Health-related quality of life in children and adolescent with different types of scoliosis: A cross-sectional study

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Abstract

Background: The health-related quality of life (HRQoL) was affected in children and adolescents with scoliosis. However, there was lack of study to compare the HRQoL among patients with different types of scoliosis. We aimed to investigate whether the HRQoL differs among patients with idiopathic, congenital, neuromuscular, and syndromic scoliosis.

Methods: Children and adolescents with scoliosis were recruited from a single tertiary hospital. The HRQoL, as assessed by the child health questionnaire 50-item parent form, was compared with a reference health sample group using the effect size (ES). Intergroup differences related to scoliosis subtype and severity were explored.

Results: A total of 67 participants with scoliosis (24 idiopathic, 15 congenital, 15 neuromuscular, and 13 syndromic) were analyzed. The HRQoL in patients with neuromuscular scoliosis was affected the most, in both physical (ES range: 0.97–2.4) and psychosocial domains (ES range: 0.92–2.58). To a lesser extent, the physical (ES range: 0.99–1.13) and psychosocial (ES range: 0.8–1.18) domains were also affected in patients with syndromic scoliosis. The domains of family activities (ES = 1.1), role/social-emotional/behavioral (ES = 0.99), general health perception (ES = 0.94), and self-esteem (ES = 0.87) were affected in patients with idiopathic scoliosis. In contrast, only the general health perception domain (ES = 1.27) was affected in patients with congenital scoliosis. Scoliosis severity correlated with scores in the physical domains and some psychosocial domains, while treatment type correlated with scores in the physical domains only. Scoliosis subtype and severity both affected the physical and psychosocial domains, with a stronger impact for subtype.

Conclusion: Differences in the HRQoL exist among scoliosis subtypes, with neuromuscular scoliosis being most affected. Although the scoliosis subtype and severity both affect the HRQoL, the subtype is more influential than severity.

Keywords: Congenital scoliosis; Health-related quality of life; Idiopathic scoliosis; Neuromuscular scoliosis; Syndromic scoliosis

1. INTRODUCTION

Scoliosis is a three-dimensional deformity of the spine, clinically defined as a curvature of the spine greater than 10 degrees in the coronal plane. It is often accompanied by a rotation of the spine in the axial plane. Scoliosis can be classified into congenital, idiopathic, neuromuscular, syndromic, and functional types.1

Congenital scoliosis (CS) is a malformation resulting from a prenatal disruption of vertebral formation or segmentation, leading to imbalanced longitudinal growth and rotation of the vertebrae. The symptoms of CS are usually not observed at birth; however, impaired ambulation may be observed following vertebral rotation during development.2

Idiopathic scoliosis (IS) is the most common type of scoliosis, accounting for more than 80% of scoliosis cases.3 The diagnosis requires exclusion of other anatomic anomalies. IS may be divided into three subtypes according to the age of onset: infantile, juvenile, and adolescent. The subgroups differ in their progression and treatment. The infantile type occurs within the first 3 years of life and often resolves spontaneously. The juvenile type occurs between 3 and 9 years of age, and most of these children require intervention.4 Adolescent IS (AIS) has an onset after 10 years of age and is the most common type of IS; in most of these patients, the clinical course is not serious, with intervention required in only 10%.5

Neuromuscular scoliosis (NMS) is of multiple etiologies and the incidence is variable. The severity of the spinal curve deformity is related to the degree of neuromuscular involvement. Cerebral palsy (CP), muscular dystrophy, spinal cord injury, and spinal dysraphism are common etiologies. NMS usually develops early and may progress quickly in certain conditions, such as tethered spinal cord syndrome, hydrocephalus, or intraspinal tumor.

Syndromic scoliosis (SS) co-occurs with many genetic and nongenetic syndromes, including VACTERL association, Marfan syndrome, Ehlers-Danlos syndrome, neurofibromatosis, Rett syndrome, and Down syndrome. The cause, symptoms, and progression of SS vary depending on the disease context.

While the etiology, onset, prognosis, and treatments vary among these classifications, the possible outcomes of scoliosis are similar: respiratory compromise, seating compromise, pain, gait impairment, difficulty with activities of daily living,
The health-related quality of life (HRQoL) may thus be jeopardized. Although the HRQoL is an important issue in current clinical practice, its quantitation has not been standardized. Several questionnaires, such as the SRS-22, EQ-D-5, SPF-36, and Muscular Dystrophy Spine Questionnaire, have been used in evaluating the HRQoL of patients with scoliosis. Early studies have demonstrated that AIS can lead to increased physical, psychological, and social problems in patients. Factors such as pain, restricted physical activities, poor body image, maladjustment in school, and poor peer relationships may consequently contribute to a decreased quality of life in adolescents with scoliosis.

The child health questionnaire (CHQ) is a HRQoL measurement tool comprising physical and psychosocial domains. The CHQ is based on the parents' perceptions, which represent a subjective vision of the patient's HRQoL. The CHQ has been translated into several languages, and its validity and reliability have been evaluated in the United Kingdom, Germany, Francophone Canada, Australia, Norway, Italy, and the Netherlands. The CHQ may be applied in the general population and in chronically ill children. Nixon et al. confirmed the validity of the CHQ among survivors of childhood cancer. Westendorp et al. surveyed the responsiveness of the CHQ in adolescents with chronic pain or fatigue. Other studies have demonstrated that the CHQ-PF50, which is a parent-completed questionnaire, is useful for evaluating the HRQoL of children and adolescents with different types of scoliosis. Therefore, the primary aim of the present study was to evaluate whether the HRQoL, as assessed by the CHQ-PF50, differs among different types of scoliosis. Furthermore, we assessed the relationships between CHQ-PF50 scores and potential correlated factors and examined the interaction between scoliosis type and severity in the affected CHQ-PF50 domains.

2. METHODS

2.1. Study population

The study was conducted in a tertiary medical center in Taiwan, in design of cross-sectional study. Patients with a confirmed diagnosis of IS, CS, NMS, or SS, 5–18 years of age, were enrolled during their appointments at the scoliosis clinic, when hospitalized for surgery, or when under surveillance for scoliosis. The patients provided informed consent. The recruitment period was between May 2014 and December 2016. The data of control group were derived from a earlier Taiwanese study. A total of 129 healthy children (95 boys, 103 girls), 6–15 of age, by means of written requests and mail-back questionnaires in 2006, were chosen as the control group. The study protocol was approved by the local Institutional Review Board.

2.2. Measures

The severity of the scoliosis was defined using Cobb's angle (mild: 10º–24º; moderate: 25º–40º; and severe: >40º). The HRQoL was measured using the traditional Chinese (Taiwan) version of the CHQ-PF50 (HealthActCHQ Inc., Boston, MA). The CHQ-PF50 consists of 50 questions to assess the health status of children, according to 12 physical and psychosocial domains. Each question was answered according to a five-point scale, and was intended to assess the previous 4 weeks’ performance, except when there was a change in health, in which case it covered the most recent year of the child's life. Each domain was scored from 0 to 100, with a higher score indicating better health. The domains were further transformed into two main scores: the physical summary score (PhS) and psychosocial summary score (PsS), with a mean of 50 and a standard deviation of 10. Physical health comprises four components: physical functioning, role/social–physical, general health perception, and bodily pain. Psychosocial health comprises four components: role/social–emotional/behavioral, self-esteem, mental health, and behavior. Two scales (parental impact-time and parental impact-emotion) contribute to both dimensions but have a stronger correlation with psychosocial health. The final two subscales focus on family activities and family cohesion.

2.3. Procedure

Each patient underwent an image survey to confirm the scoliosis diagnosis; Cobb's angle was measured by the same specialist for all patients. The types of treatment were documented. The parents then completed the CHQ-PF50.

2.4. Statistical analyses

Demographic characteristics were summarized using descriptive statistics. Continuous variables are presented as means and standard deviation. Categorical variables are presented as numbers and percentage. Effect sizes (ESs) were compared with those in an earlier study involving healthy Taiwanese children. An ES > 0.8 was considered to be large. Spearman's correlation analyses were performed to identify associations between CHQ-PF50 scores and age, severity, and treatment type. A multivariable analysis was performed to evaluate the interaction between scoliosis type and severity and to clarify the possible confounding effects. The multivariable analysis was focused on domains with a significant intergroup difference as assessed using the Kruskal-Wallis test. Statistical analyses were performed using SPSS (Version 23, IBM Corporation, Armonk, NY) and a p < 0.05 was considered statistically significant for a two-tailed test.

3. RESULTS

Sixty-seven children and adolescents (45 girls and 22 boys) with scoliosis were enrolled. Twenty-four patients had IS, 15 patients had CS, 15 patients had NMS, and 13 patients had SS. The NMS group included patients with hydromyelia (n = 1), spinal tumor (n = 1), spinal dysraphism (n = 4), peripheral neuropathy (n = 1), neurofibromatosis (n = 1), CP (n = 4), spinal muscular atrophy (n = 1), Duchenne muscular dystrophy (n = 1), and basal ganglia germinoma (n = 1). The SS group included patients with Rett syndrome (n = 1), Marfan syndrome (n = 3), VACTERL association (n = 3), Apert syndrome (n = 1), Holt-Oram syndrome (n = 1), Angelman syndrome (n = 1), achondroplasia (n = 1), cerebro-costo-mandibular syndrome (n = 1), and cri-du-chat syndrome (n = 1).

There were no significant differences among the scoliosis subtypes for mean age, Cobb's angle, sex distribution, curve severity, or treatment used (Table 1). Table 2 shows the CHQ-PF50 scores of the study sample in comparison to those in the reference sample of healthy children. Patients with scoliosis had significantly lower scores in physical functioning, role/social–physical, role/social–emotional/behavioral, self-esteem, general health perception, and family activities, and lower PhS scores compared with those in healthy controls. In the CS group, only health perception scores were lower compared with those in healthy controls. The NMS and SS groups had lower scores compared with those in healthy controls in both physical and psychosocial multiple domains, while the IS group had lower scores in the psychosocial domains and general health perception of physical domain.

No domains or summary scores were correlated with age. Scores in the physical functioning (p < 0.001), role/social–physical (p = 0.002), bodily pain (p < 0.001), role/social–emotional/behavioral (p = 0.006), and family activities (p = 0.026), categories, as well as the PhS (p < 0.001), were significantly correlated with...
Table 1
Demographic characteristics

<table>
<thead>
<tr>
<th>Age (years, mean ± SD)</th>
<th>IS (N = 24)</th>
<th>CS (N = 15)</th>
<th>NMS (N = 15)</th>
<th>SS (N = 13)</th>
<th>Control (N = 129)</th>
</tr>
</thead>
<tbody>
<tr>
<td>12.8 ± 3.6</td>
<td>9.8 ± 4.6</td>
<td>11.7 ± 3.5</td>
<td>12.8 ± 3.7</td>
<td>10.1 ± 2.3</td>
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</tr>
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</table>

Table 2
Mean value and effect size of CHQ-PF50 domain and summary scores compared to that in a reference sample of healthy Taiwanese children

<table>
<thead>
<tr>
<th>Physical construct</th>
<th>IS (N = 24)</th>
<th>CS (N = 15)</th>
<th>NMS (N = 15)</th>
<th>SS (N = 13)</th>
<th>Total</th>
<th>Control</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical functioning</td>
<td>69.35 17.56</td>
<td>39.04 26.10</td>
<td>5.33 35.88 2.40</td>
<td>64.53 38.06 1.13</td>
<td>72.72 37.16</td>
<td>0.88</td>
</tr>
<tr>
<td>Role/social–physical</td>
<td>63.80 22.93</td>
<td>64.24 28.50</td>
<td>38.89 33.33</td>
<td>66.67 39.09</td>
<td>0.98</td>
<td>71.30 35.89</td>
</tr>
<tr>
<td>Bodily pain</td>
<td>86.11 23.91</td>
<td>80.00 16.04</td>
<td>64.00 26.94</td>
<td>70.00 25.17</td>
<td>0.74</td>
<td>57.34 16.62</td>
</tr>
<tr>
<td>General health perception</td>
<td>60.00 15.17</td>
<td>54.28 16.81</td>
<td>42.83 15.69</td>
<td>51.28 16.36</td>
<td>0.94</td>
<td>33.33 16.83</td>
</tr>
</tbody>
</table>

Table 3
Significant correlation between certain domains with severity and treatment

<table>
<thead>
<tr>
<th>Severity</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>R</td>
<td>P</td>
</tr>
<tr>
<td>Physical functioning</td>
<td>-0.421</td>
</tr>
<tr>
<td>Role/social–emotional/behavioral</td>
<td>-0.332</td>
</tr>
<tr>
<td>Bodily pain</td>
<td>-0.460</td>
</tr>
<tr>
<td>Role/social–physical</td>
<td>-0.364</td>
</tr>
<tr>
<td>Family activities</td>
<td>-0.272</td>
</tr>
<tr>
<td>Physical summary score</td>
<td>-0.417</td>
</tr>
</tbody>
</table>

4. DISCUSSION

This cross-sectional study used the CHQ-PF50 to determine whether differences in the HRQoL existed among child and adolescent patients with different types of scoliosis. The results demonstrate that NMS was the most affected type, with reduced scores in both physical and psychosocial domains, followed by SS, IS, and CS.

Strength of the current study includes first applying of CHQ-PF50 in evaluation of patients with different type of scoliosis in both physical and psychosocial aspect. Limitations of the present study include, first, a relatively small number of study participants, which may have resulted in a selection bias. Furthermore, the NMS and SS groups represented various underlying diseases, and our sampling may not reflect the true composition of scoliosis subtypes in the natural population. Beyond the spine disease, accompanying conditions such as mental retardation and socioeconomic status were not examined, which

with severity (Table 3). In addition, scores in the physical functioning (p = 0.004) and bodily pain (p < 0.001) categories, as well as the PhS (p = 0.006), were significantly associated with treatment type.

Nine CHQ-PF50 domains (physical functioning, role/social–physical, bodily pain, general health perception, role/social–emotional/motorial, mental health, parent impact–time, family activities, and PhS) were significantly affected as assessed by the Kruskal-Wallis test, and were submitted to the multivariable analysis. Among these domains, physical functioning, role/social–physical, role/social–emotional/motorial, and PhS were affected by both scoliosis subtype and severity. The other domains were significantly influenced by scoliosis subtype only.
may have contributed to the observed differences in HRQoL. Additionally, the study was cross-sectional in nature. Several scoliosis-related factors not examined by our study may have affected the HRQoL (e.g., the age of onset, recent progression of Cobb's angle, or curve type).

Overall, the scores for physical functioning, role/social–physical, role/social–emotional/behavioral, self-esteem, general health perception, family activities, and PhS were lower in patients with scoliosis compared with those in healthy subjects. The general health perception was the most affected domain as reported by parents, followed in order by the family activities, self-esteem, role/social–emotional/behavioral, physical functioning, and role/social–physical domains.

Generally, observation rather than treatment is suggested for mild scoliosis, whereas watchful waiting, bracing, and surgery are suggested for moderate to severe scoliosis. In the present study, there were fewer surgery cases than patients with severe scoliosis, suggesting that the patients and their families preferred nonsurgical treatments. This might be related to cultural differences; Asians tend to be more concerned with and less willing to accept surgery. In a study examining patient ethnicity and treatment decisions, Asians had a lower preference for surgery compared with those in other ethnicities.

While physical and psychosocial domains were both affected in NMS and SS, all of the physical and psychosocial domains, except mental health and family cohesion, were affected in the NMS group. Considering the nature and severity of the underlying diseases, it is not surprising that there were more impacted HRQoL domains in patients with NMS or SS than in patients with IS or CS. Diseases such as CP, spinal dysraphism, and VACTERL association have been reported to significantly decrease the HRQoL relative to that in healthy subjects. Presumably, the comorbidities associated with the scoliosis may have dominated the HRQoL scores in the current study. Patients with CP, spinal dysraphism, or hydromyelia would experience a more profound impact physically, which may lead to reduced HRQoL. The heterogeneity of the SS group might account for the smaller reduction in the HRQoL compared with that in the NMS group; the HRQoL might be only mildly affected in patients with Marfan syndrome, whereas it may be more severely affected in patients with cerebro-costo-mandibular syndrome or cri-du-chat syndrome. Although the sampling of disease in NMS and SS may have led to some errors, there is a lack of large-scale studies that have investigated the prevalence of each syndrome among scoliosis subtypes.

Most of the significantly affected categories in the IS group were psychosocial in nature. Using the SRS-22 questionnaire, Lee et al. observed that self-image was significantly decreased in AIS while the overall HRQoL was not significantly affected. Our similar results are not surprising given that appearance and body image are major concerns in the adolescent population. In addition, these results indicate that physicians should be more aware of this psychosocial issue. Not only should health education and communication with the patient and family be emphasized, but patients with IS should also be screened for potentially psychological diseases.

In the CS group, only the general health perception was significantly affected. Moreover, the CS group was the only group without affected family activities. Although there were no significant differences in age or Cobb's angle, patients with CS were slightly younger (mean age: 9.8 years) compared to those in the other three groups, which might explain this observation. Before adolescence, physical demands and expectations are lower, and self-image is not so important. Therefore, in patients with CS, the PhS was affected to a lesser extent (ES = 0.12) than the PhS (ES = 0.60).

Based on the results in Table 2, one might conclude that the general health perception was negatively impacted in all types of scoliosis. However, the general health perception was affected the least in patients with IS and the most in patients with NMS. Bodily pain was only significantly affected in patients with NMS, which may have been confounded by factors such as spasticity in CP or joint contracture caused by neuromuscular disease.

The present results also suggest that treatment type and severity correlate with certain HRQoL domains, mainly those in the physical construct. Thus, psychosocial issues in the domains significantly affected in the present survey, such as role/social–emotional/behavioral and family activities, should be emphasized regardless of sex, treatment type, or severity. Psychological support and possible referrals for all patients with scoliosis are suggested for a comprehensive approach. In addition, the multivariable analysis confirmed that both the subtype and scoliosis severity affected the HRQoL in certain domains, and that the type of scoliosis appears to have more impact. Therefore, special medical attention, in terms of related physical and psychosocial issues, is highly recommended when caring for patients with NMS/SS and severe scoliosis in clinical practice.

In conclusion, the current study used the CHQ-PF30 questionnaire to evaluate differences in the HRQoL among four types of scoliosis in children and adolescents. Among the four types, the HRQoL was most affected in patients with NMS, with significantly lower scores in both physical and psychosocial domains compared with those in healthy controls. To a lesser extent, patients with SS were affected in both physical and psychosocial domains. In patients with IS, role/social–emotional/behavioral, self-esteem, general health perception, and family activities scores were lower than those in healthy controls. In contrast, patients with CS were only affected by decreased general health perception scores. Treatment type and severity correlated with the HRQoL. Although the scoliosis subtype and severity both affected the HRQoL, the scoliosis subtype demonstrated a greater influence. Special medical attention in certain physical and psychosocial issues should be considered and integrated into clinical practice.

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REFERENCES


