PROGRESSIVE SCOLIOSIS AND SYRINGOMYELIA: CHARACTERISTICS OF SURGICAL TACTICS

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Objective. To analyze the results of surgical correction of scoliosis in patients with syringomyelia.
Material and Methods. The study included 33 patients with syringomyelia and the enlarged spinal canal. Neurosurgical intervention was performed in 20 patients. The following operations were carried out: formation of a large occipital tank, draining and emptying of cysts, and resection of terminal filament. Nineteen patients were operated on using the CDI. Four patients are undergoing stage correction with VEPTR. In one case, instrumentation was not successful, and nine patients are receiving treatment at the Department of Neurosurgery.
Results. The study did not reveal any statistically significant relationship between sex, age, deformity apex, side of scoliotic curve, rate of progression, neurological symptoms and the presence of syringomyelia. It was noted that the neurological deficit begins to develop when a cyst width exceeds 4.5 mm, yet its length and location do not play a significant role. In the presence of cysts of up to 9 mm in width, clinical manifestation and neurological symptoms may be absent.
Conclusion. The proposed approach to the treatment of patients with spinal deformities, which have syringomyelia as a concomitant vertebral pathology can allow to achieve satisfactory results in scoliosis correction and to avoid neurological complications.
Key Words: syringomyelia, idiopathic scoliosis, correction of spinal deformity.

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Modern technologies, in particular those used at the preoperative preparation stage, are sufficient to identify most comorbidities, which helps choose an appropriate treatment option as well as predict and significantly reduce the risk of intra- and postoperative complications. Despite this, the problem of choosing a treatment option for scoliosis combined with syringomyelia still remains topical. While the issues of surgical treatment outcomes and diagnosis of these conditions are well represented in the literature, there is no common approach or algorithm of actions. A variety of techniques and procedures have been developed for treatment of syringomyelia, but none of them, except surgery, provides outcomes sufficient for early correction of the scoliotic deformity with a minimum risk of neurological complications. Although cysts may collapse spontaneously, prolonged ineffective conservative treatment may lead to severe progression of scoliosis. Neurosurgical interventions, in turn, also do not guarantee a perfect result; often, re-operations are required. Therefore, the patient with syringomyelia and scoliosis is always a particular case in terms of choosing a treatment approach. The main questions that the surgeon asks himself are as follows: how safe is correction of the spinal deformity associated with syringomyelia; which conditions require neurosurgery to be performed before correction; which surgical treatment approach should be used in the absence of indications for neurosurgery; if intraoperative traction is applicable?

The rate of scoliosis in syringomyelia patients ranges from 25.0 to 74.4 % [4, 19]. In turn, syringomyelia occurs in 1.2 to 9.7 % of scoliosis patients [3, 4, 6, 7, 8, 10, 11, 16]. It should also be noted that syringomyelia is associated with Arnold – Chiari malformation in 30–86 % of cases [1, 2, 9, 10]. This combination makes it necessary to extend the amount of both preoperative examination and neurosurgery.

An analysis of the literature demonstrates that some authors prefer to perform neurosurgical treatment first, arguing that neurosurgery under the age of 10 years may completely eliminate the need for scoliosis correction [12, 13, 15]. Other authors tend to believe that prior neurosurgical operation may significantly reduce the risk of neurological complications associated with scoliosis correction, but does not stop deformity progression [5, 14, 15]. The number of reports on scoliosis correction performed without any neurosurgical treatment is limited, but these interventions have been carried out using intraoperative neuromonitoring [17, 18, 21, 22]. Despite the difference in approaches, almost all authors agree that the risk of complications associated with scoliotic deformity correction is very significant. However, we know only one report of postoperative complica-

tions. R.A. Ozerdemoglu et al. reported that only 3 of 38 (8 %) patients developed neurological complications after scoliotic deformity correction [14].

The study objective was to evaluate results of surgical correction of the scoliotic deformity in syringomyelia patients.

Material and Methods

Three thousand and one hundred twenty patients with scoliosis of various etiologies underwent treatment at the Clinic for Child and Adolescent Vertebrology of the Novosibirsk Research Institute of Traumatology and Orthopedics in the period between 1996 and 2015. Syringomyelia was diagnosed in 33 (1.05 %) patients. Syringomyelia associated with idiopathic scoliosis was detected in 21 out of 2,334 (0.9 %) patients; syringomyelia associated with scoliosis of other (congenital and paralytic) etiologies was identified in 12 out of 786 (1.52 %) patients. The study included patients with syringomyelia (n = 24) and a dilated central canal, referred to as hydromyelia, (n = 9). The reason to combine these groups was a potential risk of intra- and postoperative complications due to correction of scoliosis associated with any intracanal lesions, accompanied by a change in the shape of the spine and spinal canal caused by spine traction.

Neurosurgery was performed in 20 patients. The performed procedures included formation of the cisterna magna, drainage and evacuation of cysts, and resection of the filum terminale. Fourteen (42.4 %) patients had syringomyelia combined with Chiari malformation. Two patients with a tethered spinal cord underwent resection of the filum terminale.

Nineteen patients were operated on using CD instrumentation. Four patients underwent staged correction using VEPTR instrumentation. Placement of instrumentation failed in one case. Nine patients were receiving treatment at the Department of Neurosurgery.

At admission to the clinic, all patients underwent a standard preoperative examination that involved MRI of the thoracic spine and the region comprising the curve apex as well as an examination by a neurologist. If patients having gross deformities (>80°) were detected with spinal cord cysts, they additionally underwent a traction test with the patient's total body weight as well as electroneuromyography and evoked potentials. Patients detected with cysts in the thoracic or upper thoracic spinal cord underwent MRI of the cervical spine and craniovertebral junction.

In 17 cases, syringomyelia was diagnosed incidentally during preoperative MRI. Neurological deficits of varying severity, which were the basis to suspect spinal cord malformations, occurred in 13 patients; in one patient, a neurological deficit was detected only during registration of evoked potentials (there were no clinical manifestations). In 3 patients, syringomyelia was diagnosed before application to the clinic. Only two patients had the complete classical clinical picture of syringomyelia: dissociated sensory loss, pain, and trophic disorders. The other patients presented either with idiopathic scoliosis or with an initial neurological deficit associated with other causes (Arnold – Chiari malformation, diastematomyelia).

Results

We conducted a retrospective analysis of cases where syringomyelia was combined with scoliosis requiring surgical correction (Table). We evaluated the initial neurological symptoms, cyst size, effect of previous neurosurgical interventions on progression of the spinal deformity, risk factors for neurological deficits, and character of performed orthopedic procedures. The resulting data were used to define the basic principles of a treatment approach.

At the time of paper preparation, 9 patients received treatment and were followed-up by neurosurgeons. Two of the patients had no neurosurgery due to the absence of neurological symptoms, small cavity size, absence of the indications for immediate scoliosis surgery (scoliotic curves of 38° and 40°, respectively), and lack of deformity progression. The patients were being followed-up. In 8 patients without scoliosis correction, syringomyelia was associated with Arnold – Chiari malformation. Furthermore, two patients undergoing staged treatment with the VEPTR instrumentation did not undergo neurosurgery due to the absence of an initial neurological deficit and severe malignant progressive scoliosis requiring immediate surgical correction. One of the patients had hydromyelia in the upper thoracic spine, with an expansion of the central canal of up to 4 mm; the other patient had a syrinx of 4.6 mm in width in the lumbar spine. Also, 9 patients who underwent scoliosis correction using the CD instrumentation did not undergo neurosurgery; of these, 5 patients had an expansion of the central canal, and 4 patients had syringomyelia cysts. A neurological deficit (myelopathy) was detected in one case in the form of a reduced conduction velocity in the left lower limb during recording of evoked potentials. Given small sizes of the cysts and central canal of the spinal cord as well as no neurological symptoms and no need for immediate orthopedic care, scoliosis correction was performed without neurosurgical interventions.

The performed neurosurgical interventions included:

1) cisterna magna formation and cyst evacuation (n = 9);

2) isolated evacuation of a cyst (n = 8);

3) cisterna magna formation without cyst evacuation (n = 3).

Re-operation was required in three cases (repeated evacuation and reconstruction with dura mater fragments).

Two patients underwent neurosurgery under the age of 10 years. After neurosurgery, the scoliotic deformity continued to progress in both cases: from 81° to 105° and from 119° to 130°, respectively, for one year. Among patients who underwent neurosurgery at the age of 10 to 17 years (11 patients), the scoliotic deformity also continued to progress. The other patients (n = 7) underwent surgery after the completion of active bone growth (at the age of 17 years or older); for this reason, the scoliotic deformity

in these patients was in a stable condition, without clinically significant progression. Neurosurgical treatment was not performed.

At the time of paper preparation, four patients with congenital spinal deformities were undergoing treatment with the VEPTR instrumentation; the mean age at the time of surgery was 5.8 years (range, 2.5 to 8.5 years); the mean baseline deformity of 100.3° (67° to 130°) was reduced to 78.8° (51° to 105°), on average, after primary correction. An 1.5–2 cm distraction was performed during successive treatment stages (Fig. 1). There was no neurological deficit worsening associated with primary correction and successive distractions. Neurosurgical treatment was required in 2 patients. MSCT revealed diastematomyelia in both patients: at the L2–L3 level in the first case and at the T11–12 level in the second case. At baseline, both children were diagnosed with lower flaccid paraparesis, with pelvic organ dysfunction being present in one case. The patients underwent evacuation of syrinxes without resection of bony septa. The intersurgical interval was 6 months. In both cases, the spinal deformity continued to progress after neurosurgical treatment.

The mean age of patients who underwent spinal deformity correction using various instrumentation was 15.0 ± 6.7 years (2.5 to 34.3 years). There was no statistically significant relationship among the gender, age, curve apex, scoliotic curve direction, rate of progression, neurological symptoms, and presence of syringomyelia. A neurological deficit was noted to start developing at a cyst width of over 4 5 mm. In this case, the length and location of the cyst did not play a significant role. Interestingly, in the case of cysts of up to 9 mm in width, the clinical signs and neurological symptoms may be absent. The distribution of syrinx localizations was as follows: cervical spine – 8 cysts; cervicothoracic spine – 15 cysts; thoracic spine – 8 cysts; lumbar spine – 1 cyst; filum terminale – 1 cyst.

If there was no neurological deficit, and cysts were less than 4–5 mm in width, deformity correction through the posterior approach was performed with skeletal traction (Fig. 2, 3). Posterior correction alone was used in 10 cases. In three cases, placement of the CD instrumentation was complemented with releasing discectomy. Also, three-stage surgery (releasing discectomy, skeletal traction, and deformity correction with dorsal instrumentation) was performed in 3 cases. If after neurosurgery, neurological symptoms did not regress completely, or the syrinx size was larger than 4 5 mm, surgical correction of the spinal deformity was performed without skeletal traction (Fig. 4). Twenty patients underwent surgery using the CD instrumentation; their mean age was 16.9 ± 5.7 years, and the mean follow-up period was 4.2 ± 2.8 years (range, 1.6 to 11.9 years). Preoperatively, all patients underwent a traction test with the patient's total body weight, which was not accompanied by worsening of neurological symptoms. Patients consulted with neurosurgeons who jointly decided on the tactics and amount of surgery. The major scoliotic curve was located in the thoracic spine in 28 cases and in the lumbar spine in 5 cases. The mean magnitude of the major scoliotic curve before surgery was 72.8° ± 21.3°, and the mean compensatory curve was 29.3° ± 21.7°. Preoperatively, the mean thoracic kyphosis was 41.6° ± 20.1°, and the mean lumbar lordosis was 69.1° ± 17.5°. Implantation of an endocorrector failed in one patient. The dural ectasia signs were detected during preoperative examination. Intraoperatively, excessive liquorrhea developed upon an attempt to place laminar hooks, which resulted in the decision to refuse scoliosis correction.

Postoperatively, the mean major curve was 33.3° ± 19.0°; the mean compensatory curve was 11.1° ± 12.2; the mean thoracic kyphosis amounted to 28.3° ± 17.8°; the mean lumbar lordosis was 52.7° ± 8.7°. Therefore, the major curve correction was 39.5° ± 13.6° (56.3 ± 18.2 %); the secondary curve correction was 19.0° ± 17.0° (43.5 ± 22.1 %). At the end of the follow-up period, postoperative progression was 2.1° ± 4.8° (3.3 ± 7.3 %) for the major curve and 3.0° ± 6.0° (4.1 ± 9.0 %) for the secondary curve. Surgical correction of the spinal deformity did not lead to worsening of neurological symptoms in any of the cases.

Discussion

A comparison between our results for correction of scoliosis associated with syringomyelia and the data of other authors [18, 20, 21, 22] reporting correction of the major curve in a range of 63 to 80 % who used shortening vertebrotomy techniques, vertebral column resection (VCR) and pedicle subtraction osteotomy (PSO), demonstrates that the achieved results are quite satisfactory. In this case, the absence of neurological complications is the major indicator underlying the choice of a treatment option. Perhaps, a more aggressive surgical approach may provide more significant correction of the major scoliotic curve, but the risk of neurological deficit worsening would also increase. The use of modern intraoperative neuromonitoring that enables real-time tracking of the effect of any corrective procedure may be helpful in achieving a greater degree of spinal deformity correction.

The absence of neurological deficits in studies by authors who have achieved scoliosis correction of up to 80 % of the initial curve magnitude without preliminary neurosurgery, using in particular shortening vertebrotomy techniques, makes this approach very attractive.

Another question arising before the surgeon performing scoliosis correction is postoperative control of the spinal cord condition. An implanted metal device complicates MRI visualization of the spinal cord. A MSCT study, even with contrast enhancement, may also lack sufficient data for complete evaluation of the cyst size. The size of cysts and the central canal lumen can vary over time, and not only reduce. Upon enlargement of lesions and compression of the spinal cord structures, when the structures exhaust their compensatory potential, the neurological status may worsen. In this case, it is necessary to remove corrective instrumentation, perform additional examination, and make a decision on the need for neurosurgical care. We

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Table
Characteristics of patients with syringomyelia

Patient	Age of application to clinic	Etiology	Neurological deficit before surgery	Chiari malformation	Neurosurgery*	Time to orthopedic surgery (years)	Orthopedic surgery**	Cobb angle before surgery	Cobb angle after surgery	Cobb angle at the latest examination
CD instrumentation (n = 20)										
1	14.9	Idiopathic	–	+	K	0.6	Z	43	12	18
2	7.6	Idiopathic	–	–	–	–	T + C + Z	61	44	44
3	13.4	Idiopathic	–	+	K + C	1.2	Z	72	31	28
4	18.0	Idiopathic	–	+	K + C	4.8	T + Z	110	68	66
5	19.0	Congenital	+	–	–	–	C + Z	67	35	47
6	18.1	Idiopathic	+	+	K + C	1.1	T + Z	90	40	42
7	16.9	Idiopathic	–	–	C	1.7	T + C + Z	74	20	22
8	19.1	Congenital	–	–	–	–	C + Z	50	30	27
9	14.3	Idiopathic	–	–	–	–	Z	48	4	6
10	18.3	Congenital	–	–	–	–	C + Z	44	17	17
11	18.5	Idiopathic	+	+	K + C	1.2	Z	70	36	36
12	15.1	Idiopathic	+	–	C + T	0.7	P + Z	85	61	65
13	34.3	Idiopathic	+	+	–	–	Z	75	26	26
14	11.8	Idiopathic	–	–	–	–	T + C + Z	79	39	43
15	24.1	Idiopathic	+	–	–	–	Z	69	22	37
16	14.3	Idiopathic	–	–	C	1.1	Z	51	10	12
17	12.0	Congenital	–	–	–	–	Z	104	73	75
18	14.1	Idiopathic	–	–	C	0.9	Z	115	43	44
19	10.8	Congenital	–	–	–	–	Z	77	22	18
20 (without instrumentation)	22.4	Paralytic	+	–	C	11.2	–	95	–	116
VEPTR instrumentation (n = 4)										
1	8.5	Congenital	+	–	C	1.4	130	130	105	116
2	4.9	Congenital	–	–	–	–	67	67	51	62
3	7.3	Congenital	+	–	C	1.3	105	105	77	74
4	2.5	Idiopathic	–	–	–	–	99	99	82	81
No surgery (n = 9)										
1	15	Idiopathic	+	+	K + C + T	–	–	53	–	59
2	17	Idiopathic	+	+	K + C	–	–	72	–	75
3	13	Idiopathic	–	+	K	–	–	95	–	106
4	28	Congenital	–	–	–	–	–	38	–	38
5	29	Neurofibromatosis	+	+	K + C	–	–	82	–	84
6	18	Idiopathic	–	+	K + C	–	–	80	–	81
7	18	Paralytic	+	+	K	–	–	106	–	109
8	17	Idiopathic	–	+	K+C	–	–	40	–	48
9	14	Idiopathic	–	+	–	–	–	40	–	41

*K – cisterna magna formation, C – cyst evacuation, T – resection of the filum terminale;
**T – thoracotomy, mobilizing discectomy, interbody fusion, C – skeletal traction through the skull and lower legs, Z – spinal deformity correction with CD instrumentation through the posterior approach.

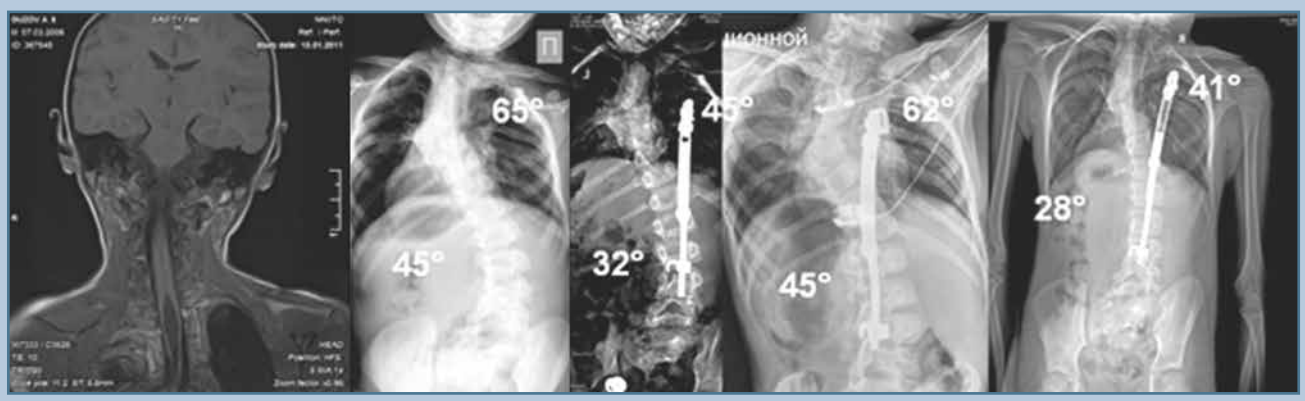

Fig. 1
MR and X-ray images of a 5-year-old male patient G. with a syrinx of 8 mm in size; no initial neurologic deficit: stages of surgical correction using VEPTR instrumentation.

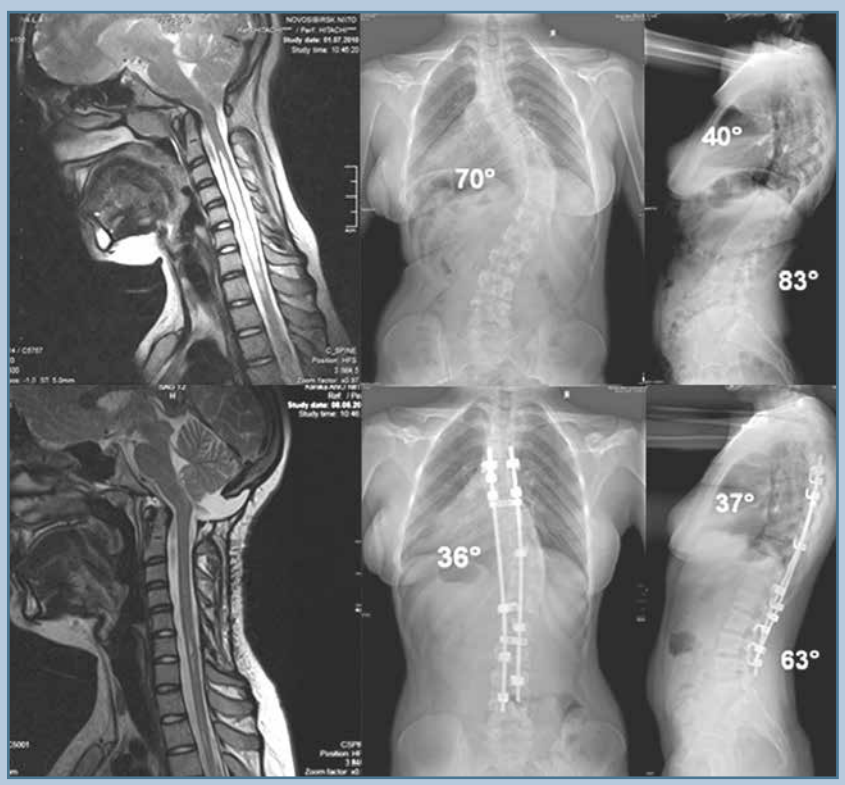

Fig. 2
MR and X-ray images of an 18-year-old male patient M.: the cerebellar tonsils are located 4mm below the Chamberlain line; a syringomyelic cavity of 8 mm in width, with smooth internal walls and separate incomplete septa; minor upper paraparesis; after syrinx drainage and formation of the cisterna magna, a local cyst of 2 mm in width; neurological symptoms completely regressed; the spinal deformity was corrected with skeletal traction

did not observe such cases. Removal of instrumentation may be associated with a significant loss of correction. In this regard, the quality of dorsal spine fusion is of particular importance. A secure bone block is a prerequisite for preservation of the achieved correction.

Despite the fact that neurological deficits may develop if a central canal expansion is 4–5 mm or more, it is impossible to predict neurological status changes caused by scoliosis correction. Probably, even the maximum correction may not lead to neurological deficit worsening. On the other hand, in the case of a syrinx size of 8–9 mm and the absence of neurological symptoms at baseline, even the minimum correction associated with a spinal cord dislocation may impair compensation of the spinal cord condition.

The rate of scoliosis-associated syringomyelia is 1.05 %; therefore, syringomyelia is one of the most common concomitant vertebral pathologies. A standard plan of preoperative patient examination should include consultation with a neurologist and MRI of the thoracic spine and the region comprising the deformity apex. If syringomyelia is detected, the patient should undergo an examination by a neurosurgeon, a traction test with the total body weight, neurophysiological examinations, and MRI of the cervical spine and craniover-

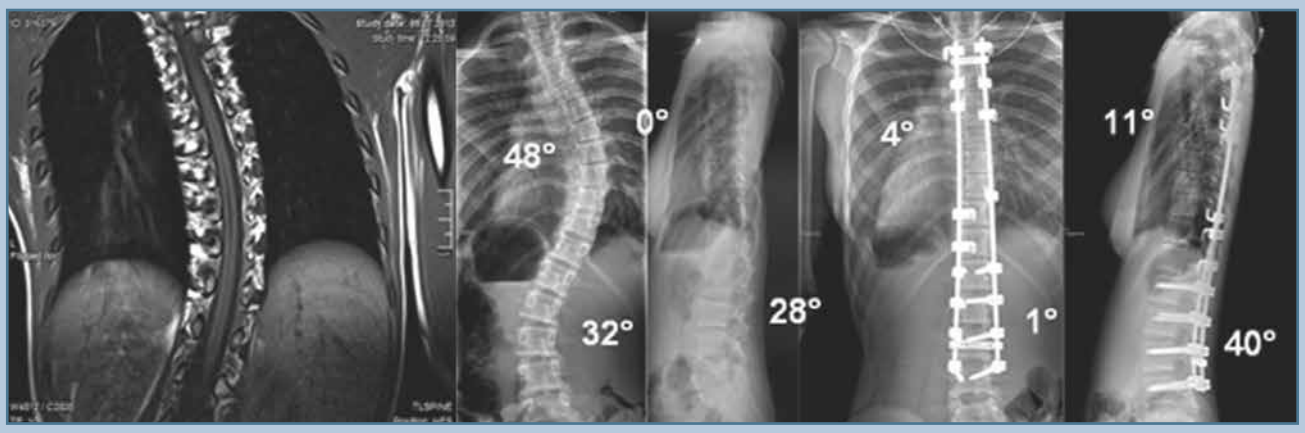

Fig. 3
MR and X-ray images of a 14-year-old female patient P.: expansion of the central canal of up to 2–3 mm in the thoracic spine; no initial neurologic deficit; the scoliotic deformity was corrected with skeletal traction, without preliminary neurosurgical intervention

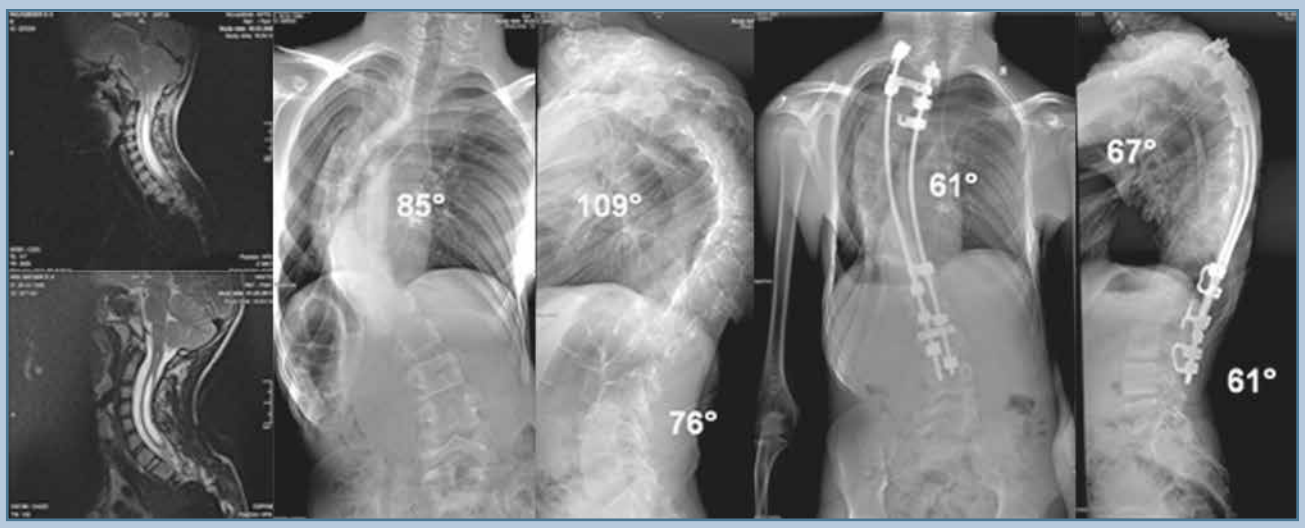

Fig. 4
MR and X-ray images of a 15-year-old male patient R.: an intramedullary cyst of up to 12 mm in width, with smooth inner walls and separate septa, is seen; lower central paraparesis (reflex paraparesis on the right; slight paraparesis, up to 4 points, on the left); formation of the cisterna magna and drainage of the cyst were performed; partial regression of neurological symptoms after surgery; control MRI: the cyst of up to 9.5 mm in width, with clear and smooth contours and homogeneous contents; the spinal deformity was corrected without intraoperative traction

tebral junction. It may be supposed that a neurological deficit may develop when the syrinx reaches 4–5 mm in width. In the presence of a neurological deficit, large syrinxes (>10 mm in width), or Arnold – Chiari malformation, we consider neurosurgery as the first stage treatment. If the outcome of neurosurgical treatment is positive (regression of neurological symptoms), the maximum amount of orthopedic surgery may be planned. If neurological symptoms do not regress completely, corrective surgery without intraoperative traction would be less traumatic. If neurological symptoms are absent at baseline, and the size of the central canal or syringomyelia cysts is less than 4 mm in width, the spinal deformity may be corrected with skeletal traction. In this case, mobilizing discectomy may also be used. To prevent the development and worsening of neurological symptoms, intraoperative neuromonitoring should be a prerequisite.

Conclusion

In our opinion, neurosurgical treatment at the first stage, even if it is performed under the age of 10 years, does not stop progression of the spinal deformity. Progression of the scoliotic curve may be terminated or delayed only after the completion of primary bone growth.

The proposed approach to the treatment of patients with spinal deformities and syringomyelia, as a concomitant vertebral pathology, may provide satisfactory results in scoliosis correction and avoid neurological complications.

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