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## Original Article

## Natural history of scoliosis in cerebral palsy and risk factors for progression of scoliosis

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## ABSTRACT

**Background:** Scoliosis in cerebral palsy (CP) often occurs and causes a disturbance in daily life. The purpose of this study was to investigate the natural history of scoliosis in cerebral palsy and determine risk factors for the progression of scoliosis using multivariate analyses.

**Methods:** We revised 113 patients with CP (47 males and 66 females) who had scoliosis with a curve of at least 10° were reviewed and retrospectively investigated these cases of scoliosis and analyzed the risk factors for the progression of this condition.

**Results:** The mean follow-up period was 16.5 years and the mean age at onset of scoliosis was 6.6 years (range: 1–16 years). In 59 patients (52%), the age at onset of scoliosis was under 6 years. On the final radiographs, the mean Cobb angle was 55.1° (range: 10° to 169°). After the age of 20 years, 13 of 40 patients (32.5%) had a progression of over 10° in scoliosis. Multivariate analyses showed the risk factors for the progression of scoliosis to be hip displacement ( $p = 0.0038$ ), the onset of scoliosis before the age of 6 years ( $p = 0.0024$ ), and 30° of the Cobb angle before the age of 10 years ( $p < 0.001$ ). A subtype of CP (spastic quadriplegia) was identified as a potential risk factor.

**Conclusions:** After the age of 20 years, 32.5% patients had a progression of over 10° in scoliosis. Risk factors for the progression of scoliosis in CP included hip displacement, early-onset scoliosis, and Cobb angle of 30° before the age of 10 years.

**Level of evidence:** Prognostic level IV - case series.

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## 1. Introduction

Scoliosis in cerebral palsy (CP) frequently occurs; its incidence has been reported from 25% to 69% [1–4]. Severe scoliosis often causes a disturbance in daily life and places a high demand on nursing care. Treatment for scoliosis in CP is primarily includes brace treatment or surgical operation. Brace treatment in CP is often difficult to continue. Recently, surgical treatment has been recommended and performed for various indications. However, there is no consensus on the treatment of scoliosis in CP and indications of surgical treatment [4,5]. This may be because there are few studies on the natural history of scoliosis in CP and on risk factors for the progression of scoliosis [6–8]. Many factors may

influence the progression of scoliosis, and the condition can continue to progress even after growth maturity. Previously We reported the natural history of scoliosis with 89 patients [9]. We divided into 2 groups and univariate analysis of several factors was performed. We concluded the risk factors for the progression of scoliosis were Gross Motor Function Classification System (GMFCS) [10] level V, Spastic Quadriplegia, hip displacement, the early-onset scoliosis and Cobb angle of 30° before the age of 10 years. Because our previous study used univariate analysis, we needed to increase the number of cases and use multivariate analysis.

In this study, we investigated the natural history of scoliosis in CP in depth and analyzed risk factors for the progression of scoliosis using multivariate analysis.

## 2. Materials and methods

A retrospective medical review of 113 patients with CP who had scoliosis with a curve of at least 10° as analyzed by the Cobb method on total spine radiographs at Osaka Rehabilitation Hospital

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for Children was performed. None of the patients received continuous treatment for scoliosis, such as brace treatment or surgical operation. The study was approved by the institutional review board and informed consent was obtained from all individual participants included in this study.

All patients were followed up by orthopedists and had several radiographs of the total spine in the supine position to monitor the progression of the curvature. The severity of scoliosis was measured by the Cobb method, and the pattern of the spinal curve, location of the curve, and number of vertebrae involved were examined. In cases with a double curve, the major curve was measured. The progression of the curvature after growth maturity was also measured. In this study, 40 patients had radiographs of the total spine after the age of 20 years. In this study, the progression of scoliosis was defined as more than 10° progression after the age of 20 years.

Risk factors for the progression of scoliosis regarding CP subtype, physical mobility, hip dislocation, scoliosis onset, and the Cobb angle before growth maturity in patients who had a follow-up after the age of 18 years were investigated. Regarding the subtype of CP, patients were divided into spastic, dyskinetic, and ataxic subtypes. Spastic CP included spastic quadriplegia, diplegia, and hemiplegia. These were classified into two groups: spastic quadriplegia and others. Physical mobility was classified according to GMFCS. Patients were classified into two groups based on the following: 1) those classified as GMFCS level V (bedridden) and 2) others. In addition to spinal radiographs, hip dislocation via anteroposterior radiographs of the hip was examined. In this study, a normal hip was defined as when the migration percentage of the hip was under 50% and a displacement hip was defined when the migration percentage was over 50%. Patients were classified into three groups as follows: 1) normal bilateral hip, 2) unilateral hip displacement, and 3) bilateral hip displacement. For age at onset of scoliosis, patients were divided into two groups: 1) before the age of 6 years and 2) after the age of 6 years. In analyzing risk factors for the progression of scoliosis regarding age and the Cobb angle, patients were divided into two groups: 1) under 30° and 2) over 30° before the age of 10 years.

### 2.1. Statistical analysis

Statistical analysis was performed using JMP Statistical Software, Version 11 (SAS Institute Inc., Cary, NC). Each parameter was analyzed using the Mann–Whitney test. Cobb angle between normal hip and unilateral and bilateral hip displacement was tested by one-way ANOVA. Differences were considered statistically significant at  $p < 0.05$ . Factors significant on univariate analysis were evaluated by multiple regression analysis. The threshold for including variables in the multiple linear regression analysis was  $p = 0.05$ .

## 3. Results

### 3.1. Demographic data

Forty-seven of the 113 (42%) patients were males, and 66 (58%) were females. The mean age at the first radiograph was 4.1 years (range: 0–17 years) and the mean follow-up period was 16.5 years (range: 6–32 years). There were 73 patients who had a follow-up over the age of 18 years. Forty patients who had a follow-up over the age of 20 years and mean follow-up after 20 years was 6.0 years (range 1–14 years). Eleven patients (10%) were able to walk (GMFCS level I ~ III). 19 patients (17%) were classified as level IV, and 83 patients (73%) were level V in GMFCS. A total of 72 patients (64%) had spastic quadriplegias (16 dyskinetic, 12 spastic diplegia, 6 ataxia, and 7 others).

### 3.2. Scoliosis characteristics

Eighty-four patients (74%) had single curves, and 29 patients (26%) had double curves. In the major curves, 46 patients (41%) had thoracic curves, 41 patients (36%) had thoracolumbar curves, and 26 patients (23%) had lumbar curves. The number of vertebrae involved in the major curves was an average of 7.1 (4–14).

The mean age at onset of scoliosis was 6.6 years (range: 1–16 years). In 59 patients (52%), the age at onset of scoliosis was under 6 years. On the final radiographs, the mean Cobb angle was 55.1° (range: 10° to 169°) (Fig. 1).

### 3.3. Progression of the curvature after growth maturity

After the age of 20 years, 13 of 40 patients (32.5%) had a progression of over 10° in scoliosis. The mean Cobb angle at the growth maturity was 60.3° (range 22–169°) with the patients who had no progression of over 10° and 84.9° (range 12–126°) with the patients who had a progression over 10°. There was no significant difference in the progression after growth maturity for the degree of curvature at growth maturity, the subtype of cerebral palsy, and the GMFCS level.

### 3.4. Risk factors for the progression of scoliosis

To determine the important prognostic factors for the progression of scoliosis, ten factors were evaluated by uni- and multivariate analyses. Table 1 shows the distribution of sex, the pattern of the curve, the location of the major curve, the subtype of CP, the GMFCS level, hip displacement (unilateral or bilateral hip displacement), the onset of scoliosis, and Cobb angle at the age of 10 years. In patients with spastic quadriplegia, GMFCS level V, hip displacement (both unilateral and bilateral hip displacement), the onset of scoliosis before the age of 6 years and Cobb angle of 30° before the age of 10 years, a significant progression was revealed by univariate analysis. In the multiple linear regression analysis, the important prognostic factors for progression were hip displacement ( $p = 0.0018$ ), the onset of scoliosis before the age of 6 years ( $p = 0.0001$ ) and Cobb angle of 30° before the age of 10 years ( $p < 0.001$ ) (Table 2). The subtype of CP ( $p = 0.071$ ) were identified as potential risk factors for progression.

## 4. Discussion

Several studies have reported the frequency of scoliosis in CP. Balmer et al. [1] reported that 21 of 100 (21%) children in an outpatient clinic had scoliosis. Madigan et al. [3] reported 64%, and Koop et al. [11] reported 77%. However, there are few reports of the natural history of scoliosis in CP. Saito et al. [4] reported a detailed investigation of the natural history of scoliosis in CP from childhood to adult life. They reported that 54 of 79 (68%) institutionalized patients with spastic CP had scoliosis. They analyzed 37 patients via radiographic examination and the details of the patients (e.g., mental retardation, nutritional status, joint contractures, and frequency of pneumonia occurrence). They also reported that 11 of 13 (85%) patients had a spinal curve of more than 40° at the age of 15 years, and their scoliosis progressed to more than 60°. They also reported that scoliosis of more than 60° developed in patients with total body involvement (67%), those who were bedridden (100%), and those with thoracolumbar curves (57%). In their conclusions, risk factors for the progression of scoliosis in spastic CP included a spinal curve of 40° before the age of 15 years, total body involvement, bedridden, and thoracolumbar curve. Gu et al. [5] investigated the natural history of scoliosis in 110 patients with nonambulatory spastic tetraplegic CP. They reported that the risk

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