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# COMPARATIVE CHARACTERISTICS OF THE EFFICIENCY OF DIFFERENT METHODS OF OPERATIONAL TREATMENT FOR PECTUS EXCAVATUM IN CHILDREN: A MULTICENTER STUDY

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**Background.** Congenital malformations of the chest are observed in 1%–4% of the population, and the most common among these is pectus excavatum (90%).

**Aim.** We aimed to conduct a retrospective multicenter study to compare the effectiveness of various methods of operative removal of pectus excavatum in children.

**Material and methods.** We retrospectively analyzed the results of the surgical treatment of funnel-like deformity of the thorax in children conducted in clinics of pediatric surgery in seven regions of Russia (1,226 patients). The ratio of boys to girls in the study population was 2.2:1. The study population was divided as per their age into the following groups: 4-7 years (n = 180, 14.7%), 8-14 years (n = 731, 59.6%), and > 14 years (n = 315, 25.7%). The average age at which most children were operated was  $11.83 \pm 1.24$  years. All children underwent a standard preoperative laboratory examination, including a general blood test, urine tests, a biochemical blood test, a hemostasiogram; radiographic diagnostic methods were used with the calculation of the Gizycka index; functional methods of investigation, such as electrocardiography, spirography, and radioisotope scintigraphy of the lungs were also used. Children with second-or third-degree pectus excavatum underwent surgical treatment almost exclusively. The symmetrical forms of the pectus excavatum were more prevalent. In most cases, the main pathological course was complicated.

**Results and discussion.** The operated patients were divided into the following 3 groups: the first (n = 62): operations with the resection of the curved cartilages and external fixation of the sternum-rib complex (Bairov's operation), the second (n = 374): thoracoplasty with the resection of warped cartilages using internal metal fixators by Timoshchenko, Ravitch, Paltia, Kondrashin), and the third (n = 790): minimally invasive operations without resection with internal fixation (Nuss operations: original and modified). In the first group, favorable results of surgical treatment were noted in 80.6% of the patients, satisfactory results were observed in 6.5%, unsatisfactory results were seen in 12.9%, and the overall effectiveness of operative correction was 87.1%. In the second group, good results of surgical treatment were recorded in 88% of the patients, satisfactory in 6.4%, and unsatisfactory in 5.6%; the overall efficiency of operative

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correction was 94.4%. In the third group, good results of surgical treatment were recorded in 95.3%, satisfactory in 3.8%, and unsatisfactory in 0.9%; the efficiency of operative correction was 99.1%.

Conclusion. Today, the authors consider acceptable the operations by Timoshchenko and Paltia for complex of indicators, the optimal operation is Nuss.

Keywords: funnel chest deformation; pectus excavatum; surgery; children.

### СРАВНИТЕЛЬНАЯ ХАРАКТЕРИСТИКА ЭФФЕКТИВНОСТИ РАЗЛИЧНЫХ СПОСОБОВ ОПЕРАТИВНОГО ЛЕЧЕНИЯ ВОРОНКООБРАЗНОЙ ДЕФОРМАЦИИ ГРУДНОЙ КЛЕТКИ У ДЕТЕЙ: МУЛЬТИЦЕНТРОВОЕ ИССЛЕДОВАНИЕ

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Актуальность. Врожденные пороки развития грудной клетки отмечаются у 1-4 % населения, наиболее часто из них встречается воронкообразная деформация грудной клетки (90 %).

Цель: в ходе ретроспективного мультицентрового исследования определить эффективность различных способов оперативного устранения воронкообразной деформации грудной клетки (ВДГК).

Материалы и методы. В статье ретроспективно анализируются результаты оперативного лечения ВДГК у детей, проведенного в клиниках детской хирургии семи российских регионов (1226 больных). Соотношение мальчики/девочки составило 2,2:1. По возрасту больные распределились следующим образом: от 4 до 7 лет —  $180\ (14,7\ \%)$  детей, от 8 до  $14\ -\ 731\ (59,6\ \%)$  ребенок, старше  $14\ \text{лет}\ -\ 315\ (25,7\ \%)$  детей. Наиболее часто были подвергнуты оперативному лечению пациенты в возрасте 11,83 ± 1,24 года. Всем детям проводили стандартное предоперационное лабораторное обследование: общий анализ крови, мочи, биохимический анализ крови, гемостазиограмму; применяли методы лучевой диагностики с расчетом индекса Гижицкой, функциональные методы исследования: электрокардиографию (ЭКГ), спирографию, радиоизотопную сцинтиграфию легких. Оперативное лечение получили пациенты практически исключительно со 2-й и 3-й степенями ВДГК. Преобладали симметричные формы ВДГК. В большинстве случаев течение основной патологии являлось осложненным.

Результаты и обсуждение. Прооперированные больные были разделены на три группы: первая (62) — операции с резекцией искривленных хрящей и наружной фиксацией грудино-реберного комплекса (операция Г.А. Баирова), вторая (374) — торакопластики с резекцией искривленных хрящей с использованием внутренних металлических фиксаторов (операции В.А. Тимощенко, М. Ravitch, V. Paltia, Н.И. Кондрашина) и третья (790) — минимально инвазивные операции без резекции с внутренней фиксацией (операции D. Nuss — оригинальная и в модификациях). В первой группе хорошие результаты хирургического лечения зафиксированы у 80,6 % пациентов, удовлетворительные — у 6,5 %, неудовлетворительные — у 12,9 %, общая эффективность оперативной коррекции составила 87,1 %. Во второй группе хорошие результаты хирургического лечения зафиксированы у 88 % больных, удовлетворительные — у 6,4 %, неудовлетворительные — у 5,6 %, общая эффективность оперативной коррекции составила 94,4 %. В третьей группе хорошие результаты хирургического лечения зафиксированы у 95,3 %, удовлетворительные — у 3,8 %, неудовлетворительные — у 0,9 %, эффективность оперативной коррекции составила 99,1 %.

**Заключение.** На сегодняшний день приемлемыми по комплексу показателей авторы считают операции В.А. Тимощенко и V. Paltia, оптимальной — операцию D. Nuss.

Ключевые слова: воронкообразная деформация грудной клетки; оперативное лечение; дети.

#### Background

Congenital malformations of the chest are reported in 1%-4% of the population, with pectus excavatum (PE) being the most common accounting for 90% of all cases [1-4]. Since performing the first surgical intervention for PE in 1911, over 100 corrective methods have been proposed for the deformity now. Significant changes in surgical correction methods and indications for surgical treatment in recent decades have facilitated the use of modern designs for internal fixation of the sterno-costal complex, which considerably enhanced the functional and cosmetic results of the treatment [5]. Despite the improvement of surgical treatment methods, the proportion of unsatisfactory results, according to various authors, ranges 10%–40%, with complications occurring in 30% of thoracoplasty cases [6]. However, relevant literature has insufficiently elucidated the dependence of the number of postoperative complications and the efficacy of the surgical intervention itself on the type of surgical aid for pediatric patients with PE.

Therefore, this retrospective multicenter study aims to determine the efficacy of various surgical elimination methods of PE: with resection of curved cartilages and external fixation of the sterno-costal complex; thoracoplasty with resection of curved cartilage using internal metal fixators; and minimally invasive surgeries (without resection with internal fixation).

#### Materials and methods

In this retrospective multicenter study, we reviewed the treatment results of 1226 pediatric patients with PE from specialized medical

institutions in seven regions of the Russian Federation. Notably, specialists in each institute used a single diagnostic algorithm to examine patients of this category and adhered to unified approaches in determining the optimal timing of the operative treatment as indicated. All patients (or their legal representatives) voluntarily signed informed consent for participation in this study, processing of personal data, and performing a surgical intervention.

Based on the generalized data of this study, the ratio of boys and girls in the study cohort was registered as 2.2:1 (the prevalence of boys among all patients was over two times). Based on the age, we distributed patients as follows: 180 (14.7%) pediatric patients aged 4–7 years, 731 (59.6%) pediatric patients aged 8–14 years, and 315 (25.7%) pediatric patients aged >14 years. Mostly, children were operated at the age of  $11.83 \pm 1.24$  years.

All pediatric patients underwent a standard preoperative laboratory examination, including a general blood test, urine test, biochemical blood test, and a hemostasiogram. In addition, we used radiation diagnostics, such as plain radiography of the thoracic organs, to evaluate the degree of deformity and the presence of displacement of the mediastinal organs; the tests comprised computed tomography (CT) of thoracic organs in the bone and pulmonary regimen, ultrasound examination (US) of the abdominal cavity organs and the retroperitoneal space, and echocardiography as indicated. CT of the bone regimen revealed the anatomical and topographic interrelations of the internal organs of the thoracic cavity, the displacement of the mediastinal organs, and the degree of the sternum retraction and its rotation. Furthermore,

CT of thoracic organs in the pulmonary regimen determined the comparative density of the lung parenchyma at the maximum and minimum levels of the sternum retraction.

Radiographically, we calculated the degree of deformity using the Gizhitskaya index (GI; sternovertebral index is the ratio of the smallest distance between the sternum and the anterior surface of the vertebral bodies to the largest one), as assessed by plain radiography of the thoracic cavity in the lateral projection (degree 1, GI > 0.7; degree 2, GI = 0.7-0.5; degree 3, GI < 0.5). Since 2015, in some regions, the developed computer program PectExcavPro (Federal State Budgetary Educational Institution of Higher Education Tyumen State University, Tyumen, Russia) was applied to calculate the degree of deformity [7]. In this study, however, we used the classification of N.I. Kondrashin. We distinguished the shape as conical (the sterno-costal complex retracts in the form of a cone with the base facing outward) and flat-funnel (the sterno-costal complex retracts evenly in the form of a platform relative to the anterior surface of the thorax).

In addition, we used the functional methods of investigation, namely electrocardiography (ECG), spirography, and radioisotope scintigraphy of the lungs. Notably, ECG and spirography were performed at the preoperative stage for all pediatric patients, facilitating the assessment of the degree of functional disorders in the cardiovascular and respiratory systems. Furthermore, radioisotope lung scintigraphy detected pulmonary tissue perfusion abnormalities, which were especially significant to indicate surgery in patients with degrees 1–2 of PE with a mild cosmetic defect that did not cause psychological discomfort.

In this study, statistical analyses were performed on a personal computer using the Microsoft Excel application and statistical data analysis package Statistica 5.1 for Windows (StatInc.). We considered P < 0.05 as statistically significant.

#### Results and discussion

In this study, we identified the following indications for surgical treatment: (a) functional (manifested in pathological changes in the cardiovascular and respiratory systems that were objectively assessed by ECG, spirography, and radioisotope scintigraphy of the lungs);

(b) orthopedic (caused by a progressive postural disorder and curvature of the spine, based on visual examination, radiography, and chest CT); and (c) cosmetic (associated with the presence of a defect and the psychological discomfort caused by it, mainly in the older age group).

Pediatric patients almost exclusively with the degrees 2 and 3 of PE underwent surgical treatment. In addition, 0.6% of operated pediatric patients had PE of degree 1, 65.3% had degree 2, 33% had degree 3, and 1.1% presented with the extreme severity of the deformity (the thoracic cavity was almost bisected). Of note, the symmetrical forms of the PE prevailed.

In several cases, the deformity developed and proceeded with the deformity of the skeleton in the form of postural disorder or scoliosis (72% of patients), Marfan syndrome and Marfan-like anomalies (8%), Ehlers-Danlos syndrome (7.5%), planovalgus deformity (7%), congenital heart defects (7%, including mitral valve prolapse, open oval window, and aortic valve stenosis), and kidney duplication (2%). In addition, most patients (69%) had laboratory manifestations of the syndrome of undifferentiated connective tissue dysplasia, namely the disorder of the platelet-vascular unit of hemostasis, in the form of a disorder of platelet adhesive and aggregating function, structural and chronometric hypocoagulation associated with a deficiency of the prothrombin complex factor, and moderate thrombocytopenia.

Data from the Russian Federation regions, presented in this study, revealed the use of various surgical techniques in the treatment of PE in pediatric patients. Based on our experience, we assigned all patients to one of the following three groups: group 1, surgeries with resection of curved cartilages and external fixation of the sterno-costal complex (G.A. Bairov surgery); group 2, thoracoplasty with resection of curved cartilages using internal metal fixators (surgeries of V.A. Timoschenko, M. Ravitch, V. Paltia, and N.I. Kondrashin); and group 3, minimally invasive surgeries without resection with internal fixation (surgeries of D. Nuss, original and modified by the authors). Furthermore, the term for wearing internal metal fixators was 2-11 years, and the average removal period was 4.2 years after the placement.

Of note, two regions provided data for the first half of the 1990s, concerning the surgical treatment

Methods of surgical treatment of patients with pectus excavatum

Region	Method	Number of patients				
With resection and external fixation (62; 5.06%)						
Kirov region	According to Bairov 32					
Tyumen Region	According to Bairov	30				
With resection and internal fixation (374, 30.50%)						
Kirov region	According to Timoschenko	25				
	According to Paltia	17				
Moscow — Krasnoyarsk	According to Ravitch	19				
Moscow	According to Paltia	90				
Sverdlovsk region	According to Paltia	191				
Tomsk region	According to Kondrashin	32				
Without resection with internal fixation (790, 64.44%)						
Kirov region	According to Nuss	49				
Moscow — Krasnoyarsk	According to Nuss and modified Nuss	47				
Moscow	According to modified Nuss	452				
Tomsk region	According to modified Nuss	47				
Tyumen region	According to Nuss and Nuss-Vinogradov	126				
Chelyabinsk region	According to Nuss	69				

of PE in pediatric patients with resection of curved cartilages and external fixation of the sterno-costal complex (62 patients, all were operated according to Bairov). In addition, five regions provided the results of surgical treatment of patients in this category with resection of curved cartilages and internal fixation (374 cases: 25 according to Timoschenko, 19 according to Ravitch, 298 according to Paltia, and 32 according to Kondrashin) and six regions provided the results of surgical treatment without resection of curved cartilages with internal fixation (790 patients operated by Nuss and modified Nuss; Table 1).

In this study, all patients were enrolled for a follow-up observation for a period of 3–10 years. The results of surgical treatment by all specialists were evaluated according to the same rules as "good" (the deformity is completely corrected, and the cosmetic result fulfills the expectations of parents and patients), "satisfactory" (sternal hypercorrection [postoperative keeled deformity] and incomplete correction [decrease in the degree of PE]), and "unsatisfactory" (relapse and progression of PE). In addition, we considered the frequency and severity

of postoperative complications and the average bedday in each group.

In group 1, good results of surgical treatment were observed in 80.6% of patients, satisfactory in 6.5%, and unsatisfactory in 12.9%. The overall efficiency of surgical correction (good and satisfactory results) was 87.1%. In group 2, good results of surgical treatment were recorded in 88% of patients, satisfactory in 6.4%, and unsatisfactory in 5.6%. The overall efficiency of surgical correction was 94.4%. In group 3, good results of surgical treatment were recorded in 95.3% of patients, satisfactory 3.8%, and unsatisfactory in 0.9%. Thus, the overall efficiency of surgical correction was 99.1% (Table 2).

Of note, the analysis of the surgical treatment results of PE depending on the methods used was of great practical interest (Table 3). The least effective (good and satisfactory results) method was Bairov surgery, which, in fact, should be considered obsolete now. In group 2, Kondrashin surgery was the least effective (71.9%). In addition, Timoschenko and Paltia surgeries should be recognized comparable in efficiency (96%). However, it seems unfair to

Table 2
Results of surgical treatment of patients with pectus excavatum by regions (absolute figures)

	Method of surgical treatment								
Region	with resection and external fixation (62)		with resection and internal fixation (374)			without resection with internal fixation (790)			
Results of surgical treatment									
	Good	Sat.	Bad	Good	Sat.	Bad	Good	Sat.	Bad
Kirov region	25	2	5	36	3	3	48	0	1
Tyumen region	25	2	3				120	4	2
Moscow — Krasnoyarsk				15	4	0	42	5	0
Moscow				90	0	0	442	9	1
Sverdlovsk region				172	10	9			
Tomsk region				16	7	9	35	10	2
Chelyabinsk region							66	2	1

Table 3
The efficiency of various surgical treatment methods for patients with pectus excavatum (absolute numbers and efficacy in percentage terms)

Method	Resu	Efficiency (%)			
Method	Good	Sat.	Bad	Efficiency (70)	
According to Bairov	50	4	8	87.1	
According to Timoschenko	24	0	1	96.0	
According to Paltia	274	13	11	96.3	
According to Ravitch	15	4	0	100.0	
According to Kondrashin	16	7	9	71.9	
According to Nuss and modified Nuss	753	30	7	99.1	

Table 4
Recorded postoperative complications and duration of inpatient treatment of patients with pectus excavatum

	n	Postoperative co		
Method		hemo- (pneumo-, hydro) thorax (%)	Suppuration (%)	Bed-day
According to Bairov	62	16.5	-	34.1 ± 3.5*
According to Timoschenko	25	4.0	-	10.0 ± 1.1*
According to Paltia	298	18.0	1.5	11.1 ± 1.8*
According to Ravitch	19	5.3	-	11.2 ± 0.9
According to Kondrashin	32	3.1	15.6	11.3 ± 1.3*
According to Nuss and modified Nuss	790	12.1	_	$8.4 \pm 0.9$

Note. \*p < 0.05 compared to Nuss surgery.

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conclude the efficacy of Ravitch surgery because of the small number of observations. Furthermore, the minimally invasive methods, such as Nuss and modified Nuss, exhibited the highest efficiency (99%) in this study.

In this study, we also performed data analysis of postoperative complications of surgical treatment of PE in pediatric patients and a hospital bed-day, which revealed some regularities, depending on the method of surgical intervention (Table 4). Thus, the highest percentage of early postoperative complications (16.5%) was observed in group 1. Among methods used in group 2, Kondrashin surgery exhibited the smallest number of complications (3.1%), whereas Paltia surgery exhibited the highest number of complications (18.0%). In group 3, finally, the percentage of early postoperative complications was moderate (12.1%).

We recorded cases of suppuration of postoperative wounds only after performing Paltia and Kondrashin surgeries (1.5% and 15.6%, respectively). The best method in the least number of days spent in the hospital was the Nuss method (original and modified) with 8.4 days.

Thus, in pediatric patients with PE, surgeries with resection of curved ribs and external fixation of the sterno-costal complex (according to Bairov) are the least effective, characterized by a large number of postoperative complications and unnecessarily prolonged bed-days. Among surgical techniques that we assigned to the second group (Timoschenko, Paltia, Ravitch, and Kondrashin), the first two exhibited adequate efficacy, the first of which is characterized by the smallest number of postoperative complications. In addition, minimally invasive surgeries without resection of curved cartilages with internal fixation of the sterno-costal complex (Nuss and modified Nuss) were favorably characterized with the highest clinical and esthetic efficiency, a low percentage of postoperative complications, and the least number of bed-days.

#### **Conclusions**

This study derived the following conclusion. PE is a common congenital pathology that affects boys two times more often, and the most optimal treatment period is the age of 8–12 years. In most cases, the course of pathology is burdened, mostly with the

undifferentiated connective tissue dysplasia (69%). Surgeries with resection of curved ribs and external fixation of the sterno-costal complex (Bairov type) are characterized by the lowest efficiency (87.1%), a large number of postoperative complications (16.5%), and an unreasonably high number of beddays (34). In addition, surgeries performed with resection of curved cartilages and internal fixation of the sterno-costal complex (Timoschenko and Paltia) exhibit significant efficacy (96%), fewer bed-days (10–11), and Timoschenko surgery also presents least postoperative complications (4%). Finally, this study establishes that Nuss surgery is characterized with optimal esthetic results, the highest clinical efficiency (99%), and the least number of bed-days (8.4), although it is inferior to some surgeries of group 2 in the number of postoperative complications (12%).

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