplasia were evaluated. Clinical information and radiographic images were obtained from hospital records. Factors potentially associated with the frequency of scoliosis were identified from the data. Multiple linear regression analysis was performed to estimate the variables affecting scores. The Enter Model was used in the regression analysis. The statistical significance level was set as $p<0.05$. **Results:** A total of 64 (27 female/37 male) individuals were included in the study. The mean age of the individuals was 13.06±8.14 years. The presence of scoliosis was 43.7% and developmental hip dysplasia was 12.5%. There was a statistically significant difference between the Cobb angle and gross motor functional classification system (GMFCS) and developmental hip dysplasia (p=0.002, p=0.001). There was no statistically significant difference between the other parameters (p>0.05). The model is significant (F=21.96; p=0.001). This model explains 72% of the predictors. The severity of scoliosis in individuals was influenced by GMFCS, affected body region, and scoliosis-related surgeries (p<0.05). **Conclusion:** In CP, the low level of motor function and the presence of hip dysplasia increase the severity of scoliosis.

Keywords: Cerebral palsy; Cobb angle; scoliosis

ÖZET Amaç: Bu çalışmanın amacı, serebral palsili (SP) bireylerde skolyozun varlığını belirlemek ve skolyoz gelişimini etkileyen faktörleri araştırmaktır. **Gereç ve Yöntemler:** Bu retrospektif, tanımlayıcı kesitsel çalışmada; CP tanısı konulmuş çocuklar incelenmiştir. Demografik veriler, Risser evresi, triradiate fibrokartilaj, skolyoz tipi, Cobb açısı, önceki ameliyatlar ve gelişimsel kalça displazisi değerlendirilmiştir. Klinik bilgiler ve radyografik görüntülere hastane kayıtlarından ulaşılmıştır. Verilerden skolyoz sıklığı ile ilişkili olabilecek faktörler belirlenmiştir. Ölçek puanlarını etkileyen değişkenleri tahmin etmek için çoklu doğrusal regresyon analizi yapılmıştır. Regresyon analizinde "Enter Model" kullanılmıştır. İstatistiksel anlamlılık düzeyi $p<0.05$ olarak kabul edilmiştir. **Bulgular:** Çalışmaya toplam 64 (27 kadın/37 erkek) birey dâhil edilmiştir. Bireylerin ortalama yaşı 13,06±8,14 yıldır. Skolyoz görülme oranı %43,7, gelişimsel kalça displazisi görülme oranı ise %12,5 idi. Cobb açısı, "kaba motor fonksiyon sınıflama sistemi (GMFCS)" ve gelişimsel kalça displazisi arasında istatistiksel olarak anlamlı bir fark vardı (p=0,002, p=0,001). Diğer parametreler arasında istatistiksel olarak anlamlı bir fark yoktu (p>0,05) (F=21,96; p=0,001). Bu model, tahmin edicilerin %72'sini açıklamaktadır. Bireylerdeki skolyozun şiddeti GMFCS, etkilenen vücut bölgesi ve skolyozla ilgili ameliyatlardan etkilenmiştir (p<0,05). **Sonuç:** SP'de motor fonksiyon düzeyinin düşük olması ve kalça displazisinin varlığı skolyoz şiddetini artırmaktadır.

Anahtar Kelimeler: Serebral palsi; Cobb açısı; skolyoz

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Cerebral palsy (CP) is a common physical and developmental disorder whose severity varies between individuals. According to the international definition, CP defines several disorders in movement and posture development that cause activity limitation as a result of permanent but non-progressive damage to the developing fetal or infant brain.[1,2] In individuals diagnosed with CP, upper motor neuron damage in the brain affects the motor system negatively. Affection of the motor system in CP leads to various problems, including selective motor control disorder, abnormal postural tonus, and poor trunk control.[3,4]

CP is the most common congenital/neonatal central nervous system disease associated with spinal deformities. The likelihood of spinal curvature increases with the severity of CP.[5,6] Spinal deformities, including scoliosis, kyphosis, or lordosis, are frequently observed in CP. Kyphosis is associated with weakness in the spinal extensor muscles and tension in the hamstrings and may lead to posterior pelvic tilt. The possibility of scoliosis may increase with the severity of CP. Regardless of skeletal maturation, curves greater than 40 degrees tend to progress. While the average incidence of scoliosis in CP is 20%, the incidence reaches 68% in quadriplegic children. The risk of progression is higher in patients with quadriparesis, increased spasticity, large curves, young age, poor sitting balance, and pelvic obliquity.[5,7,8]

Scoliosis observed in CP is divided into 2 groups. In the first group, single or double thoracic curvature is observed, especially in patients with II-III according to the gross motor functional classification system (GMFCS). In the second group, long or C-shaped thoracolumbar curvature accompanied by pelvic obliquity is observed, and this condition is encountered more frequently in nonambulatory patients.[5] The term neuromuscular scoliosis (NMS) defines the non-congenital spinal deformity seen in patients with any pre-existing neuromuscular diagnosis. Progressive deformity may affect ambulation, sitting, and cardiopulmonary functions over time.[9] The incidence of NMS is higher than the incidence of adolescent idiopathic scoliosis (0.35-14%) in the general population. In CP, this rate varies between 21% and 64%.[10]

Many studies have reported the frequency of scoliosis in CP. In these studies, the relationship between scoliosis and gross motor function level was mostly investigated.[11,12] Although musculoskeletal complications in CP have been extensively investigated in the literature, the factors determining the development of scoliosis in this population remain insufficiently elucidated. Therefore, the aim of this study was to determine the prevalence of scoliosis in children with CP and to systematically examine the clinical and demographic factors associated with its development.

Research Questions

1. What is the frequency of scoliosis among individuals with CP?
2. Which factors are associated with scoliosis severity among individuals with CP who have scoliosis?

MATERIAL AND METHODS

STUDY DESIGN

This retrospective, descriptive cross-sectional study included individuals who applied to the physical therapy outpatient clinics of Kayseri City Hospital between January 2020 and September 2023 with a diagnosis of CP and who underwent postero-anterior chest X-ray and/or scoliosis X-ray and/or pelvis X-ray. Ethics committee approval was obtained from Kayseri City Hospital Clinical Research Ethics Committee on August 13, 2024 with decision number 167, and the study was conducted following the Declaration of Helsinki.

PARTICIPANTS

Children who presented to physiotherapy and pediatric outpatient clinics with a diagnosis of CP and had scoliosis examination in the last 1 year were included in the study. Children with concomitant neurological disease, metabolic disease, connective tissue disease, congenital scoliosis, history of spinal operation, lower extremity inequality, torticollis, and brachial plexus injury were excluded (Figure 1).

Procedures

Age, sex, height, weight, type of CP, ambulation level, gross motor function level, Risser stage,

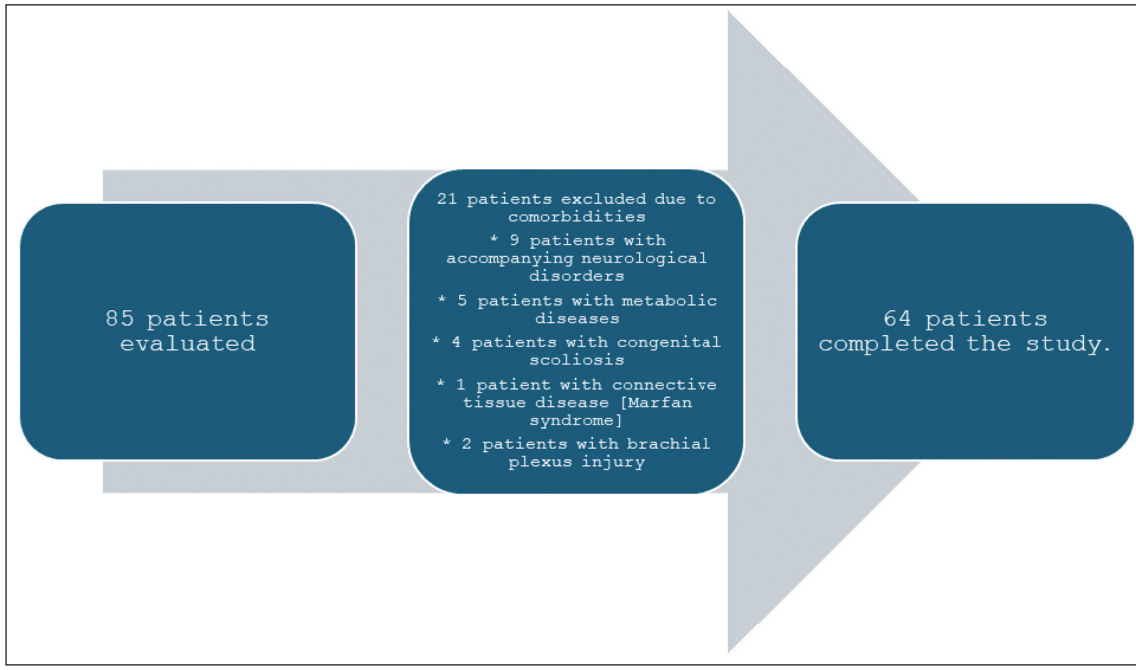


FIGURE 1: Flow chart

whether the triradiate cartilage was open or not, scoliosis type, Cobb angle, previous surgeries, and presence/absence of developmental hip dysplasia (DHD) were recorded.

DATA ANALYSIS

SPPS 26 (IBM Corp. (2019). IBM SPSS Statistics for Windows, Version 26.0. Armonk, NY: IBM Corp.) statistical package program was used to evaluate the data. Descriptive statistics (mean, standard deviation, minimum value, maximum value, number, and percentile) were given for categorical and continuous variables. Before regression analysis, model assumptions were evaluated. The normality of residuals was assessed using visual inspection (histogram and Q-Q plot) and the Shapiro-Wilk test. Multicollinearity was examined using variance inflation factor and tolerance values. Homoscedasticity was evaluated by inspecting scatterplots of standardized residuals versus predicted values. Multiple linear regression analysis was performed to predict the variables affecting the scores. Enter Model was used in the regression analysis. Regression coefficients (B), standardized beta (β), 95% confidence intervals, and p values were reported. The regression model was performed only in individuals with scoliosis. Due to the limited number of cases in the subgroups, scoliosis type was classified as thoracolumbar and other. Statistical significance was set at $p<0.05$.

RESULTS

A total of 64 (27 females and 37 males) individuals were included in the study. The mean age of the individuals was 13.06±8.14 years. The prevalence of scoliosis was 43.7%, and that of DHD was 12.5%. Demographic and clinical characteristics of the individuals are shown in Table 1.

The highest number of children was in the 3rd stage of the GMFCS (38.5%), while the highest number of children were in the 1st and 5th stages of the Risser stage (43.1% and 35.4%), respectively. The GMFCS and Risser stage scores of the individuals are shown in Table 2.

A statistically significant difference was found between scoliosis type and type of scoliosis surgery in terms of the Cobb angle ($p<0.001$). However, no statistically significant differences were found for the other parameters ($p>0.05$) (Table 3).

TABLE 1: Demographic and clinical characteristics of individuals with cerebral palsy (n=64)

	X̄±SD	n	%
Age (years)	13.06 ±8.14	-	-
Cobb angle	11.50 ±16.08	-	-
Gender	Female	27	42.2
	Male	37	57.8
CP type	Hemiplejic	8	12.3
	Diplejic	28	43.1
	Quadriplejic	28	43.1
Presence of scoliosis	None	36	56.3
	+	28	43.7
Scoliosis type	Torocolomber C	21	75
	Other	7	25
Developmental hip dysplasia	None	56	87.5
	+	8	12.5
Operation story	None	45	70.3
	+	19	29.7
Scoliosis operation	None	62	96.9
	+	2	3.1

Other: S-type scoliosis (n=4, 14.3%), thoracic C-type scoliosis (n=1, 3.6%), lumbar C-type scoliosis (n=1, 3.6%), and three-curve scoliosis (n=1, 3.6%). SD: Standard deviation

TABLE 2: GMFCS and risser stage values of individuals

		n	%
GMFCS	1	3	4.6
	2	19	29.2
	3	25	38.5
	4	9	13.8
	5	8	12.3
Risser phase	1	28	43.1
	2	7	10.8
	3	1	1.5
	4	5	7.7
	5	23	35.4

GMFCS: Gross motor functional classification system

TABLE 3: Comparison of clinical characteristics of individuals with cerebral palsy according to scoliosis parameters

	GMFCS	Scoliosis type	Scoliosis operation
Cobb angle	0.105***	**<0.001*** **	**<0.001****
Scoliosis +/-	0.277*	-	0.103*
Developmental hip dysplasia	0.309***	0.811***	0.277*

*Pearson chi-square; **Independent Samples Test;***One way analysis of variance; GMFCS: Gross motor functional classification system

In the regression analysis (n=28), the model was found to be statistically significant (F=21.96; p=0.001), and the model's explanatory power is 72%. According to the model, it was determined that the severity level of scoliosis in individuals was significantly influenced by the functional level (GMFCS). (β=2.72; p=0.023), the number of affected segments (β=7.20; p=0.001), and scoliosis surgery (β=21.90; p=0.002). In this regard, individuals with severe scoliosis had higher GMFCS levels, a history of scoliosis surgery, and a greater number of affected segments (Table 4, Table 5).

DISCUSSION

In this study, scoliosis was detected in 43.7% of individuals diagnosed with CP. Increasing severity of scoliosis was shown to negatively affect motor function levels and hip dysplasia. Risser stage, presence of triradiate cartilage, and previous orthopedic release surgery were less determinative factors in scoliosis severity. The severity of scoliosis in individuals was influenced by GMFCS, affected body region, and scoliosis-related surgeries.

Spasticity, poor muscle control, muscle weakness, and muscle imbalance around the spinal column are thought to play a primary role in the etiology of spinal curvatures in CP. However, there is insufficient evidence to support this. It is not clear whether spinal deformity is caused by the primary pathology causing CP or by spasticity, muscle weakness, or other secondary problems.[13] In a large study conducted by Persson-Bunke et al., it was emphasized that there was a statistically significant relationship between GMFCS level and development of scoliosis, and 50% of children with GMFCS IV-V developed severe scoliosis.[11] In this study, scoliosis severity and GMFCS level were found to be significant in CP. The significant difference between CP-related scoliosis and GMFCS that scoliosis may cause increased difficulties in motor skills by impairing body posture and motor control.

Developmental hip dysplasia (DHD) and scoliosis are common in individuals with CP. DHD is observed in 15-20% of children with CP.[14] Similar to this study, Garg et al. showed that there was no dif-

TABLE 4: Descriptive statistics of Cobb angle in children with cerebral palsy and scoliosis

	n	Minimum	Maximum	$\bar{X}$	SD	Skewness		Kurtosis	
					SD	statistics	SE	statistics	SE
Cobb angle	28	10	55.0	26.25	14.2325	0.653	0.441	-0.877	0.858

SD: Standard deviation; SE: Standard error

TABLE 5: Combined evaluation of independent variables affecting scoliosis severity level: multiple regression analysis results (n=28) comparison with regression analysis

			Standardised			95% confidence interval	
Predicted variable	Predictor variables	B	Beta	t value	p value	Lower limit	Upper limit
Cobb angle	GMFCS	2.72	0.18	2.34	**0.023**	0.39	5.05
	Cerebral palsy type	-1.95	-0.083	-1.091	0.280	-5.53	1.63
	Risser phase	0.700	0.08	0.55	0.57	-1.80	3.20
	Deelopmental hip dysplasia	-4.69	-0.09	-1.39	0.16	-11.44	2.05
	Gender	-0.49	-0.01	-0.21	0.83	-5.14	4.16
	Scoliosis type	7.20	0.72	9.99	**0.001**	5.76	8.64
	Operation story	2.43	0.07	0.93	0.35	-2.77	7.64
	Scoliosis operation	21.90	0.23	3.32	**0.002**	8.69	35.10
Model	**F=21.96**	**P=0.001**	**R^2=0.762**	**Adj. R^2=0.727**	**R=0.87**	**Durbin-Watson=2.05**	

Model assumptions (normality, multicollinearity, and homoscedasticity) were checked and met. GMFCS: Gross motor functional classification system

ference between DHD and scoliosis.[15] Another study in 337 patients showed that pelvic obliquity developing after DHD was strongly associated with scoliosis severity in patients with CP.[16] Studies investigating the relationship between DHD and scoliosis are quite limited. In our study, the rate of DHD was 12.5%, and the effect of this condition on the development of scoliosis is in parallel with other studies in the literature. This suggests that hip dysplasia is a factor that affects postural balance and changes the load distribution on the spine.

There are limited studies in the literature showing that previous limb surgery in CP may be related with scoliosis. In this study by Bertoncelli et al., it was not clearly explained whether the history of hip surgery was related with CP and/or spasticity.[17] In our study, no relation was found between tendon and/or bone operations for spasticity and development of scoliosis. Extremity muscle or tendon surgeries are usually performed at an earlier age and focus only on certain muscle groups. However, scoliosis usually becomes evident in later periods, and the curvature of the spine may occur over time. It was thought that such orthopedic surgeries target only the muscles and tendons and do not directly affect the spine.

This study provides valuable information on the impact of scoliosis in individuals with CP. GMFCS level, scoliosis type, and lower extremity surgery history were associated with Cobb angle. Furthermore, the parameters affecting scoliosis were analyzed in detail by regression analysis. Although we found a direct correlation between scoliosis and developmental dysplasia of the hip, the retrospective design and the relatively small sample size in our study mean that this finding needs to be supported by further research. However, the study also has limitations such as relatively little emphasis on the role of factors such as Risser stage, triradiate cartilage, and previous orthopedic surgeries, which may warrant further investigation. Another limitation of our study is that the relatively high R^2 value may be partly due to sample characteristics and the interrelated nature of clinical variables. Furthermore, the cross-sectional nature of the study limits causal inferences and longitudinal re-

search would provide a better understanding of the long-term progression and effects of scoliosis in individuals with CP.

CONCLUSION

Early diagnosis and treatment of scoliosis are extremely important, especially in patients with low GMFCS. For treating scoliosis, motor function levels and co-occurring conditions such as DHD should be considered. Although the coexistence of DHD and scoliosis is frequently observed, this association should be further investigated in large-scale prospective studies involving individuals with CP.

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