THE USE OF MRI IN THE STUDY OF PATIENTS WITH IDIOPATHIC SCOLIOSIS: A SYSTEMATIC REVIEW OF THE LITERATURE

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Objective. To analyze the frequency of hidden neuraxial pathology in idiopathic scoliosis (IS), to substantiate the need for MRI in IS and to identify promising areas for the use of MRI in the examination of patients with IS.

Material and Methods. The literature review was carried out using the PubMed and Google Scholar databases. Of the 780 papers on the research topic, 65 were selected after removing duplicates and checking for inclusion/exclusion criteria. As a result, 49 original studies were included in the analysis. Level of evidence – II.

Results. According to modern literature, the main direction of using MRI in idiopathic scoliosis is the search for predictors of latent pathology of the spinal cord and craniovertebral junction. The frequency of neuraxial pathology in idiopathic scoliosis is 8 % for adolescent IS and 16 % for early IS. The main predictors of neuraxial pathology are male sex, early age of deformity onset, left-sided thoracic curve and thoracic hyperkyphosis. MRI in IS may be a useful addition to radiological diagnostic methods to identify risk factors and to study degenerative changes in the spine.

Conclusion. MRI of the spine should be performed in the early stages of IS to detect latent spinal cord tethering. In type I Chiari anomalies, there is a possibility that early neurosurgery can prevent the development of scoliosis. The main signs of latent neuraxial pathology in IS are early progression of spinal deformity, left-sided thoracic curve, male gender and thoracic kyphosis over 40° according to Cobb. MRI can be used as an effective non-invasive tool in research aimed at identifying risk factors for IS, including helping to track early degeneration of intervertebral discs.

Key Words: idiopathic scoliosis, hidden spinal cord pathology, hidden neuraxial pathology.

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Scoliosis is a three-dimensional deformity of the spine and its main diagnostic criterion is the magnitude of the curve in the frontal plane greater than 10° according to Cobb. Idiopathic scoliosis (IS) is a diagnosis by exclusion when history, clinical, and instrumental findings fail to provide reliable evidence of a specific etiology. Despite the terminology indicating the absence of a cause, idiopathic scoliosis is currently considered a multifactorial disease caused by various epigenetic factors associated with genetic predisposition [1].

Latent, "mute", or asymptomatic neuraxial pathology is found in some cases of suspected IS. The most common ones are syringomyelia, Chiari malformation and spinal cord tethering [2]. Spinal cord tumors are extremely rare but the most dangerous pathology, manifesting as scoliosis without primary neurologic impairment in some cases [3–19].

May scoliosis be caused by spinal pathology and be considered as neuromuscular condition? May scoliosis be considered as idiopathic with asymptomatic mild cerebellar tonsillar ectopias, mild syringomyelia, an enlarged medullispinal canal, or lipoma of the terminal filament with the normal medullary cone position? May syringomyelia be secondary to scoliosis?

The expediency of spinal MRI in children with IS, especially in adolescence, is still a matter of debate. The objective is to analyze the frequency of latent neuraxial pathology in idiopathic scoliosis (IS), to substantiate the need for MRI in IS, and to identify promising areas for the use of MRI in the examination of patients with IS.

Material and Methods

The literature review was carried out using the PubMed and Google Scholar databases. The following queries were used: idiopathic scoliosis MRI, neuraxial pathology idiopathic scoliosis, neural axis pathology idiopathic scoliosis, spinal cord pathology idiopathic scoliosis, tumors idiopathic scoliosis, Chiari idiopathic scoliosis, tethered cord idiopathic scoliosis, syringomyelia idiopathic scoliosis. The evidence level is II.

Statistical analysis. Statistical calculations were performed using Microsoft Excel 2010 software with the integrated data analysis package Attestat (13.2, 16/02/2015) and SPSS Statistics (version 21.0.0.0).

For indicators described by quantitative values (age, deformity, percentage of correction, duration of observation), the type of statistical distribution of the collected data was tested. Statistical significance of differences between groups was assessed using Wilcoxon signed-rank test and Mann-Whitney U test for independent and paired samples at $p < 0.05$. For qualitative characteristics, its binomial proportion (D %) and error of binomial propor-

tion (md) were calculated using the formula, where N is the sample size:

$$m = \sqrt{\frac{D \cdot (1 - D)}{N}}.$$

The significance of intergroup differences was evaluated by the non-parametric criterion χ^2 at $p < 0.05$.

Results

In the first half of 2018, three reviews on the problem of latent pathology of the spine in IS were published almost simultaneously. Faloon et al. [20], Heemskerk et al. [2] and Tully et al. [21] analyzed studies on the frequency of identification of neuraxial pathologies in IS and came to the similar results. The frequency of latent neuraxial pathologies ranged from 8 % to 16 %. The authors concluded that MRI of the whole spinal cord with detection of the craniovertebral junction is necessary in all patients with suspected IS, regardless of the absence of neurological symptoms.

We conducted a new systematic review based on current data. Out of 780 papers on the research topic, 65 were selected after removing duplicates and checking for inclusion/exclusion criteria. As a result, 49 original studies were included in the analysis. They presented the frequency of detection of latent pathology of the spinal cord in patients with suspected IS without neurological symptoms who underwent MRI of the whole spinal cord (Table).

All original studies can be divided into three groups according to the patient's age of scoliosis onset. They are studies of adolescent idiopathic scoliosis (AIS) [4, 6, 7, 12, 22–30], early onset scoliosis (EOS) [5, 8, 10, 32–39] and a mixed group (EOS + AIS) [3, 11, 40–55]. It is worth noting that the detection rate of neuraxial pathology in the AIS group was 8.18%, in the mixed group – 8.29 %, and in the EOS group – 16.35 % ($p < 0.001$). Moreover, when analyzing uneven-aged groups, some investigators have shown that neuraxial pathology are detected more frequently in EOS than in AIS [2, 7, 11, 40–42, 44, 45, 47, 53].

Separate studies have been conducted on severe IS (uneven-aged group; more then 80° according to Cobb) [9, 56, 57] and IS with left-sided thoracic/right-sided lumbar curve (uneven-aged group) [58, 59]. The frequency of neuraxial pathology was 37.43 and 40.21%, respectively (Table). Many studies have confirmed these patterns [2, 5, 7, 11, 44, 47, 58].

Additional signs of latent neuraxial pathology, may be male gender [2, 8, 24, 40, 44, 45, 47, 58, 60], thoracic kyphosis more than 40° according to Cobb [2, 24, 25, 42, 44, 47], chronic back pain [44], pathology of reflexes [7, 43, 45, 53], the apex of the deformity in the thoracolumbar spine [47] and the rotation of the vertebrae at the apex of the deformity [11].

Discussion

A review of the literature has revealed five major areas of research regarding the use of MRI in IS. They are the following: 1) search for predictors of latent neuraxial pathology; 2) investigating the need for preventive neurosurgical intervention in combined neuraxial pathology and IS; 3) MRI as an additional technique to study scoliosis and reduce radiation exposure; 4) identification of factors predisposing/combining with the development and progression of IS (MR anatomy of the central nervous system, muscles, bones, etc.); 5) studies of degenerative changes of the spine.

Risk factors for neuraxial pathology in IS

Risk factors for the presence of neuraxial pathology are male gender, age, early onset of deformity, thoracic kyphosis more than 40° according to Cobb, and atypical curve (a left-sided thoracic and/or a right-sided lumbar). Thus, if a male patient, with suspected IS and/or with IS developed under the age of 10 years, with a left-sided thoracic or a right-sided lumbar curve and a thoracic kyphosis more than 40° according to Cobb, has applied to you, then, with a high probability, the patient may also have a latent neuraxial pathology.

In our opinion, it is very difficult to be guided by the severity of the deformity, degree of vertebral torsion and decreased or increased tendon reflexes as signs of latent neuraxial pathology. An MRI should be performed in the early stages of disease. This is because, for some types of latent neuraxial pathology, early neurosurgical intervention can prevent the progression of scoliosis.

Does a patient with asymptomatic neuraxial pathology need surgery before the deformity correction?

It is obvious that when treating scoliosis caused by a tumor, the focus is on the treatment of tumors. Spinal cord tumors require special treatment and must be treated before the deformity correction.

There are studies showing that some patients with no neurological symptoms do not need previous neurosurgery. Deformity correction can be performed independently, mainly with type II diastematomyelia and terminal filament lipoma with a normal level of the medullary cone and without any symptoms [41, 49, 61].

Neurosurgery is not required for mild idiopathic or secondary to scoliosis syringomyelia [3, 24, 26, 36, 37, 41, 46, 49, 51, 55, 61]. If syringomyelia is primary and severe, surgical treatment should be performed before scoliosis correction [6, 7, 40, 51].

The presence of concomitant type I Chiari malformation and syringomyelia is an indication for preliminary neurosurgery before deformity correction. Meanwhile, some patients experience a reduction or stabilization of the scoliotic curve after craniovertebral junction surgery [3, 7–10, 24, 35–38, 40, 44, 46, 51, 62–66]. However, some surgeons perform surgeries on patients with type I Chiari malformation without syringomyelia or specific symptoms before the deformity correction. Brockmeyer et al. [63] noted that in some patients, the scoliotic curve decreased or stabilized during a follow-up period of more than two years [7, 8, 37, 40]. However, most authors consider the preventive surgery unnecessary in this case [3, 24, 26, 35, 36, 46, 49, 55, 67].

Table
The incidence of spinal cord pathology in patients with idiopathic scoliosis (according to the literature data)

Authors	Detected neuraxial pathology, n	Patients, n
Patients of uneven-aged group		
Maiocco et al. [50]	2	45
Emery et al. [51]	25	100
Winter et al. [49]	4	140
Morcuende et al. [41]	11	72
Inoue et al. [42]	44	250
Saifuddin et al. [50]	9	73
Benli et al. [39]	7	104
Hooker et al. [11]	15	78
Rajasekaran et al. [51]	15	94
Oztruk et al. [44]	20	249
Diab et al. [40]	39	923
Nakahara et al. [43]	18	472
Singhal et al. [3]	20	206
Qiao et al. [45]	35	446
Zaveri et al. [31]	32	300
Ameri et al. [38]	27	271
Shen et al. [46]	2	72
Mohanty et al. [52]	25	296
Pazarlis et al. [53]	8	129
Total number	358 (8.29 %)	4320
Patients with AIS		
Cheng et al. [28]	14	164
Do et al. [22]	6	327
Hausmann et al. [27]	3	100
Richards et al. [25]	36	529
Unnicrishnan et al. [29]	14	235
Lee et al. [30]	15	171
Karami et al. [23]	17	143
Lee et al. [24]	24	378
Swarup et al. [6]	18	210
Scaramuzzo et al. [4]	28	140
Xu et al. [7]	68	714
De Oliveira et al. [12]	25	198
Total number	302 (8.18 %)	3690
Patients with EOS		
Lewonowsky et al. [32]	5	26
Evans et al. [33]	8	31
Gupta et al. [10]	6	34
Dobbs et al. [34]	10	46
Pahys et al. [37]	7	54
Koc et al. [35]	8	72
Martin et al. [36]	7	43
Zhang et al. [8]	94	504
Pereira et al. [38]	4	71
Kouri et al. [5]	13	53
Williams et al. [39]	42	314
Total number	204 (16.35 %)	1248

In the presence of latent spinal cord tethering (type I diastematomyelia, thickened terminal filament, or lipoma of the terminal filament with a low location of the medullary cone), most authors recommend preventive neurosurgery [7, 8, 37, 40, 61]. The safety of deformity correction without spinal cord untethering is doubtful, since the neurological symptoms of tethered spinal cord syndrome may manifest in the postoperative period [68]. Additionally, in patients with asymptomatic tethered spinal cord syndrome and scoliosis after untethering, a decrease in scoliosis with a curve of less than 45° according to Cobb has been described [69].

In our clinic, we have the following approaches for the treatment of latent neuraxial pathology in patients with suspected IS. We perform deformity correction with subsequent observation in asymptomatic type I Chiari malformation without syringomyelia, with syringomyelia without tension, type II diastematomyelia, and terminal filament lipoma with the normal position of the medullary cone. Preliminary neurosurgery is recommended for patients with type I Chiari malformation and syringomyelia, with tense syringomyelia, type I diastematomyelia, and a low-lying medullary cone associated with a terminal filament lipoma and thickened terminal filament. Moreover, if a patient has a tumor, Chiari malformation, or syringomyelia, a brain MRI is also recommended.

MRI as an alternative to radiography and CT

In children, CT is associated with a higher dose of ionizing radiation than in adults [70, 71]. Since the 2000s, a number of techniques have been developed and implemented to reduce radiation exposure in children with scoliosis: special pediatric CT settings, low-dose configurations, EOS, scoliometers, and replacing CT with MRI and ultrasound [72–76].

Currently, there is a growing number of studies showing an increased risk of malignancy after CT in children, including girls with AIS [76–84]. Radiation not only increases the risk

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Authors	Detected neuraxial pathology, n	Patients, n
Patients with severe idiopathic scoliosis		
O'Brien et al. [56]	0	33
Freund et al. [9]	21	37
Zhang et al. [8]	43	101
Total number	64 (37.43 %)	171
Patients with idiopathic scoliosis and left-sided thoracic curve		
Mejia et al. [59]	2	29
Wu et al. [58]	37	68
Total number	39 (40.21 %)	97

AIS — adolescent idiopathic scoliosis; EOS — early onset scoliosis.

of tumors but also affects the female reproductive system. In girls with AIS, an association between exposure to CT radiation and failed pregnancy attempts, spontaneous abortions, and congenital malformations was found [85].

Another way to reduce radiation exposure is to avoid or minimize the use of intraoperative CT navigation and C-arm fluoroscopy, as well as free-hand screw insertion techniques. Postoperative CT is widely used to check screw position but is used not in all clinics.

There are a number of studies devoted to comparison of the effectiveness of X-ray, CT, and MRI in the assessment of scoliosis and to preoperative planning of the size and pathway of screws: both positive [86–91] and negative results are described [92, 93].

MR-anatomy of the spinal cord, craniovertebral area, muscles, discs and bones in IS

In addition to the study of genetic etiology in patients with scoliosis, there is a parallel direction of research on the MR structure of tissues and anatomical structures. The most popular directions of research are the following: asynchronous neuro-bone growth [94–96], MR-morphology of the craniovertebral junction and cerebellum [97–100], MR-morphology of paravertebral muscles [101–107], MR-cerebrospinal fluid dynamics [108–111], morphological changes of intervertebral discs and vertebrae according to MRI [112–117], enlargement of the anterior spinal areas according to MRI data [118], and latent anatomical changes of the spinal cord [119–121].

The importance of these studies is that they aim to identify the causes of scoliosis development or deterioration, and so they can help determine ways to prevent disease progression.

Degenerative changes of the spine in IS

It has been proven that degenerative changes occur faster in the spinal tissues in scoliosis, and the degree of these changes is related to the severity of scoliosis. This was detected, among other things, with the help of MRI [122–124]. In addition, greater mobility in the lumbar region is associated with less degeneration of spinal structures and lower back pain. Therefore, maintaining or improving lumbar mobility may be beneficial in the prevention or treatment of back pain in IS [125].

Conclusion

In our opinion, an MRI of the spine should be performed in the early stages of IS. A number of patients with IS may have latent neuraxial pathology, and neurosurgery can prevent the progression of scoliosis in the early stages. This pathology includes type I Chiari malformation, spinal cord tethering, some types of syringomyelia, and tumors. Early onset of IS, a left-sided thoracic curve, male gender and a thoracic kyphosis more than 40° according to Cobb are the main signs of latent neuraxial pathology in IS.

MRI can be used in surgical planning and as an effective non-invasive method in studies dedicated to determining the causes of the development and progression of IS. The main directions of an IS study using MRI are the investigation of structural changes in muscles, bones, intervertebral discs, the nervous system, vascular blood supply, and cerebrospinal fluid dynamics. They are considered as factors that may be related to the development and progression of IS and the investigation of degenerative changes of the spine during medical and surgical treatment of IS.

The study had no sponsors. The authors declare that they have no conflict of interest.
The study was approved by the institution's local ethics committee.
All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.

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