

Single-group Study to Explore the Optimal Management of Neuropathic Scoliosis Caused by Neural Axis Abnormalities Based on its Clinical Features

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Abstract: Although it is not easy to differentiate neuropathic scoliosis caused by neural axis abnormalities (NAAs) from idiopathic scoliosis for lack of neurological signs, neuropathic scoliosis caused by NAAs has some specific clinical findings. The purpose of this study was to investigate the characteristics of neuropathic scoliosis caused by NAAs and to discuss the essential points of diagnosis and treatment. From 1998 through 2008, 622 new patients visited our scoliosis outpatient clinic. Of those, 12 patients with neuropathic scoliosis caused by NAAs diagnosed by cervical MRI were involved in this study. They included 5 boys and 7 girls with a mean age of 10 years and 5 months. We investigated the relationship of NAAs, treatment methods for scoliosis, and effects of neurosurgical treatment for prognosis of outcome. There were 10 cases of syringomyelia with Chiari malformation type I, 1 case of syringomyelia alone, and 1 case of spinal cord tumor. Surgery was performed in 3 cases, an under arm brace was prescribed in 6 cases, and observation was selected in 3 cases. Scoliotic curves were improved in 4 out of the 11 cases on whom neurosurgical intervention for the relationship of NAAs were performed. The Cobb angles before surgery of these 4 cases were less than 30° and 3 of the 4 cases were 8 years old or less. We would emphasize that early treatment for the relationship of NAAs at younger age (≤ 8 yrs) and before curve progression ($< 30^\circ$) should be actively enforced for favorable prognosis of neuropathic scoliosis caused by NAAs.

Keywords: abdominal skin reflex, Chiari malformation, magnetic resonance imaging (MRI), neural axis abnormality, neuropathic scoliosis, syringomyelia.

INTRODUCTION

Neuropathic scoliosis caused by neural axis abnormalities (NAAs) often lacks neurological signs and expresses the spinal deformity as the only symptom [1-3]. Accordingly, it is not easy to differentiate neuropathic scoliosis caused by NAAs from idiopathic scoliosis. However, neuropathic scoliosis caused by NAAs has some specific clinical findings, for example; abnormality of abdominal skin reflex [1,4], infantile or juvenile onset [5-8], atypical curve pattern [1, 9-11], thoracic hyperkyphosis [12] and so on.

The purpose of this retrospective study was to investigate the characteristics of neuropathic scoliosis caused by NAAs and to discuss the essential points of diagnosis and treatment for such kind of pathological condition.

MATERIALS AND METHODS

From 1998 through 2008, 622 new patients visited our scoliosis outpatient clinic. Of those, 12 patients (1.9%) with neuropathic scoliosis caused by NAAs diagnosed by cervical MRI were involved in this study. They included 5 boys and 7 girls ranging in age from 5 years 4 months to 17 years 1 month, with a mean age of 10 years and 5 months.

We investigated the relationship of NAAs, detection of scoliosis, treatment methods for scoliosis and the clinical course, surgeries for NAAs and their effects for scoliosis, and clinical features; including ages of scoliosis detection, gender, curve pattern represented by King classification [13], curve magnitude assessed by Cobb angle, thoracic kyphosis measured between T2 and T12, and neurological findings. Further, 272 cases of idiopathic scoliosis patients, whose Cobb angles were more than 25°, detected during same period were set up as a control group. Then, clinical features were compared between neuropathic scoliosis caused by NAAs and idiopathic scoliosis. The clinical course of the treatment for scoliosis was considered progression if more than 5° curvature increase occurred and improvement if more than 5° curvature decrease occurred. Statistical analysis was performed using a two-tailed unpaired *t*-test and Chi square test. $P < 0.05$ level was considered statistically significant.

Written informed consent was obtained from the patients for publication of this report. And all procedures were in accordance with the Helsinki declaration.

RESULTS

There were 10 cases of syringomyelia with Chiari malformation type I, 1 case of syringomyelia alone, and 1 case of spinal cord tumor. With respect to detection of scoliosis, 5 cases were pointed out by other persons, 3 cases were discovered by school screening, and 4 cases were indicated

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Table 1. Clinical Data of Each Patient

Case	Age at Scoliosis Diagnosis (yr + mo)	Gender	Scoliosis Pattern (King)	NAAs	Occasion of Scoliosis Detection	Treatment for Scoliosis (Initial)	Treatment for Scoliosis (Final)
2	12 + 8	F	III	Chiari type I & syrinx	pointed out by parents	surgery	surgery
3	8 + 1	M	R-III *	spinal tumor	nonscheduled med exam	observation	observation
4	5 + 11	F	R-I *	Chiari type I & syrinx	pointed out by parents	brace	brace
5	5 + 8	F	III	Chiari type I & syrinx	nonscheduled med exam	brace	surgery
6	7 + 11	F	III	Chiari type I & syrinx	nonscheduled med exam	brace	surgery **
7	17 + 1	M	R-III *	Chiari type I & syrinx	school screening	surgery	surgery
8	15 + 8	F	IIB	Chiari type I & syrinx	school screening	surgery	surgery
9	11 + 3	F	R-III *	syrinx	school screening	brace	surgery
10	14 + 3	M	R-III *	Chiari type I & syrinx	pointed out by parents	observation	observation
11	5 + 7	F	III	Chiari type I & syrinx	nonscheduled med exam	observation	brace
12	5 + 4	M	V	Chiari type I & syrinx	pointed out by parents	brace	brace

NAAs: neural axis abnormalities

* R: atypical inverted curve (left thoracic, right lumbar)

** case 6: growing rod

Table 1. contd...

Case	Surgery for NAAs	Effect of Surgery for NAAs to Scoliosis	Neurological Findings		Thoracic Kyphosis (°)
			Abdominal Skin Reflex	Neurological Symptom	
1	FMD+SS shunt	none	-- / --	motor dysfunction of right arm	27
2	FMD	N/A	++ / ++	none	34
3	tumorectomy	yes	++ / ++	acute left hemiplegia	35
4	FMD+SS shunt	yes	-- / ++	gait abnormality	37
5	FMD+SS shunt	none	-- / ++	none	38
6	FMD	none	-- / --	none	43
7	FMD+SS shunt	N/A	++ / --	none	48
8	FMD	N/A	-- / --	none	44
9	none	N/A	++ / --	none	41
10	FMD	yes	+ - / --	none	37
11	FMD	yes	-- / ++	none	54
12	FMD	none	++ / --	none	47

N/A: surgery for scoliosis was performed within 1 year after surgery for original NAAs

Table 1. contd...

Case	Curve Magnitude (°)				Length of Follow-Up after Surgery for NAAs	Length of Follow-Up after Surgery for Scoliosis	
	At First Visit	Before Surgery for NAAs	After Surgery for NAAs	Before Surgery for Scoliosis			At Latest Visit
1	40	55	68	N/A	68	6y3m	N/A
2	66	95	98	98	46	3.5m	4y

Table 1. contd....

Case	Curve Magnitude (°)					Length of Follow-Up after Surgery for NAAs	Length of Follow-Up after Surgery for Scoliosis
	At First Visit	Before Surgery for NAAs	After Surgery for NAAs	Before Surgery for Scoliosis	At Latest Visit		
3	20	20	12	N/A	12	5y8m	N/A
4	27	27	3	N/A	3	12y3m	N/A
5	30	30	64	64	24	8y10m	1y6m
6	45	40	67	67	32	3y2m	1y
7	70	70	68	68	47	2.5m	4y3m
8	90	90	90	90	64	10m	5y
9	41	N/A	N/A	63	27	N/A	1m
10	28	28	23	N/A	23	4y	N/A
11	24	24	19	N/A	19	3y5m	N/A
12	48	48	56	N/A	56	2y10m	N/A

Table 2. Summary of Clinical Features of Neuropathic Scoliosis Caused by NAAs

Clinical Features	Rate of Abnormality
Abnormality of abdominal skin reflex *	10 / 12 (83.3%)
Atypical inverted curve pattern	5 / 12 (41.7%)
Younger age (less than 10 yrs)	6 / 12 (50.0%)
Severe scoliosis (40 degrees or more)	7 / 12 (58.3%)
Neurological symptom **	3 / 12 (25.0%)

* laterality: 7 cases / loss: 3 cases

** motor dysfunction of right arm, acute left hemiplegia, gait abnormality

NAAs: neural axis abnormalities

Table 3. Comparison Between Neuropathic and Idiopathic Scoliosis

	Neuropathic	Idiopathic	Statistics
Age (ave.)	10 yr 5 mo	13 yr 10 mo	P<0.001*
Gender (M / F)	5 / 7	22 / 250	P<0.001**
Rate of female	58.3%	91.9%	
Cobb angle	44.1 ± 21.4°	35.2 ± 11.0°	P<0.001*
Thoracic kyphosis	40.0 ± 8.2°	24.9 ± 10.6°	P<0.001*

*unpaired t-test, **chi square test

by nonscheduled medical examination. With regard to initial treatment methods for scoliosis and the clinical course, surgery was performed in 3 cases, an under arm brace was prescribed in 6 cases, and observation was selected in 3 cases. All 4 patients who started brace treatment at 40° and over became worse. Finally, 3 cases initially received brace treatment went to surgery and 1 case under observation switched to brace treatment due to worsening of curve magnitude. Surgeries for the relationship of NAAs were carried out for all cases, except a case of syrinx alone. Scoliotic curves were improved in 4 out of the 11 cases undergoing neurosurgical intervention. The Cobb angles measurement at initial presentation for these 4 cases were less than 30° (Table 1). Moreover, 3 of the 4 cases were 8

years old or less. As for the typical findings of neuropathic scoliosis caused by NAAs, 10 cases showed the abnormalities of abdominal skin reflex, 5 cases indicated an atypical inverted curve patterns, 6 cases were diagnosed before 10 years old, 7 cases had severe curves over 40°, 3 cases had neurological symptoms, and 6 cases had thoracic hyperkyphosis over 40° (Table 2).

The results of comparing about clinical features between neuropathic scoliosis caused by NAAs and idiopathic scoliosis patients is recorded in Table 3. Neuropathic scoliosis caused by NAAs patients included 5 boys and 7 girls with a mean age of 10 years and 5 months. Idiopathic scoliosis patients included 22 boys and 250 girls with a mean age of 13 years and 10 months (ranging in age from 4 years

Table 4. Rate of NAAs by Screening Cervical MRI

Reasons for Screening Cervical MRI	No of Cases	YES	NO	Rate of Abnormality
Progressive curve ($40^\circ <$)	30	7	23	23.3%
Younger age (10yr $>$)	17	6	11	35.3%
Abnormality of abdominal skin reflex	17	10	7	58.8%
Unusual inverted curve pattern	29	6	23	20.7%
Triple curve pattern	9	0	9	0.0%
Neurological sign (numbness, paralysis)	3	3	0	100.0%
Others (headache, progression after maturity)	4	0	4	0.0%
All cases	78	12	66	18.2%

NAAs: neural axis abnormalities

7 months to 30 years 4 months). Average initial Cobb angles of neuropathic scoliosis caused by NAAs and idiopathic scoliosis patients were $44.4 \pm 21.4^\circ$ and $35.2 \pm 11.0^\circ$, respectively. Average thoracic kyphosis of neuropathic scoliosis caused by NAAs and idiopathic scoliosis patients were $40.0 \pm 8.2^\circ$ and $24.9 \pm 10.6^\circ$, respectively. The neuropathic scoliosis group was younger in age, lower rate of female, predominant more in degree of magnitude, and had larger thoracic kyphosis than idiopathic scoliosis.

DISCUSSION

In the treatment of neuropathic scoliosis caused by NAAs, intervention in the relationship of NAAs should be given preference over that in the spinal deformity because neuropathic disorder is the main cause of scoliosis. Cardoso *et al.*, [14] reported that a Chiari decompression leads to a reduction or resolution of the syrinx and may result in a concomitant improvement in scoliosis. Zadeh *et al.* [4] also stated that the primary and the most important step in the successful treatment of scoliosis with syringomyelia is early surgical decompression of the syrinx. Therefore, although it is critical to differentiate neuropathic scoliosis caused by NAAs from idiopathic scoliosis owing to lack of neurological signs, we should make efforts to accomplish early detection of scoliosis attributed to neuropathic disorders by turning up some specific clinical findings in neuropathic scoliosis caused by NAAs.

Abnormality of abdominal skin reflex is one of the most important features in neuropathic scoliosis caused by NAAs. Arai *et al.*, [1] reported that abnormalities of abdominal skin reflex were shown in 36 of the 43 (83.7%) of scoliosis patients associated with syringomyelia. Zadeh *et al.* [4] indicated that syringomyelia was discovered by MRI screening in 10 of the 12 (83.3%) scoliosis patients who were loss of an abdominal skin reflex. In our clinical experience, spinal cord disorders were similarly discovered in 10 of the 17 (58.8%) scoliosis patients who had abnormality of an abdominal skin reflex (Table 4) [15].

Most scoliosis patients presenting NAAs tend to be infantile or juvenile. Gupta *et al.* [5] reported that there were spinal cord disorders in 6 of the 34 (17.6%) of juvenile cases and in 3 of the 6 (50.0%) of infantile cases. Lewonowski *et al.* [6] mentioned that in 26 patients with idiopathic scoliosis

under 11 years of age without neurological signs, MRI showed abnormal intraspinal pathology with Chiari malformation type I in 5 (19.2%). Dobbs *et al.* [7] recommended a total spine MRI evaluation at the time of presentation for all patients with infantile idiopathic scoliosis who have a curve measuring $\geq 20^\circ$ because 10 of the 46 (21.7%) cases with an age of 3 years or less were found to have a NAA on MRI. Evans *et al.* [8] also stated that MRI of all patients with scoliosis of juvenile onset should be performed on the basis of their prospective study in which 8 of the 31 patients aged 4-12 years cases (25.8%) had evidence of NAAs. In our current study, patients with neuropathic scoliosis caused by NAAs were younger than those with idiopathic scoliosis (Table 3).

Furthermore, atypical curve pattern, including left thoracic curve is an additionally characteristic feature in neuropathic scoliosis caused by NAAs. Arai *et al.* [1] reported that in their series, 15 of the 43 (34.9%) scoliosis patients associated with syringomyelia had left thoracic curve. Muhonen *et al.* [11] indicated that 4 of the 11 (36.4%) scoliosis patients with pediatric Chiari malformation exhibited a left thoracic curvature pattern. Charry *et al.* [9] reported 10 of the 25 (40.0%) patients with scoliosis and associated syringomyelia had left thoracic curvatures which did not resemble typical idiopathic scoliosis. Coonrad *et al.* [10] advocated that the left thoracic curve in itself might indicate the presence of intraspinal abnormalities. In our clinical experience, 6 of the 29 (20.7%) scoliosis patients with unusual inverted curve pattern showed abnormalities of spinal cord by cervical MRI (Table 4) [15]. However, Diabs *et al.*, [12] recently reported that the thoracic hyperkyphosis ($>40^\circ$) and juvenile onset (≤ 10 yrs) were risk factors for NAAs, whereas, apex left thoracic curve was not recognized as an independent predictor of NAAs. Certainly, our results showed that neuropathic scoliosis caused by NAAs was younger age with larger thoracic kyphosis than idiopathic scoliosis. But it is still debatable with regard to the relation between the direction of the apex and the presence of NAAs.

Based on these characteristics conducted from previous and current studies, for early detection of neuropathic scoliosis caused by NAAs, we routinely perform a screening cervical MRI for the patients with scoliosis who meet the following conditions; under 10 years old, abnormal of

abdominal skin reflex, atypical curve pattern, and progressive scoliotic curve over 45°.

The other points for early diagnosis of original NAAs related to scoliosis besides MRI screening are education of pediatricians about neuropathic scoliosis caused by NAAs and its features because they may have more opportunities to identify these younger scoliosis patients than spine surgeons. A more liberal use of MRI screening for all suspected younger idiopathic scoliosis patients may be effective. Winter *et al.* [16] and Do *et al.*, [17] stated that routine MRI evaluation of neurologically normal adolescents with idiopathic scoliosis is not warranted from their clinical study and that few patients were noted to have an abnormality on MRI and no patient required a neurosurgical intervention or further evaluation. However, Cheng *et al.* [18] emphasized that some adolescent idiopathic scoliosis patients who had progressive deformity may have asymptomatic spinal disorders regardless of an age of onset.

Meanwhile, not only prompt detection but also early intervention in the relationship of NAAs is mandatory for favorable prognosis of neuropathic scoliosis. Eule *et al.* [19] reported that early decompression of Chiari malformation type I with syringomyelia and scoliosis resulted in improvement or stabilization of the spinal deformity in 5 of the 5 (100%) cases who underwent decompression before 8 years of age and before the curve was severe. Charry *et al.*, [9] described that 5 of the 8 neuropathic scoliosis cases underwent neurosurgical treatment at age 8 or younger had no worsening or improvement of their scoliosis. Muhonen *et al.* [11] also reported on two scoliosis patients with Chiari malformation under 10 years of age who had resolution of their scoliosis, despite preoperative curves measurements of more than 40°. Flynn *et al.* [20] identified that progression of spinal deformity despite neurosurgical management of Chiari malformation was noted with older age at neurosurgical decompression and initial neuralgic symptoms, double scoliosis curve pattern, kyphosis, rotation, and large curve presentation. In our study, spinal deformities of the 4 cases on whom surgeries for the relationship of NAAs were performed at the Cobb angles less than 30° were improved. Further, 3 of the 4 cases were 8 years old or less. On the basis of these facts, we presume early treatment for NAAs at young age below 8 years old and before curve progression under 30° are essential points for desirable prognosis of neuropathic scoliosis caused by NAAs. With respect to treatment of scoliosis due to syringomyelia alone or syringomyelia associated with Chiari malformation, Phillips *et al.*, [21] advocated bracing as a temporizing measure to delay a posterior spinal fusion, and Farley *et al.* [22] stated bracing was not effective in preventing curve progression. Therefore, corrective surgery may be warranted in management of scoliosis. In the cases of corrective surgery, they should be performed after neurosurgical treatment because forceful surgical correction of scoliosis in association with syringomyelia, without treating the underlying pathology, is potentially disastrous. There have been some reports of paraplegia and quadriplegia after a posterior spinal fusion without neurosurgical intervention [23-26]. Phillips *et al.* [21] suggested that intraoperative neuropathic injury might be due to direct compression on the dilated syrinx, vascular compromise from traction on the spinal cord, cerebrospinal fluid pressure changes, and

adhesions. Diabs *et al.* [12] stated in his report that there were no neural and other complications in surgical treatment of neuropathic scoliosis if neural examination was normal. This is a highly controversial issue because neural complications can be extremely critical and difficult to reverse.

Unfortunately, the number of cases enrolled in current study was small. Therefore, further investigation using much more patients must be necessary to confirm the validity of our stories, but we believe that our study will contribute to some improvement for the future management of neuropathic scoliosis caused by NAAs.

CONCLUSIONS

In order to manage neuropathic scoliosis caused by NAAs securely, treatment for the relationship of neuropathic disorders should take precedence over that for spinal deformities. Early detection by cervical MRI screening, education for all physicians about the existence and characteristics of neuropathic scoliosis caused by NAAs is important. Prompt treatment of NAAs at younger age (≤ 8 yrs) and before curve progression ($< 30^\circ$) may lead to favorable prognosis of neuropathic scoliosis caused by NAAs.

CONFLICT OF INTEREST

The authors confirm that this article content has no conflicts of interest.

ACKNOWLEDGEMENT

Declared none.

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Received: July 05, 2012

Revised: October 31, 2012

Accepted: November 02, 2012

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