Dynamic Compression therapy for pectus carinatum in children and adolescents: Factors for success

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Highlights

- What is currently known about this topic?
- What new information is contained in this article?
- Pectus carinatum is increasingly treated with dynamic compression therapy. Factors for success for are relatively unknown.
- The pressure of initial correction and height of deformity are predictors for the total treatment time and the time to correction. Chest scores improved in the first months after start of treatment. Scores remained high.
Dynamic Compression therapy for pectus carinatum in children and adolescents: Factors for success

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Competing Interests: None
Abstract:

Background: Pectus carinatum (PC) is a congenital chest wall deformity. In childhood, it is increasingly treated with dynamic compression therapy. Factors for success for dynamic brace therapy are relatively unknown.

Methods: Between 2013 and 2020, 740 patients treated with the Dynamic Compression System (DCS), were studied. This included the effect of age, gender, pectus height, symmetry and pectus rigidity on treatment time and symptoms with linear multiple regression analyses.

Results: Carinatum height and high pressure of initial correction at the start of treatment were associated with a prolonged duration of treatment. For each cm increase in carinatum height, the total treatment duration increased with 1.9 months (p-value= 0.002, 95% CI: 0.70-3.13). An initial correction pressure of ≥7.6 pounds per square inch (psi), increased the treatment duration with 3.5 months (p-value 0.006, 95% CI: 1.04-6.01) compared to an initial correction pressure of ≤5.0 psi.

A high initial pressure of correction of ≥7.6 psi increased the odds of having somatic symptoms with 1.19 (p-value= 0.012, 95% CI: 1.04-1.45) and psychosocial symptoms with 1.13 (p-value= 0.04, 95% CI: 1.01-1.27) compared to a low initial pressure of correction of ≤5.0 psi. An initial pressure of correction of 5.1-7.5 psi increased the odds of having somatic
symptoms with 1.14 (p-value 0.046, 95% CI: 1.00-1.29) compared to an initial pressure of correction of ≤5.0 psi. Patients with asymmetric chests were more likely to abandon therapy

**Conclusions:** High carinatum height and high initial pressure of correction are associated with prolonged bracing treatment and a higher failure rate.

**Keywords:** Pectus; Pectus carinatum; Pigeon breast; Orthosis; Bracing; Dynamic bracing; Dynamic compression brace

**Level of Evidence:** III

**Introduction:**

Pectus carinatum (PC), or pigeon chest, is a congenital chest wall deformity with protrusion of the sternum due to overgrowth of rib cartilage [1]. The prevalence of PC in the general population is 0.3-0.7% and it is the second most common chest wall deformity in children after pectus excavatum [2, 3]. Typically, the deformity develops in puberty, however it may present at younger age if there is a genetic predisposition or as part of a syndromic disorder such as Marfan or Noonan syndrome [4]. The pathogenesis of PC remains unclear, although it is widely accepted that the deformity is caused by a defect in the costochondral cartilage [5].
The conventional treatment of pectus carinatum is surgical, including the Ravitch procedure, in which the excess cartilage is removed and the sternum is repositioned, and the Abramson procedure (or reversed Nuss procedure) in which a steel bar is placed subcutaneously over the sternum and fixated bilaterally to the ribs, in order to compress the sternum and restore the normal thoracic shape [6-7]. Disadvantages of these surgical procedures are pain, hospitalization, costs, scars and risks of adverse events (such as surgical site infection or pneumothorax) [8].

Brace therapy, in which the protruding sternum is redressed by pressure to the anterior sternum, has been done since the 1970’s but was only popularized after the introduction of the Dynamic Compression System in 2008 [8-12]. Due to the high pressure that was used with the former braces, dropout rates of up to 40% were not uncommon and were largely caused by high percentage of adverse side events such as pain and skin lesions [9-11]. With the dynamic compression technique, the sternum is gradually redressed to its normal position with a lower acceptable fraction of the initial correction pressure (PIC), called the pressure of treatment (POT). This allows for more wearing comfort, less pain and therefore a higher success rate.

Despite its gaining popularity it is still unknown in which percentage of patients with pectus carinatum bracing treatment is successful, if there are predictors for success and what the duration of treatment is.

**Methods:**

*Design*
All patients in whom the Dynamic Compression System (DCS) was used after March 2013 to October 2020 entered the study at our Pediatric Pectus Center in Amsterdam. This included also patients with concomitant connective tissue disorders and very young patients.

During their first visit to the outpatient clinic, all patients were examined and carinatum height and chest asymmetry were assessed. Initial measurements were taken and prospectively collected in a database, after consent of the patient and one or both parents was obtained. For each patient a personal treatment plan was made. A custom brace was made and applied and instructions for use were given. Patients were asked to wear the brace as much as possible in order to achieve the best results and follow-up consultations were planned initially with 6-8 weeks intervals and later with 3-4 months intervals until correction of the chest was obtained (correction phase). The correction phase is defined as ‘time to correction’. After correction of the chest, patients went into the retainer mode (or weaning phase) and gradually decreased the wearing time of the brace until the end of treatment [14, 15]. The correction phase and weaning phase together are defined as ‘total treatment time’. The moment of total correction of the chest and completion of treatment was determined by both the surgeon and the patient (after assessing the chest wall together). If patients were not satisfied or no longer motivated to wear the brace, termination of brace therapy and possible surgical options were discussed.

Materials

The FMF Dynamic Compressor System (FMF-DCS), manufactured by Pampamed, was used in all patients. It is a custom-made aluminum brace with an anterior plate for compression, to which a pressure measuring device can be attached. The plate can be moved sideways to redress either symmetrical or asymmetrical deformities.
The pressure of initial correction (PIC) is the total pressure in pounds per square inch (psi) required to position the sternum to its anatomical normal or desired position. It is measured by pressing the pressure-measuring device against the anterior chest wall of the patient (with the patient upright against a wall) until complete correction. This distance from protrusion to position of correction was measured and defined as height of the deformity.

The pressure of treatment (POT) is the pressure that is used during brace therapy and it is ideally kept between 2.0 and 3.0 psi. Initially, adjustment of the brace was done by the surgeon only. When patients became more experienced with the device, sometimes instructions were given on how to slightly increase POT at home.

Statistics

Data were analyzed using IBM SPSS statistics 26. Linear multiple regression analyses were performed with time to correction and total treatment time as outcome variables.

Rigidity of the chest wall was stratified by height of the PIC into three groups: Group 1 (low pressure): PIC ≤5.0 psi, group 2 (intermediate pressure); PIC 5.1 – 7.5 psi and group 3 (high pressure); PIC ≥7.6 psi. Other variables in the equation included: age, gender, pectus height (in cm) and symmetry of the deformity. Symmetry was defined as symmetrical, asymmetrical to the left or asymmetrical to the right. Pectus height was defined as the measured protrusion in cm of the sternum at the first visit. For these analyses, only patients who completed treatment successfully were included (n=406).

The risk of somatic or psychosocial symptoms was assessed with a multiple regression analysis with log-transformations because of a non-normal distribution, with odds ratios. All
patients who finished treatment successfully were used in this analysis and no missing data occurred in this group (n=406).

A sub-analysis was done for patients that were lost to follow-up.

Sum scores were made of initial symptoms for every patient. These symptoms were obtained through questionnaires patients received before starting brace treatment, implicating symptoms were self-reported. Symptoms were divided in two separate groups: somatic and psychosocial complaints. The most frequent reported symptoms and their incidence assessed in our study are shown in Table 2.

Patients were asked to score their chests esthetically before treatment with the brace and after 6, 12, 18 and 24 months. For the analysis of the esthetic self-assessment of the chest at baseline and during treatment, paired sample t-tests were performed.

**Results:**

740 patients started dynamic compression brace therapy between March 2013 and October 2020. At time of analysis 203 patients were still being treated. The remaining patients either finished treatment successfully (n=406), stopped treatment prematurely (n=74) - because of a lack of motivation or insufficient results - or were lost to follow-up (n=57). The lost to follow-up group was excluded from analysis. Of the patients who stopped prematurely, 16 (21%) received surgical correction. 85% of patients successfully finished treatment (Figure 3).

The patient characteristics of patients who were treated successfully or unsuccessfully with dynamic compression are described in table 1.
The follow-up period of these patients was the same as the total treatment time. Unless patients came back with a recurrence or complaints, they were not followed. The recurrence rate of pectus carinatum in successfully treated patients was 1.5%.

Somatic complaints were reported 377 patients (51%). The most common complaints included: thoracic pain during exercise (9.6%), shortness of breath during exercise (9.2%) and thoracic pain during rest (7.4%).

Psychosocial complaints were reported in 458 patients (62%). The most frequently reported psychosocial complaints were: shame of appearance (38.2%), esthetic problems (13.1%) and

The correction time was significantly longer in patients with a high PIC: 4.9 months in patients with a PIC of ≥7.6 psi compared to low PIC of ≤5.0 psi (2.539-7.286, p<0.001).

Pectus height or a right sided asymmetrical chest were no risk factors for prolonged correction.

The total treatment time was longer in patients with large pectus height and high PIC. One cm increase in pectus height increased the treatment duration with an average of 1.9 months; a high PIC of ≥7.6 psi extended the duration of treatment with 3.5 months compared to a low PIC of ≤5.0 psi (Table 2).

If we divide our patients in two subgroups divided by median age, it results in an outcome for children 0-14 and children 15 and older. In the younger age group (0-14) a significant result was found only for the height of the deformity (p=0.006). One cm increase in pectus height will increase treatment time with 2.6 months. We found similar results in the older age group, however a significant result in this group was also found for rigidity of the chest wall deformity (p=0.039). A patient with a high PIC will have an average treatment time...
extension of 3.1 month compared to a patient in the low PIC group. In contrast to our hypothesis, no evidence was found for correlation of asymmetry with total treatment time.

**Incidence of symptoms:**

Furthermore, we assessed the influence of rigidity, symmetry and pectus height on the incidence of somatic and psychosocial symptoms (Table 3).

A significant result was found for rigidity in patients with somatic symptoms as well as for psychosocial symptoms.

Both the high PIC group and intermediate PIC group are associated with a significantly more somatic complaints versus the low PIC group (resp. p= 0.01 and p= 0.05). The high PIC group is associated with more psychosocial complaints versus the intermediate and low PIC group (p=0.04).

No significant results were found for pectus height or an asymmetrical chest in correlation to symptoms.

**Unsuccessfully treated and lost to follow up**

Patients who were unsuccessfully treated had a significantly higher PIC than patients with successful treatment (7.6 +/- 1.8 psi vs. 6.7 +/- 1.7 psi) (Table 2).

This group of patients on average had a significantly higher PIC of 7.6 PSI (SD 1.8) compared to 6.7 (SD 1.7) in the successful group (p-value > 0.001). 53.3% of these children had a high PIC compared to 35.2% in the successful group. Pectus height was not significantly different: 3.5 cm (SD 0.94) vs 3.3 (SD 0.74) in the successful and unsuccessful group respectively (p=0.11). In short, patients with a rigid chest are more likely to end treatment prematurely and unsuccessfully.
Patients who were lost to follow-up also had a higher PIC than patients with successful treatment (7.4 +/- 1.8 psi vs. 6.7 +/- 1.7 psi). Also in this group there were relatively more patients with a PIC ≥7.6 psi (49.2% vs. 35.2%, p=0.001). Patients lost to follow-up were also more likely to have an asymmetrical pectus carinatum (44.2%); 18.0% left sided and 26.2% right sided. Pectus height did not differ significantly; 3.3 cm (SD 0.79, p=0.91), compared to the successful group.

**Patients with a pressure of initial correction of >10 PSI**

In our study group 29 patients participated with a PIC > 10.0 PSI. In 12 of these patients (41%) treatment was successfully completed, eight patients are still being treated, six ended treatment prematurely and three were lost to follow-up. The total treatment time was not significantly longer than for adolescents with a lower PIC; 21.4 months compared to 18.4 months in the successful group (p-value 0.096). When we consider the shape of the deformity of these High PIC patients, we can conclude that more patients have a right sided asymmetrical deformity (34.5%) rather than a left sided (6.9%). Moreover, these patients had a significantly higher protrusion of the sternum; 3.7 cm (SD 0.75, p-value 0.002).

**Patients with connective tissue disease**

In our study six patients had known connective tissue disease (Marfan or Poland syndrome). Two patients finished brace therapy successfully, three were lost to follow-up and one failed treatment. In patients with Marfan or Poland syndrome surgery might be considered the treatment of first choice because of poor results after bracing.

**Self-assessment of the chest**
Patients esthetically scored their chests before and during treatment. This was done using a scale of 1-10. We found a significant increase in scores after six months of bracing ($p<0.001$). At baseline, before the start of brace treatment, the mean self assessment chest score was 4.0. After six months of treatment, this score increased to 7.8. At 12, 18 and 24 months the scores remained stable and significant better compared to the baseline measurement.

**Discussion:**

Patients with pectus carinatum are increasingly treated with a dynamic compression brace. We have shown that this treatment is successful in a vast majority of patients. The time to correction depends on the initial pressure necessary to correct the chest wall, whereas the total duration of treatment depends on the initial pressure of correction and pectus height.

In our study, the dynamic compression therapy took between 1 and 2 years [14, 15]. This was relatively long compared to other studies as we used a longer period for the retainer phase [12, 14, 19, and 21]. We aimed for a retainer period of three months, however, the average time of the retainer phase turned out to be 9.8 months. Others described a median time of the retainer phase of 5.5 months. The period of time of the retainer phase was mainly determined by compliance of the brace therapy [20].

Patients experience a rapid decrease in the severity of the deformity in the first months after the start of brace treatment. Subsequently, when they enter the retainer mode they are keen to redress any remaining minimal protrusion to a perfect and completely symmetrical
flat chest, although they might already be content with the achieved results. This could explain why in some patients the retainer phase takes longer. Perfect symmetric results are not always feasible and patients will often end up with a minimal protrusion or relapse. The endpoint of the retainer phase is dependent on judgement of the treating physician and the patient and his/her family. It appears that with a higher initial correction pressure, the retainer phase takes longer.

Furthermore, we noticed that, especially in the male teenage group, the longer patients were using the brace, the more they feared stopping treatment after correction.

It may be difficult to determine the exact moment that the retainer phase was started. Some patients were not motivated enough and wore the brace just a few hours a day initially. In these cases, the active correction phase gradually merged into the retainer phase. In low rigidity chest walls, wearing the brace just a few hours a day can be enough to correct the deformity so that after correction, patients could continue the original frequency of wearing the brace until the end of their treatment.

Most studies limited the PIC to treat patients with the DCS brace to 10.0 PSI or less [12, 14, 19, and 21]. In our study group 29 patients participated with a PIC > 10.0 PSI and treatment was successful in 41%. Most of these patients had right sided asymmetrical deformity and a significantly higher protrusion of the sternum. Our experience in these high PIC patients is that when they are motivated frequent use of the brace decreases their PIC under the level of 10.0 PSI within weeks to months. Caution needs to be taken not to start with high treatment pressures because of the risk of skin lesions and pain.
In our group of patients, 24% of patients had an unsuccessful treatment or were lost to follow up. In this group, there were more asymmetrical deformities and this suggests that asymmetrical deformity may be associated with a longer DCS brace treatment.

As described in our previously published articles, we initially started brace treatment at young age (as young as 4 years) [8, 15]. We considered that the flexible chest at young age would lead to a faster correction and better compliance of treatment. However, in our recent experience it seems preferable to postpone brace treatment in youngest patients. Despite correction is easier at young age, there is a large risk of recurrence during growth spurt with the need to start brace therapy again or to prolong the retainer phase until the end of the growth spurt.

It remains difficult when treatment can be considered successful. Esthetic self-assessment is very subjective. In our study, results were objectified by the patient and the treating physician. Independent surgeons or peers were not used to objectively score the end results. In the future, there may be a role 3D scanning in assessing progress and to objectify end results. We just started using the 3D camera technique and derivative graphics in our clinic for objective measurements.

The largest series of patients with dynamic compression bracing was described by Dekonenko et al. They included 460 patients but there was no correlation between initial correction pressure or pectus height and the time to correction [20]. In our study, there was a significant correlation between pressure and time to correction and for pressure and pectus height on total treatment time.

Our study shows that the pressure of initial correction (PIC) is an important predictor for the total treatment time and the time to correction in brace patients. Pectus height of the
deformity seems to be a predictor in our successfully treated patients but not in the unsuccessfully treated group of patients. A high PIC is associated with more somatic and psychosocial complaints in patients with PC.

Despite our own subjective experience that asymmetric deformities were more rigid, we found no evidence for an effect of chest asymmetry of the deformity on treatment duration. However, there were significantly more patients with an asymmetrical chest who were lost to follow-up. This suggests that there might be a correlation of asymmetry on treatment success rate. In addition, we found a positive correlation between the incidence of symptoms and a high PIC.

Chest scores were used to evaluate the subjective opinion of the patient. These scores improved in the first 6 months after the start of the treatment. Thereafter the scores remained high for one to two years, which shows the benefits of brace treatment.

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Table 1: Patient characteristics of successfully treated and unsuccessfully treated patients.

<table>
<thead>
<tr>
<th></th>
<th>Successfully treated</th>
<th>Unsuccessfully treated</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Number of patients</strong></td>
<td>406</td>
<td>74</td>
</tr>
<tr>
<td><strong>Mean Age ± SD [range]</strong></td>
<td>$13.8 ± 2.44 [4-19]$</td>
<td>$14.9 ± 2.5 [5-21]$</td>
</tr>
<tr>
<td><strong>Male (%)</strong></td>
<td>370 (91.1)</td>
<td>69 (93.2)</td>
</tr>
<tr>
<td><strong>Pressure of initial correction in PSI ± SD [range]</strong></td>
<td>$6.7 ± 1.72 [1.9-12.5]$</td>
<td>$7.6 ± 1.88 [4.0-13.3]$</td>
</tr>
<tr>
<td><strong>Pressure of initial correction:</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1. Low; ≤ 5.0 PSI (%)</td>
<td>80 (19.7)</td>
<td>7 (9.5)</td>
</tr>
<tr>
<td>2. Intermediate; 5.1-7.5 PSI (%)</td>
<td>183 (45.1)</td>
<td>28 (37.8)</td>
</tr>
<tr>
<td>3. High; ≥7.6 PSI (%)</td>
<td>143 (35.2)</td>
<td>39 (52.7)</td>
</tr>
<tr>
<td><strong>Carinatum height in cm ± SD [range]</strong></td>
<td>$3.3 ± 0.74 [1.5-6.0]$</td>
<td>$3.5 ± 0.9 [1.5-6.0]$</td>
</tr>
<tr>
<td><strong>Symmetry</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>- symmetrical</td>
<td>294 (72.4)</td>
<td>39 (52.7)</td>
</tr>
<tr>
<td>- asymmetrical</td>
<td>112 (27.6)</td>
<td>35 (47.3)</td>
</tr>
<tr>
<td><strong>Initial pressure of treatment in PSI ± SD [range]</strong></td>
<td>$2.47 ± 0.34 [1.6-6.0]$</td>
<td>$2.5 ± 0.52 [1.7-4.8]$</td>
</tr>
<tr>
<td><strong>Family with PC (%)</strong></td>
<td>80 (19.7)</td>
<td>13 (17.6)</td>
</tr>
<tr>
<td>Family with PE (%)</td>
<td>41 (10.1)</td>
<td>6 (8.1)</td>
</tr>
<tr>
<td>--------------------</td>
<td>----------</td>
<td>--------</td>
</tr>
<tr>
<td>Time to correction (months)</td>
<td>7.85 ± 7.81 [1-36]</td>
<td>-</td>
</tr>
<tr>
<td>Total treatment time (months)</td>
<td>18.4 ± 8.15 [1-45]</td>
<td>13.7 ± 10.0 [1-38]</td>
</tr>
</tbody>
</table>

Table 2: logistic regression analysis of risk factors for time to correction and total treatment time in patients that successfully finished treatment.

<table>
<thead>
<tr>
<th>Risk factors</th>
<th>N=406</th>
<th>OR (95% CI)</th>
<th>P-value</th>
<th>OR (95% CI)</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female</td>
<td>0.14 (-2.99-2.71)</td>
<td>0.92</td>
<td>0.44 (-2.58-3.45)</td>
<td>0.78</td>
<td></td>
</tr>
<tr>
<td>Age at start brace</td>
<td>0.33 (-0.03-0.67)</td>
<td>0.07</td>
<td>-0.09 (-0.46-0.28)</td>
<td>0.64</td>
<td></td>
</tr>
<tr>
<td>Pectus height (cm)</td>
<td>0.87 (-0.29-2.02)</td>
<td>0.14</td>
<td>1.91 (0.70-3.13)</td>
<td>0.0002 *</td>
<td></td>
</tr>
<tr>
<td>Intermediate vs low PIC group</td>
<td>1.55 (-0.65-3.76)</td>
<td>0.17</td>
<td>1.64 (-0.68-3.95)</td>
<td>0.17</td>
<td></td>
</tr>
<tr>
<td>High vs low PIC group</td>
<td>4.91 (2.54-7.29)</td>
<td>&lt;0.001 *</td>
<td>3.52 (1.04-6.01)</td>
<td>0.006 *</td>
<td></td>
</tr>
</tbody>
</table>

*Significant (p-value <0.05)

Table 3: logistic regression analysis of risk factors for somatic and psychosocial outcomes.

<table>
<thead>
<tr>
<th>Risk factors: N=505</th>
<th>Somatic symptoms</th>
<th>psychosocial symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>OR (95% CI)</td>
<td>P-value</td>
</tr>
<tr>
<td>Intermediate vs low PIC group</td>
<td>1.14 (1.002-1.293)</td>
<td>0.05 *</td>
</tr>
<tr>
<td>High vs low PIC group</td>
<td>1.19 (1.038-1.354)</td>
<td>0.01 *</td>
</tr>
<tr>
<td>Asymmetrical to</td>
<td>1.05 (0.920-1.195)</td>
<td>0.47</td>
</tr>
<tr>
<td>the left</td>
<td>1.212)</td>
<td></td>
</tr>
<tr>
<td>--------------------------</td>
<td>------------</td>
<td></td>
</tr>
<tr>
<td>Asymmetrical to the right</td>
<td>1.01 (0.894-1.135)</td>
<td>0.90</td>
</tr>
<tr>
<td>Pectus Height in cm</td>
<td>0.97 (0.913-1.028)</td>
<td>0.30</td>
</tr>
</tbody>
</table>

*significant (p-value <0.05)

Figure Legends

**Figure 1**: Measuring the pressure of intitial correction (PIC) with the pressure measuring device.
Figure 2: (A) Dynamic Compressor System brace with the docking holes for the pressure-measuring device. (B) Measuring the pressure of treatment (POT) on a patient wearing the brace with the detachable pressure measuring device
Figure 3: Flow diagram of study population.

- 740 patients
  - 203 still in treatment
  - 537 patients
    - 57 lost to follow-up
    - 480 finished treatment
      - 74 not successfully
      - 406 successfully finished treatment