



Bracing in pediatric patients with pectus carinatum is effective and improves quality of life

Sara Colozza, Andreana Bütter*

Division of Pediatric Surgery, Children's Hospital, The University of Western Ontario, London, Ontario, Canada, N6A 4 G5

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Abstract

Purpose: The purpose of this study was to examine the effectiveness and patient satisfaction with bracing for pectus carinatum (PC).

Materials and Methods: Twenty-five PC patients were treated between August 2007 and October 2011. Most patients were male (21/25,84%) with a mean age of 14.4 ± 2.0 yrs. A lightweight, patient controlled, external brace (Braceworks, Calgary, AB) was used. Monthly follow-up with anterior–posterior (AP) width measurements occurred until bracing was completed. Three quality of life (QOL) questionnaires were used: SF-36, SSQ, and PEEQ.

Results: Group 1 involved twenty patients who successfully completed bracing (12/25,56%) or who are still bracing (8/25,32%). Group 2 comprised five patients who failed bracing (2/25,8%) or who were noncompliant (3/25,12%). One patient who failed bracing underwent successful Ravitch repair. AP width decreased more in those with successful bracing (2.31 vs 0.64 cm, $p=0.05$). Questionnaires were completed by 19/25 (76%) patients. Pre-bracing, the SF-36, and PEEQ revealed that few patients were symptomatic, although most still avoided activities which showed their chest. The SSQ revealed that the majority of patients were very satisfied with their post-bracing appearance, experienced minimal discomfort while bracing, and would use the brace again. Self-esteem increased significantly after bracing (7.5 vs 8.7, $p=0.01$).

Conclusions: Bracing in PC patients is very effective in a compliant patient with close follow-up. Surgical repair remains feasible if bracing fails.

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Pectus carinatum (PC) is characterized by protrusion of the sternum resulting from overgrowth of the costal cartilages [1–3]. The overall prevalence of PC is 0.6%, with 80% of cases being male [1,3]. PC is usually considered a cosmetic deformity [4]. Despite its lack of physiological symptoms, PC tends to affect patients psychologically [3,5], instilling embarrassment due to the appearance of their chest

[6]. PC can be present since birth or may develop and worsen during adolescence [7]. PC patients become aware of their chest wall deformity and its lack of attractiveness. Most PC patients will avoid any activity in which their chest will be seen, e.g. swimming. Correction of their PC is speculated to improve self-esteem and confidence [8]. Until recently, the only methods of correction for PC patients involved aggressive surgical intervention in the form of an open or modified Ravitch or minimally invasive procedures [5,9,10]. Intraoperative risks, such as excessive bleeding and

* Corresponding author. Tel.: +1 519 685 8401; fax: +1 519 685 8421.
E-mail address: andreanabutter@gmail.com (A. Bütter).

pneumothorax, as well as postoperative complications, such as scarring, pain, and asphyxiating thoracic dystrophy can be significant [2,11].

Given that the chest wall is still compliant during adolescence, as demonstrated by Nuss bar correction of pectus excavatum [12], remodeling is possible using external pressure by a brace [3,9]. Non-surgical treatment of PC avoids operative intervention and its possible complications, is scarless, and is less costly than surgery [6,9]. Although compressive bracing has been found to yield highly satisfactory results in adolescents [1,5–7,10], the ideal treatment of PC remains controversial. In addition, no study has examined the psychological effects of bracing. The aim of this study was to examine the effectiveness and patient satisfaction of bracing to correct PC in pediatric patients.

1. Materials and methods

After obtaining IRB approval, the charts of all PC patients treated with a lightweight, patient-controlled, external brace (Braceworks, Calgary, AB) (Fig. 1) at the Children's Hospital in London, Ontario, between August 2007 and October 2011 were reviewed. With specific measurements taken by the surgeon and a tracing of the chest deformity performed, a custom-designed brace was created. The brace was adjusted accordingly to ensure a perfect fit. Patients wore a snug fitting undershirt under the brace at all times to prevent skin breakdown. Patients were then seen monthly for anterior–posterior (AP) width measurements and to ensure that there were no difficulties with the brace. We followed the Calgary protocol for the duration of the study [1]. Briefly, patients wear the brace for 23 h per day (correction phase) until the chest is flat (approx. 4–8 months). During the maintenance phase (approx. 4–8 months), patients brace for 8 h per day (usually at nighttime). Upon completion of



Fig. 1 Patient wearing the lightweight, external brace (picture courtesy of Braceworks, Calgary, AB). Black arrow points to one of the adjustable portions of the brace.

Table 1 Patient characteristics.

	N	Mean±SD
Duration of PC prior to diagnosis (yrs)	22	2.25 (0.5–13) ^a
Age at the start of bracing (yrs)	25	14.4±2.0
Length of correction phase (mos)	22	4.5±2.7
Length of maintenance phase (mos)	14	3.9±3.0
Total change in AP diameter (cm)	22	2.13±1.19

SD=standard deviation, AP=anterior–posterior.

^a Median value (range).

bracing, patients continue to be followed to ensure that their chest remains flat.

Variables analyzed included patient demographics and the total change in AP diameter. Three quality of life (QOL) questionnaires were used to assess patient satisfaction with bracing. Due to lack of PC QOL questionnaires, we slightly modified 3 existing ones: (a) SF-36 (health related QOL questionnaire) and (b) PEEQ (Pectus Excavatum Evaluation Questionnaire) and SSQ (Single Step Questionnaire) (both disease specific QOL questionnaires for pectus excavatum patients) (Tables 1, 2 and 3). These questionnaires have been previously validated [8,13,14]. All questionnaires were mailed to each patient with an explanatory letter as well as a letter of consent. For those patients who had not returned the questionnaire after 1 month, we contacted them by telephone. A second mailing was done if they were interested. A few patients preferred to complete the questionnaires by telephone.

All data were analyzed using SPSS software (version 11, Chicago, IL) and included the Student's t-test, paired t-test or non-parametric Mann–Whitney test where appropriate. Statistical significance was considered to be *p* less than 0.05.

2. Results

Of the twenty-five patients with PC in this study, 21 (84%) were male. The median duration of PC prior to diagnosis was 2.25 years (range 0.5–13 yrs) (Table 1). The mean age at the start of bracing was 14.4±2.0 years (range 11.3–17.4 yrs). By the end of the study, twenty-two patients (88%) had completed the correction phase of bracing. The average length of the correction phase was 4.5±2.7 months (range 1–13 months). Eight patients (32%) were in the maintenance phase. Of the 14 patients who completed bracing, the average maintenance phase length was 3.9±3.0 months (range 1–11 months). The median range of follow-up in clinic was 0.25 yrs (range 0.83). However, almost all patients were contacted recently by telephone. No recurrences were reported by the 14 patients who have completed bracing.

Three patients (12%) never returned to clinic after their 1st or 2nd followup visit and were deemed noncompliant. One of these patients braced for only 3 months yet his AP

Table 2 Modified Pectus Excavatum Evaluation Questionnaire (PEEQ).

Question stem	Scoring				Mean		P
	1	2	3	4	Group 1	Group 2	
1. Looks in general	V happy	M happy	M unhappy	V unhappy	1.6±0.5	2.3±1.2	0.08
2. How chest looks without shirt	V happy	M happy	M unhappy	V unhappy	2.1±0.8	2.7±0.6	0.24
3. Spending rest of life with chest as it looks now	V happy	M happy	M unhappy	V unhappy	1.9±0.9	2.7±1.5	0.26
4. Kids make fun of child because of chest	V often	Often	S	Never	4.0±0.0	3.7±0.6	0.02
5. Avoids doing things because of chest	V often	Often	S	Never	3.4±0.9	2.3±1.2	0.09
6. Hides chest	V often	Often	S	Never	3.3±0.9	1.7±0.6	0.008
7. Bothered because of the way chest looks	V often	Often	S	Never	3.3±0.7	2.3±1.2	0.06
8. Feels shy/self-conscious because of chest	V often	Often	S	Never	3.4±0.7	2.0±1.0	0.01
9. Feels bad about self	V often	Often	S	Never	3.8±0.4	3.3±0.6	0.11
10. Has trouble exercising	V often	Often	S	Never	3.8±0.8	4.0±0.0	0.67
11. Chest causes shortness of breath	V often	Often	S	Never	3.7±0.6	3.7±0.6	1.0
12. Chest causes child to be tired	V often	Often	S	Never	3.7±0.8	4.0±0.0	0.58

V=very, M=mostly, S=sometimes.

Group 1=successfully completed bracing/still bracing (n=16), group 2=failed bracing/noncompliant (n=3).

Total number of questions=12. This questionnaire was modified from the Child Pectus Excavatum Questionnaire by Lawson et al. [8].

width decreased by 2.5 cm. However, he stopped bracing shortly after, never returned to clinic and did not fill out the questionnaires.

Two of the 14 patients failed bracing. One had Marfan's syndrome with a severe PC and only started bracing at 16 years of age. His chest wall was already fairly rigid and his PC was the most severe of all patients in this series. Given his aortic root dilatation, we attempted bracing in an effort to avoid surgical intervention. However, his brace kept slipping off due to his severe sternal protrusion. After medical clearance by his cardiologist, he eventually underwent a modified Ravitch repair with excellent results. The other patient braced for 2 years although he only wore the brace

for a few hours each day, even during the correction phase. Consequently, he never achieved good correction of his PC. He was offered surgery but never returned to clinic.

AP width decreased more in patients who successfully completed or are still bracing compared to those who failed bracing (2.31 ± 1.17 cm vs 0.64 ± 0.90 cm, $p=0.05$) (Fig. 2). Of the 12 patients who successfully completed bracing, their AP width decreased significantly (2.22 ± 1.19 cm, $p<0.01$).

Satisfaction questionnaires were completed by nineteen (76%) patients. Patients were categorized into 2 groups. Group 1 consisted of patients who had successfully completed bracing or who were still bracing (n=20) while Group 2 patients had failed bracing or were noncompliant

Table 3 Select questions from the modified Single Step Questionnaire (SSQ).

Question stem	Scoring	Mean		P
		Group 1	Group 2	
3. Extent that chest interferes with pre-bracing social activity	Not at all=5, Slightly=4, Moderately=3, Quite a bit=2, Extremely=1	2.5±1.2	3.0±1.4	0.6
4. Extent that chest interferes with post-bracing activity	Not at all=5, Slightly=4, Moderately=3, Quite a bit=2, Extremely=1	4.5±0.8	2.5±2.1	0.01
5. Satisfaction with overall post-bracing appearance	ES=5, VS=4, S=3, D=2, VD=1	3.9±0.9	2.0±0.0	<0.01
8. Pre-bracing self-esteem	1–10	7.3±2.0	7.0±3.0	0.82
9. Post-bracing self-esteem	1–10	8.6±1.5	6.0±3.6	0.05
10. Pain during bracing	None=6, V mild=5, Mild=4, Moderate=3, Severe=2, V Severe=1	4.5±1.1	3.7±0.6	0.2
14. Overall satisfaction with final result	ES=5, VS=4, S=3, D=2, VD=1	4.1±0.8	2.0±0.0	<0.01
15. How different chest looks after bracing	Major improvement=5, improved=4, no change=3, worse=2, a lot worse=1	4.4±0.8	3.0±0.0	0.01
16. Going back, would you use the brace again?	Yes=3, un-sure=2, no=1	2.7±0.6	1.7±1.2	0.03

ES=extremely satisfied, VS=very satisfied, S=satisfied, D=dissatisfied, VD=very dissatisfied, V=very.

Group 1=successfully completed bracing/still bracing (n=16), group 2=failed bracing/noncompliant (n=3).

Total number of questions=16. This questionnaire was modified from the SSQ by Krasopoulos et al. [14]. Questionnaire assumes patients had completed bracing; if they were currently bracing, they were instructed to answer questions based on their current thoughts or feelings.

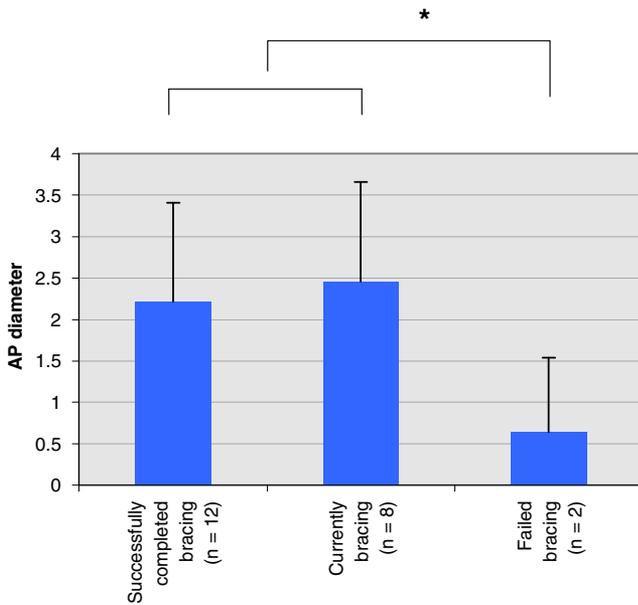


Fig. 2 Total change in anterior-posterior (AP) diameter with bracing. AP width decreased significantly more in patients who successfully completed or who are still bracing compared to those who failed bracing (* $p=0.05$).

($n=5$). Pre-bracing, the modified SF-36 revealed that few patients in either group were symptomatic from their PC. Post-bracing, the PEEQ showed that both groups remained asymptomatic (Table 2: Q10–12, $p=NS$). However, Group 1 patients only sometimes hid their chest while Group 2 patients often hid their chest (Table 2: Q6, $p<0.01$). Additionally, Group 1 patients sometimes or never felt shy or self-conscious about their chest while Group 2 patients often felt shy or self-conscious (Table 2: Q8, $p=0.01$). Pre-

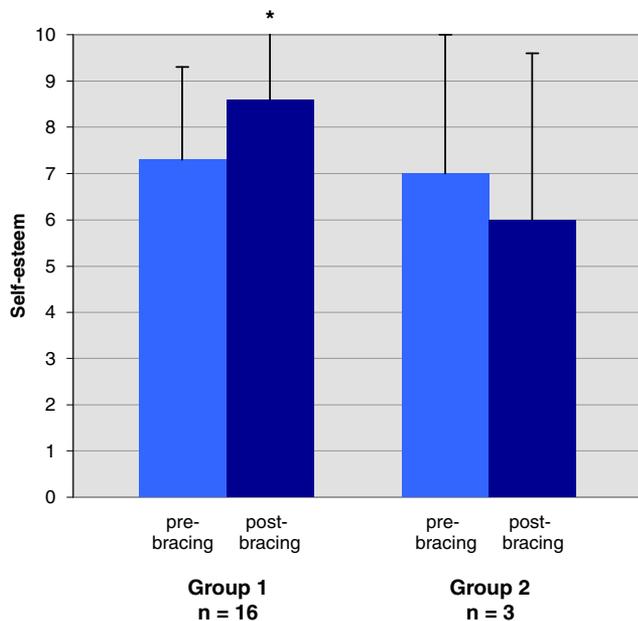


Fig. 3 Self-esteem before and after bracing. Self-esteem increased significantly after bracing in Group 1 (* $p<0.01$).

bracing, the SSQ found that social activity was moderately affected by chest appearance for both groups (Table 3: Q3, $p=0.6$). However, after bracing, Group 1 responded that chest appearance no longer affected social activity whereas Group 2 patients felt that their chest deformity continues to affect their social activities (Table 3: Q4, $p=0.01$). Group 1 patients noted significant improvement in chest appearance and were very satisfied while Group 2 patients remained dissatisfied (Table 3: Q5 and Q14, $p<0.01$). Interestingly, both groups experienced minimal discomfort while bracing (Table 3, Q10, $p=0.2$). Most patients in Group 1 would use the brace again whereas most patients in Group 2 would not (Table 3, Q16, $p=0.03$). Finally, self-esteem increased significantly after bracing for Group 1 (7.3 vs 8.6, $p=0.005$) whereas it remained similar in Group 2 (7.0 vs 6.0, $p=0.5$) (Fig. 3).

3. Discussion

Our data demonstrate that bracing is an effective means of correcting PC in a compliant patient. AP width decreased significantly in patients who successfully completed bracing. Until recently, operative correction of PC, whether by open or modified Ravitch repair or by newer minimally invasive techniques, has been the standard treatment [2,15–19]. Haje proposed nonoperative treatment of PC with a compressive brace [20]. However, this initial brace was large and awkward to use. The brace used in our series is lightweight and easily adjustable by patients (Braceworks, Calgary, Alberta). Despite the lack of prospective data and very few papers on the use of bracing for PC in children, a recent national survey by Emil et al. demonstrated that bracing is the preferred first-line treatment of PC by Canadian pediatric surgeons [21]. The majority of these surgeons judged their bracing results as good or excellent.

There is scant literature concerning patient satisfaction with bracing for PC. Kravarusic et al. employed a simple 5-point patient satisfaction scale and reported a baseline satisfaction of 2.3 (out of 5) with an increase to 4.7 after bracing [1]. Martinez-Ferro et al. employed a 10-point scale judging the final outcome of bracing, and 88.4% of patients reported a 7- to 10-point correction from bracing [9]. The survey by Emil et al. reported that 74% of Canadian pediatric surgeons felt that most or all patients braced were satisfied [21]. Recently, two papers explored psychological characteristics of PC patients prior to surgical correction but not prior to bracing. Steinmann et al. collected quality of life and body image information using questionnaires and rating of self-esteem for both pectus excavatum (PE) and PC patients [22]. PC patients reported highly impaired body image, decreased mental quality of life, and significant restrictions in social activities prior to surgery. PC patients were less satisfied with their appearance than PE patients [22]. Using an 11-point rating scale, Krille et al. found that pre-

operatively, PC patients reported a significantly greater self-perceived impairment in physical appearance than adults [23]. The scarcity of data concerning the psychological status of PC patients undergoing bracing led us to modify the validated SF-36, SSQ and PEEQ to create PC QOL questionnaires. We recognize that the SSQ and PEEQ were originally designed to assess PE. However, given that PC is also a chest wall deformity and affects young males, we felt that these questionnaires, rather than new, non-validated ones, were better assessment tools of PC quality of life pre- and post bracing. In our study, patients who successfully completed bracing, or who were still bracing, noted such a significant improvement in the appearance of their chest that their social activities were no longer affected. Post-bracing, they were no longer being teased because of their chest and were less likely to hide their chest. Consequently, their self-esteem increased significantly. On the other hand, patients who failed bracing or who were noncompliant did not notice any change in their chest appearance or in their self-esteem. Interestingly, both groups experienced minimal discomfort with the brace. In the failed/noncompliant group, minimal change in chest appearance was cited as the reason for discontinuing the brace. Both patients who failed bracing stated that they would use the brace again if the outcome was improved.

We recognize that our study has several limitations. Firstly, small numbers of patients were studied retrospectively with relatively short followup. Secondly, the separation of Group 1 and Group 2 patients may present selection bias. Although patients in both groups had moderate to severe PC, one of the 5 patients in Group 2 had the most severe PC of both groups. Thus, successful bracing might have been more difficult in Group 2. This may account for their insignificant AP width decrease and their dissatisfaction with bracing results, rather than the fact that they did not follow stringent bracing protocol.

In conclusion, bracing in PC patients is very effective in a compliant patient with close follow-up. We found that successful bracing significantly improves chest appearance and self-esteem of pediatric PC patients. Given its non-invasive nature, minimal discomfort and high patient satisfaction, we believe that bracing is an effective first line treatment in many PC patients. Surgical repair remains feasible if bracing fails [24].

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