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Pectus excavatum in children: modern aspects of the problem

Natallia V. Motorenko[1], Alexandr V. Vinnik[2]

[1]*Gomel State Medical University, Gomel, Belarus*

[2]*Gomel Regional Children's Clinical Hospital, Gomel, Belarus*

Abstract

The article provides a historical background of the etiopathogenesis, approaches to diagnosis and surgical treatment of pectus excavatum (PE). Foreign and domestic literary sources on morphological changes in the chest and the functional state of internal organs in patients with intracranial hypertension and correlations with diseases of other organs and systems were studied. Data on diagnostics and treatment tactics for PE are summarized and systematized.

Keywords: *congenital anomalies of the chest, pectus excavatum, instrumental research methods, thoracoplasty*

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Воронкообразная деформация грудной клетки у детей: современные аспекты проблемы

Н. В. Моторенко[1], А. В. Винник[2]

[1]*Гомельский государственный медицинский университет, г. Гомель, Беларусь*

[2]*Гомельская областная детская клиническая больница, г. Гомель, Беларусь*

Резюме

В статье представлена историческая справка, содержащая информацию о этиопатогенезе, подходах к диагностике и оперативному лечению воронкообразной деформации грудной клетки (ВДГК). Проанализированы зарубежные и отечественные литературные источники, описывающие результаты исследований морфологических изменений грудной клетки и функционального состояния внутренних органов при ВДГК, корреляции с заболеваниями других органов и систем. Обобщены и систематизированы данные по диагностике и лечебной тактике при ВДГК.

Ключевые слова: *врожденные аномалии развития грудной клетки, воронкообразная деформация грудной клетки, инструментальные методы обследования, торакопластика*

Вклад авторов. Моторенко Н.В., Винник А.В.: концепция исследования, обзор публикаций, написание текста, редактирование, утверждение рукописи для публикации.

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Introduction

Currently, there is a large group of congenital anomalies of the sternocostal complex, 90% of which are pectus excavatum deformity (PE). Pectus excavatum deformity (sunken chest, funnel chest, cobbler's chest, pectus excavatum) is the most common malformation of the chest; according to various authors it ranges from 0.6% to 2.3% [1]. PE is a curvature of the sternum and anterior ribs of varying depth and shape, leading to a decrease

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in the volume of the chest, compression and displacement of the mediastinal organs and causing functional disorders of the cardiovascular and respiratory systems, manifested by cosmetic defects of varying severity [2, 3]. This pathology is a problem not only for pediatric orthopedics, but also a problem for pulmonologists, cardiologists, psychologists, rehabilitation specialists, and geneticists. Taking into account not only the cosmetic defect, but also cardiorespiratory changes, which are aggravated with an increase in the degree of deformation of the chest, it is necessary to conduct a maximum comprehensive examination of patients with PE with all diagnostic methods (laboratory, instrumental) and the involvement of specialized specialists to determine the indications and timing of reconstructive thoracoplasty taking into account the preservation of the body's compensatory capabilities, and functional disorders of the lungs and heart, which will ultimately lead to the best cosmetic and physiological result.

Etiology of PE

PE has long been known to mankind. So, for example, in the drawing by Leonardo da Vinci, dated 1490, the Vitruvian man is depicted with a clearly visible funnel chest deformity. Currently, there are many hypotheses and theories of the origin of PE. For the first time, PE was described by the Swiss anatomist J. Bauhinus in 1596 [4]. He developed the theory of retraction — pull in the dorsal direction of the diaphragm causes the development of this anomaly. One of the proofs of this theory is the development of PE in children operated on for diaphragmatic hernias [5]. Another suggestion was made by R. Y. Sweet. According to this theory, the sternum is displaced posteriorly towards the spine by unusually long, inwardly curved costal cartilages [6]. There are theories that the development of PE is based on a decrease in the strength of costal cartilages as a result of changes in the quantitative and qualitative content of collagen, glycosaminoglycans, and water [7]. This leads to an inability to support the anterior chest wall with the costal cartilages during breathing, resulting in progressive depression of the sternum. A number of authors believe that the reason for the retraction of the sternum is the excessive growth of the sternocostal cartilages, which outstrips the growth of the ribs, thereby screwing the sternum into the chest. The process is based on a violation of the structure of cartilage tissue due to changes in the structure of cartilage cells and an intermediate substance. Cellular and nuclear polymorphism, the presence of connecting elements in the cartilage prove the presence of primary embryonic cartilaginous structures. Oshesner and DeBakey were the first to come to this conclusion back in 1939, believing that it is the excessive growth of the costal cartilages that prevents the chest from developing physiologically, thereby causing funnel-shaped deformity of the sternum [8].

In the middle of the XX century, scientists were inclined to the hypothesis of a shortening of the anterior sections of the diaphragm or a short sternophrenic ligament, as a result of which the sternum is "pulled up" to the spine and a funnel-shaped retraction occurs in the sternum [9–11].

Currently, scientists consider PE as one of the phenotypic manifestations of connective tissue dysplasia [12,13]. The term "dysplasia" refers to the abnormal growth/development of a tissue or organ. Connective tissue dysplasia is a genetically determined process, which is based on a mutation of the genes responsible for the synthesis of collagen structures. The occurrence of dysplastic changes in the cartilage is associated with the pathology of enzymes that ensure the catabolism of glycosaminoglycans of the main substance of the connective tissue [14]. A distinct redistribution between the content of glycosaminoglycans and glycoproteins towards an increase in the latter, a change in the type of collagen in the form of the appearance of III and IV of its types, not found in full-fledged cartilage, was revealed. Changes in the structure, type of collagen, packing density and their relationship with glycosaminoglycans and glycoproteins can cause the formation of unstable abnormal collagen structures and affect the biomechanical strength of the costal cartilage. The significant role of the abnormal structure of the costal cartilage in the pathogenesis of PE is proved by the high incidence of deformity of the anterior chest wall in patients with connective tissue anomalies, such as Marfan syndrome and scoliosis [14–15].

Abalmasova E.A. and Luzina E.V., in 1976 focused on the importance of heredity as a leading factor in the etiology of congenital chest deformities [16]. They believe that this pathology is a manifestation of congenital systemic processes, since most often PE occurs in children with Marfan syndrome, neurofibromatosis, and dysraphic status [11–13].

Pathogenesis of PE

As a result of deformity of the sternum and ribs in patients with PE, the distance from the sternum to the spine decreases, the chest flattens, the ribs take on an oblique or oblique position, and the location of the pectoral muscles and diaphragm changes. A decrease in the volume of the chest leads to hypertension in the pulmonary circulation, chronic hypoxia, and functional disorders of the organs of the chest cavity. As a result of incomplete expansion of the lungs, the "anatomical dead space" increases, alveolar ventilation decreases, and the drainage function

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of the bronchi is disturbed with the formation of thoracophrenic syndrome.

As a result, patients suffer from frequent respiratory diseases with a protracted course. Compression of the lungs, respiratory failure, sputum stagnation in the bronchi lead to shortness of breath, obsessive cough. Clinically, this is manifested by a picture of tracheitis, bronchitis, and pneumonia more often in lower parts of the lungs. The frequency of respiratory movements and the value of the respiratory volume increase, due to which the compensation of the respiratory function is achieved with normal indicators of the vital capacity of the lungs (VC), maximum ventilation of the lungs (MVL), respiratory reserve. With age, as the compensatory mechanisms are depleted, respiratory function decreases, that leads to chronic oxygen starvation and the formation of dystrophic changes in systems and organs [17].

A change in shape of the chest leads to displacement and rotation of the heart, “torsion” of the vascular trunks. As a result of an increase in intrathoracic pressure, the inflow and outflow of blood is disturbed, which contributes to the occurrence of cardiac arrhythmias, increased pressure in the pulmonary circulation system, pulmonary hypertension, and dilatation of the right chambers of the heart. The pumping function of the heart decreases, despite the increase in myocardial mass and a compensatory increase in the active filling of the ventricles. The shock and minute volume of blood decreases. Patients of this group often have minor anomalies in the development of the heart, in the form of mitral valve prolapse, an open foramen ovale, and additional chords in the heart chambers [18]. The degree of deformation determines the nature and severity of changes in the morphofunctional parameters of the heart and lungs.

Clinic PE

In the neonatal period funnel-shaped retraction in the sternum can be insignificant and manifest itself as paradoxical breathing due to underdevelopment of the sternal leg of the diaphragm, with retraction of the sternum and ribs on inspiration. Frequent acute respiratory viral infections, which tend to be complicated by pneumonia, should draw attention. This is due to the fact that the deformation of the chest affects the functional state of the respiratory system.

At the age of 3 to 7 years various changes in the shape can be detected, which are mainly localized in the sternum and look like depressions, the edges of the costal arches protrude forward and a transverse groove forms under them — “Harrison’s pseudo-groove”. At the same time, there is no violation of the function of external respiration. The form depends on the type of changes, which are determined in accordance with the modern classification. Such children are prone to colds, but physical and psychomotor development does not suffer.

In children older than 7 years (usually up to 10 years), the deformity is well-defined with the naked eye. However, PE reaches its greatest severity in children at puberty. Parents note the active growth of the child, at the same time accompanied by an increase in the retraction of the sternum and sternocostal cartilages, which were previously ignored. As the child grows and the deformity progresses, other external signs appear. Children have a rather typical appearance: their head and neck protrude forward, their abdomen is protruding. In severe cases, there is a noticeable lag in physical development. Such children complain of constant fatigue, headaches, dizziness, increased sweating, heart failure, shortness of breath on the slightest physical exertion [19]. Against the background of severe deformity, the functional state of the heart may suffer, which is accompanied by arrhythmia, an increase or decrease in blood pressure [20]. This often causes discomfort in the chest in the form of tingling or burning. The deformity becomes fixed, the symptom of the “inspiration paradox” disappears. Respiratory excursions of the chest are reduced to 1–2.5 cm (instead of 4–7 cm in the norm). The dimensions of the funnel increase: its depth reaches 7–8 cm, and the volume — 250–300 cm^2.

Such children are prone to frequent tracheitis, chronic bronchitis, pneumonia and other respiratory diseases [2]. The pathogenesis is based on two reasons. The first is associated with compensated chronic compression of the retrosternal organs and lung tissue. Compression of the bronchi leads to stagnation of secretions in the alveoli, disruption of their evacuation, and as a result, a frequent inflammatory process. The second is related to the etiology of the disease. Connective tissue dysplasia is multi-organ in nature and affects not only cartilage and ribs, but also bronchopulmonary tissue. The lung tissue and bronchial tree affected by dysplasia are susceptible to such diseases as bronchiectasis, tracheobronchomalacia, broncho-obstructive syndrome, anomaly in the development of the bronchial tree, and bullous lung disease. Connective tissue dysplasia leads to multiple changes in the musculoskeletal system. Patients are characterized by [13]:

- asthenic type of body structure;
- arachnodactyly (“spider fingers”);
- flat feet;
- myopia;
- malocclusion;
- pathological joint mobility;
- decrease in indicators of muscle strength.

Many children with pectus excavatum have psychosocial problems due to a cosmetic defect, which leads to various behavioral disorders, withdrawal,

unwillingness to do sports and requires the help of a psychologist.

Diagnosis and treatment

Diagnosis of the disease is usually not difficult on examining the patient. Therefore, the examination aims not only to identify the deformity, but also to assess the general condition of the patient, the function of the respiratory and circulatory organs. To identify functional disorders of the chest organs, assess the compensatory capabilities of the body and its general condition, identify complications from the vital organs (heart and lungs), analyze the progression of the pathology, examination and consultation of specialist doctors is necessary.

The nature and degree of deformity is determined using thoracometry and various indices of the chest, taking into account the depth of deformity at the level of the xiphoid process, the width of the chest and chest circumference at the same level, the elasticity of the chest and the volume of the infundibulum.

To assess the degree of deformity, the choice of the method of correction and the amount of surgical intervention, it is necessary to conduct a number of diagnostic studies. The severity of chest deformity is most objectively determined using radiation methods of investigation — chest x-ray in 2 projections and/or multislice computed tomography [21].

X-ray examination of the chest organs in a direct projection allows you to determine the pathology of the ribs, sternum and lung tissue. Lateral projection, before the advent and widespread introduction of multislice computed tomography, was widely used to determine the degree of sternum deformity using the I. Gizhitska, which is equal to the ratio of the smallest distance between the sternum and the spine to the largest one (Figure 1) [22].

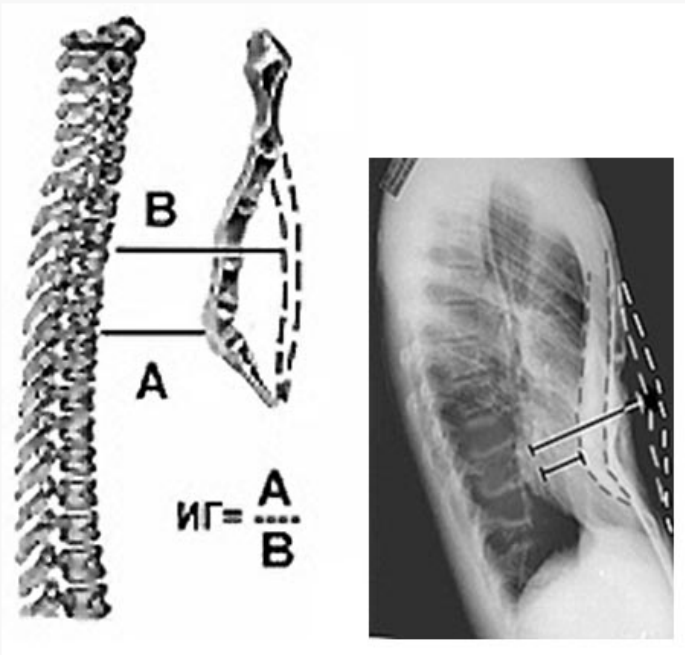

Figure 1. Definition of I. Gizhitska. Note. A - the smallest distance is determined between the posterior surface of the sternum and the anterior surface of the vertebral bodies; B is the distance between the true position of the sternum [22]

Considering this index, there are 3 degrees of chest deformity: at the 1st degree, this index is 0.9 to 0.7, at the 2nd degree — 0.7 to 0.5, at the 3rd degree — 0.5 to 0.

Multislice computed tomography of the chest and organs of the chest cavity allows assessing the deformation of the ribs, sternum and cartilage, shows the degree of compression and displacement of the heart and lungs. The Haller index is used to assess the severity of pectus excavatum. Computed tomography images measure the horizontal distance between the inner surface of the ribs at the level of the greatest curvature (Figure 2). The resulting value is divided by the anterior-posterior size (the distance between the inner surfaces of the spine and sternum). Normally, the Haller index is 2.5–2.7. With indicators of 3.25 or more, surgical treatment is indicated [22–23]. Based on the Haller index, 4 degrees of severity of deformity are distinguished:

- I (3.0–3.9);
- II (4.0–4.9);
- III (5.0–5.9);
- IV (6.0 and more).

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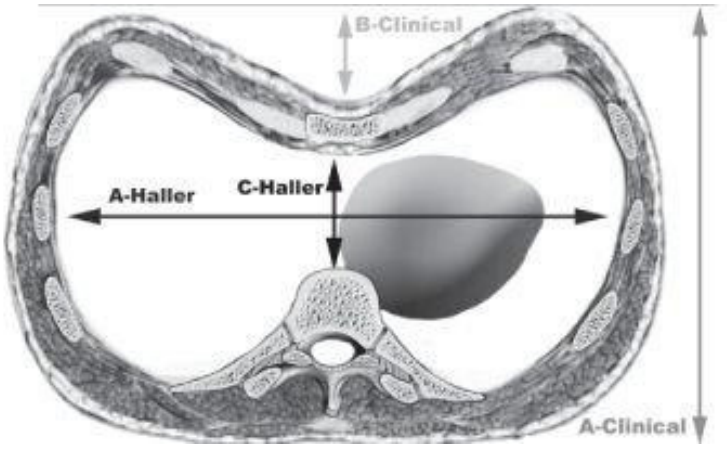

Figure 2. Haller index definition [24]

Based on the computed tomography data and for the purpose of diagnosis and successful surgical correction of PE H.J. Park et al. (2008) proposed, the processing of 3D images of patients with sternum deformity with the calculation of depression indices DI (Depression Index), asymmetry AI (Asymmetry Index), eccentricity EI (Eccentricity Index) and imbalance UI (Unbalance Index). In the anatomical classification according to Park, groups of patients are distinguished [25]:

• symmetrical types:

— classic − the most common type, CT shows a concave deformity of the chest along the midline;

— wide flat — retraction has a smaller depth and a larger area;

• asymmetrical shapes:

— eccentric local type — displacement of the center of deformation to the right or left, at the same time observe the inclination of the sternum towards the curvature;

— asymmetric wide flat — differs from the first type in a smaller defect depth;

— eccentric long deep type — has a large length of the funnel, can start from the sternoclavicular joint;

— unbalanced view — combines the curvature of the front with an asymmetric structure of the right and left sides of the chest;

— combined type — combined with keeled deformation of varying severity.

The classification according to Zeng Q. is a simplified classification of H.J. Park. The author divided the types of funnel deformity into the following groups [26]:

1. Symmetrical type
2. Eccentric type
3. Unbalanced type

In the stages of diagnosis after the radiograph, the main thing in terms of significance is the assessment of the functional activity of the organs of the cardiorespiratory system based on a number of instrumental and laboratory studies, such as:

— electrocardiography (to assess the state of conduction, the presence of metabolic changes in the myocardium, dilatation of the heart);

— echocardiography and dopplerography of the main vessels (to detect the severity of cardiac depression);

— holter monitoring (to assess transient rhythm and conduction disturbances);

— ultrasound examination of the costal cartilages (to determine the extent of the hyperechoic zone in the structure of the costal cartilage);

— study of the function of external respiration with the determination of the main functional parameters: VC, % — vital capacity, FVC, % — volume of forced vital capacity of the lungs, FEV1, % — forced expiratory volume in 1 second, Tiffno index, % — ratio of forced expiratory volume in 1 second to vital capacity, expressed as a percentage. An exercise stress test is performed to assess exercise tolerance.

— clinical blood test — assess the indicators of “red” blood (erythrocytes, hemoglobin, hematocrit) reflecting the transport of oxygen.

— acid-base state of the blood (partial pressure of oxygen, carbon dioxide, saturation, lactate level) — as the main marker of tissue (cellular) hypoxia.

— determination of the level of vitamin D — to determine the mineralization of the skeleton [27].

Thus, the problem of diagnosing patients with PE requires emphasis from the point of modern positions. It is advisable, taking into account childhood age, to use minimally invasive, non-traumatic methods that provide maximum data on the morphology of abnormally altered areas of the chest, to assess the state of blood supply and aerodynamics in pathologically compressed lungs and hemodynamics in the heart, with an assessment

of their functionality, by which it will be possible to determine indications to surgical treatment and specify the period of correction.

Based on the results of instrumental and laboratory research methods, patients with PE are consulted by an orthopedist to determine the tactics of surgical treatment, in the presence of secondary changes in the cardiorespiratory system, they are examined by a pulmonologist, a cardiologist to correct impairments. The psychological counseling of these patients is of great importance.

According to literary sources, at present conservative treatment (physiotherapy exercises, massage, kinesitherapy, manual therapy) is possible only with PE of the I degree, but it does not lead to a noticeable restoration of not only the surface of the chest, but also the anatomical position of the mediastinal organs. Operations for PE I degree are rarely performed, only at the insistence of patients and their parents, and they are aimed at eliminating a cosmetic defect (functional disorders of the cardiovascular and respiratory systems are most often absent). Absolute indications for thoracoplasty are II and III degree deformities in the decompensated and subcompensated stages. Contraindication to thoracoplasty is: comorbidity, which significantly increases the risk of early postoperative and anesthetic complications; severe mental illness.

The history of the development of surgical interventions in children with pectus excavatum has several decades. Thus, the operation was performed by the German surgeon L. Meyer in 1911, and by another German surgeon, the founder of thoracic surgery F. Sauerbruch in 1920 [28]. The essence of their operations was to remove compression of the ribs on the heart, in this connection 2–3 ribs were resected on both sides. In 1949 thoracic surgeon M.M. Ravitch proposed a surgical correction technique based on resection of the costal cartilages that form the deformity zone, while preserving the perichondrium, and osteotomy of the sternum [29].

Previously, complex traumatic interventions associated with resection of the ribs and sternum were often used. However, they have been replaced with new minimally invasive techniques. Currently, more than 100 methods of thoracoplasty and their modifications are known for the operational correction of PE. All surgical interventions include two stages: mobilization of the sternocostal complex and its fixation in the correction position [19].

In 1998 D. Nuss proposed a completely new method of surgical treatment of pectus excavatum, which made it possible to correct simultaneously all components of multiplanar chest deformity without resection thoracoplasty. Correction of chest deformity according to Nuss's method is achieved by inserting a special corrective plate under the sternum, at the site of maximum deformity. Myotomy of the intercostal muscles is performed at the intended points of insertion of the plate, after which the parietal pleura is mobilized from the posterior surface of the sternum and ribs, and a tunnel is formed in the retrosternal space [30, 31]. A premodeled corrective plate is fixed, the plate is set with traction by the conductor in the direction from right to left, along the formed tunnel, bending backwards, without contact with the lungs and pleura. At the last stage, the corrective plate is fixed with clamps on the distal sections on both sides, after which the 180˚ rotation is performed and the plate is placed on the anterior sections of the ribs in strict accordance with the new contour of the chest. The correction plate remains in the patient's body for 1 to 4 years and is then removed.

Correction by D. Nuss's method today allows you to get the best cosmetic result compared to other operations and is the method of choice for the treatment of PE. This type of thoracoplasty has its advantages: correction is performed through small incisions, minimal blood loss, good cosmetic result, short operation time, the possibility of long-term wearing of endofixators until complete chest remodeling [32]. Many published studies have confirmed the improvement in the quality of life, both in children and adults, as a result of surgical correction of PE [30–32].

Conclusion

Modern clinical diagnostic studies make it possible to assess comprehensively the state of the cardiorespiratory system in children with PE, and analyze the cosmetic and social aspects of the problem. Taking into account the obtained indicators, a differentiated approach, with regard to all factors, to minimally invasive surgical correction of this pathology, which in the delayed period allows obtaining a positive physical, psychological and social effect.

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Information about the authors / Информация об авторах

Natallia V. Motorenko, Assistant Lecturer at the Department of Pediatrics, Gomel State Medical University, Gomel, Belarus
ORCID: https://orcid.org/0000-0001-6729-446X
e-mail: nmotorenko31@mail.ru

Alexandr V. Vinnik, Head of the Orthopaedics and Traumatology Department, Gomel Regional Children's Clinical Hospital, Gomel, Belarus
ORCID: https://orcid.org/0000-0001-8624-5225
e-mail: avinnik22@mail.ru

Моторенко Наталья Викторовна, ассистент кафедры педиатрии, УО «Гомельский государственный медицинский университет», Гомель, Беларусь
ORCID: https://orcid.org/0000-0001-6729-446X
e-mail: nmotorenko31@mail.ru

Винник Александр Васильевич, заведующий ортопедо-травматологическим отделением, У «Гомельская областная детская клиническая больница», Гомель, Беларусь
ORCID: https://orcid.org/0000-0001-8624-5225
e-mail: avinnik22@mail.ru

Corresponding author / Автор, ответственный за переписку

Natallia V. Motorenko
e-mail: nmotorenko31@mail.ru

Моторенко Наталья Викторовна
e-mail: nmotorenko31@mail.ru

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