YOUNG PATIENT WITH SPINA BIFIDA OCCULTA – ASSESSMENT AND REHABILITATION

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Abstract

Aim. Spina bifida, one of the frequent congenital anomaly of spine, appears through variable closure defect of vertebral arch, with incomplete development of neural tube, in lumbar or sacral vertebral regions. Imaging examination (mainly radiological) is more essential in diagnosis than clinical evaluation. The objective of this study is to determine if patients suffering from spina bifida occulta and low back pain (without neurological signs) benefit from rehabilitation program.

Materials and Methods. 13 young patients (6 women, 7 men, aged between 14 - 19 years) were observed (clinical-functional evaluations and radiological investigations) and treated during 2017 - 2018. All subjects were evaluated at beginning and after 4 week kinetic program.

Results. All patients improved in pain and quality of life, the differences between mean score values (visual analogue scale score for pain and the quality life score) were significant better in the end of rehabilitation program.

Conclusions. Lumbar spine is one of the frequent sites of spina bifida occulta. This abnormality generates pain, stiffness and functional clinical disabilities in association with other spine congenital syndromes (scoliosis, kyphosis). The objective of any preventive or therapeutic rehabilitation program is to teach patients how to help themselves.

Key words: spina bifida occulta, assessment, rehabilitation program
INTRODUCTION

The vertebral column – part of the axial skeleton, flexible, durable and elastic, made from a succession of functional elements (Junghans motor segments, or functional spinal units, that consist of two adjacent vertebrae, the intervertebral disc and all adjoining ligaments between them and excludes other connecting tissues such as muscles) – has roles in protection (mainly of the nervous system), insertion, support and mobility. The anomalies of the spine are congenital or acquired.[1, 2].

The “spina bifida” term (a literal translation from Latin would mean “an open spine”) refers to a malformation, a defect of the spine that encapsulates various lesions of different gravity and prognosis, that require an adapted therapeutic assessment.

Spina bifida is a congenital disorder where there is incomplete closure of the vertebral arch, resulting in an incomplete development of the neural tube, that usually happens at the end of the first month of embryonic development [3]. Its usual location is in the lower lumbar and sacral region, but can also be located at a cervical level. It is a disease that causes dysfunctions of the locomotor apparatus in children, being ranked in terms of incidence immediately after cerebral palsy [4].

Spina bifida (or posterior rachischisis) is a generic term for several diseases with various degrees of severity, as follows (Image nr. 1):

- Spina bifida occulta (SBO), when the surface skin is intact – the easiest form,
- Spina bifida cystica, when the margins of the neural tube are in direct contact with the skin, meaning there is a mieloschisis:
  - meningocele (the defect of the vertebral arch allows the meninges to herniate between the vertebrae) and
  - myelomeningocele or spina bifida aperta (open) (the unfused portion of the vertebral arch allows the spinal cord to protrude through an opening, forming a sac enclosing the spinal elements, such as meninges, cerebrospinal fluid, and parts of the spinal cord and nerves).

SBO is an easy form of the disease, with mild symptoms, frequently even asymptomatic, consisting only of a closure defect in some vertebrae without nervous tissue herniation, affecting 10% of the population, with no consequences, discoverable only by imaging techniques.

Meningocele (currently named spina bifida) is the moderately severe form, the spinal cord is not injured, can be resolved surgically with or without neurological sequelae in the spinal nerves. Myelomeningocele – located in the lumbar or sacral region, is the most severe form of the disease, as a portion of the spinal cord protrudes through the opening in the vertebral arch, sometimes being covered by skin, sometimes not, with full exposure of nervous tissues. [4, 5].

There are many factors that can lead to the presence of spina bifida, and the genetic predisposition can be influenced by several environment factors. Several studies emphasized the negative impact of a low level of folic acid in mothers before conception. The brain and the spinal cord form in the first 28 days of the pregnancy. Although from reasons yet to be
understood, several factors interfere with the normal development of nervous tissue [2,3]:

- geographical or ethnic factors;
- specific pregnancy events – fever in the first trimester, medication use, nutritional deficiency in zinc and folates (especially folic acid);
- alcohol abuse in the first trimester of pregnancy;
- mother with diabetes mellitus;
- family history of spina bifida.

Clinically speaking, SBO is most of the times asymptomatic, being a frequent discovery of a radiologic examination asked for a patient accusing lower back pain, with the limitation of mobility of the respective vertebral segment, accompanied or not by other complaints.

When associated with other congenital anomalies (transitional vertebra, scoliosis, flat feet), the symptoms may start earlier, thus the age the disease is discovered may be earlier [6,7]. Low lumbar pain in a child or a young person with SBO accompanied by other congenital vertebral anomalies is explained, from a biomechanical point of view, by the abnormal mobility and torque of the intervertebral spaces.

Imagistic examination (especially the radiologic examination) is the main element for a positive diagnosis. On a radiography the area is described as having a reduced intensity, located in the middle, vertically or obliquely. (Image nr. 2).

Currently there is no treatment for spina bifida, but there are several options to relieve the symptoms of patients with spina bifida aperta. In most of the patients a surgical intervention is needed to ease the pain and all problems associated with spina bifida.

The best way to prevent spina bifida is a prenatal correct behaviour and prophylactic treatment: it is recommended for women to have daily supplements of 400mg of folic acid (for example, each portion of bread, pasta or rice contain up to 400mg of folic acid) [3, 6].

The present study is a complete evaluation attempt – clinical, paraclinical, functional – of pupils with SBO, as well as of the particularities of the complex healthcare program.

SUBJECTS AND METHOD

We studied a lot of 13 patients (6 girls and 7 boys), from schools from the city of Craiova, that were followed up from 2017 to 2018 both clinically, paraclinically and functionally. They were all diagnosed with SBO and had episodes of low lumbar pain (LBP).

The patients had ages between 14 and 19 (mean value of 15.3 years old).

None of the patients had in their medical history traumas (falling or aggression) and neither orthopedic or surgical interventions on their vertebral column. Pupils with associated visceral pathology have been excluded from the study.

Each child was carefully questioned for a thorough anamnesis, in order to have the complete profile of their lumbar pain. In all patients the pain started insidious, with progressive accentuation, and with temporary improvement after rest and / or usual painkillers. The pain had a mechanical characteristic, was located at a lumbar-sacral level, with no dermatomes or lower limbs irradiation. The intensity of the felt pain was evaluated using the Visual Analogue Scale (VAS) for pain, with values that range from 0, meaning no pain, to 10, meaning maximum pain. The mean value of spontaneous pain was 7.6 (SD=2.1). None of the patients had neurological events (muscle strength deficiency, with significant impairment of prehension or walking).

The clinical evaluation consisted of a complete clinical exam followed by a careful examination of the vertebral column, including a neurological exam that showed no pathological modifications.
The paraclinical evaluation consisted of a radiological examination of the according vertebral segment (Image nr 3), with the amendment that none of the patients had had a prior radiological exam.

Image nr. 3. Spina bifida L5 (marked with a black arrow)

The functional evaluation was conducted with the help of the Oswestry Low Back Pain Disability Questionnaire LBP (anexa) [9].

All pupils followed an ambulatory kinetic rehabilitation program for four weeks, with the following objectives:

- general, nervous and muscular relaxation;
- suppression of pain, contraction and cervical / lumbar sprain;
- relieving muscle dis-synergies;
- combating static and vertebral dynamics while maintaining a correct posture;
- stabilizing the vertebral segment under the conditions of joints' flexibility and appropriate muscle balance;
- improving quality of life.

Kinetic and massage measures have been judiciously established in accordance with clinical-functional status and patient compliance, respecting the principle of painlessness. Each kinetotherapy session lasted for 30 minutes and was held daily for 5 days / week.

No drug therapy was indicated because we considered that for the clinical-functional status of the study subjects the medication was inexpedient.

Patient evaluation was initially made after 1 and 4 weeks, respectively, based on the average scores of the VAS and the Oswestry questionnaire. The overall score of this questionnaire is obtained by summing the quotes from 0 to 5 for each item of the scale, with a maximum value of 50 and percent expression. In the statistical processing we used the quoted sum, the ratio being 50 for all subjects.

Statistical data processing to assess the efficiency of the kinetic program was done taking into account the regression curve for each of the two studied parameters (VAS and LBP).

RESULTS

The study was conducted on a group of young patients, randomly diagnosed with SBO using the radiological exam, who accused a lumbar pain with minimal functional disorders, in the absence of neurological manifestations.

Although in the specialty literature rachischisis is more commonly located in the cervical spine, in the studied group of patients we observed a lumbar localization.

The presence of these congenital abnormalities contributes, through the perturbation of the biomechanics of the respective vertebral segment, along with other pathological aspects, to the tissue irritation which causes the onset of pain with a state of constant muscle tension. Pain and muscle tension open the pathogenic cascade of inflammation (tissular ischemia, oedema, accumulation of metabolites) that through the final fibrous reaction generates disturbances of soft paravertebral structures, with mobility limitation, muscle tone change, and secondary functional consequences.

All subjects presented an insidious debut of pain for which they had not requested a specialist medical checkup, but the exacerbations of algo-functional phenomena (fatigue, functional impotence at exaggerated mobilization) determined the present consultation.

The objective exam noted the absence of significant changes in the physiological curves of the spine. The paravertebral extensor muscles showed a tendency to retract, in varying degrees in all patients, in contrast to the tendency to hypotonia of the flexor muscles, aspects that were taken into account in the kinetic program.

The VAS score improved in the end by about 50% in all patients, with the regression curve proving this (Image nr.4).
Pain was influenced immediately after the initiation of kinetic treatment, which is supported by the fact that the clinico-functional status of soft paravertebral structures is predominantly influenced by massage and physiotherapy. The regression curve justifies this favorable trend, being confirmed by the optimal predictive values (R square 0.599).

Classical massage associated with physical exercise has the role of fighting pain and muscle contraction, and Cyriax massage allows the physical therapist to control the functionality of perivertebral connective structures (predominantly interspinal ligaments).

The LBP questionnaire had an average value for the studied group of just 18% at the initial stage, meaning a small disability according to the values of this questionnaire. At the final evaluation, after a month of kinetic program, pupil disability decreased to 11%, confirmed by the regression curve aspect, with very high predictability (R squared 0.9) (Image nr. 5).
We found the patient cooperation in the recovery program to be of extreme importance, especially for getting the correct posture, as well as for continuing at home the exercises included in the “back school”. We regard this aspect of kinetic prophylaxis as particularly important because all patients are young, active, socially integrated persons who carry out daily activities that are demanding to the vertebral column.

**DISCUSSION**

The number of studies regarding SBO in young patients is quite limited compared with those regarding symptomatic spina bifida [10].

Diagnosis is often the consequence of a routine imaging exam, when the lumbosacral pain requires complete patient paraclinical exploration [11].

Existing studies emphasize the importance of the functional aspect of the congenital anomaly, with a major impact when the student becomes of adult age and will perform a professional activity that is demanding on the lumbosacral vertebral segment [12].

In the patient who associates SBO with other congenital locomotor malformation, the complexity of infirmity impacts on multiple dysfunctions, which requires a complex evaluation for the formulation and application of a proper health care program [13]. Among other congenital vertebral malformations, SBO is most commonly associated with the transitional vertebra, ie the last lumbar vertebra is abnormally positioned, with the transverse apophyses having anatomical contact with the iliac crest and even with nerve threads, with significant clinical-functional consequences [14].

It is unanimously believed that the early diagnosis of SBO, with consequently explaining the particularities of this entity to both the patient and the family, is of great importance. Under the conditions of an adequate daily life, respecting the rules of orthopedic hygiene of the spine and lower limbs, the young patient who is properly educated about the disease he has, will have no obstacle to the maintain a normal life style [12].

The child and young patient with SBO need to be explained the importance of the kinetic program for a normal functional status of the spine. Through the physical exercise, the optimal parameters of the lumbar and abdominal paravertebral muscles are restored and maintained, while also maintaining the optimal muscular corset.[15].

**CONCLUSIONS**

Lombalgia is a real public health issue described in the various types of spinal pathologies, causing a variety of malfunctions, with a direct impact on the quality of life.

The data we obtained corresponds to those in the specialty literature, according to which congenital vertebral abnormalities, such as SBO, are occasional discoveries in a radiological examination often performed to determine a positive diagnosis in a patient with an inconclusive clinical picture.

The young patient needs to be explained, according to his degree of understanding, the nature of his “suffering” and the importance of the “back school”. Only a compliant patient, mentally prepared for a possible painful episode at any level of the spine, which also proves self-control in managing pain, can be included in the multidisciplinary healthcare team for vertebral sufferings.

Kinetic methods – components of the “back school” – are easy to perform when knowing and understanding the anatomy and biomechanics of the vertebral segments, clearly contributing to the amelioration and prevention of pain symptoms, and to the improvement of muscle strength and of the amplitude of mobilization of the segment (cervical or lumbar).
Anexa. Oswestry Low Back Pain Disability Questionnaire

The Oswestry Disability Index (also known as the Oswestry Low Back Pain Disability Questionnaire) is an extremely important tool that researchers and disability evaluators use to measure a patient’s permanent functional disability.

The test is considered the ‘gold standard’ of low back functional outcome tools

**Scoring instructions**

For each section the total possible score is 5: if the first statement is marked the section score = 0; if the last statement is marked, it = 5. If all 10 sections are completed the score is calculated as follows:

- Example: 16 (total scored) / 50 (total possible score) x 100 = 32%
- If one section is missed or not applicable the score is calculated: 16 (total scored) / 45 (total possible score) x 100 = 35.5%
- Minimum detectable change (90% confidence): 10% points (change of less than this may be attributable to error in the measurement)

<table>
<thead>
<tr>
<th>Interpretation of scores 0% to 20%: minimal disability:</th>
<th>The patient can cope with most living activities. Usually no treatment is indicated apart from advice on lifting sitting and exercise.</th>
</tr>
</thead>
<tbody>
<tr>
<td>21%-40%: moderate disability:</td>
<td>The patient experiences more pain and difficulty with sitting, lifting and standing. Travel and social life are more difficult and they may be disabled from work. Personal care, sexual activity and sleeping are not grossly affected and the patient can usually be managed by conservative means.</td>
</tr>
<tr>
<td>41%-60%: severe disability:</td>
<td>Pain remains the main problem in this group but activities of daily living are affected. These patients require a detailed investigation.</td>
</tr>
<tr>
<td>61%-80%: crippled:</td>
<td>Back pain impinges on all aspects of the patient’s life. Positive intervention is required.</td>
</tr>
<tr>
<td>81%-100%:</td>
<td>These patients are either bed-bound or exaggerating their symptoms.</td>
</tr>
</tbody>
</table>

**Instructions**

This questionnaire has been designed to give us information as to how your back or leg pain is affecting your ability to manage in everyday life. Please answer by checking ONE box in each section for the statement which best applies to you. We realise you may consider that two or more statements in any one section apply but please just shade out the spot that indicates the statement which most clearly describes your problem.

<table>
<thead>
<tr>
<th>Section 1 – Pain intensity</th>
<th>I need some help but manage most of my personal care</th>
</tr>
</thead>
<tbody>
<tr>
<td>I have no pain at the moment</td>
<td>I need help every day in most aspects of self-care</td>
</tr>
<tr>
<td>The pain is very mild at the moment</td>
<td>I do not get dressed, I wash with difficulty and stay in bed</td>
</tr>
<tr>
<td>The pain is moderate at the moment</td>
<td></td>
</tr>
<tr>
<td>The pain is fairly severe at the moment</td>
<td></td>
</tr>
<tr>
<td>The pain is very severe at the moment</td>
<td></td>
</tr>
<tr>
<td>The pain is the worst imaginable at the moment</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Section 2 – Personal care (washing, dressing etc)</th>
<th>I cannot lift or carry anything at all</th>
</tr>
</thead>
<tbody>
<tr>
<td>I can look after myself normally without causing extra pain</td>
<td>Pain does not prevent me walking any distance</td>
</tr>
<tr>
<td>I can look after myself normally but it causes extra pain</td>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□□□□□□□□□</td>
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<tr>
<td>It is painful to look after myself and I am slow and careful</td>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□□□□□□□□□</td>
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<tr>
<td></td>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□ □□□□□□□□□</td>
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<tr>
<td></td>
<td>I can only walk using a stick or crutches</td>
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<tr>
<td></td>
<td>I am in bed most of the time</td>
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<table>
<thead>
<tr>
<th>Section 3 – Lifting</th>
<th></th>
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<tbody>
<tr>
<td>I can lift heavy weights without extra pain</td>
<td>I need some help but manage most of my personal care</td>
</tr>
<tr>
<td>I can lift heavy weights but it gives extra pain</td>
<td>I need help every day in most aspects of self-care</td>
</tr>
<tr>
<td>Pain prevents me from lifting heavy weights off the floor, but I can manage if they are conveniently placed eg. on a table</td>
<td>I do not get dressed, I wash with difficulty and stay in bed</td>
</tr>
<tr>
<td>Pain prevents me from lifting heavy weights, but I can manage light to medium weights if they are conveniently positioned</td>
<td></td>
</tr>
<tr>
<td>I can lift very light weights</td>
<td></td>
</tr>
<tr>
<td>I cannot lift or carry anything at all</td>
<td></td>
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</tbody>
</table>

<table>
<thead>
<tr>
<th>Section 4 – Walking*</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain does not prevent me walking any distance</td>
<td>I cannot lift or carry anything at all</td>
</tr>
<tr>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□□□□□□□□□</td>
<td></td>
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<tr>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□□□□□□□□□</td>
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<tr>
<td>Pain prevents me from walking more than □□□□□□□□□□□□□□□□□□□□□</td>
<td></td>
</tr>
<tr>
<td>I can only walk using a stick or crutches</td>
<td>I need some help but manage most of my personal care</td>
</tr>
<tr>
<td>I am in bed most of the time</td>
<td>I need help every day in most aspects of self-care</td>
</tr>
</tbody>
</table>
### Section 5 – Sitting
- I can sit in any chair as long as I like
- I can only sit in my favourite chair as long as I like
- Pain prevents me sitting more than one hour
- Pain prevents me from sitting more than 30 minutes
- Pain prevents me from sitting more than 10 minutes
- Pain prevents me from sitting at all

### Section 6 – Standing
- I can stand as long as I want without extra pain
- I can stand as long as I want but it gives me extra pain
- Pain prevents me from standing for more than 1 hour
- Pain prevents me from standing for more than 30 minutes
- Pain prevents me from standing for more than 10 minutes
- Pain prevents me from standing at all

### Section 7 – Sleeping
- My sleep is never disturbed by pain
- My sleep is occasionally disturbed by pain
- Because of pain I have less than 6 hours sleep
- Because of pain I have less than 4 hours sleep
- Because of pain I have less than 2 hours sleep
- Pain prevents me from sleeping at all

### Section 8 – Sex life (if applicable)
- My sex life is normal and causes no extra pain
- My sex life is normal but causes some extra pain
- My sex life is nearly normal but is very painful
- My sex life is severely restricted by pain
- My sex life is nearly absent because of pain
- Pain prevents any sex life at all

### Section 9 – Social life
- My social life is normal and gives me no extra pain
- My social life is normal but increases the degree of pain
- Pain has no significant effect on my social life apart from limiting my more energetic interests eg, sport
- Pain has restricted my social life to my home
- Pain has restricted my social life and I do not go out as often
- I have no social life because of pain

### Section 10 – Travelling
- I can travel anywhere without pain
- I can travel anywhere but it gives me extra pain
- Pain is bad but I manage journeys over two hours
- Pain restricts me to journeys of less than one hour
- Pain restricts me to short necessary journeys under 30 minutes
- Pain prevents me from travelling except to receive treatment
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