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TREATMENT OF FUNNEL CHEST IN CHILDREN

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Background. Funnel chest has a relatively high prevalence in the Russian population. Given the high percentage of the unsatisfactory results of thoracoplasty, further research for the development of new treatment methods is needed.
Aim. To improve the treatment results for funnel chest in children.
Materials and methods. We analyzed the treatment results of 230 children with funnel chest after thoracoplasty. We used 2 surgical techniques: classic thoracoplasty by Nuss (114 children) and its modified version by the authors (116 children). The modified technique included two-sided thoracoscopy, partial resection of the deformed rib cartilages, and endoscopic longitudinal transection of the sternal cortical plate, resulting in subcutaneous emphysema along the sternum.
Results. The comparison of the 2 surgical techniques showed no significant difference in terms of duration and invasiveness of the procedure. Recurrent episodes of funnel chest were observed in children who had undergone thoracoplasty before 7 years of age, regardless of the technique used.
Conclusion. This study revealed that the author's modified thoracoplasty method was more effective in children >14 years of age with rigid funnel chest because it allowed the surgeon to decrease the thoracic pressure on the plate and the plate itself on the ribs, facilitating the repositioning of the sternum and preventing the deformation and development of pressure sores on the ribs.

Keywords: pectus excavatum, Nuss technology, an authorial method of the correction.

ЛЕЧЕНИЕ ВОРОНКООБРАЗНОЙ ДЕФОРМАЦИИ ГРУДНОЙ КЛЕТКИ У ДЕТЕЙ

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Актуальность. Частота заболевания, высокий процент неудовлетворительных результатов после торакопластики по поводу воронкообразной деформации грудной клетки являются причиной дальнейшего поиска новых технических приемов оперативного лечения.
Цель исследования: улучшение результатов лечения воронкообразной деформации грудной клетки у детей.
Материал и методы. Анализируются результаты торакопластики у 230 детей с воронкообразной деформацией грудной клетки. Применялись две методики операций: классическая торакопластика по Nuss (у 114 детей) и ее авторская модификация (у 116 детей), предусматривающая торакоскопию с обеих сторон, надсечение хрящей деформированных ребер и эндоскопическое продольное частичное рассечение кортикальной пластинки грудины путем создания подкожной эмфиземы в проекции грудины.
Результаты. Установлено, что статистически значимого различия по продолжительности выполнения операции, ее травматичности в анализируемых группах не выявлено. Частичный рецидив заболевания развивается у детей, перенесших торакопластику в возрасте до 7 лет, независимо от используемого метода торакопластики.

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Выводы. Авторский метод торакопластики наиболее эффективен у детей с ригидной формой грудной клетки в возрасте старше 14 лет, так как позволил уменьшить давление грудино-реберного комплекса на металлоконструкцию и самой пластины на опорные ребра, что облегчало выведение грудины в физиологическое положение, предупреждало деформацию/пролежни опорных ребер.

Ключевые слова: воронкообразная деформация грудной клетки, Nuss-технология, авторский способ коррекции.

Introduction

Cobbler's chest (CC) is one of the most frequent chest malformations, which is accompanied by cosmetic defects, severe functional changes in the thoracic organs [1-7], and psychosocial problems [8]. The literature describes more than 80 different variants of surgical interventions and their modifications for the correction of CC [9-16]. Despite the vast number of surgical treatment methods, long-term results do not always satisfy the surgeon and the patient [17-20]. The widespread minimally invasive method of thoracoplasty by D. Nuss also does not solve all problems associated with this condition, including the development of partial relapse of deformity in pediatric patients who underwent thoracoplasty around preschool age, deformity/pressure-related injuries of the supporting ribs under the surgical hardware, and numerous intra- and postoperative complications.

The study aimed to improve the results of CC treatment in children by differentiating the method of surgery depending on the age of the patient and the degree of chest rigidity.

Material and methods

The study was based on an analysis of the results of the surgical treatment of 230 patients with CC, that were treated in the Irkutsk State Regional Children's Clinical Hospital from 2002 to 2016. We conducted a prospective comparative study to evaluate the results of surgical treatment of CC. All patients were divided into two groups: TG1 and TG2.

Clinical examination of pediatric patients was conducted according to the generally accepted scheme. Degree of deformity was determined by the Gyzicka index based on chest x-ray examination. The preoperative in-depth examination included spirometry, echocardiography, and the state of the blood coagulation testing.

In TG2, the correction of the CC was performed according to the classical Nuss method, proposed in 1998 and consisting of substernal conduction of a C-shaped plate with abutment on the ribs at the edge of the CC deformity. In TG1, the techniques of the Nuss procedure were complemented with two endoscopic stages (patent of the Russian Federation No. 2883052): bilateral thoracoscopy, partial incision of the cartilages of the deformed ribs and partial longitudinal corticotomy of the sternum under the optical control by creating a subcutaneous emphysema to facilitate elimination of the sternal-costal complex deformity. Surgical interventions in all pediatric patients were performed under intubation apparatus anesthesia, the main parameters of the cardiovascular and respiratory systems were monitored. The second stage of the surgical intervention included the removal of surgical hardware after 12 and 24 months. The short-term results were estimated in all 230 patients according to criteria including surgery duration, volume of blood loss, duration of postoperative pain requiring anesthesia, and intra- and early postoperative complications. Long-term results were traced in 184 patients with the catamnesis duration (after the removal of the surgical hardware) from 2 to 12 years. Evaluation criteria included long-term complications, partial relapse, and complete relapse of the disease. Parents of the patients provided informed consent for surgical thoracoplasty. The study protocol was approved by the ethical committee of the Irkutsk State Medical Academy of Postgraduate Education, during meeting No. 7, min dated 12.09.2007. The results of the studies were analyzed by variational statistics. The variational series were compared according to the Student's t-criterion. The value of $p < 0.05$ was accepted as indicating statistical significance. Statistical processing of data was performed with the Statistica for Windows software package.

Results and discussion

The first group (TG1) included 116 (50.4%) patients who received surgical treatment for CC by the author's method from 2002 to 2009. The second group (TG2) included 114 (49.6%) patients with

a similar pathology, who received surgical treatment according to the classical Nuss method from 2010 to 2016, inclusive.

In the study groups, boys were slightly predominant (n = 120, 52.2%) compared to girls (n = 110; 47.8%). Distribution by age was the following: there were 21 patients of 3–6 years (9.1%), 31 patients of 7–9 years old (13.4%), 84 patients were of 10–14 years old (36.5%), 94 patients were of 15–18 years old (40.8%). Manifestations of connective tissue dysplasia (CTD) (deformity of bones of the skeleton, Marfan-like and other dysplastic syndromes) were noted in 53 (24.0%) pediatric patients. In most patients (n = 144, 62.2%), the degree III of deformity was determined. The distribution of patients by the terms of immobilization with the surgical hardware of the sternal-costal complex was the following: 1 year in 59 pediatric patients (26.0%), and 2 years in 171 (74.0%) pediatric patients. In the groups analyzed, there were no statistically significant differences in gender, age, and degree of deformity.

The significant criteria for surgical efficiency are amount of blood loss, duration and injury rate of surgical intervention, which is determined by the duration and intensity of the pain syndrome requiring anesthesia during the postoperative period. These indicators are presented in Table 1.

Table 1

Duration of the surgery, the amount of blood loss and the duration of the pain syndrome requiring anesthesia in the postoperative period in the age groups

Comparison criterion	TG1 (n = 116)	TG2 (n = 114)	T-criterion	p
Surgery duration	32.28 ± 2.16	24.65 ± 4.27	1.972	> 0.05
Amount of blood loss (ml). depending on the age				
3–6 years	27 ± 1.13	14.66 ± 2.05	2.093	< 0.05
7–9 years	30.14 ± 2.3	18.52 ± 3.17	2.045	< 0.05
10–14 years	31.27 ± 4.32	20.42 ± 3.7	1.99	> 0.05
14–18 years	36.42 ± 3.81	22.74 ± 4.24	1.987	< 0.05
Duration of pain syndrome (day), depending on age				
Up to 15 years	3.36 ± 1.12	2.54 ± 1.15	1.984	> 0.05
Older than 15 years	3.87 ± 1.16	3.76 ± 0.82	1.984	> 0.05

The surgical period was slightly shorter when using the classic Nuss procedure and the blood loss was also less. The volume of blood loss was greater in the group of pediatric patients who received surgical treatment by the author's method. This was due to chondrotomy, incomplete sternotomy. However, blood loss for school age pediatric patients (30 ml) is significant and allows this surgery to be considered minimally invasive. By age 10–14, there was no statistically significant between-groups difference in the volume of blood loss. Changing the shape of the chest in both methods of thoracoplasty was associated with equivalent pain syndrome severity.

In the analysis of intra- and early postoperative results, the following complications were revealed in the compared groups, which required additional therapeutic measures (Table 2). In TG1, bleeding from the supracostal vessel (n = 1), postoperative hemothorax (n = 2), and pneumonia (n = 1) were identified. In TG2 we observed hemothorax development (n = 4), hypoventilation (n = 1) and postoperative pneumonia (n = 1).

Since the significance level was greater than 0.05, there was no connection between the factor and the resulting sign. It can be concluded that the presence of intra- and postoperative complications

Table 2

Intra- and early postoperative complications in TG1 and TG2

Comparison group	Number of complications	Absence of complications	χ^2	p
TG1	4	112	0.455	> 0.05
TG2	6	108		

in clinical groups does not depend on the choice of treatment method.

Late postoperative complications occurred in both groups, but there were significantly more of them in the group of pediatric patients who received surgery according to the classical Nuss method. The number of complications is presented in Table 3.

Table 3

Number of delayed complications

Comparison group	Number of delayed complications	Absence of complications	χ^2	p
TG1	1	115	4.945	< 0.05
TG2	7	107		

In the group TG1 there was one bleeding in the bed formed for the surgical hardware. There were seven complications in TG2, including one fracture of the sternum at the level of the articulation of the manubrium and the body. In one case a tense hematoma developed in the fibrous capsule formed around the surgical hardware. Another patient demonstrated incarceration of a portion of the broadest muscle of back under the surgical hardware, and another patient exhibited partial compression of the sub-clavicular vascular bundle. Three children developed marked pain syndrome (intercostal neuralgia) 3–5 months after the surgery and continuing until removal of the surgical hardware.

The first complication was a fracture of the sternum at the level of articulation of its manubrium and body, which arose 1.5 months after thoracoplasty in a 15 year old patient. Third degree CC was one of the manifestations of severe dysplastic marfan-like syndrome. The patient had Degree IV scoliotic deformity of the spine, pronounced hypoplasia of all muscles, and pathologically increased volume of movements in all joints. After the surgical spine stabilization with the elimination of its rough deformity, the parents and the adolescent himself insisted on thoracoplasty of CC. The surgery was performed using a single plate according to the classical Nuss method. In the early postoperative period, a good cosmetic result was noted. A fracture of the sternum at the level of the articulation of the body and manubrium appeared spontaneously, without any provoking factors, three months after thoracoplasty. The thorax, in its lower half, has become keel-shaped. The surgical hardware stabilizing the sternum was removed as per protocol; however, the keel-shaped deformity of the thorax remained. The parents refused a second surgery, and the young man continues to live with the acquired keel-shaped deformity of the chest.

The second complication, a massive hemorrhage into the cavity of the fibrous capsule formed around the surgical hardware, arose two years after the thoracoplasty. Being completely healthy, pains in the chest, in the projection of the surgical hardware appeared in the adolescent, and he noted swelling of the tissues in the region of the subcutaneous terminal ends of the plate. Clinically, a fluid tense low-painful lesion with clear boundaries was determined in the projection of the plate, on both sides. This was confirmed by ultrasound examination. On the roentgenogram of the chest, deep deformities of the supporting ribs were revealed. During the surgery to remove the surgical hardware, when dissecting the fibrous capsule formed around the plate, approximately 300 ml of lysed blood was released. There was no continued bleeding. Most likely, the source of bleeding was the intercostal vessel. Two years after the thoracoplasty, a pressure injury occurred, which included erosion followed by heavy bleeding into the plate capsule. During removal of the plate, the bleeding stopped spontaneously.

The third patient complication arose 1.5 years after thoracoplasty. This involved incarceration of the portion of the broadest muscle of back under the "shoulder" of the surgical hardware. The cause of this complication can be considered the installation of a plate of excessive length, with ends reaching the rear axillary line. Actions of the adolescent, when he got into a small car, contributed to incarceration of the muscle in the space between the plate and the rib. Strong pain syndrome, increasing edema of tissues near the incarceration served resulted in hospitalization. But in the first h after hospitalization, the infringement was eliminated itself, the pain alleviated, and the patient experienced no pain relapse during the next six months until the plate was removed.

Table 4

The number of deformities/pressure injuries of support ribs

Comparison group	Presence of deformities, pressure injuries of supporting ribs	Absence of complications	χ^2	*p*
TG1	2 (1.7 %)	114 (98.3 %)	38.563	< 0.01
TG2	37 (32.4 %)	77 (67.6 %)		

The fourth complication was in an 18-year-old patient with severe canyon-type CC. His whole sternum and adjacent ribs were retracted. We used thoracoplasty (according to Nuss) with two plates to displace the sternal-costal complex to its physiological position. After activation of the patient in the postoperative period, he began to complain about the cyanosis of the left hand and the distal forearm, which appeared when the arm was lowered. If the patient raised his hand, the symptoms of blood flow disturbance stopped. Dopplerography revealed a significant violation of venous outflow at the level of the subclavian vein and partial compression of the subclavian vein. We attributed the development of these symptoms to changing the shape of the chest, narrowing the gap between the collarbone and the rib. Dynamic observation of this patient during the year revealed that over time there was adaptation of the main blood flow and the circulatory disorder in the left arm stopped. The intercostal neuralgia-type pain syndrome in three children was the result of excessive pressure of the surgical hardware on the supporting ribs. At the same time, the supporting rib changed its habitual position in the area of the articulation with the vertebrae, leading to compression of the intercostal nerve.

Following retrospective analysis, we concluded that all of these complications could be avoided by taking an individual approach to each patient, taking into account the degree of CTD and age.

1. Surgical hardware should not reach the broadest muscle of back 1.5–2.0 cm, which can prevent its incarceration.

2. In the presence of dysplastic Marfan-like syndrome, in the rigid thorax in young men aged 17-18 years and in the canyon-type CC, two plates should be used in thoracoplasty, which can reduce the excessive pressure on one supporting rib and does not lead to a secondary fracture of the sternum or damage to the intercostal vessels.

3. In older children with a rigid thorax, it is advisable to use a thoracoplasty technique developed by us, that provides for partial chondrotomy of deformed ribs. This reduces the load on the supporting ribs.

One known thoracoplasty complication, according to the classical Nuss method, is deformity of the ribs on which the surgical hardware abuts. We observed this late complication in a number of patients, as shown in Table 4.

As follows from Table 4, in 39 children the deformity/pressure injuries of support ribs appeared where there was pressure from surgical hardware. The forming callus covered the metal plate almost in a muff-shaped manner, which made it much more difficult to perform the surgical hardware removal. In this case, it was necessary to traumatically mobilize the plate from the bone tissue. This was accompanied by bleeding, with access from both sides. In this group of children, the postoperative period was characterized by prolonged pain syndrome. In one patient, a tense hematoma appeared in the fibrous capsule that formed around the surgical hardware during the postoperative period. We can also attribute this to pressure injuries on the rib and erosion of the supracostal vessel. Of the 42 patients who received the Nuss method surgery at the age of 15–18 years, deformities and pressure injuries of supporting ribs developed in 36 (85.7%); while in children under 15 years there was only one case of pressure-related deformity (1.4%). Of the patients who received the surgery on by the method we proposed, these late complications arose in two people (3.85%) at the age of 15–18 years. Furthermore, in patients who received the surgical treatment at an earlier age, they were not observed. Such a significant between-groups differences are explained by use of our additions to the technique of thoracoplasty, which reduce the force of the surgical hardware load on the supporting ribs.

Long-term results were traced in 184 people: 98 of them were in TG1 (53.2%) and 86 (46.8%) in TG2. The durations of catamnesis are presented in Table 5. Only one 5 year old child from TG1 had a complete relapse of the disease. Its cause was a gross violation

of the postoperative regime (jumping from a hill four months after thoracoplasty), which caused the plate displacement into the pleural cavity and required emergency removal. The parents refused repeat surgery. Taking into account that the cause of the relapse was not a consequence of the surgery itself, we believe that this event should not be considered in evaluation of treatment outcomes.

Table 5

Distribution of children by the duration of catamnesis after the second stage of treatment

Group	Age			Total
	2–4 years	5–10 years	> 10 years	
TG1	4	67	27	98
TG2	25	61	–	86

Since the author's technology was used at the first stage of treatment of children, the observation catamnesis was also longer. The presence in both analyzed groups of the equal number of children with a follow-up period of 5–10 years is also important, since during this period all long-term complications were revealed to the fullest.

The most important problem in treatment of children with CC was the development of partial relapse. This typically occurred 3–5 years after removal of the surgical hardware, along with the continued growth of the child. This sternum retraction appeared up to a Degree I deformity in 26 children (14 children in TG1 and 12 pediatric patients in TG2). Analysis was performed to determine the dependence of the relapse frequency on the age of the children who received surgery, the type of surgery, the presence of concomitant pathology, and the timing of immobilization of the sternal-costal complex. Table 6 shows the age of children with partial relapse, when thoracoplasty was performed.

Table 6

The frequency of partial relapse, depending on the age of patients at the time of surgery

Amount of children (n = 184)	Age at the time of surgery (years)	Number of partial relapses
19	3–6	19 (100 %)
19	7–9	3 (15.8 %)
74	10–14	2 (2.7 %)
72	15–18	2 (2.8 %)

Table 7

Frequency of partial relapse of CC in pediatric patients with connective tissue dysplasia

Treatment group	Children with connective tissue dysplasia	Partial relapse occurred	χ^2	p
TG1 (n = 116)	31	15	0.005	< 0.05
TG2 (n = 114)	22	11		
Total (n = 230)	53	26		

Table 8

Frequency of partial relapse of CC in pediatric patients, depending on the degree of deformity

Treatment group	Children with partial relapse	Initial degree of the chest deformity		χ^2	p
		II	III		
TG1 (n = 116)	15	2	13	0.112	< 0.05
TG2 (n = 114)	11	1	10		
Total (n = 230)	26	3	23		

Regardless of the surgical method chosen, 100% partial relapse was observed in pediatric patients who received surgical treatment at preschool age. In children of primary school age it occurred in only 15.8%, and episodical cases were observed in older children of the older age group, who had severe primary CTD. There was no statistically significant difference in the frequency of partial relapse depending on the type of thoracoplasty method used (Tables 7, 8).

The degree of deformity (Grade III) and the presence of concomitant pathology (connective tissue dysplasia) influenced the development of partial relapse. At the same time, a partial relapse did not develop in almost half of the pediatric patients with CTD. Its development most likely results from a combination of three prognostically unfavorable factors: preschool age, Grade III deformity of the chest, and CTD.

Conclusion

The Nuss method for the treatment of CC is most justified if the thorax is elastic, which is noted in children under 14 years of age. In older children, with rigid thoraces, it is more rational to use the author's technique of thoracoplasty. To prevent partial relapse of chest deformity in the long-term postoperative period, it is desirable to perform thoracoplasty in children over the age of 12 years. In children of preschool age with Grade III thoracic deformities and apparent functional impairments of the respiratory and heart organs, the surgery should not be postponed until this age. These children should anticipate repeated thoracoplasty in adolescence to achieve the ideal correction of the developmental chest defect.

Information about the contribution of each author

V.N. Stalmakhovich – performing the surgeries, the concept and design of the study, the editing of the article.

V.V. Dudenkov – participation in surgeries, collection and processing of materials, analysis of the materials obtained, writing the text.

A.A. Dyukov – performing the surgeries, collecting and processing materials, analysis of the materials obtained, writing the text.

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