Bracing is an effective therapy for pectus carinatum: Interim results

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Haller index;
Pubertal growth;
Spurt

Abstract

Background: Pectus Carinatum is a common congenital chest wall malformation. Until recently the mainstay of treatment was surgical remodeling of the deformed chest wall. Initial results suggest that non-operative bracing may be an effective therapy, but the optimal strategy for correction is not known. Herein we report the results of a self-adjustable low profile bracing system worn continuously until the defect is corrected (correction phase), then worn at night (8 h/day) until completion of axial growth (maintenance phase)—the Calgary Protocol.

Methods: Patients referred to a pediatric surgery chest wall clinic were prospectively asked to join an IRB approved outcomes monitoring study. 124 patients were evaluated from 2007 to 2011, and 98 were prescribed a brace and counseled to follow the protocol.

Results: 98 patients consented to follow-up at starting bracing age: 14.4 ± 1.9 years, Tanner stage: 3.6 ± 0.5, protrusion: 2.1 ± 1.0 cm, self-rating of appearance: 2.9 ± 1.1, and exercise tolerance: 4.4 ± 1.1 (1–5 with 5 = normal). 10 patients are in correction phase, and 44 patients have completed correction after 7.0 ± 7.3 months: Tanner stage: 3.8 ± 0.1, protrusion: 0.5 ± 0.6 cm*, appearance: 4.3 ± 0.3* and exercise tolerance 4.6 ± 1.0. Correction occurred more quickly in patients prior to achieving Tanner stage IV (4.2± 0.9 months) vs. Tanner stage IV (8.0 ± 7.1 months) at the beginning of bracing. 21 patients completed maintenance bracing after 17.9 ± 19.0 months: Tanner stage: 3.9 ± 0.2, protrusion 0.5 ± 0.7 cm*, appearance: 4.3 ± 0.9*, and exercise tolerance: 4.8 ± 1.4. Average follow-up after bracing is 13.9 ± 16.0 months (mean ± S.D., *P < .05). There was one recurrence, likely due to early discontinuation of maintenance. This responded to an additional 6 months of bracing. 42 patients failed therapy secondary to non-compliance or were lost in follow up, while 2 patients did not respond to bracing and required open operation.

Conclusions: If patients are compliant, a self-adjusting brace system can give rapid correction of the pectus carinatum protrusion with excellent patient satisfaction. These interim results suggest that continued bracing until skeletal maturity gives long term durability to the correction. Further studies will be required to further refine this promising therapy.

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Disclosures: MS is the inventor of the brace described here and the owner of Braceworks, an independent company making the brace. None of the remaining authors have any conflicts of interest.

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Pectus carinatum (PC) is a relatively common pediatric congenital chest wall deformity. It is characterized by an abnormal overgrowth of the costal cartilages resulting in anterior protrusion of the sternum and adjacent costal cartilages [1,2]. There are two main types of PC deformity: the more typical in mid-sternum is gladiolar, while if confined to the upper sternum it is known as a manubrial prominence. In North America, PC has an estimated incidence of 1 in 1500 live births, an overall prevalence of 0.6%, and a male predominance (4:1 ratio) [3,4]. Although a clear genetic linkage has not been identified, a familial occurrence is frequently observed, suggesting a genetic predisposition [4].

The fundamental cause of PC is unknown and the most common presentation is as an isolated finding in otherwise well teenaged male. It can be associated with certain genetic disorders or syndromes [5], most commonly musculoskeletal (especially spinal curve defects) and connective tissue abnormalities [6–8]. The natural history of PC is of a mild defect seen in infancy, which is stable during childhood, but then with the growth spurt of puberty the deformity often worsens dramatically, drawing medical attention [6]. Aside from the appearance, the majority of patients present with relatively mild symptoms; the most commonly reported are tenderness, bone pain or mild exercise intolerance [9]. The cardiopulmonary derangement seen in pectus excavatum (PE) is not typically seen in PC patients [10,11]. The most common concerns of patients and families are the appearance and psychosocial issues secondary to body image [12].

The mainstay of treatment over the past 50 years has been a modification of the original Ravitch technique wherein the deformed costal cartilages are surgically resected along with reconfiguration of the chest wall [13–16]. The procedure is long and tedious and is associated with risks and morbidities that are common to major surgical procedures [17]. Long term results have been mixed, with reports of worsening cosmetic results and decreased chest wall compliance over time [18–20]. Therefore, surgical repair has been reserved for the most severe cases; this leaves many patients and families with a mild to moderate deformity in a dilemma between undergoing extensive surgery and being left untreated [21].

The recent surge in activity using the minimally invasive approach for the repair of pectus excavatum deformity has increased our understanding of the chest wall and its maturation pattern; the chest wall is typically malleable and plastic during puberty [22–24]. This concept led to the development of a non-surgical external compression bracing device, to essentially mirror the effects of the internal bar in excavatum patients. This is theorized to remodel the growth pattern of the deformed chest wall cartilage, which is the fundamental cause of pectus carinatum, and over time to correct the defect. Initial results have demonstrated that the method is effective in the short term [25–28]. However, the optimal protocol to achieve stable long term correction, and the durability of the repair are not known. We hypothesized that in order to achieve long term remodelling of the chest wall contour, a phase of maintenance bracing would be required, so long as axial growth was occurring. We report herein our initial cohort of patients treated with this protocol, examining the time to achieve correction, the time required for maintenance of the correction, and preliminary results of the longevity of the correction.

1. Material and methods

Since October 2003, our institution has adopted the Calgary Protocol to treat PC patients; this uses a lightweight patient-controlled adjustable chest brace made of aluminum with padded back support and a dense foam pressure pad (Fig. 1). With ethics board approval, all PC patients referred for evaluation at our chest wall clinic were prospectively asked to join an outcome monitoring study. At each visit, patient demographics, and the characteristics of the protrusion (Fig. 2) were recorded, including the extent of maximal protrusion (distance from the point of maximum protrusion to the estimated normal level of chest wall), craniocaudal length (craniocaudal length of protruding zone, measured through the point of maximal protrusion) and lateral length (length of protruding zone, again measured through the point of maximum protrusion in the transverse direction). Patient self-reported appearance satisfaction (Fig. 3) and exercise tolerance were also recorded. After recording of baseline data, patients were fitted with the adjustable tension brace (Fig. 4).

In August 2011, an interim survey was sent to all patients, in the varying phases of the bracing protocol, regarding the efficacy of improvement: maximum protrusion, craniocaudal length, and lateral length.
length and lateral length of the PC defect, as well as patient self-reported appearance, exercise tolerance, and compliance were also reported. Patients were either reviewed by telephone or at a return clinic visit.

2. Results

One hundred twenty-four PC patients were evaluated from October 2003 to April 2011 at the pediatric congenital chest wall clinic at the Alberta Children’s Hospital. There were 109 (88%) males and 15 (12%) females identified with a true carinatum defect, with a mean age of 14.4 years (range 7–18) and Tanner Stage of 3.6 at the time of evaluation. Patient characteristics are presented in Table 1.

After clinic review, 98 patients were prescribed bracing according to the protocol. The remaining patients refused bracing (4), were judged not mature enough to begin bracing (15), or not appropriate for bracing (7). Of the patients who started bracing, all consented to follow-up with the study protocol. At the time of review, August 2011, 10 (10%) patients were in the CP, 44 (45%) patients had completed the corrective phase (CP) and of these, 21 (21%) patients completed the entire treatment protocol. There were 44 (45%) patients deemed treatment failures. Of those, 28 (29%) patients were lost to follow-up during CP and an additional 14 (15%) were non-compliant. 2 (2%) patients did not achieve significant improvement despite 6 months of compliant bracing and went on to surgical correction. The relationship between skeletal maturity and time to achieve correction was examined; in patients who started bracing prior to attaining Tanner stage IV, the time to correction was $4.2 \pm 0.9$ months (Range: 1.6–5.4), while in those who achieved Tanner stage IV correction time was $8.0 \pm 7.1$ months (Range: 1.9–27.7) ($P < .02$ by Student’s t-test).

Of the 44 patients who completed CP, 21 completed the entire protocol and 23 are currently in MP. 1 patient developed recurrence during MP secondary to non-compliance but responded, with good correction once the patient resumed the night time sessions.

There was a significant improvement in both subjective measures and objective measures at different stages of the protocol. The protrusion, lateral measurement and craniocaudal measurement (objective measures) of the defect improved significantly in both the completed CP and the completed MP group (Table 2). Patient self-reported subjective chest appearance also improved significantly from $2.9 \pm 1.1$ out of 5 at the beginning of bracing protocol to $4.3 \pm 0.8$ out of 5.

<table>
<thead>
<tr>
<th>Scale</th>
<th>Patient’s Appearance Satisfaction</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>I am very disappointed and dissatisfied with the result</td>
</tr>
<tr>
<td>2</td>
<td>I am somewhat disappointed with the result; it does not look that great</td>
</tr>
<tr>
<td>3</td>
<td>The result is just OK</td>
</tr>
<tr>
<td>4</td>
<td>I am very pleased with the result and notice considerable improvement</td>
</tr>
<tr>
<td>5</td>
<td>I am completely satisfied with the result</td>
</tr>
</tbody>
</table>

Fig. 2 Objective characteristic measurements of pectus carinatum protrusion.

Fig. 3 Scale of patient self-reported satisfaction [21].
5 ($P < .02$) when completed CP and 4.3 ± 0.9 out of 5 ($P < .02$) at the end of MP respectively. There was no significant change in patient self reported exercise tolerance at any stage. Using September 1, 2011 as the reference date, the 21 patients who have completed therapy have been off bracing for 13.9 ± 16.0 months and there were no reports of recurrence at this final follow up evaluation, nor was there a change in the average chest wall protrusion (Table 3).

### 3. Discussion

Our data support the hypothesis that bracing is an effective treatment for pectus carinatum, improving both the objective protrusion and the appearance, as judged by the patient. Haje and Raymundo first reported non-operative bracing to correct a PC defect in 1979 [25]. Subsequent reports have demonstrated that compressive bracing to

![Fig. 4](https://example.com/fig4.png)

Calgary Protocol bracing therapy time line. The protocol timeline for using the brace is shown in Fig. 4: initially patients were instructed to wear the brace for 23 h daily (correction phase, CP). They were evaluated every 2–3 months during this phase. After the defect was corrected to the satisfaction of the surgeon and the patient, bracing was reduced to 8–12 h daily (typically overnight) until cessation of axial skeletal growth (maintenance phase, MP). Once the patient’s height was stable for 6 months the bracing was discontinued.

#### Table 1  Patient characteristics at evaluation, and at each phase of follow-up.

<table>
<thead>
<tr>
<th></th>
<th>Beginning of Bracing</th>
<th>Completed CP</th>
<th>Completed MP</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Gender:</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>90 (92%)</td>
<td>41 (93%)</td>
<td>21 (95%)</td>
</tr>
<tr>
<td>Female</td>
<td>8 (8%)</td>
<td>3 (7%)</td>
<td>1 (5%)</td>
</tr>
<tr>
<td><strong>Age (years):</strong></td>
<td>14.4 ± 1.9</td>
<td>14.3 ± 1.8</td>
<td>16.6 ± 1.8</td>
</tr>
<tr>
<td></td>
<td>[7.3–18.2]</td>
<td>[7.3–18.2]</td>
<td>[11.4–19.5]</td>
</tr>
<tr>
<td><strong>Tanner Stage:</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I</td>
<td>1 (1%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>II</td>
<td>1 (1%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>III</td>
<td>40 (41%)</td>
<td>14 (32%)</td>
<td>4 (19%)</td>
</tr>
<tr>
<td>IV</td>
<td>38 (39%)</td>
<td>19 (43%)</td>
<td>16 (76%)</td>
</tr>
<tr>
<td>NR</td>
<td>18 (18%)</td>
<td>11 (25%)</td>
<td>1 (5%)</td>
</tr>
</tbody>
</table>

Data: Mean ± Std. Dev. [range].

#### Table 2  Results of patients treated with bracing for PC.

<table>
<thead>
<tr>
<th></th>
<th>Beginning of Bracing</th>
<th>Completed CP</th>
<th>Completed MP</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>N:</strong></td>
<td>98</td>
<td>44</td>
<td>21</td>
</tr>
<tr>
<td><strong>Time to complete CP (months) [Range]:</strong></td>
<td>N/A</td>
<td>7.0 ± 7.3 [1.0–29.5]</td>
<td>N/A</td>
</tr>
<tr>
<td><strong>Time to complete MP (months) [Range]:</strong></td>
<td>N/A</td>
<td>N/A</td>
<td>17.9 ± 19.0 [4.4–87.5]</td>
</tr>
<tr>
<td><strong>Months since completion of protocol (to Sep 1, 2011) [Range]:</strong></td>
<td>N/A</td>
<td>N/A</td>
<td>13.9 ± 16.0 [0.3–71.4]</td>
</tr>
<tr>
<td><strong>Protrusion (cm):</strong></td>
<td>2.1 ± 1.0 [0.5–5]</td>
<td>0.5 ± 0.6$^<em>$ [0.5–0.7$^</em>$]</td>
<td>0.5 ± 0.7$^*$ [0–1.5]</td>
</tr>
<tr>
<td><strong>Lateral (cm):</strong></td>
<td>9.5 ± 3.5 [2.5–18]</td>
<td>4.1 ± 4.0$^<em>$ [3.2 ± 2.7$^</em>$]</td>
<td>3.2 ± 2.7$^*$ [0–10]</td>
</tr>
<tr>
<td><strong>Cranial–caudal (cm):</strong></td>
<td>11.4 ± 2.8 [5–24]</td>
<td>5.5 ± 4.7$^<em>$ [4.6 ± 3.9$^</em>$]</td>
<td>4.6 ± 3.9$^*$ [0–11]</td>
</tr>
<tr>
<td><strong>Chest appearance (out of 5):</strong></td>
<td>2.9 ± 1.1 [0–5]</td>
<td>4.3 ± 0.8$^<em>$ [4.3 ± 0.9$^</em>$]</td>
<td>4.3 ± 0.9$^*$ [2–5]</td>
</tr>
<tr>
<td><strong>Exercise tolerance (out of 5):</strong></td>
<td>4.4 ± 1.1 [0–5]</td>
<td>4.61 ± 1.0 [4.8 ± 0.4]</td>
<td>4.8 ± 0.4 [0–5]</td>
</tr>
<tr>
<td><strong>Compliance (out of 5):</strong></td>
<td>4.4 ± 1.1 [0–5]</td>
<td>4.3 ± 1.4 [4.3 ± 1.4]</td>
<td>4.3 ± 1.4 [0–5]</td>
</tr>
</tbody>
</table>

Data: Mean ± Std. Dev. [range].

$^*$ Student’s t-test, $P < .05$ when compared to beginning of bracing.

$^*$ Mann–Whitney U-test, $P < .02$ when compared to beginning of bracing.
correct a PC defect is effective, but the time to correction and the durability of the effect have not been well described [27,29,30]. Our current results provide insight into these factors, and validate the approach of using a staged correction and maintenance phase while skeletal growth is ongoing. The initial correction phase is aimed at redirecting the contour of the chest wall cartilages, and so a near continuous pressure (23 h/day), at the maximum force that the patient can tolerate is suggested. It is interesting to observe the response of the patients; if they are ambivalent to the protocol, once they begin to see results, which typically occurs in 3–4 weeks, they enthusiastically increase the force used, and their compliance. The present results suggest that this protocol results in an initial correction in 95% of compliant patients. Our findings also validate the notion that the younger, more malleable chest wall responds more rapidly; patients prior to reaching Tanner stage IV achieved correction in half the time of the more mature subjects. There is also a perception that the final appearance was improved with bracing beginning prior to Tanner stage IV; this will require more sophisticated evaluation methods to validate.

If an initial correction is achieved, this can be maintained through the remainder of the pubertal growth phase in almost all patients using bracing for 8 h per day. It was instructive to note that many patients independently described a worsening of their appearance during a growth spurt, especially if they had reduced their compliance. This underscores the importance of ongoing monitoring, with the treating team playing as both evaluators and supporters of the patient and their family. In the current protocol, bracing was only discontinued once axial growth stopped for a demonstrated 6 month period. The durability of the correction by bracing through to young adulthood has not been extensively reported; importantly, no patients reported recurrence after completing the maintenance phase. This description of phases in correction along with the expected timelines provides a new understanding of the process of bracing in PC patients.

In our initial study [27], the time to complete CP was reported to be 4.3 ± 2.1 months which is shorter than that was found in our current data (7.0 ± 7.3 months). We observed that compliance is one of the major issues with this particular age group. Bracing affects patient activity in and out of school; this can be particularly intrusive if they engage in sports. Although the self reported compliance of patients who completed CP is 4.4 ± 1.1 (out of 5), one common “practice” of patients and families reported was using the brace less than the time prescribed during day time, which appeared to result in lengthening of the correction phase. Although we found, as previously reported, that bracing achieves more rapid correction in younger patients at an earlier stage of puberty [28], we also found that in our population this age group was also more difficult to maintain compliance in. Further, these families were very resistant to the idea of a ‘locked in’ brace, preferring to allow the patient to control the tension of the device. Further direct study of the relationship between compliance, the tension of the brace, the rapidity of correction, and strategies to improve compliance is the subject of our ongoing work.

A limitation of this study was the lack of a control group, or randomization. This is a common problem in the evaluation of novel therapies, without a comparator treatment. In this study, most of the patients had mild to moderate defects without significant physical symptoms. Traditionally, this group of patients and families is left with a decision between radical corrective surgery and being left untreated. When given the option of non-operative adjustable compressive chest brace, it became the option of choice for almost all patients.

The current finding provides an interim report of the outcome following treatment of PC patients with an adjustable compressive chest brace. As more patients complete the protocol, we will be able to further analyze the long term effect of non-operative correction of a PC defect, including the relationships between compliance, skeletal maturity, activity, and the type of defect and results.

In conclusion, our data support that bracing is an effective non-operative alternative to improve self image in patients with PC. Bracing provides significant short to midterm subjective and objective improvement to patients’ PC. Our result also suggests that patients prior to achieving Tanner Stage IV at the beginning of treatment according to our protocol achieve correction in a significantly shorter period of time when compared to those who started treatment after attaining Tanner Stage IV.

References


Discussion

"Bracing is an Effective Nonoperative Therapy in Patients with Pectus Carinatum: An Interim Report of the Calgary Protocol.” Presented by Richy T. Lee, M.D., Calgary, Alberta, CANADA.

Discussant: Robert Shamberger, M.D., (Boston, MA):
Thank you, Dr. Lee, for an outstanding report. It is very encouraging to see your results.

You mentioned you had a picture of one child that seemed to have the chondromanubrial configuration. I am interested specifically if you had success dealing with those patients and also if you did an analysis between the children that had the symmetric versus the asymmetric carinatum protrusion. It has been my probably biased impression that the children that just had unilateral protrusion are harder to get repaired with the brace than those that have the symmetric defect.

Response: Dr. Lee: Thank you for the comment. We concur with your observation. From our study we did not actually do a formal analysis between the groups but we do find that for most patients who did not have success they were mostly nonsymmetric and also the manubrial type of defect.

Discussant: Rebecka Meyers, M.D., (Salt Lake City, UT): I really enjoyed that presentation. My first question was exactly Dr. Shamberger’s question. The asymmetric patients are the ones that really present a challenge with this therapy. My second question is what do you do with the children that are less than Tanner stage II that come in. Do you have them wait until Tanner stage III, or do you do them earlier and put them on longer maintenance therapy?

Response: Dr. Lee: We tend to wait until later on and observe. That is the reason we have 124 patients that we screened or evaluated in the initial clinic visit and we only have 98 patients prescribed the brace.

Discussant: Bryan Dicken, M.D., (Edmonton, Alberta, Canada): I use this same protocol and the same brace. I have been using vertical height as well as bone growth as an opportunity to determine the bracing length, and my orthopedic colleagues tell me that looking at wrist x-rays and pelvic x-rays can help with that determination. Have you looked at that?

Response: Dr. Lee: We have not looked at that. We also use the height and the weight of the patient. As soon as they reach a stable height and weight for about 4–6 months, then we determine that as the skeletal maturation.

Discussant: Michael Goretsky, M.D., (Norfolk, VA): I also enjoyed your talk. With the compliance issue, obviously
if they are compliant this is a great technique, no matter what modality. The question with compliance — was there a way to figure out why half were non-compliant? Was it the number of hours? We have found that even 12–16 h gets good success rate. The other thing is if it is an adjustable brace and they can monitor it for the discomfort with the pressure control, they seem to be more compliant because it does not hurt as much. We found even doing the brace in younger children 10–11 years of age, they get good results and a majority of them if you follow them will not get a recurrence, so I would not say there is an absolute age range for that.

Response: Dr. Lee: Thank you for your comments. We still are in the process to see how we can improve compliance of our patients. However, when talking to the parents who were involved in the bracing process, they comment that they treasure the fact that the brace is self-adjustable because the patient can actually adjust the tension of the brace according to their comfort level and that seems to help improve compliance a little bit.