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ORIGINAL ARTICLE

Clinical manifestations and radiological characteristics in patients with idiopathic syringomyelia and scoliosis

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Abstract

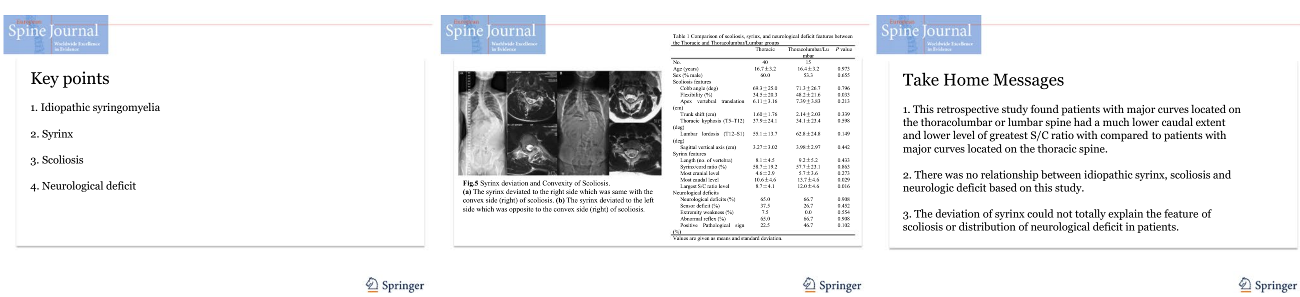
Purpose To clarify the clinical manifestation and radiological characteristics of idiopathic syringomyelia (IS) and to investigate the relationship between syrinx and scoliotic curves in IS-related scoliosis patients.

Methods Fifty-five patients with IS and scoliosis were identified and reviewed retrospectively from June 2009 to December 2016. Radiographic features of syrinx, scoliosis and clinical manifestations of neurological deficits were collected. The syrinx/cord (*S*/*C*) ratio was defined as the anteroposterior diameter of syrinx divided by the diameter of spinal cord at the same level. Patients were classified into two groups, the thoracic group (T group, apex vertebra located from T2 to intervertebral disk of T11–T12) and the thoracolumbar/lumbar group (TL/L group, apex vertebra located from T12 to L5).

Results There was no correlation between the radiological features of idiopathic syrinx and scoliotic curve parameters. The TL/L group had a lower level of most caudal extent (13.7 compared with 10.6, $P=0.029$) and lower level of largest *S*/*C* ratio (12.0 compared with 8.7, $P=0.016$) than that in T group. The deviated side of syrinx was not coincident with major curve convexity (27.2% concordance rate, $P=0.522$) or dominant side of neurological deficit (16.3% concordance rate, $P=0.212$).

Conclusions Patients with major curves located on the thoracolumbar or lumbar spine had a much lower caudal extent and lower level of greatest *S*/*C* ratio compared to patients with major curves located on the thoracic spine. No significant relationships were detected between syrinx features, scoliotic curve parameters and neurological deficits.

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.



Keywords Idiopathic syringomyelia · Syrinx · Scoliosis · Neurological deficit

Introduction

Idiopathic syringomyelia (IS) referred to syrinx not caused by conditions such as Chiari malformation, tethered cord syndrome, myelomeningocele, diastematomyelia, spinal cord tumor, trauma or infectious adhesive arachnoiditis [1–5]. About half of IS patients presented with scoliosis according to previous study [5]. However, limited research

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focusing on the relationship between IS and scoliosis had been reported [6–8].

One possible theory of causing scoliosis in IS patient was that the imbalance of trunk muscles, which was caused by asymmetric pressure to gray matter from a deviated syrinx in the spinal cord, contributed to the development of scoliosis [6, 9]. Some researchers confirmed this theory by finding the side of convexity in scoliosis coinciding with the side of syrinx deviation [6, 10], while this finding was not as obvious as reported according to our clinical experience. Even if the above theory held true, did the neurological deficits in IS patients have the similar features of being presented on the dominant side due to the asymmetric syrinx in spinal cord? Nevertheless, the relationship between the dominant side of neurological deficit and the deviated syrinx in IS collective was still not clear. On the other hand, the syrinx could be located from cervical to thoracolumbar spine with different diameters and lengths [5, 6, 11, 12]. It was important to clarify whether the range of scoliosis could be influenced by the sagittal location of syrinx. However, to the best of our knowledge, there had been no related study published, especially focus on the correlation between the syrinx location and the range of scoliosis.

Therefore, the objective of the current study was to summarize the clinical manifestation and radiological characteristics of IS, to investigate the role of syrinx deviation on the occurrence of scoliosis and to clarify the relationship between the sagittal location of syrinx and the range of scoliosis.

Materials and methods

Subjects

The medical records of patients with IS and scoliosis admitted to our spine center for surgical treatment of scoliosis from June 2009 to December 2016 were retrospectively reviewed. Inclusion criteria were (1) IS with scoliosis, (2) syrinx length more than two vertebra levels and diameter larger than 1 mm, (3) complete medical records and (4) no congenital spinal deformity or history of spine surgery. Patients with syringomyelia secondary to other causes, such as Chiari malformation, tethered cord syndrome, myelomeningocele, diastematomyelia, spinal cord tumor, trauma or infectious adhesive arachnoiditis, were excluded [2]. Fifty-five patients were enrolled in this study. There were 32 male and 23 female patients with an average age of 16.5 years (range 11–23 years) at the time of admission. Each patient received a detailed neurological examination, including sensation, extremity muscle strength, superficial abdominal reflexes (SAR), tendon reflexes and Babinski sign, by one experienced spine surgeon. The dominant side of neurological deficits was defined as the side of body with abnormal findings. If the above neurological deficits existed on both sides, the involvement of both sides was considered. All the neurological findings and the determinations of dominate sides were corroborated by a second experienced spine surgeon. Patients were classified into two groups based on the apex of major curve, the thoracic group (T group, apex vertebra located from T2 to intervertebral disk of T11–T12) and the thoracolumbar/lumbar group (TL/L group, apex vertebra located from T12 to L5).

Radiographic measurement

The Cobb angle, trunk shift (TS), apical vertebral translation (AVT), thoracic kyphosis (T5–T12, TK), lumbar lordosis (T12–S1, LL) and sagittal vertical axis (SVA) were measured on standing posteroanterior or lateral radiographs [3, 13]. Flexibility of the coronal curves was calculated based on the bending X-ray [14]. All patients underwent magnetic resonance imaging (MRI) of the entire spine to measure syrinx parameters. Length of the syrinx was measured as the number of vertebral segments traversed (Fig. 1). The maximal syrinx/cord (*S*/*C*) ratio was defined as the anteroposterior diameter of the syrinx divided by the diameter of the spinal cord at the level of maximum expansion to assess the axial width of

Fig. 1 Syrinx length. The syrinx length was defined as the number of vertebral segments spanned by the syrinx

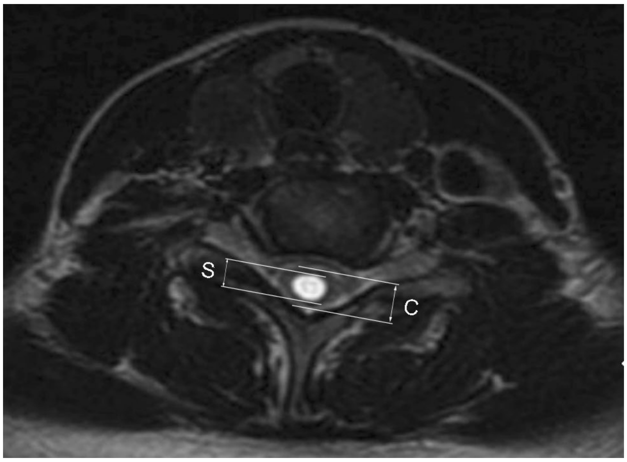


Fig. 2 Syrinx/cord ratio. The maximal syrinx/spinal cord ratio was measured as the maximal diameter of the syrinx (*S*) divided by the diameter of the spinal cord (*C*) at the same level

the syrinx [15] (Fig. 2). Syrinx deviation was calculated as (right-side distance at the upper end + right-side distance at the lower end)/(left-side distance at the upper end + left-side distance at the lower end); syrinxes with values of more than 1.10 or less than 0.90 were defined as eccentrically located [6]. For statistical calculations, all vertebral levels were converted into numeric values based on number of vertebra from the foramen magnum. Thus, the level of foramen magnum was set to 0, and C1 was set to 1, and so forth through L5 which was set to 24. The disk space level was measured as the average of the upper and lower vertebra. All radiological parameters and the determinations of syrinx deviation sides were measured and corroborated by a second experienced spine surgeon.

Statistical analysis

Statistical analysis was performed using SPSS 19.0 software for Windows. Data were compared between groups using independent-samples *t* tests, Chi-square tests, Fisher's exact tests and Pearson's and Spearman's correlations, depending on the parametric qualities of the variable analyzed. The significance level was < 0.05.

Results

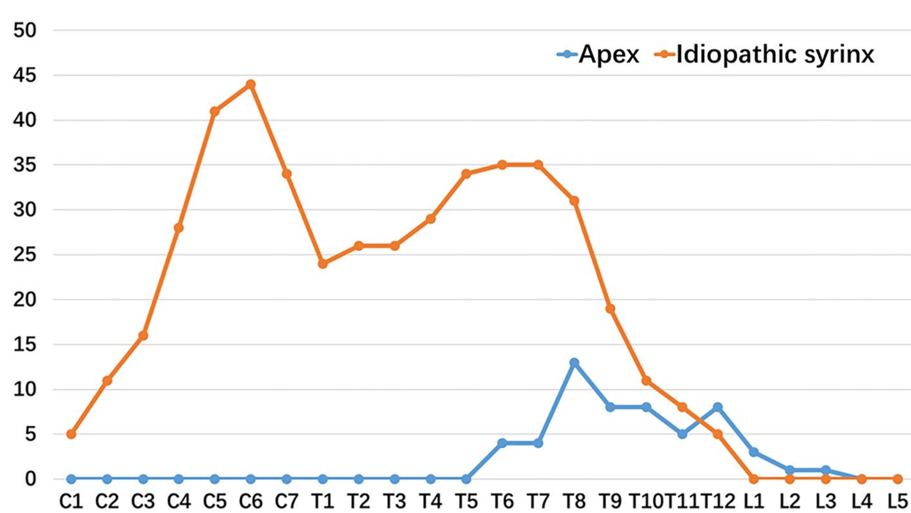
Radiological features of IS and scoliosis

Of a total of 55 patients enrolled into this study, 24 (43.6%) syrinxes were located in the cervical spine, 8 (14.5%) crossed the cervicothoracic junction, and 23 (41.8%) were located in the thoracic region (Fig. 3). The proximal syrinx level ranged from C1 to T8, while the distal level varied from C5 to T12. The maximal *S*/*C* and length of the syrinx averaged 0.58 (range 0.20–0.98) and 8.4 (range 2–19) vertebral levels, respectively. Thirty (54.5%) syrinxes were centrally located, 14 (25.5%) on the left and 11 (20.0%) on the right side. There were 41 thoracic (13 left and 28 right) and 14 thoracolumbar/lumbar (6 left and 8 right) curves. The major curve Cobb averaged 69.8° (range 33°–132°), with flexibility of 38.3% (range 5–90.4%), with TS of 1.75 cm (range 0–9.00 cm) and with SVA of 3.47 cm (range 0–12.20 cm).

Relationship between syrinx and spinal deformity

All patients were divided into two groups: the thoracic group (T group) and the thoracolumbar/lumbar group (TL/L group) based on the location of primary curve. Comparison of scoliosis, syrinx features and neurological deficits between these two groups showed that the TL/L group had

Fig. 3 Locations of idiopathic syrinxes and the apices of the major curves in all patients. Most idiopathic syrinxes were located at the cervical and thoracic region. The apices of major curves were located below where syrinxes were located. This discrepancy demonstrated that the location of syrinxes was not coincident with the range of major curves



a lower caudal extent (13.7 compared with 10.6, $P = 0.029$) and lower level of largest *S*/*C* ratio (12.0 compared with 8.7, $P = 0.016$) than those in the T group (Table 1, Fig. 4). Taking all patients into analysis, the radiological features of scoliosis, including major curve Cobb, flexibility, length, AVT, TS, TK, LL and SVA, were not related to maximal *S*/*C* ratio or syrinx length. There was no concordance between largest *S*/*C* ratio level and apex level of the major curve, and no significant relationship between location of syrinx and range of major curve was found (Table 2, Fig. 3). The side of convexity in scoliosis coincided with the side of syrinx deviation at rates of 27.2% (15/55). However, no association was found between the side of syrinx deviation and convex side of the major curve in T group, TL/L group or total patients (Table 3, Online Resource 1, Online Resource 2).

Patient characteristics and relationship between syrinx and neurological deficits

Nineteen (34.5%) patients were neurologically intact, while the absence of SAR was found in 28 patients (50.9%, 8 with absent in left, 9 with absent in right, 11 with absent in both sides), abnormal tendon reflex in 22 (40.0%, 19 with abnormal knee and ankle reflex, 3 with abnormal biceps reflex), sensory deficit in 19 (34.5%), positive Babinski sign in 16 (29.1%) and extremity weakness in 3 (5.4%, 1 with grade III strength in left upper extremity, 2 with grade III strength in right lower extremity). Radiological parameters of syrinx and scoliosis were not significantly different between patients with or without neurological deficits, while three patients with extremity weakness had a larger Cobb than patients with normal muscle strength (115.0° compared with 67.2°, $P = 0.019$). No significant differences of radiological parameters of syrinx or scoliosis were found between other subgroups of sensory deficit, abnormal SAR, abnormal tendon reflex or positive Babinski sign. The dominant side of neurological deficits coincided with the side of syrinx deviation at rates of 16.3% (9/55) and coincided with the convex side of major curve at rates of 30.9% (17/55). However, dominant side of neurological deficit was not correlated with syrinx deviation (Table 4) or convexity of major curve (Table 5). There was no association between syrinx deviation and dominant side of neurological deficit in T group or TL/L group (Online Resource 3, Online Resource 4).

Table 1 Comparison of scoliosis, syrinx and neurological deficit features between the thoracic and thoracolumbar/lumbar groups

	Thoracic	Thoracolumbar/lumbar	*P* value
No.	40	15	
Age (years)	16.7 ± 3.2	16.4 ± 3.2	0.973
Sex (% male)	60.0	53.3	0.655
Scoliosis features			
Cobb angle (°)	69.3 ± 25.0	71.3 ± 26.7	0.796
Flexibility (%)	34.5 ± 20.3	48.2 ± 21.6	0.033
Apex vertebral translation (cm)	6.11 ± 3.16	7.39 ± 3.83	0.213
Trunk shift (cm)	1.60 ± 1.76	2.14 ± 2.03	0.339
Thoracic kyphosis (T5–T12) (°)	37.9 ± 24.1	34.1 ± 23.4	0.598
Lumbar lordosis (T12–S1) (°)	55.1 ± 13.7	62.8 ± 24.8	0.149
Sagittal vertical axis (cm)	3.27 ± 3.02	3.98 ± 2.97	0.442
Syrinx features			
Length (no. of vertebra)	8.1 ± 4.5	9.2 ± 5.2	0.433
Syrinx/cord ratio (%)	58.7 ± 19.2	57.7 ± 23.1	0.863
Most cranial level	4.6 ± 2.9	5.7 ± 3.6	0.273
Most caudal level	10.6 ± 4.6	13.7 ± 4.6	0.029
Largest *S*/*C* ratio level	8.7 ± 4.1	12.0 ± 4.6	0.016
Neurological deficits			
Neurological deficits (%)	65.0	66.7	0.908
Sensor deficit (%)	37.5	26.7	0.452
Extremity weakness (%)	7.5	0.0	0.554
Abnormal reflex (%)	65.0	66.7	0.908
Positive pathological sign (%)	22.5	46.7	0.102

Values are given as means and standard deviation

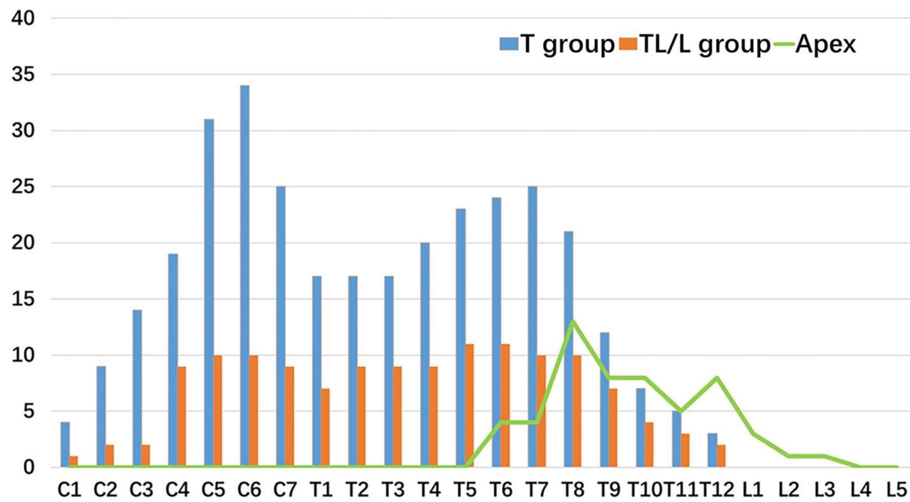


Fig. 4 Locations of idiopathic syrinxes and the apices of the major curves separately in thoracic group (T group) and thoracolumbar/lumbar group (TL/L group) and the locations of apices in all patients. The idiopathic syrinxes of T group were mostly located at the cervical and thoracic region with more cases in the cervical region. Most idiopathic syrinxes of TL/L group were located at the cervical or thoracic region too, however, with almost equal proportions between these two regions. Thus, the TL/L group had a higher proportion of syrinxes, which were located caudally, than T group. The *y*-axis shows the numbers of patients, and the *x*-axis shows the vertebral level

Table 2 Correlations between scoliosis curve range and location of syrinx

Characteristics of major curve	Characteristics of syrinx				
	Most cranial level	Most caudal level	Level of largest *S/C*	Length	Largest *S/C* ratio
Level of upper end vertebra	$c^*=0.119$ $P^\#=0.388$	$c=0.166$ $P=0.226$	$c=0.247$ $P=0.069$	$c=0.004$ $P=0.979$	$c=-0.139$ $P=0.313$
Level of lower end vertebra	$c=0.166$ $P=0.226$	$c=0.249$ $P=0.066$	$c=0.180$ $P=0.187$	$c=0.128$ $P=0.350$	$c=0.036$ $P=0.793$
Level of apex vertebra	$c=0.050$ $P=0.717$	$c=0.154$ $P=0.263$	$c=0.177$ $P=0.197$	$c=0.043$ $P=0.755$	$c=-0.023$ $P=0.868$
Length	$c=-0.099$ $P=0.474$	$c=0.061$ $P=0.660$	$c=-0.154$ $P=0.261$	$c=0.159$ $P=0.247$	$c=0.263$ $P=0.053$
Cobb	$c=-0.062$ $P=0.652$	$c=-0.037$ $P=0.789$	$c=0.034$ $P=0.808$	$c=0.067$ $P=0.627$	$c=0.015$ $P=0.916$

*c = correlation coefficient

#P = P value

Table 3 Correlation between major curve direction and side of deviated syrinx

Major curve direction[a]	Syrinx deviation	
	Left	Non-left
Left	6	13
Right	8	28
Major curve direction[b]	Syrinx deviation	
	Right	Non-right
Right	9	27
Left	2	17

[a]Fisher's exact test, $P=0.522$

[b]Fisher's exact test, $P=0.295$

Discussion

The pathogenesis of syringomyelia was associated with alteration in the cerebrospinal fluid flow (CSF) dynamics and related chronic inflammation in the region of craniovertebral junction [16]. Several conditions could cause the abnormal CSF dynamics, such as Chiari malformation, tethered cord syndrome. However, IS was defined as an entity not associated with any of the previously mentioned conditions. Although some studies reported the atlantoaxial instability, abnormal cervical spinal canal narrowing was found in IS patients and might be associated with change of CSF dynamics [17, 18], and more powerful evidence was lacking to support these above findings. Other theory, the reduction in the compliance of the veins draining the spinal cord,

Table 4 Correlation between dominant side of neurological deficits and syrinx deviation

Syrinx deviation	Neurological deficits		
	Left	Both	Right
Left	3	3	3
Middle	6	7	5
Right	3	0	6

Fisher's exact test, $P = 0.212$

underlay the syringomyelia but also needed further investigations to be proved [19]. Thus, the underlying pathogenesis of IS was still unclear.

IS and scoliosis

Scoliosis was common in IS patients, and Rodriguez et al. [5] found that there were 49.1% (30/61) cases of "idiopathic" scoliosis in IS patients. And many studies reported that the IS was unintentionally found in "presumed" idiopathic scoliosis, especially in the scoliosis with Cobb larger than 90 degrees [1, 20, 21]. Although the relationship between the IS and scoliosis seemed close, limited studies indicated that there was no correlation between *S/C* ratio and syrinx length with scoliosis [6]. Results from our study further confirmed this view. According to previous studies, the influence of syrinx location on scoliosis was still controversial. Zhu et al. [6] and Ozerdemoglu et al. [8] reported that the level of greatest syrinx size was correlated with apex vertebral level, and the most caudal level of syrinx was related to the level of the lower end of the major curve, indicating a possible relationship between syrinx location and scoliosis in the sagittal plane. However, other researchers did not think the range of scoliosis was correlated with syrinx location [10, 22]. Based on results of this study, patients with thoracolumbar/lumbar curve had a lower most caudal level and lower level of greatest *S/C* ratio than patients whose major curve was located in the thoracic spine (Table 1, Fig. 4). This result suggested that the lower the major curve was located, the more caudally the syrinx might extend, supporting the opinions of Zhu et al. and Ozerdemoglu et al. in some extent.

Table 5 Correlation between dominant side of neurological deficits and major curve direction

Major curve direction[a]	Dominant side of neurological deficits	
	Left	Non-left
Left	5	6
Right	7	18
Major curve direction[b]	**Dominant side of neurological deficits**	
	Right	Non-right
Right	12	13
Left	2	9

[a]Fisher's exact test, $P = 0.446$

[b]Fisher's exact test, $P = 0.142$

The mechanism underlying the development of scoliosis secondary to syringomyelia remained unclear. One possible theory was that the imbalance of trunk muscles, which was caused by asymmetric pressure to gray matter from a deviated syrinx, contributed to the development of scoliosis [9]. Some researches have confirmed it by finding convexity of scoliosis coinciding with the side of syrinx deviation at rates of 83–100% [6, 9, 10]. While our study did not show such high coincidence rate, only 25 (45.5%) of all 55 syrinxes were eccentrically located, and the side of syrinx deviation was not all the same as the side of convexity in primary curve (Table 3, Fig. 5). It meant syrinx deviation might not be correlated with the convexity of scoliosis. On the other hand, the syrinx deviation theory could not explain all scoliosis, especially those syrinxes located centrally or opposite which accounted for 72.8% (40/55) in this study. Thus, results from our study indicated that syrinx deviation might not contribute to the pathogenesis of scoliosis in all cases, and there might be an underlying mechanism influencing the imbalance of trunk muscles apart from deviated syrinx.

IS and neurological deficits

Classical symptoms and physical examination in syringomyelia patients included absence of SAR, sensory deficit and upper/lower extremity weakness. According to the studies from Saifuddin et al., the abnormal SAR was one significant indicator for syringomyelia in "presumed" idiopathic scoliosis [23]. Likewise, these neurological indicators were also found in our patients, especially the abnormal SAR (50.9%), which was firstly reported as far. Zhu et al. and Zadeh et al. reported the neurological deficits of patients were not correlated with syrinx features [6, 24], and our study further supported this finding. Ozerdemoglu et al. [8] reported that neurological deficits in patients with syringomyelia were more dominant on the convex side of the curve. However, our results were opposite to the above findings, for only 47.2% (17/55) of patients had dominant neurological deficits on the convex side (Table 5). In addition, there was no correlation between syrinx deviation with dominant side of neurological deficits (Table 4), and the incongruity between side of syrinx deviation and neurological deficits further weakened the theory that pathogenesis of scoliosis was influenced by asymmetrical syrinx.

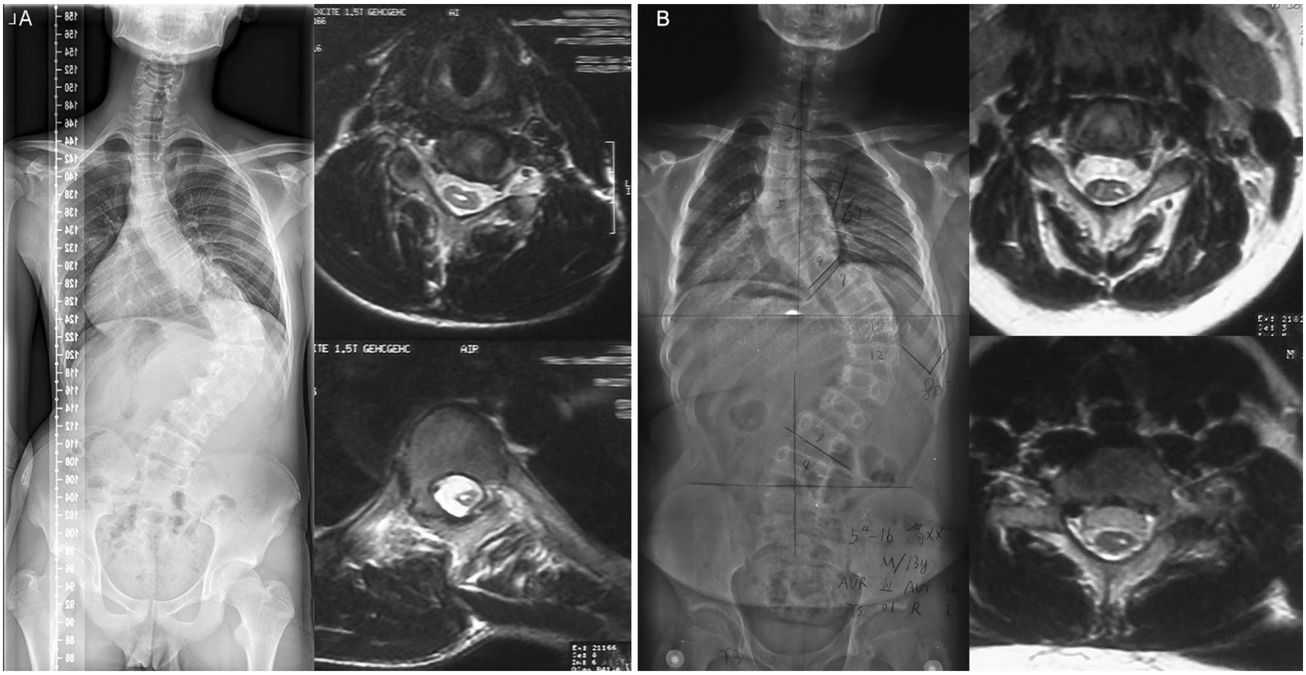


Fig. 5 Syrinx deviation and convexity of scoliosis. **a** The syrinx deviated to the right side which was the same with the convex side (right) of scoliosis. **b** The syrinx deviated to the left side which was opposite to the convex side (right) of scoliosis

In our study, three patients with extremity weakness had larger major curve Cobb than patients with normal muscle strength. And all these three patients had syrinx deviation and neurological deficits on the convex side of primary scoliosis. The existence of scoliosis relatively increased the length of spinal canal compared to the spinal cord, causing the outside tension force to the spinal cord on the sagittal plane, while the deviated syrinx provided the inner asymmetric compression on the spinal cord on the cross section. So, this result might hint that the combination of intraspinal abnormality (deviated syrinx) and severe spinal deformity (large Cobb angle) might increase the tension of the spinal cord from various dimension, causing the major neurological deficits (such as decrease in extremity strength) in IS patients.

Limitations of the current study were the retrospective, cross-sectional design, and these IS-related scoliosis patients were enrolled because of their will of corrective surgery. The syrinx was one three-dimensional space-occupying lesion in the spinal cord, and the three-dimensional parameter, such as volume, might represent feature of syrinx much better than *S*/*C* ratio or length theoretically, and related study should be carried out further.

To our knowledge, this was the first study to evaluate the relationship between IS, scoliosis and neurological deficits. The present results, which were opposite to previous reporting, questioned the hypothesis of asymmetric syrinx in the pathogenesis of syringomyelia-related scoliosis. Further investigation of underlying mechanisms was valuable to be explored.

Conclusions

Patients with a major curve located on the thoracolumbar or lumbar spine had much lower levels of caudal extent and lower level of greatest *S*/*C* ratio compared to patients with a major curve on the thoracic spine. No significant relationships were detected between syrinx features, scoliotic curve parameters and neurological deficits. Other mechanisms besides deviated syrinx needed to be explored further to determine the pathogenesis of scoliosis in IS patients.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval For this type of study formal consent is not required.

Informed consent Informed consent was obtained from all individual participants included in the study.

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