Is it safe and effective to correct congenital scoliosis associated with multiple intraspinal anomalies without preliminary neurosurgical intervention?

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Abstract

The aim of the study was to determine if multiple intraspinal anomalies increase the risk of scoliosis correction compared to the normal intraspinal condition or 1 or 2 intraspinal anomalies in congenital scoliosis (CS) and whether correction for multiple intraspinal anomalies need to be performed with preliminary neurosurgical intervention before scoliosis correction.

A total of 318 consecutive CS patients who underwent corrective surgery without preliminary neurosurgical intervention at a single institution from 2008 to 2016 were retrospectively reviewed, with a minimum of 2 years of follow-up. The patients were divided into 3 groups according to different intraspinal conditions. In the normal group (N group; n = 196), patients did not have intraspinal anomalies. In the abnormal group (A group; n = 93), patients had 1 or 2 intraspinal anomalies. In the multiple anomaly group (M group; n = 29), patients had 3 or more intraspinal anomalies including syringomyelia, split cord malformation [SCM], tethered cord, low conus, intraspinal mass, Chiari malformation or/and arachnoid cyst. The occurrence of complications as well as perioperative and radiographic data were analyzed.

The incidence rate of multiple intraspinal anomalies in CS patients was 9.1% (29/318). No significant difference was observed in the perioperative outcomes or radiographic parameters at the final follow-up. There were no significant differences among the 3 groups in the total, major or neurological complication rates (all $P > .05$). Two patients (1 in the N group and 1 in the A group) experienced transient neurological complications, whereas no patient experienced permanent neurological deficits during surgery or follow-up.

To our knowledge, the current study reported the largest cohort of intraspinal anomalies in patients with CS that has been reported in the literature. The results of our study demonstrated that patients with congenital scoliosis associated with intraspinal anomalies, even multiple intraspinal anomalies that coexist with more complex intraspinal pathologies, may safely and effectively achieve scoliosis correction without preliminary neurological intervention. More complex intraspinal pathologies do not seem to increase the risk of neurosurgical complications during corrective surgery.

Abbreviations: CS = congenital scoliosis, MC = major coronal, SCM = split cord malformation.

Keywords: congenital scoliosis, multiple intraspinal anomalies, intraspinal pathology

1. Introduction

Congenital scoliosis occurs due to abnormal development of the vertebrae (failure of formation and/or segmentation) and is frequently associated with intraspinal anomalies.[1–3] The incidence of intraspinal anomalies reported in patients with congenital scoliosis ranges from 15% to 38%.[4–7] These intraspinal anomalies include diastematomyelia, syringomyelia, tethered spinal cord, Chiari malformation and other findings. Furthermore, some patients have multiple anomalies.[2,3]

The treatment of congenital scoliosis associated with intraspinal anomalies has not reached a consensus, mainly because the intraspinal anomalies may increase the risk of corrective surgery in CS patients.[4,5,8,9] In the past, to prevent potential neurologic complications, most surgeons have agreed that intraspinal anomalies need to be addressed before the treatment of scoliosis because of the increased risk of neurologic deterioration during scoliosis correction. The classic advocated approach in such patients is first to perform surgery for intraspinal pathologies and then to perform surgery for correction and stabilization of the deformity 3 to 6 months later; alternatively, simultaneous surgical treatment for the congenital deformity and intraspinal anomalies can be used.[5,10–15] However, as the pathology of intraspinal anomalies has become better understood in recent years, some investigators have put forward other ideas. Zhang et al described 29 CS patients with syringomyelia and coexisting diastematomyelia and/or tethered cord who were treated safely without neurosurgical intervention.[16] Shen et al reported a large cohort of 214 CS patients with split cord malformation and demonstrated that patients with CS associated with split cord malformation safely and effectively achieved spinal deformity correction without neurological intervention.[17] Tao et al reviewed 41 patients with congenital spinal deformity and tethered cord and concluded that it was possible to correct congenital spinal deformity associated with tethered cord without prophylactic intradural detethering.[18] Nonetheless, according to the literature, only 1 or 2 specific intraspinal anomalies are typically assessed. Theoretically, concurrent multiple intraspinal anomalies (coexistence of 3 or more intraspinal anomalies including syringomyelia, split cord malformation [SCM], tethered cord, low conus, intraspinal mass, Chiari malformation or/and arachnoid cyst) can increase the risk of neurologic injury during surgical correction in CS patients compared with the normal intraspinal condition or CS patients with 1 or 2 intraspinal anomalies due to the presence of more complex intraspinal pathologies.[5,10–15]

To our knowledge, no study has been published that has identified the risk of scoliosis correction and clinical outcomes in congenital scoliosis with multiple intraspinal anomalies. We therefore sought to determine

1. if multiple intraspinal anomalies increase the risk of scoliosis correction compared to the normal intraspinal condition or 1 or 2 intraspinal anomalies in congenital scoliosis (CS) and
2. whether multiple intraspinal anomalies need to be performed with preliminary neurosurgical intervention before scoliosis correction.

2. Methods

2.1. Patients and groups

CS patients undergoing corrective surgery without preliminary neurosurgical intervention in a single institution from January 2008 to June 2016 were retrospectively reviewed. The inclusion criteria included the following:

1. patients diagnosed with CS;
2. patients who were neurologically intact or stable over the past 2 years before corrective surgery;
3. patients without preliminary neurosurgical intervention for intraspinal anomalies;
4. complete medical records; and
5. patients with a minimum of 2 years of follow-up.

The exclusion criteria included the following:

1. patients who underwent previous spinal surgery; and
2. patients treated with vertebral column resection (VCR), because of the high risk of the procedure itself.

Each patient's perioperative and radiographic data as well as complication information were extracted.

Previous studies have demonstrated that patients with congenital scoliosis associated with 1 or 2 specific intraspinal anomalies may safely and effectively achieve spinal deformity correction without neurological intervention.[16,17,19–22] In this study, some patients with congenital scoliosis were found to have multiple intraspinal anomalies. Therefore, patients were assigned to 3 groups according to different intraspinal conditions. In the normal group (N group; n=196), patients did not have intraspinal anomalies. In the abnormal group (A group; n=93), patients had 1 or 2 intraspinal anomalies. In the multiple anomaly group (M group; n=29), patients had 3 or more intraspinal anomalies including syringomyelia, split cord malformation [SCM], tethered cord, low conus, intraspinal mass, Chiari malformation or/and arachnoid cyst.

2.2. Surgical strategy

One senior surgeon from a single institution performed all surgeries. In this study, pedicle subtraction osteotomy was performed in 27 patients; Ponte osteotomy was performed in 16 patients; hemivertebral resection was performed in 31 patients; growing rods were placed in 10 patients; and posterior correction and fusion without osteotomy was performed in the other 234 patients. All patients underwent corrective surgery under combined motor-evoked potential (MEP) and somatosensory evoked potential (SEP) monitoring without preliminary neurosurgical intervention, and a wake-up test was routinely administered during the operation.

2.3. Perioperative and radiographic data

Perioperative data included patients' age and sex as well as surgery time, intraoperative blood loss, total number of blood transfusions, length of hospital stay, and cost of the procedure.
Radiographic parameters of interest included the

1. Cobb angle of the main major coronal (MC) curves;
2. apical vertebral rotation (AVR, Nash-Moe);
3. apical vertebral translation (AVT);
4. coronal balance (CB);
5. thoracic trunk shift (TTS);
6. clavicle angle (CA);
7. C7-S1 sagittal vertical axis (SVA): distance between the C-7 plumb line and the posterosuperior sacrum;
8. thoracic kyphosis (TK): T5-12 angle (positive indicates kyphosis); and

Table 1

The baseline characteristics of the 3 groups of patients.

Variable	N Group (n=196)	A Group (n=93)	M Group (n=29)	P value (N vs A)	P value (N vs M)	P value (A vs M)
Flexibility(%)	35.2 ± 1.3	35.0 ± 1.7	37.0 ± 3.5	.954	.596	.596
MC Cobb (degrees)	64.1 ± 1.4	64.8 ± 2.2	66.8 ± 4.6	.781	.499	.639
AVR(Nash-Moe)	2.8 ± 0.1	2.8 ± 0.1	2.7 ± 0.2	.832	.334	.303
AVT(mm)	6.3 ± 3.9	8.1 ± 5.9	-7.1 ± 14.6	.801	.251	.221
CB(mm)	-2.6 ± 1.7	5.5 ± 2.5	10.4 ± 5.5	.009	.008	.347
TTS(mm)	3.6 ± 1.7	7.0 ± 3.0	3.8 ± 6.0	.317	.976	.573
CA(degrees)	-5.0 ± 3.1	0.2 ± 0.4	0.9 ± 0.7	.237	.393	.921
SVA(mm)	-8.8 ± 2.6	-17.7 ± 4.4	-7.3 ± 6.5	.067	.846	.205
TK(degrees,T5-T12)	38.6 ± 1.7	37.8 ± 2.4	37.2 ± 4.1	.767	.766	.918
LL(degrees,T12-S1)	55.1 ± 1.5	58.5 ± 1.5	54.0 ± 2.9	.064	.690	.141

N = normal, A = abnormal; M = multiple anomaly, MC = indicates major coronal curves, AVT = apical vertebra translation, AVR = apical vertebral rotation, CB = coronal balance, TTS = thoracic trunk shift, CA = clavicle angle, SVA = C7–S1 sagittal vertical axis, TK = thoracic kyphosis, LL = lumbar lordosis.

9. lumbar lordosis (LL): T12-S1 angle (positive indicates lordosis) preoperation, early postoperation (2 weeks after operation), and at the latest follow-up.

The flexibility index, the correction rate of the MC curve, the change values of the MC curve, and the loss values of the MC curve were calculated from the above radiographic parameters.

2.4. Complications

Complications were classified as neurologic complications or nonneurologic complications. Major complications were defined as complications that prompted active medical intervention or return to the operating room. For example, wound infection, debridement surgery and revision surgery were considered major complications.

2.5. Statistical analysis

All data were collected and analyzed using SPSS 17.0 for Windows. For multiple comparisons across the 3 groups, a 1-way analysis of variance and the Pearson Chi-Squared analysis were adopted if the distributions of the data conformed to normality and if there was equality of variances; if not, a Kruskal–Wallis rank-sum test was used. Further analysis was performed with the Bonferroni test. A P value $<.05$ was considered statistically significant.

3. Results

The baseline characteristics of the 3 groups are presented in Table 1. A total of 318 consecutive CS patients (200 females and 118 males) were carefully reviewed. The latest follow-up averaged 3.1 years (range, 2–8 years). The comparison analysis showed no significant differences among the 3 groups in terms of the baseline parameters.

The perioperative outcomes of the 3 groups of patients are presented in Table 2. In the N group, there were 112 females and 84 males, and the mean age at the time of surgery was 14.6 ± 0.7 years. In the A group, there were 63 females and 30 males, and the mean age at the time of surgery was 15.7 ± 0.7 years (Fig. 1 and Fig. 2). In the M group, there were 25 females and 4 males, and the mean age at the time of surgery was 16.5 ± 1.2 years (Fig. 3 and Fig. 4). No significant differences were observed among the 3 groups in terms of hospital stay, surgery time, blood loss, blood transfusion, or hospital charges.

The radiographic outcomes early postoperation and at the latest follow-up are presented in Table 3. Normal, abnormal, and multiple intraspinal conditions all achieved satisfactory deformity correction, and the values of the final MC curves were 35.0, 37.1, and 38.0, respectively. No significant differences were found among the 3 groups regarding the Cobb angle of the MC curve, the change values of the MC curve, the correction rate of the MC curve, the loss of the MC curve, apical vertebral rotation, apical vertebra translation, thoracic trunk shift, clavicle angle, C7-S1 sagittal vertical axis, thoracic kyphosis, or lumbar lordosis, including early postoperation and at the latest follow-up.

The details of the coexisting intraspinal pathologies are presented in Table 4. In this study, 122 (38.4%) patients were found to have intraspinal anomalies (93 patients in the A group and 29 patients in the M group), which consisted of syringomyelia in 69 (21.7%) patients, split cord malformation in 65 (20.4%) patients (22 patients with type 1 SCM and 43 patients with type 1 SCM), tethered cord in 41 (12.9%) patients, arachnoid cyst in 19

Table 2

The perioperative outcomes of the 3 groups of patients.

Variable	N Group (n=196)	A Group (n=93)	M Group (n=29)	P value (N vs A)	P value (N vs M)	P value (A vs M)
Sex(F/M)	112/84	63/30	25/4	–	–	–
Age(years)	15.4 ± 0.5	15.6 ± 0.7	16.3 ± 1.0	.814	.506	.629
Hospital stay(day)	22.1 ± 0.8	26.0 ± 2.6	25.4 ± 3.5	.069	.335	.860
Surgery time(minutes)	321.1 ± 7.0	312.4 ± 8.5	317.8 ± 16.6	.456	.855	.786
Blood loss(ml)	1177.5 ± 65.7	1224.9 ± 111.4	1223.1 ± 159.4	.696	.812	.993
Blood transfusion(ml)	1247.7 ± 69.4	1170.1 ± 80.3	1407.4 ± 181.1	.503	.383	.226
Charges($)	16164.9 ± 409.0	16067.0 ± 641.1	14600.7 ± 870.7	.893	.175	.234

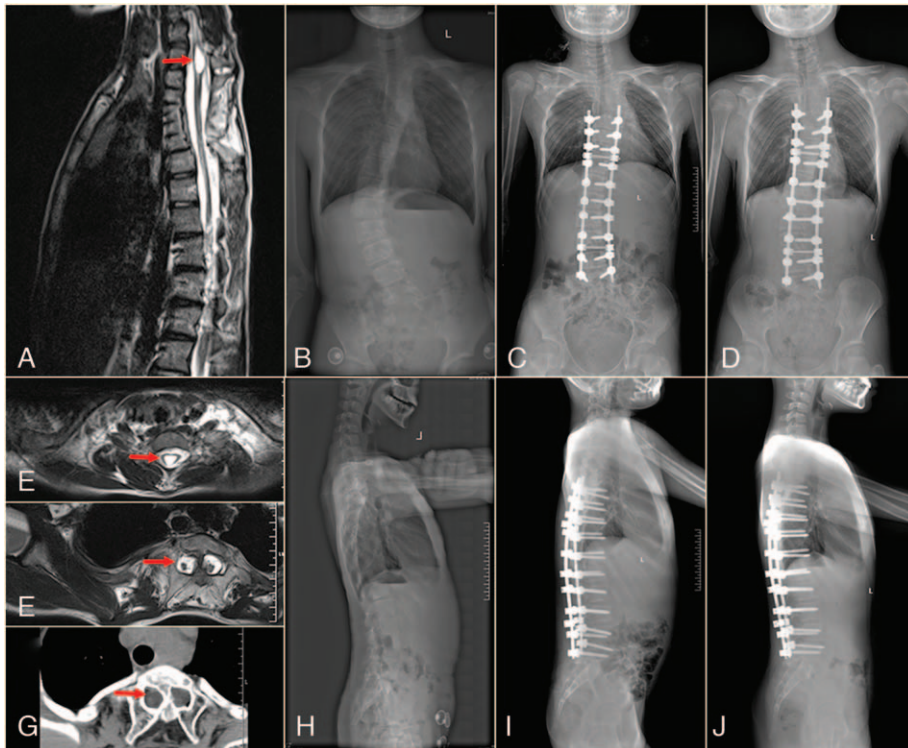N = normal, A = abnormal, M = multiple anomaly.

Figure 1. A 11-year-old girl with congenital scoliosis and 2 intraspinal anomalies (coexisting syringomyelia and type 1 split cord malformation[SCM]) received posterior correction and instrumentation without preliminary neurosurgical intervention. No neurologic deficits were found before or after surgery. (A) A syringomyelia on sagittal magnetic resonance imaging. (E,F) Syringomyelia and type 1 SCM on axial magnetic resonance imaging. (G) A consecutive osseous spur on axial CT imaging. Preoperative anteroposterior (B) and lateral (H) views. Postoperative anteroposterior (C) and lateral (I) views. Final follow-up anteroposterior (D) and lateral (J) views.
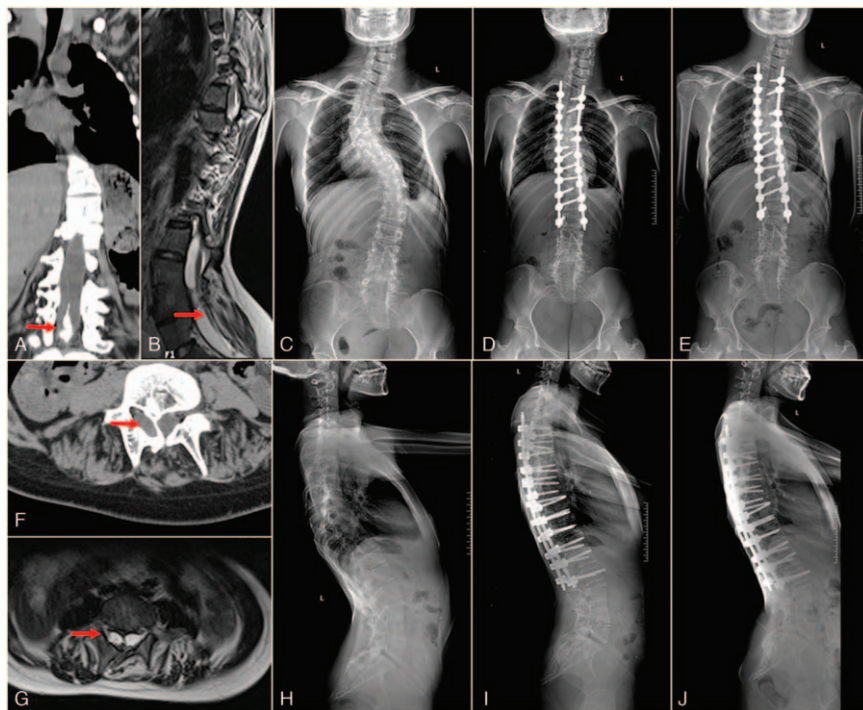
Figure 2. A 18-year-old girl with congenital scoliosis and 2 intraspinal anomalies (coexisting type 1 split cord malformation[SCM] and tethered cord) received posterior correction and instrumentation without preliminary neurosurgical intervention. No neurologic deficits were found before or after surgery. (A) An osseous spur on coronal CT imaging. (B) A tethered cord on sagittal magnetic resonance imaging. (F) A consecutive osseous spur on axial CT imaging. (G) A consecutive osseous spur with the owl sign on axial MRI imaging. Preoperative anteroposterior (C) and lateral (H) views. Postoperative anteroposterior (D) and lateral (I) views. Final follow-up anteroposterior (E) and lateral (J) views.

Figure 3. A 14-year-old girl with congenital scoliosis and 3 intraspinal anomalies (coexisting syringomyelia, tethered cord and type 2 split cord malformation[SCM]) received posterior correction and instrumentation without preliminary neurosurgical intervention. No neurologic deficits were found before or after surgery. (A) Syringomyelia and tethered cord on sagittal magnetic resonance imaging. (E,F) Syringomyelia and type 2 SCM on axial magnetic resonance imaging. Preoperative anteroposterior (B) and lateral (G) views. Postoperative anteroposterior (C) and lateral (H) views. Final follow-up anteroposterior (D) and lateral (I) views.

(6.0%) patients, low conus in 18 (5.7%) patients, intraspinal mass in 7 (2.2%) patients, Chiari malformation in 4 (1.3%) patients, and myelomeningocele in 4 (1.3%) patients. Intraspinal masses (including 4 lipomas, 2 teratomas, and 1 epidermoid cyst) were located in the lower thoracic and lumbar spine. Of the 93 patients in the A group, 55 patients had single intraspinal anomalies, and 38 patients had 2 types of intraspinal anomalies. Of 29 total patients in the M group, 20 of them had 3 types of intraspinal anomalies, and 9 had 4 types of intraspinal anomalies. Multiple intraspinal anomalies were found in 9.1% (29/318) of patients with congenital scoliosis.

Complications were recorded during the follow-up period after surgery and are summarized in Table 5. Although the total and major complication rates in the M group were higher than those in the other 2 groups, there were no statistically significant differences (both $P > .05$). In the current study, a total of 2 patients experienced transient neurological complications (including sensory deficits and weakness in the lower extremity): 1 in the N group and the other in the A group. Both patients completely recovered before hospital discharge. Among the other postoperative minor complications, pulmonary complications, including pneumonia, hydrothorax, and pneumothorax, were observed in 6 patients (4 patients in the N group and 2 patients in the A group). Cerebrospinal fluid leakage occurred in 5 patients (3 in the N group, 1 in the A group, and 1 in the M group), and all these patients recovered successfully after 4 to 7 days of drainage.

One of the patients was debrided and sutured because of poor healing. One patient in the M group had a rod breakage 31 months after corrective surgery, and a revision surgery was performed. Four patients developed wound infection and required additional surgery because the infection was a deep wound infection (2 patients in the N group, 1 patient in the A group and 1 patient in the M group). There were no significant differences among the 3 groups with regard to postoperative neurological complication rates ($P = .769$). None of the patients had pseudarthrosis or permanent complications.

4. Discussion

Multiple intraspinal anomalies in CS are still a great challenge due to the presence of more complex intraspinal pathologies. More complex intraspinal pathologies do not seem to increase the risk of corrective surgery but have not been fully identified. This study represents the largest series, to our knowledge, of intraspinal anomalies in the published literature and is the first to report the incidence of multiple intraspinal anomalies. In this study, we found that 9.1% (29/318) of patients had associated multiple intraspinal anomalies; then, we compared the complications as well as the perioperative and radiographic data of the normal, abnormal, and multiple types of different intraspinal conditions in patients with congenital scoliosis. The results indicated that patients with congenital scoliosis associated with
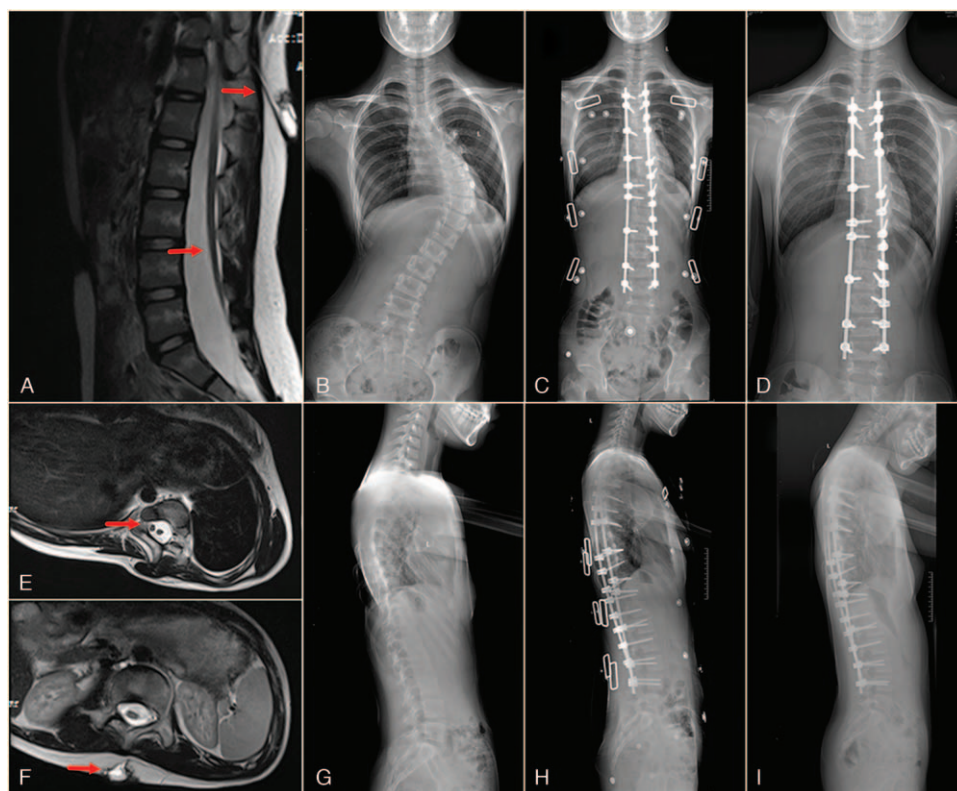
Figure 4. A 13-year-old girl with congenital scoliosis and 3 intraspinal anomalies (coexisting tethered cord, myelomeningocele and type 2 split cord malformation [SCM]) received posterior correction and instrumentation without preliminary neurosurgical intervention. No neurologic deficits were found before or after surgery. (A) Tethered cord and myelomeningocele on sagittal magnetic resonance imaging. (E,F) Type 2 SCM and myelomeningocele on axial magnetic resonance imaging. Preoperative anteroposterior (B) and lateral (G) views. Postoperative anteroposterior (C) and lateral (H) views. Final follow-up anteroposterior (D) and lateral (I) views.

Table 3

The radiographic outcomes of the 2 groups of patients.

Variable	N Group (n=196)	A Group (n=93)	M Group (n=29)	P value (N vs. A)	P value (N vs. M)	P value (A vs. M)
Early postoperation						
MC Cobb (degrees)	31.7±1.3	33.7±2.2	37.0±5.0	.466	.188	.434
Change of MC Cobb (degrees)	32.3±0.7	31.1±1.1	29.8±1.9	.334	.203	.535
Correction rate (%)	53.4±1.2	51.3±1.9	50.1±4.3	.362	.374	.770
AVR (Nash-Moe)	1.7±0.0	1.4±0.1	1.4±0.3	.009	.058	.829
AVT (mm)	4.4±2.5	0.8±4.3	−3.8±9.9	.457	.288	.580
CB (mm)	−6.9±1.4	−0.7±2.1	2.5±5.4	.019	.023	.464
TS (mm)	−2.5±1.2	−1.6±1.6	2.1±2.9	.636	.152	.288
CA (degrees)	−0.6±0.3	0.1±0.3	0.1±0.9	.163	.395	.976
SVA (mm)	1.3±2.7	5.4±4.2	1.7±5.5	.391	.956	.649
TK (degrees, T5-T12)	26.8±0.8	25.1±1.2	24.6±3.0	.261	.366	.857
LL (degrees, T12-S1)	48.5±0.8	47.1±1.1	48.0±0.6	.309	.838	.681
Latest follow-up						
MC Cobb (degrees)	33.8±1.3	35.6±2.1	39.4±4.9	.454	.144	.357
Change of MC Cobb (degrees)	30.3±0.7	29.6±1.0	27.4±1.7	.526	.119	.278
Correction rate (%)	49.6±1.1	48.6±1.8	46.2±3.8	.833	.637	.300
Loss of MC Cobb (degrees)	2.0±0.2	1.5±0.3	2.4±0.5	.160	.447	.123
AVR(Nash-Moe)	1.7±0.0	1.4±0.1	1.4±0.3	.01	.06	.829
AVT (mm)	2.8±2.3	-0.1±4.1	-5.0±10.1	.532	.291	.537
CB (mm)	−3.5±1.3	2.3±2.0	5.6±4.9	.02	.02	.438
TS (mm)	−0.7±1.1	1.1±1.6	3.8±3.0	.363	.148	.414
CA (degrees)	0.5±0.2	0.7±0.3	0.6±0.6	.442	.786	.841
SVA (mm)	-14.2±2.3	-2.1±4.1	-13.3±5.9	.006	.897	.131
TK (degrees, T5-T12)	29.2±1.0	27.2±1.3	27.4±3.1	.277	.543	.940
LL (degrees, T12-S1)	52.5±0.7	50.2±1.2	49.7±2.0	.085	.182	.820

N = normal, A = abnormal, M = multiple anomaly, MC = indicates major coronal curves, AVT = apical vertebra translation, AVR = apical vertebral rotation, CB = coronal balance, TTS = thoracic trunk shift, CA = clavicle angle, SVA = C7–S1 sagittal vertical axis, TK = thoracic kyphosis, LL = lumbar lordosis.

Table 4

Details of coexisting intraspinal pathologies in the A group and M group.

Intraspinal anomalies	No. of patients
A group(Patients with 1 or 2 intraspinal abnormalities; n = 93)	
Syringomyelia alone	23
Type 2 SCM alone	7
Type 1 SCM alone	5
Arachnoid cyst alone	12
Tethered cord alone	5
Low conus alone	2
Intraspinal mass alone	1
Type 2 SCM and tethered cord	8
Type 2 SCM and syringomyelia	8
Type 2 SCM and low conus	4
Syringomyelia and Chiari malformation	4
Type 1 SCM and syringomyelia	3
Type 1 SCM and tethered cord	2
Syringomyelia and tethered cord	2
Syringomyelia and low conus	2
Type 1 SCM and low conus	1
Syringomyelia and intraspinal mass	1
Syringomyelia and myelomeningocele	1
tethered cord and intraspinal mass	1
Arachnoid cyst and intraspinal mass	1
M Group(Patients with 3 or more intraspinal abnormalities; n = 29)	
Type 2 SCM, syringomyelia and tethered cord	7
Type 1 SCM, syringomyelia and tethered cord	3
Type 2 SCM, syringomyelia and low conus	4
Type 2 SCM, tethered cord and myelomeningocele	1
Type 2 SCM, tethered cord and arachnoid cyst	1
Type 1 SCM, syringomyelia and low conus	1
Type 1 SCM, syringomyelia and arachnoid cyst	1
Type 1 SCM, tethered cord and intraspinal mass	1
Tethered cord, arachnoid cyst and intraspinal mass	1
Type 2 SCM, syringomyelia, tethered cord and arachnoid cyst	2
Type 2 SCM, syringomyelia, tethered cord and intraspinal mass	1
Type 1 SCM, syringomyelia, tethered cord and low conus	4
Type 1 SCM, syringomyelia, tethered cord and myelomeningocele	1
Syringomyelia, tethered cord, arachnoid cyst and myelomeningocele	1

A = abnormal, M = multiple anomaly, SCM = split cord malformation.

intraspinal anomalies, even for multiple intraspinal anomalies that coexist with more complex intraspinal pathologies, may safely and effectively achieve spinal deformity correction without neurological intervention, and more complex intraspinal pathologies do not seem to increase the risk of corrective surgery.

In this study, 9.1% of patients were found to have multiple intraspinal anomalies. Several studies have demonstrated that congenital scoliosis is frequently associated with intraspinal anomalies, and the incidence associated with intraspinal anomalies among patients with CS is 15% to 38%.[2–7] In the literature, authors have only reported the incidence of each specific intraspinal anomaly and have not paid attention to or have reported the incidence of multiple intraspinal anomalies. Recently, an increasing number of studies have demonstrated that preliminary neurosurgical intervention for 1 or 2 intraspinal anomalies may be safely avoided in CS patients with intact or stable neurological status.[16–22] Clinically, some patients with congenital scoliosis have been found to have multiple intraspinal anomalies. Multiple intraspinal anomalies are widely considered to increase the neurological complications of corrective surgery in CS patients compared with CS patients with 1 or 2 coexisting intraspinal anomalies due to the presence of more complex intraspinal pathologies. Do those patients have an increased risk of scoliosis correction compared to the normal intraspinal condition or to CS patients with 1 or 2 intraspinal anomalies? Do corrections for multiple intraspinal anomalies need to be performed with preliminary neurosurgical intervention before scoliosis correction?

This study performed a multiple comparison of radiographic and perioperative outcomes among 3 different intraspinal conditions, and no significant differences were found in the correction rates of the major curve or other radiographic parameters. Similar conclusions have been reported in some studies examining the presence of 1 or 2 intraspinal anomalies. A study by Shen et al involved 214 CS patients with split cord malformation and concluded that major curve corrections in patients with type-I SCM and type-II SCM were 48.5% and 42.1%, respectively, and this result was comparable to the results of previous reports.[17] Zhao et al reviewed 241 patients with congenital scoliosis and reported comparable correction rates of the major curve between CS patients with intraspinal anomalies

Table 5

The complications of the 3 groups of patients.

Complications	N Group (n = 196)	A Group (n = 93)	M Group (n = 29)	P value
Total complications	12 (6.1%)	4 (4.3%)	3 (10.3%)	.483
Major complications	4 (2.0%)	2 (2.2%)	2 (6.9%)	.286
Neurological complications	1	1	0	.769
Transient neurological complications	1	1	0	.769
Permanent new neurological deficit	0	0	0	–
Any revision surgery	3	2	2	.184
Sensory deficit	1	1	0	.769
Claudication	0	0	0	–
Weakness in the lower extremity	1	1	0	.769
Nonneurological complications				
Implant failure	0	0	1	.07
Pulmonary complications	4	2	0	.734
Spinal fluid leakage	3	1	1	.667
Wound infection	2	1	1	.540

Total complications: any complication from the intraoperative period to final follow-up. Major complications: complications needing active medical intervention or return to the operating room.
N = normal, A = abnormal, M = multiple anomaly.

(SCM, tethered cord, and/or syringomyelia) and CS patients without any intraspinal anomalies (49.8% vs 51.3%, respectively, $P > .05$).[22] In the current study, although the correction rate was relatively lower in the multiple intraspinal anomaly group than in the normal and abnormal groups (45.3% compared with 48.5% and 47.7%, respectively; $P > .05$), the rates were still acceptable at the last follow-up. The normal, abnormal, and multiple different intraspinal conditions all provided satisfactory scoliosis correction.

Neurological complications represent a primary consideration for orthopedic surgeons. In the past, intraspinal anomalies in CS patients have been empirically thought to represent 1 critical risk factor for neurologic injury during surgical correction. Theoretically, concurrent multiple intraspinal anomalies may increase the risk of neurologic injury in CS patients compared to CS patients with 1 or 2 intraspinal anomalies due to the presence of more complex intraspinal pathologies. However, postoperative neurological complications in CS patients with multiple intraspinal anomalies have rarely been reported in the literature. Zhao et al reviewed 241 CS patients and reported that only 1 patient experienced transient neurological compromise in the CS-IA group (CS without intraspinal anomalies).[22] Shen et al reported that the rate of transient neurological complications after correction surgery in 214 CS patients with untreated SCM was 5.1% (11/214).[17] No permanent neurological complications occurred. In our study, in terms of complications, 2 patients experienced transient neurological complications, and the prevalence of postoperative neurological complications was similar among the 3 groups ($P > .05$). In our opinion, although multiple intraspinal anomalies have more complex intraspinal pathologies, there seemed to be no increase in neurological complications. Based on this, we further confirmed that patients with congenital scoliosis associated with intraspinal anomalies, even multiple intraspinal anomalies, may safely and effectively achieve spinal deformity correction without preliminary neurological intervention.

Although there have been many studies reporting the treatment of CS associated with 1 or 2 intraspinal anomalies, to our knowledge, there is no formal study in the literature involving the surgical treatment of multiple intraspinal anomalies in CS. In our opinion, there are 2 reasons for this result. The first is the low incidence of multiple intraspinal anomalies in congenital scoliosis. Although previously documented in multiple intraspinal anomalies, no explicit incidence has been reported.[2,3] The results of our study first indicated that 9.1% of patients with congenital scoliosis were found to have multiple intraspinal anomalies. The second reason is the lack of research on the nature of intraspinal anomalies, which coexist with more complex intraspinal pathologies and thus make treatment controversial. This study indicates that patients with congenital scoliosis associated with intraspinal anomalies may safely and effectively achieve spinal deformity correction without neurological intervention.

Some limitations should not be ignored in this study. First, multiple intraspinal anomalies have the 2 inherent characteristics of great variability and complex relationships among each malformation compared to 1 or 2 coexisting intraspinal anomalies. The mechanism for the occurrence of neurological deterioration remains unclear. Second, as a retrospective single-center study, the evidence level was limited compared with a prospective or multicenter trial; thus, further prospective and multicenter trials are needed.

5. Conclusion

This study represents the largest series, to our knowledge, of intraspinal anomalies in the published literature and is the first to report the incidence of multiple intraspinal anomalies. Multiple intraspinal anomalies were rarely found (9.1%) in congenital scoliosis. The results of our study challenged the traditional surgical strategy for the management of intraspinal anomalies and demonstrated that patients with congenital scoliosis associated with intraspinal anomalies, even multiple intraspinal anomalies that coexist with more complex intraspinal pathologies, may safely and effectively achieve spinal scoliosis correction without neurological intervention. More complex intraspinal pathologies do not seem to increase the risk of corrective surgery.

Author contributions

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