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Review

Impact of pectus excavatum deformity on the cardiopulmonary function: a literature review

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ABSTRACT

BACKGROUND: Pectus excavatum deformity is the most common chest wall malformation. Currently, surgeons and researchers of this problem have no consensus on whether pectus excavatum is a purely aesthetic problem or whether pectus excavatum disturbs the function of the cardiopulmonary system.

AIM: To analyze publications on the effect of pectus excavatum on the cardiorespiratory system and the functional features of the heart and lung after thoracoplasty in patients with pectus excavatum.

MATERIALS AND METHODS: Data were searched in the scientific databases PubMed, Google Scholar, Cochrane Library, Crossref, and eLibrary without language limitation. In this article, the method of analysis and synthesis of information was used. Most of the studies included in the analysis were published in the last 20 years.

RESULTS: In patients with pectus excavatum, severity of cardiorespiratory dysfunction depends on the degree of chest deformity. According to obtained data, the pulmonary function test in patients with pectus excavatum in the majority of cases revealed restrictive pattern (formed vital capacity <80% of the norm, with normal ratio of forced expiratory volume in 1 minute to forced lung capacity). In most cases, echocardiography showed compression of the right heart chambers. Comparative analysis of the pre- and postoperative study of cardiorespiratory system in most cases indicated improvement and adaptation of the cardiopulmonary system to stress after surgical intervention.

CONCLUSIONS: Funnel chest is an aesthetic problem wherein a severe degree of deformity leads to impaired respiratory mechanics and dysfunction of the cardiovascular system. Surgical restoration of the volume of the retrosternal space allows improvement of the functional capabilities of the heart and lungs.

Keywords: pectus excavatum; cardio-pulmonary syndrome; pulmonary function test; echocardiography; cardio-pulmonary exercise testing.

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Научный обзор

Влияние воронкообразной деформации грудной клетки на сердечно-легочную систему (обзор литературы)

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АННОТАЦИЯ

Обоснование. Воронкообразная деформация грудной клетки — наиболее распространенный порок развития грудной клетки. В настоящее время у хирургов и исследователей данной проблемы отсутствует единое мнение относительно того, является ли воронкообразная деформация грудной клетки исключительно эстетической проблемой, или воронкообразная деформация грудной клетки нарушает функцию сердечно-легочной системы.

Цель — проанализировать публикации, посвященные влиянию воронкообразной деформации грудной клетки на сердечно-легочную систему, а также функциональным особенностям сердечно-легочной системы у пациентов с воронкообразной деформацией грудной клетки после торакопластики.

Материалы и методы. Поиск данных осуществляли в базах научной литературы PubMed, Google Scholar, Cochrane Library, Crossref, eLibrary без языковых ограничений. В процессе написания статьи использовали метод анализа и синтеза информации. Большая часть работ, включенных в анализ, опубликована за последние 20 лет.

Результаты. У пациентов с воронкообразной деформацией грудной клетки выраженность дисфункции сердечно-легочной системы зависит от степени деформации грудной клетки. Согласно данным проанализированной литературы при исследовании функции внешнего дыхания у пациентов с воронкообразной деформацией грудной клетки в большинстве случаев выявляли рестриктивный тип нарушения дыхания (сформированная жизненная емкость <80 % нормы с нормальным соотношением форсированного выдоха за минуту к форсированной емкости легких), а при проведении эхокардиографии в большинстве случаев определялась компрессия правых камер сердца. Сравнительный анализ исследования параметров сердечно-легочной системы в до- и послеоперационном периоде в большинстве случаев свидетельствовал об их улучшении и адаптации сердечно-легочной системы к нагрузке после хирургического вмешательства.

Заключение. Воронкообразная деформация грудной клетки не только представляет эстетическую проблему, но и при выраженной степени деформации приводит к нарушению механики дыхания и дисфункции сердечно-сосудистой системы. Хирургическое восстановление объема ретростернального пространства позволяет улучшить функциональные возможности сердца и легких.

Ключевые слова: воронкообразная деформация грудной клетки; кардиореспираторный синдром; функция внешнего дыхания; эхокардиография; нагрузочное кардиопульмональное тестирование.

Как цитировать

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BACKGROUND

Pectus excavatum (PE) accounts for approximately 90% of all chest wall deformities, with prevalence rates varying across populations [1–3]. This disease may occur an isolated deformity or as a manifestation of various genetic syndromes, such as Marfan syndrome, Ehlers–Danlos syndrome [4], Poland–Moebius syndrome [5], and other hereditary diseases. It is estimated that approximately every second patient with PE has a family history of chest wall deformity. Most genetic studies of familial cases evidenced the presence of multifactorial inheritance, with the specific contributing factors remaining unknown [6].

Males are reported to be affected 3–5 times more often than females. There is no known direct racial predisposition, however PE appears to be more prevalent in Whites compared to African Americans, Hispanics, or Asians [7, 8]. The literature reports an incidence of 4–8 cases per 1000 births [8, 9]. However, the actual incidence and prevalence of PE remain uncertain and may be much higher than current estimates, since no large population-based studies have been conducted so far [10]. The lack of clearly defined PE criteria (borderline, combined deformities) and the reliance on visual examination as a primary diagnostic tool, which is heavily influenced by the professional experience of an orthopedic surgeon in diagnosing anterior chest wall deformities, present a significant challenge in making an evidence-based assessment of the prevalence [11].

There is ongoing debate as to whether PE is an essentially aesthetic problem or whether it impairs the cardiopulmonary function. Similarly, it is unclear whether the cardiopulmonary function can be improved in PE patients by thoracoplasty or whether thoracoplasty has any effect on the cardiopulmonary system [12–14]. It is also debatable whether the symptoms of shortness of breath, chest pain, and heart rhythm disorders are primarily psychological in nature and whether an aesthetic surgery procedure, such as the approach proposed by Dupuis et al. (reconstruction with silicone implants) [15], may be effective.

The study aimed to review published literature investigating the impact of pectus excavatum on the cardiopulmonary system and evaluating the cardiopulmonary functions in patients with pectus excavatum after thoracoplasty.

MATERIALS AND METHODS

The data search was performed in the scientific literature databases PubMed, Google Scholar, Cochrane Library, Crossref, eLibrary, with no language restrictions, using the keywords: “pectus excavatum,” “thoracoplasty,” “pulmonary function testing,” “echocardiography,” “cardiopulmonary exercise testing,” “exercise stress testing,” “minimally invasive repair of pectus excavatum,” “Nuss procedure,” and “Ravitch procedure.” In preparing the article, the method of data analysis and synthesis was used. Most of the studies included in the review were published within the past 20 years.

RESULTS AND DISCUSSION

The cardiovascular function of PE patients has been investigated over the past 90 years. In 1932, Eideken and Wolferth first described electrocardiographic (ECG) findings in PE patients [16]. According to the literature, ECG abnormalities are common in PE patients, the most significant of which are incomplete and complete right bundle branch block, poor *R*-wave progression, changes in *P*-wave morphology, and ECG signs associated with Brugada syndrome [17, 18]. However, these changes are nonspecific, and most of children and adolescents with PE have no ECG abnormalities [9]. Echocardiography (EchoCG) is a more specific method than ECG, which was first demonstrated by Mocchegiani et al. Compared to healthy volunteers, PE patients exhibited typical EchoCG abnormalities, including compression of the right heart and significant narrowing of the right ventricular (RV) outflow tract (with a decrease in the RV end-diastolic and end-systolic areas) [19]. Jaroszewski et al. demonstrated that deformity of the ribs and sternum can cause compression of the right heart, decreased atrial filling and venous return, resulting in diastolic dysfunction and a reduction in the cardiac output [20].

The degree of cardiac compression is correlated with the severity of chest deformity (the depth of sternal depression at the apex of the deformity), as demonstrated by Chu et al. Moreover, the cardiac rotation angle and pulmonary vein angle had a positive correlation with the depth of sternal compression [21]. Sarioglu et al. showed that the cardiac rotation angle correlated with Haller index [22]. It is noteworthy that the cardiac left lateral shift with associated with the progression of PE often contributes to the development of right-sided asymmetric PE, which is a common type of deformity [20].

Malek et al. conducted a meta-analysis of preoperative and postoperative assessments of the cardiovascular function in 169 patients who underwent a surgical repair of PE (radical or minimally invasive thoracoplasty) [23]. The authors reported a statistically significant postoperative improvement in the cardiovascular function. This finding negates the hypothesis that thoracoplasty for PE is primarily an aesthetic procedure, offering only minimal physiological improvement. However, Guntheroth and Spiers [24] identified methodological shortcomings in the meta-analysis. A more rigorous selection of the studies revealed that the reviewed literature lacked sufficient data to draw a conclusion regarding the postoperative improvement of the cardiovascular function. Meanwhile, further studies evaluating the effect of PE on

the cardiac function corroborated the findings reported by Malek et al. [9, 25, 26].

An analysis of preoperative and postoperative computed tomography images showed that at 1 month after thoracoplasty, the heart shifted significantly in the anterior and rightward directions [27].

Coln et al. assessed ECG findings (before and after the Nuss procedure) of 123 patients (a mean age of 13 years) and demonstrated that preoperative cardiac compression was observed in 95% of PE patients, while postoperative ECG results were normal in 93% of the study subjects. Moreover, the authors reported that among the 54 patients with a mitral valve abnormality diagnosed by preoperative EchoCG, only 7 patients had minor mitral valve prolapse postoperatively [28].

It should be noted that the diagnosis of mitral valve prolapse is significantly more prevalent in PE patients compared to the general population [29]. Laín et al. hypothesized that mitral valve prolapse was attributable to the heart compression by the deformed chest, and that the elimination of compression would result in the prolapse regression. To confirm their preliminary hypothesis, the authors performed intraoperative transesophageal EchoCG in pediatric patients with PE and revealed that the valve configuration had changed to normal intraoperatively immediate to the sternal elevation during the Nuss procedure [30].

Previous studies have reported that a postoperative increase in the RV stroke volume and cardiac output [31, 32]. An increase in the RV ejection fraction as early as 2 weeks after the Nuss procedure was confirmed by magnetic resonance imaging data, and the left ventricular (LV) ejection fraction was normal before surgery, however showed a further improvement at the 1-year follow-up [33].

In the study by Krueger et al., transesophageal EchoCG demonstrated an improvement in the LV ejection fraction in 17 patients with PE with a mean age of 28 years [34]. However, other authors have reported that the LV parameters do not demonstrate a decline in response to cardiac compression in PE patients [20, 35].

The LV is normally located more posterior to the RV and has a thicker muscular wall. This suggests that the LV is more effectively protected from compression by the deformed anterior chest wall, which likely explains minor improvements observed in the LV after surgery, as reported by most authors [36]. While many studies suggest that the repair procedure promotes the functional recovery of the cardiac system in PE patients [20, 26, 37], the potential for the respiratory function improvement after surgery has persistently been a topic of debate amidst the conflicting results of the respiratory function assessments before and after surgery. Additionally, the use of non-uniform diagnostic criteria and methods for the lung dysfunction assessment has contributed to this uncertainty [12, 14, 38].

Lung compression is undoubted in young children with severe PE, who experience paradoxical breathing, which occurs when the sternum and ribs are retracted backward during inhalation. This type of breathing is associated with chest wall motion dysfunction and limited expansion of the lungs, primarily in the upper lobes. These results may result in frequent and prolonged respiratory diseases. Pulmonary compression, respiratory dysfunction, and retention of bronchial secretions contribute to the development of dyspnea and a persistent cough. These clinical symptoms are frequently associated with tracheitis, bronchitis, pneumonia, which predominantly affects the lower lungs [39].

However, in most young children with a mild chest deformity, PE does not cause any symptoms due to better chest wall compliance than in adults and progresses slowly, with only 22% of cases diagnosed before 10 years of age [40]. As the child grows, the chest wall elasticity and compliance gradually decrease, leading to cardiopulmonary dysfunction associated with decreased exercise tolerance, shortness of breath, chest pain, and heart rhythm disorders [31, 41, 42]. The onset or aggravation of clinical symptoms mainly occur in adolescence as the anterior chest wall deformity progresses [43].

In the study by Ramadan et al., dyspnea on exertion and chest pain at rest were reported by 13.3% and 20% of PE patients, respectively (a mean age of 13.8 ± 2 years) [44], which was consistent with the other published data [37, 45].

The pulmonary function assessment conducted by Ramadan et al. demonstrated a restrictive pattern (forced vital capacity <80% of the reference with a normal ratio of the forced expiratory volume in the first second to the forced vital capacity) in 23% of PE patients. However, there was no correlation established between these symptoms and reduced pulmonary function parameters [44] compared to the reference values [46]. In most cases, PE is not associated with chronic lung diseases [38]. PE does not affect the airways or lung parenchyma, except by mechanical compression, which has an adverse effect on pulmonary ventilation [9, 27, 47].

Katrancioglu et al. evaluated 31 pediatric patients with PE and established a negative correlation between the Haller index and each of the forced expiratory volume in the first second and the forced vital capacity of the lungs [45]. This was consistent with the findings of the multicenter study conducted by Kelly Jr et al. [38]. A restrictive pattern was 4 times more likely in patients with a Haller index of >7 [45].

To assess symptoms, such as difficulty breathing and the feeling of being unable to take a deep breath, Redlinger Jr et al. performed optoelectronic plethysmography on 119 individuals (63 patients with PE, 56 healthy controls). The study results demonstrated a chest wall motion dysfunction in the area of the anterior chest wall deformity and increased abdominal contributions to respiration in

PE patients compared to the control group. At the level of the umbilicus, PE patients had a 147% increase in marker excursion compared to the control group ($P < 0.01$) [48]. This study was a significant advance in understanding the pathogenesis of respiratory disorders in PE patients.

Binazzi et al. used an optoelectronic plethysmography technique to assess the pulmonary function in 13 patients who underwent the Nuss procedure and established that an average 11% increase in chest volume was accommodated within the upper chest ($P = 0.0001$) and to a lesser extent within the abdomen and lower chest [49].

However, the results of pulmonary function assessments after PE repair are also highly conflicting and vary considerably depending on multiple factors, such as the severity of the PE deformity, the type of surgical procedure, the homogeneity of the analyzed group, and the time of follow-up assessments [36, 50, 51].

In their meta-analysis, Malek et al. reported no significant improvement in the pulmonary function after PE repair [14], but their study included the outcomes of thoracoplasty performed using both the Ravitch and Nuss procedures.

The analysis of pulmonary function parameters suggestive of a decline in the pulmonary function or the absence of any postoperative changes was largely based on small samples and performed in the early postoperative period. Sigalet et al. presented preliminary results of the analysis of pulmonary function parameters in 11 PE patients (a mean age of 13.5 ± 3.1 years) at 3 months after Nuss thoracoplasty. A significant decrease in forced vital capacity and vital capacity was documented after surgery [52]. Jeong et al. [53] analyzed the results of pulmonary function assessments in 18 patients with PE under 18 years of age before and 4–6 months post-surgery, which was consistent with the findings from Sigalet et al. [52].

Borowitz et al. demonstrated that at 6–12 months, the Nuss procedure caused no adverse effects on the pulmonary function, and no significant differences were seen before and after surgery [54].

Jukić et al. reported a decline in the pulmonary function in the early postoperative period, which was associated with prolonged pain syndrome. The authors found that up to 81.5% of patients required analgesics for 1 day to 6 months post-surgery [55].

M. Noguchi et al. suggested that a decrease in pulmonary function parameters after PE repair was associated with the reduced usage of respiratory muscles. In their study, the authors demonstrated that early respiratory rehabilitation can significantly improve the inspiratory volume at 3 months after the Nuss procedure as compared to the inspiratory volume of patients who did not receive a respiratory rehabilitation and to the preoperative baseline values [56].

However, the implantation of one or two correction plates presents an additional factor contributing to the reduction of the chest wall motion function. Moreover, a decrease in the chest elasticity may be worsened by the patient's growth. Additionally, the time interval between the implantation and removal of the plate is several years [57]. Large cohort studies demonstrate a significant improvement in the pulmonary function after the plate removal compared to preoperative results [58, 59]. Most authors advise that a comparative analysis of pre- and postoperative results of the pulmonary function assessment should be conducted after the removal [27, 36, 60].

O'Keefe et al. reported a significant increase in vital capacity, forced vital capacity, total lung capacity, and forced expiratory volume in the first second at 3–6 months after the plate removal in patients with a mean age of 13.9 ± 2.3 years [35]. The study by Szydlik et al. demonstrated a statistically significant increase in the pulmonary function parameters (such as forced expiratory volume in the first second and forced vital capacity) after the plate removal compared to the values before the Nuss procedure. However, there was no statistically significant differences between the severity of the initial chest deformity and spirometric improvement. Moreover, no statistically significant correlations were found between the patient's age, height, and weight and the spirometric improvement [57]. This is consistent with the findings of the meta-analysis by Wang et al., who have not observed a significant correlation between the mean age of patients at the time of surgery and the functional recovery of the lungs [61]. However, these results could have been affected by the fact that the age of patients was similar in all 13 studies included in the meta-analysis. The age in 13 studies ranged from 10.4 to 16.9 years, and from 13 to 14 years in 6 of 13 studies.

Dreher et al. presented the pulmonary function parameters, such as total airway resistance and total specific airway resistance, in 114 patients before and after repair surgery. The findings have demonstrated that the plate removal was associated with an improvement in the parameters, reaching normal values, including in those patients who had been diagnosed with obstructive and emphysematous patterns in the preoperative period [60].

The pulmonary function is typically evaluated at rest; however, PE patients experience their symptoms during physical exertion. A physiological explanation for the improved exercise tolerance cannot be provided on the basis of separate assessments of the cardiac and pulmonary systems separately at rest. Cardiopulmonary exercise testing (CPET) offers more comprehensive diagnostic and prognostic details regarding the cardiopulmonary system function than functional assessments at rest. CPET is a widely accepted tool for the clinical assessment of exercise tolerance. It provides an analysis of complex pulmonary, cardiovascular and musculoskeletal responses to physical activity [31, 36]. A comparative analysis of CPET results in

PE patients before and after surgery reveals a considerable degree of heterogeneity. This is attributable to a number of factors, including different methodologies, small samples, varying ages of patients and times to the postoperative assessment [50, 62].

Wynn et al. [63] and Castellani et al. [64] analyzed the CPET results before and after thoracoplasty in pediatric patients with PE and did not find significant differences in the CPET parameters. However, Maagaard et al., who compared the CPET results in 49 adolescents with PE before and after thoracoplasty to the values measured in 26 healthy volunteers (control group), demonstrated a statistically significant reduction in the cardiac index in PE patients compared to the control group. At 3 years post-surgery, the cardiac index statistically significantly increased and did not differ from the values in the control group. However, there was no significant difference in maximum oxygen consumption between PE patients and the control group [51]. The multicenter study by Kelly Jr et al. demonstrated a 10.1% and 19% increase in maximum oxygen consumption ($P = 0.015$) and stroke volume ($P = 0.007$), respectively, in the postoperative period [38]. This was consistent with the findings from Das et al. [65] and Sigalet et al. [52].

The cardiovascular and respiratory systems are closely interrelated, such that an effect on one system will inevitably impact the other [20]. However, most studies demonstrate that the change in the breathing pattern in PE patients after the correction plate removal does not provide an explanation for the improvement in exercise tolerance [9, 20, 50]. Humphries et al. suggested that during exercise, additional cardiac output is required, and the compressed right ventricle is unable to generate a greater stroke volume due to limited diastolic filling. This results in a compensatory increase in heart rate to meet oxygen demands. As soon as the maximum heart rate is reached, there is no further increase in cardiac output and the PE patient experiences dyspnea and, consequently, limited exercise tolerance. Elimination of the RV compression leads to an increase in its volume [66], as demonstrated in many studies [58, 60, 65].

CONCLUSION

Over the past decades, there has been a shift in understanding of PE, moving away from the exclusive focus on its aesthetic aspect. Evidence has emerged to suggest an association between the chest deformity and the heart compression or changes in the respiratory biomechanics. Comparative analysis of the pulmonary function parameters and EchoCG at rest in the pre- and postoperative period demonstrates a postoperative improvement in the cardiopulmonary function. As evidenced by previous studies, an important aspect in the evaluation of surgical outcomes is the time of assessments, especially in the case of spirometry. The published literature presents limited and heterogeneous data on CPET following retrosternal repair. However, the studies demonstrate improved load adaptation of the cardiopulmonary system after surgery. Thus, surgical repair of the retrosternal space volume improves the cardiopulmonary function.

ADDITIONAL INFORMATION

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Author contribution. All authors made a significant contribution to the study and preparation of the article, and each read and approved the final version before it was published.

The largest contribution was distributed as follows. *A.M. Khodorovskaya,* wrote the manuscript, searched and analyzed the literature; *D.V. Ryzhikov* developed the study design and performed final editing; *B.Kh. Dolgiev* searched and analyzed the literature.

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