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METHODS FOR THE PREVENTION OF SCHEUERMANN'S DISEASE IN CHILDREN WITH HEREDITARY DISORDERS OF THE DEVELOPMENT OF CONNECTIVE TISSUE

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Abstract

The research is based on 30 patients with hereditary connective tissue disorders observations over the period 2011 - 2020. This pathological condition is a skeleton systemic disease, based on the vicious development of connective tissue. In the dynamic observation course, the main clinical symptoms of the disease were characterized, among other things, by the presence of curved limbs due to previously suffered fractures (from 3 or more in each patient's medical history), muscle hypotonia, vicious posture (scoliotic, kyphotic, kyphoscoliotic, flat back), blue scleras' visualization, the presence of carious teeth. The results of laboratory, x-ray, and densitometric examination methods were the final link in making a clinical diagnosis. Early detection of connective tissue pathology allows avoiding and minimizing the most dangerous complications of the disease, such as idiopathic scoliosis, early juvenile osteochondrosis, Scheuermann's disease.

Keywords

Remedial gymnastics – Children's orthopedics – Pediatrics – Scheuermann's

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Introduction

Hereditary connective tissue developmental disorders in childhood with the manifestation of further dynamic structural pathological changes in the supporting tissues are a problem today. Pathological nosologies with gross osteogenic violations are called osteodysplasia, and the most common - systemic lesions of the skeleton[1]. «Adolescent idiopathic scoliosis (AIS), with a prevalence of 0.5%–5.2% in adolescents aged 10–16 years old, is defined as a lateral curvature of the spine combined with axial vertebral rotation and/or sagittal plane imbalance»[2]. They are classified as congenital malformations, inherited predisposed and acquired. Under hereditary connective tissue developmental disorders (connective tissue dysplasia), it is customary to consider systemic diseases of the skeleton with certain disorders in the supporting tissues and their progression during periods of children's intensive linear growth and their skeletal structure formation[3]. Against this background bone deformities, and other pathological changes are often joined[2]. One of them is the growing spine disease – Scheuermann's disease, in which the pathological process stage correlates with the growth periods. This spine disease is almost always preceded by a vicious posture formation[3].

Methods

Research aim. Sheuermann – Mau disease prevention in children with early vicious posture diagnosis on connective tissue hereditary disorders developmental background.

Rationale. Currently, in pediatric orthopedist - traumatologist practice, it is becoming increasingly obvious that further improvement of preventive measures effectiveness, diagnosis, treatment and medical examination of patients with Sheuermann-Mau disease against the background of hereditary connective tissue developmental disorders, and other complications' further development is impossible without scientifically based concepts and approaches.

Scheuermann's disease is the most common cause of hyperkyphosis of the thoracic and thoracolumbar spine that affects adolescents. After idiopathic scoliosis, Scheuermann's disease is the most common developmental disorder in patients with a spine deformity[4]. The prevalence of Scheuermann's kyphosis reportedly ranges from 0.4% to 8% across the population[5] although its true incidence is likely understated since it is often overlooked or attributed to poor posture[6].

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"Scheuermann's disease was initially described in 1920 by Scheuermann as a rigid thoracic and thoracolumbar kyphosis associated with wedging of vertebral bodies occurring in late childhood. This definition has evolved over the years, and in 1964, Sorensen proposed a new definition using specific criteria of three or more adjacent apical vertebrae that are individually wedged by an angle of 5° or more. Conversely, other authors advocated different criteria. In 1987, Ali et al. stated that two or more adjacent apical vertebrae were adequate for the diagnosis, whereas Sachs et al"[7].

This anomaly occurs more often in our daily activities[8]. There is an increase in pathological conditions directly related to dysplastic processes in the body, connective tissue dysplasia. "Adolescent idiopathic scoliosis (AIS) affects 0.47%–11.1% of the general population[9]. As the name suggests, the cause of the condition remains unclear. Several theories have been proposed, including genetic, neuro-developmental abnormalities, motor control and motor-sensory integration dysfunctions, vestibular and proprioceptive disorders, biomechanical growth modulation, uncoupled spinal neuro-osseous growth, thoracospinal concept, systemic and metabolic disorders[10] and decreased bone density[11]. The topic relevance is due to its prevalence, systemic lesions, involvement in the pathological process in many body structures, systems, including the bone and joint apparatus, to a greater extent, the development of diseases, more often, pathological structural changes in the spine[12]. "The risk of curve progression is the greatest with skeletally immature patients and thus warrants close monitoring or early intervention"[13]. In recent years, clinicians, radiologists, and pathologists have focused on issues of bone pathology, in particular, the vertebral column. The contingent of patients with musculoskeletal system diseases, the course of bone pathological processes against the background of concomitant dysplastic abnormalities in children10 has changed[14].

Lowe y M. D. Kasten, "An analysis of sagittal curves and balance after Cotrel-Dubousset instrumentation for kyphosis secondary to Scheuermann's disease: a review of 32 patients", Spine (Phila Pa 1976) num 19 (1994):1680–1685

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The term "dysplasia" should be understood as an incorrect, perverse development. The main mass of dysplastic processes are congenital malformations of the skeleton. Bone system dysplasia is diverse. These are changes in a bone as an organ and changes in the skeleton as bone tissue[15].

Clinical manifestations of connective tissue disorders in children are diverse. This is not always a classic picture (forme pleine) of pathology. The patient's specific appearance and increased fragility of the skeleton bones are combined with severe pathological changes in internal organs. In our practice, joint hypermobility and posture disorders in childhood are the basic symptoms of hereditary disorders of connective tissue development. According to the severity of lesions, connective tissue dysplasia is characterized as undifferentiated and differentiated. The most severe are collagenopathies. Undifferentiated forms are most common in practice. They are diagnosed using phenotypic features and do not fit into any of the obvious typical pathologies (for example, Marfan syndrome, osteogenesis imperfecta). These patients had a history of numerous fractures and limbs deformities.

There is an increase in the number of patients whose symptoms do not fit into the classical understanding of hereditary connective tissue developmental disorders.

It should be noted that there are few studies devoted to these cases management study.

Special attention should be paid to children with impaired posture in the frontal, sagittal plane, flat back against the background of hereditary connective tissue developmental disorders. It is noted and statistically proved that spinal osteochondropathy (Sheuermann – Mau disease) development is more common in patients with hereditary connective tissue developmental disorders. Pathological changes of the spine in children were more often observed at the age of 9-14 years. They were localized mainly in the mid-thoracic section of the spine.

Currently, there are difficulties and contradictions with such patients' treatment. There are no clear algorithms for medical examination, the patients' management, certain tactical approaches in preventive measures, diagnostics, and treatment.

Materials and methods

The research is based on 60 patients' observations (30 patients made up the main group, the same number as the comparison group), with hereditary connective tissue developmental disorders, and 30 healthy children (the control group). According to gender and age criteria, patients in the groups were distributed evenly, equally. Patients were registered at the city children's trauma bay of the krai government-owned publicly funded health care institution (KGPHCI) "Children's City Polyclinic No. 9, Barnaul" in the period from 2011 to 2020. In each group there were 15 boys and 15 girls aged from 1 month to 17 years.

L. S. Evert; S. V. Borozdun y E. I. Bobrova et al., "Diagnostics of connective tissue dysplasia using biomarkers", Journal of the Siberian Federal University. Series: Chemistry num 4 (2009): 385-390.

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The study was based on international principles, standards and research methods SQUIRE Standards for Quality Improvement Reporting Excellence[16]; STARD statement for reporting diagnostic accuracy studies[17], AGREE reporting checklist to improve reporting of clinical practice guidelines[18]; •CONSORT statement for reporting randomized controlled trials[19]; COREQ consolidated criteria for reporting qualitative research[20].

The work done does not infringe on the rights and does not jeopardize the well-being of children in accordance with the ethical standards of the Committee for the Rights of Experiments of the 2008 Helsinki Declaration[21]. Parental consent to examine children was obtained.

Research results

Signs characteristic of hereditary connective tissue developmental disorders were observed in 100% cases in patients from the main group and the comparison group. Patients were characterized by pronounced changes in the musculoskeletal system, visceral manifestations characteristic of hereditary connective tissue developmental disorders.

In anamnesis (up to a year of life), all patients (in 100% of cases) of the main group and the comparison group, during the dispensary observation period, from 2011 to 2020, were diagnosed with hip dysplasia (in 50% of cases), myogenic torticollis (in 20% of cases). A course of conservative treatment was perrmed.

The results of the study were subjected to statistical processing with the calculation of the arithmetic mean value, standard deviation, arithmetic mean error, student t-test, error probability according to the table of this coefficient [22]. A posture violation in the frontal (sagittal) plane, flat back was diagnosed in all patients of the main group and the comparison group during the child's stay in a preschool institution, at the age of 5 years on preventive examinations. For biochemical studies of blood and urine, an automatic biochemical analyzer HITACHI-911E, a semi-automatic photometer Epo11-20 from ECO-MED-POLL and a spectrophotometer SF-46 were used.

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[22] A. M. Grjibovski, "Social variations in fetal growth in Northwest Russia: an analysis of medical records", Annals of Epidemiology num 9 (2003): 599–605

The following list of methods was used: total calcium determination in blood serum and urine, inorganic phosphorus in blood and urine, alkaline phosphatase in blood, oxyproline in blood and urine.

The main laboratory values at the dispensary registration beginning in the two groups differed from healthy children ones (control group). The most significant value of the collagen metabolism state in the body is the amount of oxyproline in blood serum and urine. This examination determines connective tissue damage depth and extent. All patients had an increase in oxyproline concentration in the blood serum, oxyproline and calcium urinary excretion. This is testified to collagen metabolism violation in patients. Changes in oxyproline level in the blood serum and in renal excretion may confirm the fact of collagen fibers impaired formation, one of the connective tissue matrix main components, including growth zones. In the group of healthy children, no changes were detected (Table 1).

Values	Main group (n=30) M±m	Comparison group (n=30) M±m	Control group (n=30) M±m	P_{1-2}	P_{1-3}	P_{2-3}
	1	2	3			
Blood calcium (mmol/l)	2,38±0,02	2,37±0,01	2,38±0,02	0,977	0,981	0,832
Blood phosphorus (mmol/l)	1,54±0,02	1,58±0,01	1,57±0,02	0,319	0,677	0,946
Urine calcium (mmol/l)	3,69±0,07	3,67±0,08	2,33±0,09	0,999	**<0,001**	**<0,001**
Urine phosphorus (mmol/l)	24,89±0,39	25,37±0,51	25,32±0,64	0,841	0,922	0,999
Blood oxyproline (mcM/l)	27,20±0,46	27,21±0,36	15,09±0,07	0,999	**<0,001**	**<0,001**
Urine oxyproline (mmole/mg of creatinine)	26,21±0,37	26,58±0,33	19,48±0,04	0,843	**<0,001**	**<0,001**

Note: P - significance level when hypotheses checking, the corresponding p is less than 0.05. Values were compared between groups: 1-2; 1-3; 2-3.

Table1

Main laboratory values at the dispensary registration beginning

The most important aspect of preventive measures in patients from the main group was the orientation to relieve the damaged spine segment. If there are complaints of pain, pain syndrome should be relieved, muscle corset strengthening and possible further complications (spinal osteochondropathy (Sheuermann - Mau disease)) prevention. Diagnosis of this disease was based on clinical and radiological parameters.

There was a clear stage of the disease. The first stage of the pathological process was observed in children aged 10-11 years. The second stage was diagnosed by the age of 12-14. The third stage is by the age of 16-17. Visually, the presence of thoracic kyphosis was determined in patients – the angle of deformation according to Cobb was from 20 to 45* or more (Fig. 1).

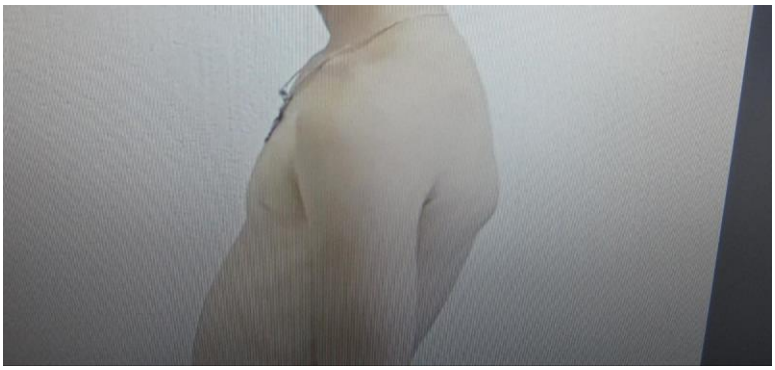

Figure 1
Thoracic hyperkyphosis[23]

Later, in patients from the comparison group, kyphotic changes progressed, in contrast to the patients of the main group (Fig. 2).

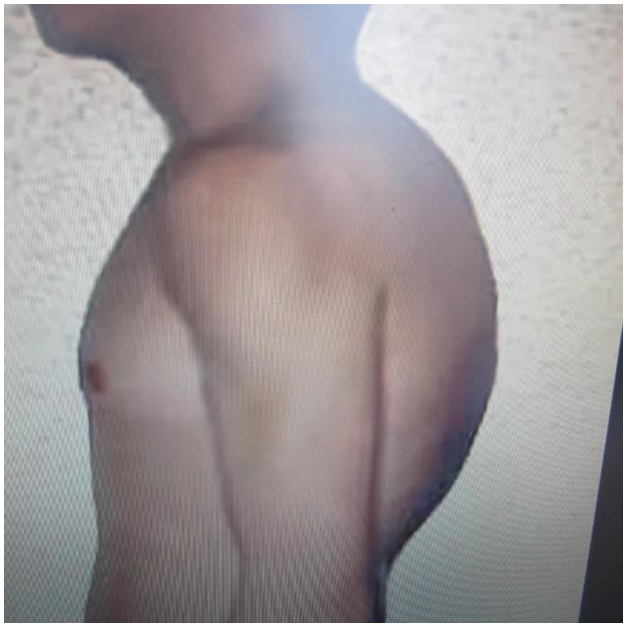

Figure 2
Pronounced kyphosis of the patient in the comparison group All x-ray examinations were performed on the Prestige 1S device

[23] WMA Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects. [Internet]. 2013. [cited 2019 Jul 12]. Available from: http://www.ub.edu/recerca/Bioetica/doc/Declaracio_Helsinki_2013.pdf

During x-ray examinations, the child was placed with the device centered on the damaged spine - lying on his stomach and on his side. X-ray signs were characterized by the wedge-shaped bodies of at least 3 vertebrae, more often, in the thoracic department, there was a protrusion of disks in the vertebral bodies – Schmorl's hernias, vertebral bodies' closing plates deformations, some narrowing of the intervertebral space (Fig. 3).

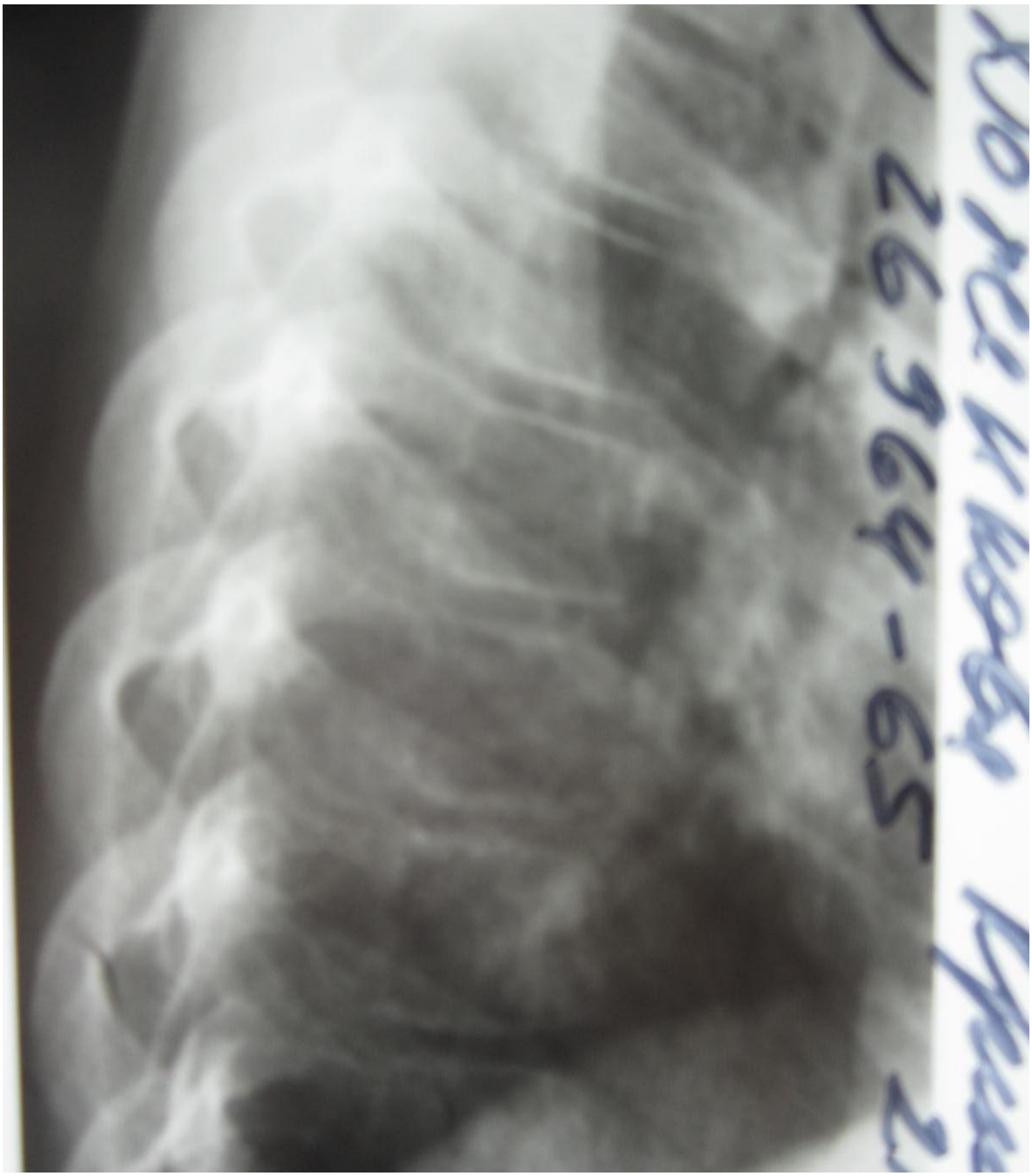

Figure 3
X-ray of Scheuermann-Mau disease initial manifestation
in the lateral projection of the patient

Under dynamic control, in patients from the comparison group, vertebral bodies adjacent to the disks surfaces appeared uneven, deep indentations and usures were determined. Localization of usurps was observed in the front parts of the surfaces (Fig. 4).

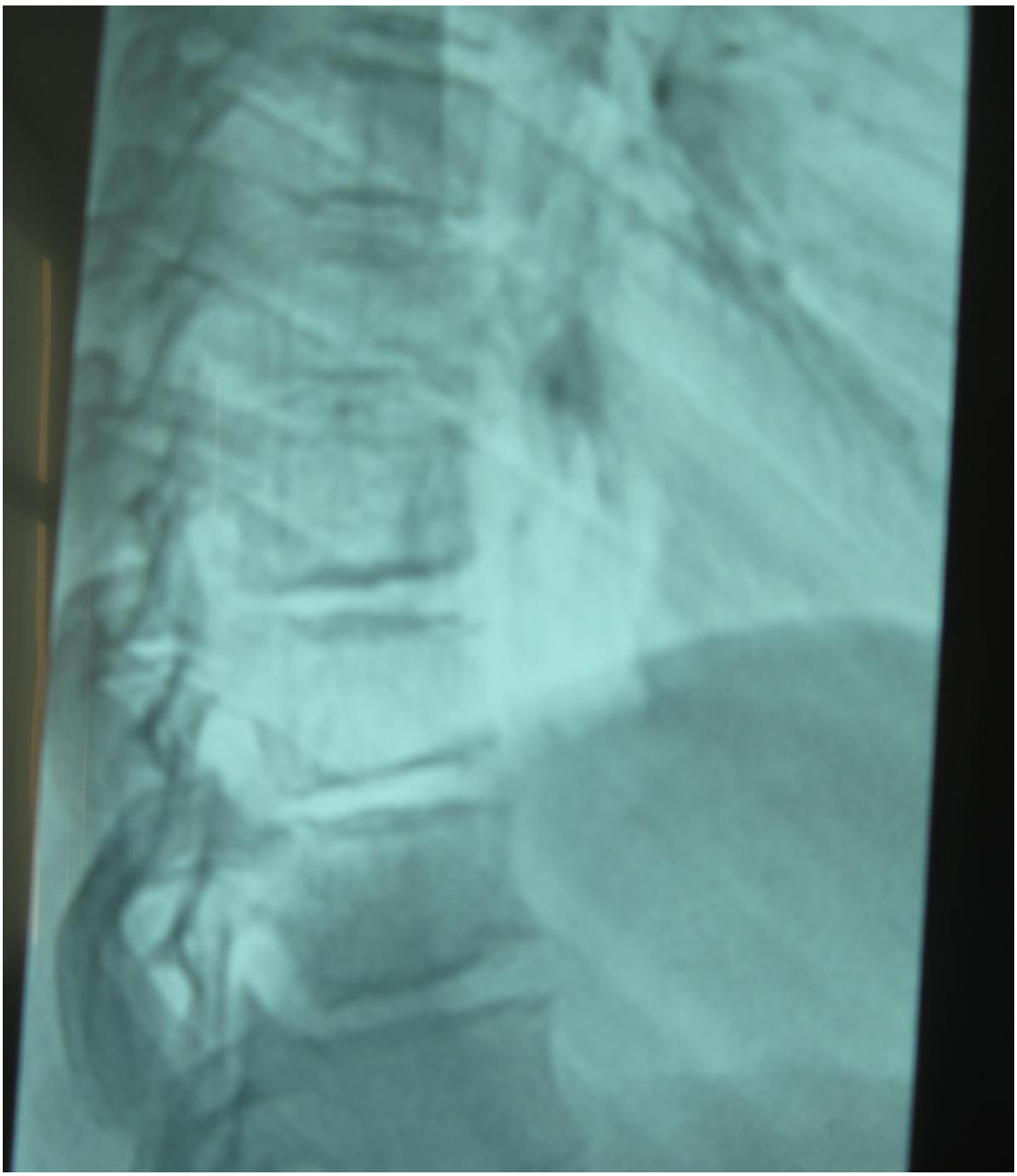

Figure 4
X-ray of a patient with Scheuermann-Mau disease in a lateral projection in dynamics

With the pathological process progression, the indentations on the vertebral bodies edges became distinct, and sclerotic compaction of bone tissue was determined. Between the vertebrae there is a fibrous ankylosis, a fibrous connection of the vertebrae (Fig. 5).

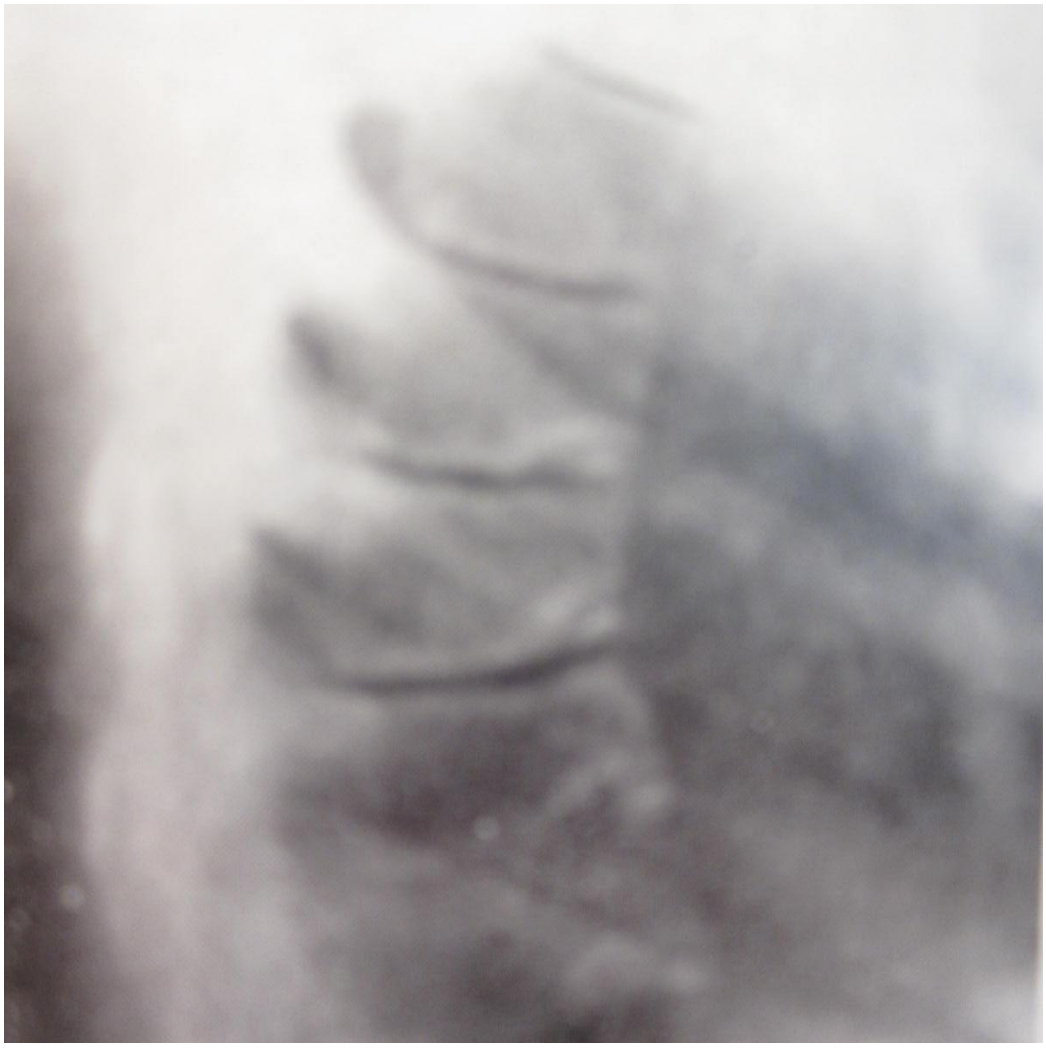

Figure 5
X-ray of a comparison group patient with third stage of Scheuermann-Mau disease in a side projection
Conducting magnetic resonance studies at the early stages of the pathological process allowed for a more accurate diagnosis of the disease in its asymptomatic course

The study was performed in direct

Lateral projections on a 0.5-TL Gyroscan T5-II MP tomograph. At the same time, a number of osteoarticular system dysplastic manifestations were detected in all patients of the main group and the comparison group: flat-valgus (flat-varus) feet position, keel-shaped (funnel-shaped) chest deformation, joints hypermobility.

Comprehensive treatment and prevention measures helped to correct and prevent the development of the pathological process. The positive dynamics are reflected in the table below (Table 2).

Symptoms	Main group (n=30)		Comparison group (n=30)		P (according to Fisher exact test)
	Absolute count	%	Absolute count	%	
Back fatigue	0	0,0	15	50,0	**<0,001**
Pain during spinous processes palpation	0	0,0	10	33,3	**0,003**
Weak core	0	0,0	25	83,3	**<0,001**

Table 2
Statics and dynamics functional disorders in children after 5 years from the dispensary observation beginning
Note: For frequencies less than 5, four-field Fischer conjugacy tables method was used (according to the author)

The child was offered an orthopedic functional bed at home. Course of remedial gymnastics was conducted in a polyclinic with the participation of an instructor and a remedial gymnastics doctor. Comprehensive preventive functional programs in children of the main group provided for improving vital organs function, external respiration, lymph circulation, blood supply, and strengthening the muscles of the back and abdomen.

If necessary, strictly individually, an orthopedic corset (type KP1-26/2 (GC-520) was prescribed for the thoracic and lumbosacral spine. Indications for it were the initial stages of Scheuermann-Mau disease. The corset basis was made of cotton fabric, which has breathability and heat-saving properties. The corrective effect of the product was carried out due to long metal stiffeners, which have an increased spring-loading property. The upper shoulder girdle was separated by two straps that crossed on the back and were fixed on the stomach. This composition of modeling the stiffening ribs by bending the vertebral column and the tension of the straps made it possible to choose the corset individually.

The corset corrective actions mechanism consisted in horizontal and vertical reclination and straps tension intensity. At the same time, the clavicles were separated, their acromial ends were lowered, and the shoulder blades were reduced and lowered.

Vertical reclination occurred due to long stiffening ribs located paravertebrally above the transverse processes of the vertebrae, as well as indirectly through the clavicles, sternum and upper ribs from the straps pressure on the shoulder joints.

The outpatient stage provided for medical supervision of the patient by an orthopedic traumatologist in the conditions of the city children's polyclinic once in 4 months, if necessary, individually, once in 3 months.

The tasks of the outpatient stage included the protective training regime creation, the muscle core formation, and the favorable conditions for vertebrae functional recovery creation in the motor segments of the vertebral column.

During the entire period of clinical observation, the patient was recommended to sleep on a hard bed. Under strict parents' supervision, they were allowed not to use the assigned corset in the morning hours within the apartment or house.

All children from the main group received a complex vitamin composition including calcium in the form of hydroxyappatite, trace nutrients, chondroitin sulfate, and vitamins. The dose of the drug and the course of administration were selected individually, taking into account the child's age and weight. Repeated treatment courses were performed 3 times a year. Prescribed therapeutic swimming in the style of "breaststroke", as well as "hangs" exercises on wall - bars with a pull-up on the hands.

Special attention was paid to food intake. The food ration included:

animal proteins (meat, fish, seafood, milk and dairy products, cottage cheese, cheese);
plant-based proteins (legumes);
strong broths, jelly, aspic dishes (meat, fish, fruit jellies);
products containing calcium (hard cheeses, fish, meat, milk, etc.);
dietary supplements with polyunsaturated fatty acids of the "Omega" class.

It was necessary to take into account vitamin D role. Several theories have been proposed to explain the etiology of adolescent idiopathic scoliosis (AIS) until present. However, limited data are available regarding the impact of vitamin D insufficiency or deficiency on scoliosis. Previous studies have shown that vitamin D deficiency and insufficiency are prevalent in adolescents, including AIS patients. Vitamin D deficiency is believed to play a role in AIS pathogenesis. This study attempts to review the relevant literature on AIS etiology to examine the association of vitamin D and various current theories. Our review suggested that vitamin D deficiency is associated with several current etiological theories of AIS.

Foreign researchers also note the great importance of the diet[11].

The following physical therapy procedures were used:

1) electrophoresis with polymineral wipes for 10 - 15 sessions 2 times a year;
1) mud treatment;
2) electrical stimulation of the spine for 10 - 15 sessions 2 times a year;
3) magnetic therapy of the spine for 10 - 15 sessions 2 times a year.

In the course of medical follow-up, patients from the main group, as a result of complex preventive programs classes, the number of spinal osteochondropathy development decreased in the adolescent period, other complications number from the vertebral column minimized (tables 3, 4).

Complications	Main group (n=30), abs. (%)	Comparison group (n=30), abs. (%)	Control group (n=30), abs. (%)	P_{1-2}	P_{1-3}	P_{2-3}
	1	2	3			
Scheuermann-Mau disease	6 (16,7)	17 (50,0)	3 (0,0)	**0,024**	0,851	**0,001**

Note: P – significance level when hypotheses checking, the corresponding p is less than 0.05. Values were compared between groups: 1-2; 1-3; 2-3.

Table 3

Scheuermann-Mau disease development in adolescence

Complications	Main group (n=30), abs. (%)	Comparison group (n=30), abs. (%)	Healthy children's group (n=30), abs. (%)	P_{1-2}	P_{1-3}	P_{2-3}
	1	2	3			
Scoliotic deformity of the thoracic spine	2 (6,7)	8 (26,7)	0 (0,0)	0,230	0,853	**0,023**
Juvenile spine thoracic section osteochondrosis	0 (0,0)	5 (16,7)	0 (0,0)	0,174	0,999	0,174

Note: P – significance level when hypotheses checking, the corresponding p is less than 0.05. Values were compared between groups: 1-2; 1-3; 2-3.

Table 4

Complications development in adolescence

Discussion

After receiving our own research results and their detailed analysis, the research work is inevitably reduced to comparing the data obtained with the experience of domestic and foreign authors. Results in patients of the main group five years after the start of comprehensive preventive programs revealed an increase in back muscle endurance, a minimum number of arthritis, arthralgic syndromes and coccygodinias manifestations. By the age of 14 a good posture, a developed muscular core, a minimal number of complications, in particular, the manifestation of Sheuermann-Mau disease were noted.

The effectiveness of preventive measures aimed at stabilizing the pathological process at initial stages, preventing the progression of the disease, the transition to the next stages of Sheuermann-Mau disease in children with impaired posture against the background of hereditary connective tissue development is noted, the expediency of early

medical examination in outpatient polyclinic conditions on the basis of a city children's polyclinic is justified. Practical recommendations:

1. When detecting early signs of hereditary connective tissue developmental disorders in children at the primary treatment stage to an outpatient doctor, it is necessary to conduct a follow-up examination, medical examination with examination of patients for possible pathological changes detection on the vertebral column part, the initial manifestations of Sheuermann-Mau disease.
2. When detecting such forms of posture disorders as a flat back, stooped back, round back-to treat at an early stage, immediately, do not use "wait-and-see" tactics.
3. Patients with impaired posture who are at risk in terms of possible further Sheuermann-Mau disease formation in preschool, school, and adolescent groups, comprehensive prevention programs should be recommended, using a special diet, vitamin calcium-containing compounds, and special gymnastic exercises.
4. Do not allow children to engage in difficult sports. As the researchers note,"In a preliminary longitudinal case-controlled study, Adolescent idiopathic scoliosis (AIS) was negatively associated with participation in dance, skating, gymnastics, karate, football, and hockey, suggesting preventative properties"[24].
5. According to foreign research works, it is important to pay attention to children's psycho-emotional state and provide them with psychological support, create a comfortable psychological environment and a friendly attitude to such children from their parents.

Conclusión

Dysplastic process, hereditary connective tissue developmental disorders include, among other things, bone and joint structure pathology.

The clinical symptoms of the disease are mainly reduced to curved limbs presence, due to fractures, muscle hypotension, impaired posture, and the presence of carious teeth.

Additional laboratory, x-ray, and densitometric methods of examination confirmed the pathological condition development.

Competent, tactical medical and rehabilitation measures helped to achieve the desired result, in the future, patients underwent a dispensary examination until adulthood.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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