



Success and duration of dynamic bracing for pectus carinatum: A four-year prospective study



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ABSTRACT

Background: This study sought to establish factors that can prognosticate outcomes of bracing for pectus carinatum (PC).

Methods: Prospective data were collected on all patients enrolled in a dynamic bracing protocol from July 2011 to July 2015. Pressure of correction (POC) was measured at initiation of treatment, and pressure of treatment (POT) was measured pre- and post-adjustment at every follow-up visit. Univariate and Cox regression analysis tested the following possible determinants of success and bracing duration: age, sex, symmetry, POC, and POT drop during the first two follow-up visits.

Results: Of 114 patients, 64 (56%) succeeded, 33 (29%) were still in active bracing, and 17 (15%) failed or were lost to follow-up. In successful patients, active and maintenance bracing was 5.66 ± 3.81 and 8.80 ± 3.94 months, respectively. Asymmetry and older age were significantly associated with failure. Multivariable Cox proportional hazard analysis of time-to-maintenance showed that asymmetry ($p = 0.01$) and smaller first drop in POT ($p = 0.02$) were associated with longer time to reach maintenance.

Conclusions: Pressure of correction does not predict failure of bracing, but older age, asymmetry, and smaller first drop in pressure of treatment are associated with failure and longer bracing duration.

Level of evidence: Prospective Study/Level of Evidence IV.

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Pectus carinatum (PC) describes a spectrum of chest wall anomalies that results in the protrusion of the anterior chest wall, including the sternum and adjacent costal cartilages [1,2]. Although historically described as being far less common than pectus excavatum, many chest wall anomaly clinics, including ours, are finding PC to be as common, or more common, than pectus excavatum [3]. Although PC does not typically result in any significant physical or cardiopulmonary symptoms, it has been shown that patients with this condition have a disturbed body image, low self-esteem and a reduced quality of life [4–6]. The condition is therefore primarily treated to decrease the resulting social disability.

Current treatment options for PC, in order of increasing invasiveness, include observation, bracing, minimally invasive techniques typically referred to as the reverse Nuss or Abramson procedures, and the classic Ravitch procedure or modifications of it. After being first reported from Brazil in 1992, bracing has gained significant popularity among surgeons and patients alike over the last decade, and has emerged as first line treatment for this anomaly [7,8].

In 2011, we started the first multidisciplinary chest wall anomaly center in Canada, and one of the few in North America, as a collaborative effort between the Montreal Children's Hospital and the Shriners Hospital for Children. The center brings together pediatric surgeons, orthopedic surgeons, plastic surgeons, nurses, physiotherapists, and orthotic specialists to offer both individualized and protocol-driven care for children with chest wall anomalies. With the mentorship of the inventors, we initiated a dynamic compression system (DCS) bracing program in July, 2011, and started collecting data prospectively on each patient in the program [3].

The decision to adopt bracing as a therapeutic option is strongly influenced by the burden of bracing, and the chances of success. Children and parents are always interested in these two issues. We conducted this study in order to investigate these issues, aiming to provide useful information regarding bracing for both parents and clinicians.

1. Methods

1.1. Bracing protocol

All new patients presenting with PC to the Chest Wall Anomaly Center of the Shriners Hospital for Children, Montreal, attended a half-day

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clinic that included group sessions with physiotherapy, nursing, and orthotics. Following these sessions, a presentation on PC was given by a pediatric surgeon. The presentation included the basic facts about PC, as well as the options available for treatment. After the presentation, each patient underwent an evaluation by one of three pediatric surgeons and a nurse, in addition to orthopedics or plastics if warranted.

Once a decision to pursue bracing as therapy for PC was made, patients underwent chest wall measurements by a certified orthotist, including chest wall diameter, circumference, width, and carinatum height. At the same time, the pressure of correction (POC), in pounds per square inch (PSI), was measured. This was done using a previously described pressure-measuring device applied over the carinatum deformity until a normal chest wall contour was obtained [3]. A customized dynamic compression brace (FMF, Buenos Aires, Argentina) was then ordered. Patients were not excluded from bracing based on POC. Standardized photos were taken by the hospital's audiovisual department of all patients prior to initiating treatment to document the baseline deformity.

Once the brace was constructed, patients returned to our clinic for a fitting with the orthotist. The pressure of treatment (POT) was set using the same pressure-measuring device, docked onto the brace. This permitted correct adjustment of the brace to a given pressure, between 2 and 3 PSI that would be tolerated by the patient and least likely to cause skin breakdown. This initiated the active phase of treatment, during which patients were instructed to wear the brace for 23 h per day, over a thin t-shirt, removing it only for showers, baths, and sporting activities. Patients were instructed to continue with an exercise program aimed at strengthening chest wall musculature and improving posture.

Patients were followed in the clinic one month and three months after initiation of therapy, and every 2 to 3 months afterwards until completion of treatment. In the interim months, they visited the orthotist monthly for adjustment. At each orthotics visit, POT was measured pre- and post-adjustment, and used to calculate a POT percent change during the treatment period $\left[\frac{(POT_{\text{previous}} - POT_{\text{current}})}{(POT_{\text{previous}})} \times 100\right]$. For example, a patient whose POT was adjusted to 2.5 PSI at initiation of treatment and returns one month later with a POT of 1.0 PSI would have a 60% change, $[(2.5 - 1.0) / 2.5 \times 100]$. A drop in POT signified movement of the chest wall inward during the treatment period. Adherence to treatment was assessed by patient self-reporting of average hours of daily wear at each appointment and documented into three categories; full, partial and noncompliance. A fully compliant patient self-reported wearing the brace at least 20 h a day, on most days, a partially compliant patient wore it between 10 and 20 h and a noncompliant patient wore the brace for less than 10 h a day. We found this highly subjective and did not include this factor in the statistical analysis.

Bracing was considered successful when the surgeon and patient agreed that normal chest contour was achieved as documented by exam and photography. At that point, chest wall measurements were repeated, and the patient was switched from active to maintenance wear, 8–12 h during the night. Maintenance continued for a minimum of 6 months. Follow-up occurred every 3 months during maintenance, and at 6 and 12 months after end of treatment. All data were gathered prospectively. Chest wall measurements and pressure data were recorded in an orthotics flow sheet.

1.2. Analysis of outcomes

All patients in the bracing program during the 4-year period, July 2011–July 2015, who had at least two bracing adjustments after initiation of bracing, formed the study cohort. Patients were divided into three groups according to their status on July 31, 2015:

Group 1: Active bracing completed and successful. This group included patients in maintenance, as well as patients who completed treatment.

Group 2: Active bracing completed and unsuccessful or patients who were lost to follow-up before success was documented.

Group 3: Active bracing in progress.

Two outcome measures were assessed: success and active bracing duration. Success was reported as a binary variable (yes vs. no), and analyzed by comparing Group 1 to Group 2. Duration of active bracing was assessed using two methods. First, patients in Group 1 were divided into two cohorts depending on duration of active bracing, ≤ 6 months vs. > 6 months, and compared. Appropriate statistics (frequencies for categorical variables, mean \pm standard deviation for numerical variables) and tests (Fisher's exact test for categorical variables, Wilcoxon rank sum tests for numerical variables) were used for comparison. Second, for patients who did not fail (Groups 1 and 3), time to maintenance was defined as the days starting from the date of initial treatment to the first date of reaching maintenance, or censored at the date end of the study (July 31, 2015). The difference in time to maintenance was tested for each possible determinant (e.g. male vs. female), using the two-sided log-rank test or Wald test based on Cox PH model at an alpha level of 0.05. Possible determinants included age, sex, symmetry, POC, and POT drop during first two follow-up visits. For all statistical analyses, $P \leq 0.05$ was considered statistically significant. The study was approved by the McGill University Faculty of Medicine Institutional Review Board (A07-M41-15B).

2. Results

2.1. Patient characteristics

During the study period, 119 patients were measured for a DCS brace, but only 114 proceeded with treatment, including 104 (91.6%) boys and 10 (8.4%) girls. The median age at presentation was 14 (mean 14.2 ± 1.8 , range 7.5–17) years, and the median age at onset of the carinatum was 12 (mean 10.4 ± 4.3 , range birth–17) years. Most patients reported noticing the carinatum at the time of their growth spurt, becoming more prominent during puberty. On a screening questionnaire assessing motivation for bracing ($n = 116$), 62 (53.4%) patients reported social concerns related to their deformity, 18 (15.5%) reported chest pain, 13 (11.2%) reported subjective exercise limitations or shortness of breath and 3 (2.6%) complained of palpitations.

When symmetry was assessed ($n = 117$), 76 (65%) had asymmetric deformities, while the remaining 41 (35%) had symmetric carinatum. Six patients with a mixed pectus excavatum–carinatum anomaly were also braced. The median height of the carinatum was 1.5 (mean 1.8 ± 0.69 , range 0.75–5.00) cm. The median POC ($n = 117$) was 5.2 (mean 5.23 ± 1.5 , range 1.8–8.6) PSI. Of note, 11 patients (9.2%) had a POC greater than 7.5 PSI.

2.2. Patient subgroups

Group 1 consisted of 64 patients (56%) who had achieved successful correction and either were in maintenance phase (52%) or had completed bracing (48%). In this group, mean duration of active bracing was 5.66 ± 3.81 months. For those who completed bracing, mean duration of maintenance bracing was 8.80 ± 3.94 months. Median follow-up in this group was 448 (range 77–1177) days. Examples of successful patients with symmetric and asymmetric anomalies are shown in Figs. 1 and 2, respectively.

Group 2 consisted of 17 patients (15%) who failed treatment [13] or were lost to follow-up and were assumed to be treatment failures [4]. In this group, median duration of active bracing ($n = 12$) was 230 (mean 302 ± 247 , range 1–897) days. None proceeded to maintenance bracing.

Group 3 consisted of 33 patients (29%) who were still in active bracing on July 31, 2015. At the time of final manuscript submission (June 6, 2016), 21 patients (64%) had succeeded, 3 patients (8%) were still in active bracing, and 9 patients (27%) either failed [3] or were lost to follow-up [6].

Therefore, the overall success rate for the entire cohort was 75% with three patients still progressing with active bracing.



Fig. 1. Correction of symmetric PC with DCS brace.

2.3. Determinants of successful bracing

Table 1 shows the comparison between Groups 1 and 2. Asymmetry was significantly associated with failed bracing. An older age at presentation was also associated with failure. The percentage of patients presenting at age < 14 was 60.9% in Group 1 vs. 29.4% in Group 2, $p = .02$. POC was not different between the groups, and there was no difference in the percent of patients with POC > 7 PSI, 12.5% vs. 17.6% in

Groups 1 and 2, respectively, $p = .51$. The first two pressure drops were also significantly higher in patients who succeeded.

2.4. Determinants of bracing duration

Table 2 shows the comparison between patients in Group I who underwent active bracing for six months or less versus more than six months. Age was lower in those who were braced for shorter periods



Fig. 2. Correction of asymmetric PC with DCS brace.

Table 1
Comparison between bracing success and failure.

Variable	Success	Failure	P
N	64	17	
Sex (% male)	93.8	94.1	.96
Age (years)	14.0 ± 1.6	14.8 ± 1.4	.03
Symmetry (%)	40.6	5.9	.005
POC (psi)	5.27 ± 1.55	5.31 ± 1.55	.96
1st POT drop (%)	79 ± 30	32 ± 67	.006
2nd POT drop (%)	67 ± 47	–11 ± 134	.011

but did not reach statistical significance. Patients who were braced for less than six months manifested a higher drop in POT one month after initiation of bracing.

Multivariable Cox proportional hazard analysis of time-to-maintenance (censored at the last treatment date if cases were still in active bracing), in which age, sex, symmetry, POC, first drop in POT, and second drop in POT were covariates, showed that asymmetry ($p = 0.01$, hazard ratio 2.1 for symmetry versus asymmetry) and smaller first drop in POT [$p = 0.02$, hazard ratio 1.12 for each 10% drop in POT (i.e. 12% increased chance of success for each 10% drop)] were associated with longer time to reach maintenance, while other covariates did not show significant association with time to maintenance.

2.5. Complications

A total of 44 (37%) patients complained of negative symptoms associated with bracing therapy. The majority of these, 23 (52.3%), were complaints of mild chest discomfort. One patient noted that the discomfort was severe enough to interfere with his ability to sleep. Thirteen patients experienced mild skin changes at the site of maximal brace pressure. Four patients experienced moderate skin changes including blistering and mild cellulitis. Of these, one patient had skin changes severe enough to require temporary discontinuation of bracing. Three patients had problems with the brace breaking, which likely prolonged their bracing time owing to delays in repair. Of interest, one patient experienced a vasovagal episode during brace fitting.

2.6. Recurrences

There were 8 patients with mild recurrences of their PC, 7 during the maintenance phase of treatment. Of these patients, 6 returned to active bracing and one increased brace wear to part-time and all achieved full resolution of their PC. Only one patient presented with a mild recurrence of their PC one month after the end of treatment and opted to wear the brace part-time for an additional 12 weeks to achieve full correction.

3. Discussion

Pectus carinatum is a chest wall deformity associated with significant social disability owing to negative perceptions of body image [4–6]. This deformity has been referred to as the “undertreated chest wall deformity” due to infrequent referrals from primary care practitioners and probable underestimation of the frequency of the deformity [9]. Until the turn of the century, the Ravitch procedure, or one of its

Table 2
Comparison of Duration of Active Bracing.

Variable	Active Bracing ≤6 mo	Active Bracing >6 mo	P
N	37	27	
Sex (% male)	91.9%	96.3%	.47
Age (years)	13.7 ± 1.8	14.4 ± 1.3	.08
Symmetry (%)	45.9	34.6	.37
POC (psi)	5.1 ± 1.5	5.5 ± 1.7	.30
1st POT drop (%)	84 ± 28	72 ± 30	.05
2nd POT drop (%)	69 ± 53	65 ± 40	.26

modifications, was considered by the pediatric surgical community to represent the only treatment option [9–11]. The invasiveness of this procedure and the ironic resulting anterior scar may have contributed to the undertreatment of PC, perceived by many to be a cosmetic problem. During the past decade, minimally invasive techniques have been described [12–15]. However, early experience revealed multiple technical challenges that had to be surmounted, and the volume of patients who have undergone these procedures still remains low. Although we offer this option in our chest wall anomaly center, we have used it quite sparingly owing to the availability of bracing.

A reliable nonoperative therapy, which avoids the risks associated with surgery, including unsightly scars, anesthesia, blood loss, recurrence, and hardware complications represents a good alternative. Nonoperative treatment of PC is not new. Vidal et al. published their experience in the French literature in 1977 [16]. They treated 52 patients with plaster casts followed by plaster jackets and exercise, and reported a high success rate [16]. Haje published a Brazilian bracing experience in the orthopedic literature in 1992 [7]. In 2006, three publications in the pediatric surgical literature helped inform pediatric surgeons regarding this noninvasive option and established it as a viable alternative [17–19]. In 2011, we surveyed surgeons who treat PC across Canada, and found that the great majority, more than 80%, preferred bracing to other options and felt that bracing should be first line therapy [8]. However, outcomes were not well defined. At the time, we also reviewed the international bracing experience through 2011, and found eight reports with variable bracing techniques and protocols, with success rates ranging from 40% to 100% [8]. In the five years since our previous study, ten more case series of bracing have appeared, attesting to the increasing popularity of this therapeutic option [15,20–28]. A recent review recommended bracing as first line treatment for PC [29].

In considering bracing therapy for PC, it is advantageous for patients, parents, and clinicians to understand the chances of success and factors associated with it. In our center, we provide a group session to patients and their parents to give the basic facts on PC and describe the treatment alternatives, prior to the individual patient evaluations. The patient's decision to proceed with bracing is intricately linked to whether they are likely to succeed, and how long they are likely to require active bracing.

The chances of bracing success have been studied mostly in the context of patient compliance with bracing. In an updated series from Calgary of 98 patients, a failure rate of 45% was reported, with all but two failures owing to noncompliance [22]. Two recent studies have looked into the issue of compliance in more detail [24,27]. Pessanha et al. found that patients were likely to be noncompliant with duration rather than frequency of brace wear, i.e. they wore the brace daily, but for shorter periods than prescribed. Compliance was highest in those who needed shorter treatment periods to reach shorter prescribed duration of daily wear [27]. Kang et al., using a bracing protocol quite similar to ours with respect to duration and frequency of wear, found that the strongest predictor of compliance was a good result during the first 2–4 weeks [24]. This is inherently logical, since a patient who notices an early change in chest wall appearance is more likely to continue wearing the brace. Compliance, success, and therapy duration are therefore closely related. We documented compliance, as reported by the patient, during each visit. However, we felt that this was a highly subjective variable, and omitted it from the analysis of success.

Since we initiated bracing in our chest wall anomaly center, we have been interested in obtaining objective data that may predict success and required duration of therapy. This led us to the choice of the FMF dynamic compression system (DCS), as it was the only brace that provided correction and treatment pressure data, allowing for precise measurements and adjustments. In the largest reported experience with the DCS, Martinez-Ferro et al. found that the pressure of correction (POC) correlated with success and therapy duration, and recommended against using the DCS in patients whose POC was higher than 7.5 PSI [3]. We did not exclude any patients based strictly on POC, and we did

not find POC to be associated with either failure or increased duration of therapy. Lopez et al. recently reported their results with the DCS, and also found POC not to predict failure, with flattening of the sternum in 8 of 14 patients with POC between 7.5 PSI and 9.0 PSI [23]. In another recent series of DCS patients, Sesia et al. also braced patients with POC up to 9.0 with good early results [28]. Therefore, a high POC should not be considered a contraindication to bracing.

Our study looked at four factors that can be assessed at initial evaluation of each PC patient: sex, age, symmetry, and POC. Of those, asymmetry and older age were associated with a higher failure rate. In a prior study, Cohee et al. excluded very asymmetric patients from DCS treatment owing to experiencing more failures in this group. In patients with symmetric PC, the pressure plate typically rests in a stable position on the sternum. Asymmetric patients often have a significant ridge, which makes bracing less stable and more difficult to apply to the most protruding area in the chest. Age also appeared to be a factor influencing bracing duration. Among patients who successfully completed the active phase of treatment, duration of active treatment was longer in older patients, approaching statistical significance. This reinforces findings from prior studies, which have shown a need for longer bracing with increasing age and Tanner stage [22,26]. In the Argentinian experience, bracing is often started quite early, when the chest wall is most compliant [3]. A recurrence at the onset of adolescence is rebraced. Most of our patients are referred several years after the PC was first reported to their pediatrician or family physician. Improvement in outcomes may be achieved by educating referring physicians, emphasizing early referrals.

We always started pressure of treatment (POT) at less than 3.0 PSI to avoid skin discomfort and skin complications, two factors which may contribute to lack of compliance and eventual failure [24]. Bracing discontinuation or suspension owing to these factors in our series was rare. The drop in POT during the first two follow-up visits, within the first three months after brace initiation, was measured to assess treatment response. Lack of a significant drop in POT signifies either non-compliance or poor response to compression. Either factor can stimulate the other. Noncompliance can lead to poor response, and an initial poor response can lead to noncompliance. To resolve which is the chicken and which is the egg, an objective monitoring of compliance is required. Harrison et al. have developed an electronic remote compliance monitoring system for DCS patients [30]. Such a system can also be combined with pressure drop data to optimize the number of hours of wear per day. Monitoring and pressure drop data can then be correlated to provide objective feedback to noncompliant patients, possibly enhancing their compliance. As expected, the first pressure drop was smaller in those who required active bracing for more than six months. It was also found to be a predictor, along with asymmetry, of longer active bracing duration in the entire cohort, when a Cox proportional hazard analysis was applied. Although this variable can be measured only after initiation of bracing, we believe it is still quite helpful. Ongoing compliance may be enhanced when patients and parents have a target date for end of active bracing, which can be estimated once the first pressure drop is measured.

Our study is limited by the inherent subjectivity of the treatment of PC. Compliance, success, and patient satisfaction with body image are not binary variables, but rather a continuum. Duration of active and maintenance bracing, while measured, is influenced by patient desire and surgeon's preference. We may have slightly underestimated success by including patients lost to follow up in the failure group. However, we felt that there is an excellent likelihood that patients who terminated their follow up before success never achieved success. Including loss to follow-up in the failure group is likely the most accurate portrayal of bracing outcomes. Despite these limitations, we have reported prospectively acquired data over a 4-year period that should help patients and clinicians to set time frames, expectations, and end points for bracing PC. As for any therapy, candidate selection is very important in optimizing successful outcomes. In the case of dynamic

bracing, patients younger than 14 years with symmetric PC and a high motivation towards correction make the best candidates.

Author contributions

Study conception and design: Emil, Sevigny, Finlay
Acquisition of data: all authors
Analysis and interpretation of data: Emil, Sevigny
Drafting of manuscript: Emil, Sevigny
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