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Review



Bracing of children with pectus carinatum: A literature review

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ABSTRACT

BACKGROUND: Conservative treatment of children with carinatum is currently an urgent and unresolved problem despite the wide range of techniques available. Conservative treatment stops the progression of deformation and reduces the degree of deformation or eliminates it. Methods of evaluating treatment results include clinical (photo–video assessment, questionnaire survey, and chest measurements) and instrumental diagnostics (radiography, computed tomography [CT], and 3D scanning); however, no method of correction and orthosis that would consider all the tasks of conservative treatment, method of evaluating conservative treatment, and uniform protocol have been established.

AIM: This study aimed to analyze the literature containing information on the conservative treatment of patients with pectus carinatum.

MATERIALS AND METHODS: The study presents the results of a search in PubMed, Cochrane, and eLibrary using combinations of operators and keywords. A total of 54 foreign and domestic sources were extracted, and the search was not limited retrospectively.

RESULTS: Orthosis is effective and positively perceived by doctors and patients. The most modern designs in the treatment of carinatum included dynamic orthoses FMF-DCS and Braceworks (Calgary). Recent studies have reported a strong correlation between 3D scanning and CT to assess treatment outcomes. However, no single option for conservative correction of deformity allowed for achieving all treatment goals. The assessment of the effectiveness of orthosis using photos, videos, and questionnaire, and chest measurement is subjective.

CONCLUSIONS: The disadvantages of prosthetics determine the need to develop a clear treatment algorithm, methodology, and objective assessment of treatment outcomes, requiring further research.

Keywords: pectus carinatum; conservative treatment; orthosis; dynamic compression; children.

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Научный обзор

Ортезирование детей с килевидной деформацией грудной клетки (обзор литературы)

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АННОТАЦИЯ

Обоснование. Консервативное лечение детей с килевидной деформацией грудной клетки на сегодняшний день — актуальная и до конца не решенная проблема, несмотря на большой выбор методик. Консервативное лечение проводят для остановки прогрессирования деформации, уменьшения степени деформации или ее устранения. Различные методики оценки результатов лечения включают клиническую (фотовидеооценка, анкетирование, измерение размеров грудной клетки) и инструментальную (рентгенография, компьютерная томография, 3D-сканирование) диагностику, но не существует ни метода коррекции и ортеза, которые учитывали бы все задачи консервативного лечения, ни метода оценки последнего и единого протокола.

Цель — проанализировать литературу, содержащую информацию о консервативном лечении пациентов с килевидной деформацией грудной клетки.

Материалы и методы. В статье рассмотрены результаты поиска в электронных базах данных PubMed, Cochrane, eLibrary с использованием комбинаций операторов и ключевых слов. Было выбрано 54 источника (иностранные и отечественные) без ретроспективного ограничения поиска.

Результаты. Ортезирование результативно, позитивно воспринимается пациентом и врачом. Наиболее современные конструкции в лечении килевидной деформации грудной клетки представлены динамическими ортезами FMF-DCS и Braceworks Calgary. Последние исследования сообщают об использовании 3D-сканирования в качестве альтернативы компьютерной томографии для оценки результатов лечения. Однако не существует единого варианта консервативной коррекции деформации, позволяющего достичь всех целей лечения. Оценка эффективности ортезирования с помощью фото-, видеосъемки, анкетирования, измерения размеров грудной клетки субъективна.

Заключение. Недостатки ортезирования определяют важность разработки четкого алгоритма лечения, а методики объективной оценки полученных результатов необходимо в дальнейшем исследовать.

Ключевые слова: килевидная деформация грудной клетки; консервативное лечение; ортезирование; динамическая компрессия; дети.

Как цитировать

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BACKGROUND

Pectus carinatum (PC) is a deformity of the sternum and ribs with their displacement above the level of the relief of the anterior chest wall and is often accompanied by asymmetry and retraction of the costal arches. It is the second most common pathology after pectus excavatum, accounting for up to 20% of all chest deformities [1]. PC occurs four times more often in boys than in girls [2–4].

S.A. Haje et al. (1999) reported the influence of sternal growth on the development of chest deformities [5]. The chondromanubrial (manubrial) type of PC is characterized by the premature closure of the growth plates of the sternum and the normally continuing growth of the costal cartilage (Fig. 1).

The corpocostal (lower) type is characterized by hypoplasia of the lower part of the sternal body and normally continuing growth of costal cartilage (Fig. 2).

In the costal (lateral) type, the disproportionate growth of the sternum is noted, causing the rotation of the sternum in the frontal plane, and retraction of the costal cartilage on the opposite side (Fig. 3).

Moreover, 25% of the PC cases have genetic causes [6, 7]. PC may also be a part of a genetic syndrome or connective tissue dysplasia [4, 6]. Numerous genetic abnormalities, such as Marfan and Noonan syndromes, are often included in the PC symptom complex [7]. Concomitant thoracolumbar scoliosis can be detected in 12%–34% of patients with PC [8], and hyperkyphosis of the thoracic spine was registered in 14% of the cases [9]. A severe deformity of the anterior chest wall before the age of 11 years was noted in <10% of patients with PC; however, some patients have significant deformity at birth, affecting the entire anterior chest. PC is usually first identified at primary school age. The deformity gradually becomes more pronounced until full skeletal maturity is achieved, after which minor changes occur throughout adult life [10]. In the case of pyramidal forms, PC may cause chest discomfort in the prone position [11] but does not usually result in severe functional cardiorespiratory impairment. However, patients often experience low self-esteem and deteriorated quality of life [12], and rarely, pain in the deformity apex is possible during growth spurt [13].

Over the past decade, PC orthosis has gained significant popularity. However, attempts at conservative treatment of this type of deformity were not new. Since the 1970s, the first publications on orthotics for patients with PC have appeared. J. Vidal et al. (1977) treated 52 patients with plaster casts, followed by plaster vests and exercise. They reported good effectiveness of therapy [14]. In 1992, S.A. Haje and J.R. Bowen shared their experience with orthoses [15]. The orthoses appeared quite bulky, which made them unpopular with both patients and doctors. In 2006, several studies on the conservative treatment of PC were published



Fig. 1. Manubrial type of pectus carinatum [5]

[16–18]. The authors described orthoses of their design, and good results (up to 90%) were noted in patients who complied with the treatment regimen, which made orthoses a significant alternative to surgical treatment [12, 19–24]. In 2011, a survey of pediatric surgeons in Canada, who treated patients with PC, revealed that >80% of doctors preferred orthotics as first-line therapy for PC [25].

In 2008, M. Martinez-Ferro et al. introduced a dynamic compression system (DCS) for PC correction, which became the prototype for subsequent dynamic orthoses [3]. DCS can be used to measure chest pressure using a digital pound per square inch (PSI) sensor and a docking device located on it, and patients were classified into chest wall stiffness categories.

The main goal of orthosis is the elimination of carinatum deformity in patients with an elastic sternocostal complex. With a rigid anterior chest wall, conservative treatment can stop the progression of the deformity, reduce

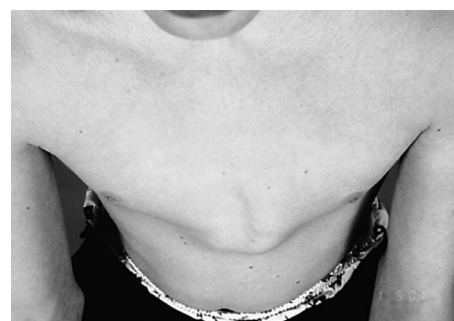


Fig. 2. Corpocostal type of pectus carinatum [12]



Fig. 3. Costal type of pectus carinatum [29]



Fig. 4. FMF-DCS brace [26]

the initial degree of deformity, and prepare the patient for surgery [26].

When correcting PC, trophic disorders of the skin of the anterior chest wall must be prevented through dosed loading and dynamic wearing of the orthosis.

Despite the increased interest of doctors in the treatment of patients with PC, no information in the literature helps establish the optimal approach to orthosis because of the lack of unified data on the mode of wearing the orthosis, duration of the correction stage, and possibility of stable compression for deformity.

This study aimed to analyze the literature containing information on the conservative treatment of patients with PC.

MATERIALS AND METHODS

A search was performed in PubMed, Cochrane, and eLibrary using combinations of operators AND and OR and keywords for English sources, namely, PC, bracing, DCS, orthosis conservative treatment and Russian sources (PC, conservative treatment, and orthosis). The retrospective search was not restricted by time.

Inclusion criteria:

- 1) Studies describing the treatment of children using external compression orthoses.
- 2) Studies describing the clinical results of orthotics and their evaluation.



Fig. 5. Braceworks Custom Orthotics Calgary [34]

Duplicate works, conference abstracts, book chapters, comments, and articles without full-text references were excluded. Fifty-four articles were selected, which included prospective cohort studies, retrospective studies, systematic reviews, and literature reviews.

RESULTS AND DISCUSSION

Orthosis options

Nowadays, numerous orthoses are used for PC correction. The main models are Fraire Martinez-Ferro DCS (FMF-DCS) (Fig. 4) [3, 25, 27–33] and Braceworks Braceworks Custom Orthotics (Calgary) (Fig. 5) [18, 34, 35]. Other orthosis models are used less frequently (Fig. 6) [16, 17, 36, 37].

The main components of orthoses for PC correction include a brace that can be adjusted to any shape of the chest, shoulder clamps to hold the orthosis on the body, a rigid support plate for compression at the top of the deformity, and an additional plate for girls whose mammary glands have begun to form. The main difference between the FMF-DCS and other orthoses is the presence of a removable pressure-measuring device (PMD) on the chest.

Age of orthotics start

The effectiveness of orthosis depends on many factors, particularly the anterior chest wall elasticity. The optimal age for starting orthotics is 6–18 years [38]. However, studies have analyzed orthotics for patients aged ≥20 years [27–30, 39].



Fig. 6. Other models of orthoses [16, 17, 36, 37]

In the identified studies, the criteria for selecting patients for orthotics are not sufficiently described; data on “elasticity” and compression strength are scattered. Patients with an elastic anterior chest wall were treated [3, 30]; however, some studies included children with rigid deformities [25, 27–29, 32, 39]. An important aspect in PC correction is the age at treatment initiation, taking into account periods of accelerated skeletal growth [40]. The appearance of severe deformity before or during the first growth spurt (7–8 years) increases the risk of the deformity progression at an older age (during the growth spurt 2). The greater the degree of deformity, the higher the risk of its progression. In children at an active growth stage, the deformity may reappear, despite regular use of the orthosis. In this case, patients are again transferred to the corrective stage to eliminate the deformity, and at the maintenance stage, they increase the time spent in the orthosis compared with the initial treatment protocol. In most children aged <10 years, the deformity is moderate; therefore, treatment is rarely prescribed during the first decade of life. In girls, the mammary glands can mask mild and moderately expressed PC, which makes orthotics unnecessary; however, if the deformity is significantly pronounced and asymmetrical, correction must be started before the age of 10 years [41]. Despite the lack of clear indications for treatment, the best results are achieved with correction of the elastic chest [22].

Assessment of chest elasticity

M. Martinez-Ferro et al. [26] developed a device to evaluate the chest elasticity. The device is integrated into the DCS, and the rigidity was measured in PSI. Patients were selected for conservative treatment of FMF-DCS by measuring the “starting pressure” (pressure for initial correction [PIC]). Initially, a PIC <7.5 PSI was used; however, patients are now eligible for orthotics if the PIC is no more than 10 PSI. If PIC values are high, patients are informed that complete restoration of the anterior chest wall relief is not probably. This treatment aims to alleviate the deformity and/or slow its progression.

Further treatment is performed at a pressure of treatment of deformity of 2.5–3 PSI to prevent skin damage and reduce pain at the site of orthosis pressure. In Russian literature, this figure ranged from 6 to 8 kg/cm² [42].

Orthotics protocol

The treatment protocol for PC deformities includes correction and maintenance. The correction stage is aimed at eliminating or reducing the deformity, whereas the maintenance stage is aimed at maintaining the correction and preventing relapse.

The Calgary orthosis protocol is the most popular [18]. The corrective stage involves wearing orthotics for 23 h a day, with 1 h left for hygiene procedures and performing a set of

exercises. The maintenance stage involves orthosis 8 h a day (mainly at night). According to most authors, orthosis should be used at least 20 h per day [21, 25, 27, 28, 31, 33–35, 37]. Generally, this wearing mode is recommended for correction with FMF-DCS orthoses and Braceworks Calgary orthoses. Some authors allowed the use of an orthosis for 8–16 h a day at the correction stage [16, 17, 30, 36, 38, 39] and mainly used orthoses of their design [16, 17, 36, 37]. However, some researchers did not pursue the maintenance stage, and treatment was completed when correction was achieved. These authors had analyzed a small number of cases and used a short evaluation period of long-term results, which did not allow them to rely confidently on their conclusions [2, 16, 36].

The treatment duration is determined not only by the type and degree of deformity but also by compliance with the wearing regimen of the orthosis. With high patient compliance, the treatment duration is reduced to 2–3 months [3, 18, 30, 31, 33]. A.S. Cohee et al. [39] used the FMF-DCS orthosis, and their corrective stage lasted an average of 6 months. For S. Emil et al. [31] (FMF-DCS) and D. Kravarusic et al. [18] (Braceworks Calgary), the deformity was eliminated 5.66 ± 3.8 and 4.3 ± 2.1 months at the corrective stage, respectively. S. Sesia et al. [29] used the FMF-DCS orthosis model and successful correction was achieved after 2.5–16 months. In most studies, patients treated with FMF-DCS and Braceworks Calgary orthoses completed treatment in an average of 24 months (end of the maintenance stage); however, some patients had to continue orthosis at the maintenance stage to prevent relapse [21, 25, 27, 28, 31, 33–35, 43].

N. Alaca et al. [44] conducted a study of the efficiency of physical exercises in the complex treatment of patients with PC. Among children with this pathology, some patients had scoliosis, kyphotic posture, and weak muscles. Other authors also believe that orthotics should be combined with exercise programs to improve function of the respiratory system, heart, and musculoskeletal system [13, 45, 46].

According to studies, the conservative method does not affect the patient's respiratory system during treatment. S. Emil et al. [31] assessed external respiration function before and after treatment and revealed no significant differences in these groups. O. Ates et al. [38] concluded that orthosis treatment did not alter pulmonary function parameters. The device does not negatively affect the respiratory system during treatment.

Methods for assessing orthotics

In most cases, treatment efficiency is determined subjectively. Using various methods for assessing the results of orthotics, establishing the efficiency of deformity correction programs is challenging [22].

Some authors used self-developed questionnaires to assess patient satisfaction with correction, including analysis of treatment results on a scale of 1–10, comfort while wearing the orthosis, and complications that emerge during orthosis treatment [2, 16, 29, 31, 34, 36, 38, 43]. Many patients were satisfied with the treatment results and recommended this treatment method to other patients with this pathology.

R.T. Lee et al. [35] took a series of photographs during orthosis treatment to evaluate treatment efficiency, and A.S. Cohee et al. [39] analyzed the anteroposterior chest size before and after treatment.

In some cases, effectiveness was determined using radiography and computed tomography (CT) [28, 37, 38, 43]. Based on CT data (Haller index, sternal rotation angle, and chest asymmetry index), the treatment results of patients with severe deformities were assessed. Positive changes in treatment efficiency were noted. The authors reported the high information content of studies when analyzing treatment dynamics but recommended CT mainly when planning surgical interventions [37].

E. Port et al. (2018) used white light scanning (WLS) [47]. It is an inexpensive, nonradiation, safe, and rapid three-dimensional imaging technique. By scanning patients before and after treatment, the authors were able to quantify the orthosis efficiency. Moreover, 13 (34%) of the 32 patients with PC examined showed an improvement of >10%, an improvement of 1%–10% in 21 (55%), and 4 (11%) had no positive dynamics. Preliminary results have revealed that WLS can be used to measure safely and accurately anterior chest wall deformity, providing an objective tool for the quantitative analysis of the dynamics of PC treatment using the Herbal-Malas index, an analog of the Haller index [47]. Data from other publications also confirm that nonradiation research methods are a good alternative to radiation methods [45, 48–52]. A. Hussain (2020) et al. also concluded that nonradiation methods are an alternative to CT for chest deformity assessment [53]. Data indicate a good correlation between the results obtained using these methods and the traditional assessment using the Haller index, and the correlation was more clearly identified in severe deformities. Nonradiation methods allow for continuous dynamic assessment of chest deformity correction during treatment.

Complications and relapses

The rate of poor outcomes with conservative treatment of PC (orthotics) reaches 38% [54]. The reasons may be damage to soft tissues in the pressure zone of the orthosis in 5.1% of cases [3, 27, 29, 31, 33, 39], pain syndrome in 12% [2, 3, 18, 32, 33], ineffectiveness of corrective treatment stage in 5.8% [27, 31, 35], relapse in 2.6% [3, 28, 31, 33, 39], tachycardia in 0.8% [39], vasovagal episodes during orthosis

installation in 1% [27, 31], and paresthesia of fingers I–III in 2.7% [29]. Damage to clothing and bedding was also described (13.2%) [27]. Patient dissatisfaction with treatment results, low compliance, pain, and skin damage are attributed to noncompliance to the orthosis protocol, failure to undergo a compression test before treatment, and/or excessive compression of the orthosis [54].

Subjective negative aspects of treatment noted by patients were the duration of correction, discomfort [29, 30], and orthosis breakdown in 5.6% of the cases [25, 29–31].

CONCLUSION

Orthotics for children with PC has proven to be a safe and effective method for eliminating the deformity and/or reducing its progression. Modern models of orthoses are less bulky and provide comfortable wearing during treatment. Nonradiation research methods, which have safe and high-quality characteristics, have begun to attract increasing attention when assessing the results of deformity correction.

Patient motivation, compliance with recommendations, and disciplined wearing of the orthosis are very important to achieve a positive effect. Thus, issues must be resolved to increase compliance and minimize complications associated with treatment inefficiency and deformity recurrence. The time for the corrective stage must be shortened; when prolonged, it increases the rigidity of the deformity. To avoid skin damage, gentle pressure on the deformed area is recommended. Comfortable wearing of the orthosis is desired, particularly in patients with the manubrial type of deformity and girls with pronounced mammary glands. However, the possibility of orthotics for patients with initial rigid chest deformity and the inclusion of this group of patients in the treatment protocol are still unaddressed.

ADDITIONAL INFORMATION

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Author contributions. All authors made significant contributions to the study and preparation of the article, and read and approved the final version before its publication.

The largest contribution was distributed as follows: A.S. *Tochilina* collected the literature data and wrote the article; D.V. *Ryzhikov* created the concept of scientific work and edited the text of the article; S.V. *Vissarionov* created the research design and edited the text of the article.

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