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ORIGINAL ARTICLE

Pectus carinatum: the effects of orthotic bracing on pulmonary function and gradual compression on patient compliance

Oğuz Ateş, Osman Z. Karakuş*, Gülce Hakgüder, Mustafa Olguner and Feza M. Akgür

Department of Pediatric Surgery, Dokuz Eylül University, Medical School, Izmir, Turkey

* Corresponding author. Dokuz Eylül Üniversitesi, Tıp Fakültesi, Çocuk Cerrahisi Anabilim Dalı., 35340 Balçova, Izmir, Turkey. Tel: +90-232-4123002; fax: +90-232-2792101; e-mail: osman.karakus@deu.edu.tr (O.Z. Karakuş).

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Abstract

OBJECTIVES: The treatment of pectus carinatum (PC) deformity has been considered to be operative. Some authors have shown that postoperative pulmonary function is worsened. They have suggested that compromised chest wall expansion secondary to surgery leads to compromised pulmonary function. Several authors have advocated an orthotic brace for the treatment of PC. Pulmonary functions after orthotic brace treatment have not been investigated.

METHODS: Between April 2006 and October 2012, 61 patients presented with PC. Orthotic braces allowing gradual compression were prepared according to the anthropometric measurements of individual patients. The brace belt was tightened gradually. The brace was worn 6 h a day during the first week and the bracing time was prolonged for an additional hour per week till 16 h per day has been reached. Pre- and post-treatment echocardiography, pulmonary function tests and thorax computed tomography (CT) were obtained. The pectus severity index (Haller index) and the angle of sternal rotation were measured using CT. Satisfaction from bracing was evaluated by parents or patients at the end of the treatment.

RESULTS: While the mean pretreatment Haller index was 1.96 ± 0.24, the mean post-treatment index was 2.26 ± 0.32. The angle of rotation was improved by 47.5%. Forced vital capacity and forced expiratory volume in 1 second were correlated with the predicted values for age. There was no statistically significant difference between pre- and post-treatment values. No skin breakdown or bruising was encountered. The overall average satisfaction score was 3.92 ± 0.27.

CONCLUSIONS: We conclude that pulmonary function tests are not affected after brace treatment and gradual progression of bracing increases the patient's compliance.

Keywords: Pectus carinatum • Orthotic brace • Pulmonary functions

INTRODUCTION

The treatment of pectus carinatum (PC) deformity has been considered to be operative. Operative treatment of PC poses surgical risks, cosmetic problems such as residual scar formation, decrease in chest compliance due to resection of costal cartilages and incision of inspiratory muscles [1–5]. Derveaux *et al*. [6] have compared preoperative and postoperative pulmonary functions in pectus deformities. They have shown that postoperative pulmonary function is worsened while postoperative radiological contours of the thorax were corrected. They have suggested that compromised chest wall expansion secondary to surgery leads to compromised pulmonary function.

Several authors have advocated non-surgical approaches for the treatment of PC. In 1992, Haje and Bowen [7] have reported custom-fitted orthotic brace treatment for PC with favourable outcomes. However, pulmonary functions after orthotic brace treatment have not been investigated.

MATERIALS AND METHODS

Between April 2006 and October 2012, 61 patients presented with PC. Five patients had chondromanubrial PC and thus had rigid thoracic cage and 3 had severe scoliosis that prevented appropriate bracing. Therefore, these 8 were excluded. The remaining 53 patients with manually compressible PC showing flexibility of the chest wall (compression test) were judged as candidates for brace treatment. Age, gender, family history and pulmonary symptoms were recorded before brace treatment.

BRACING PROTOCOL

PC deformity presents as a cosmetic problem with associated psychological and social complications. When the deformity is severe, psychological and social problems are usually the dominant factors rather than respiratory symptoms, arrhythmias and

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exercise intolerance. Psychological and social complaints based on the patient's or the parent's perception have been accepted as main indications for brace treatment.

Custom-fitted orthotic braces were produced according to the anthropometric measurements of individual patients. The orthotic brace used in the current study comprised two pieces, one piece for the anterior chest wall and the other for the posterior chest wall. The brace was made from low-weight aluminium and covered with soft padding. Extra paddings were attached for the anterior protrusion and the posterior vertebral area. Two pieces were strapped together with leather belts (Fig. 1). Initially, the brace was applied snugly without compressing PC (neutral position), the leather belt was marked at this neutral position using marker pen (line on the right side of the belt in Fig. 2). Afterwards the leather belt was tightened till PC protrusion was flattened and belt position was marked at this position (line on the left side of the belt in Fig. 2). A third marking line was drawn in the middle of these two marking lines (Fig. 2).

During the first month of bracing, the belt was tightened using the middle marking line as the reference point. Afterwards the brace was tightened up to the third line. The brace was worn 6 h a day during the first week and the bracing time was prolonged for an additional hour per week till 16 h per day has been reached (Fig. 3A and B).

After correction of PC, the brace was worn 8 h a day till linear growth has ended (Fig. 4). The patients were evaluated with monthly intervals.

Satisfaction from bracing was evaluated by parents or patients at the end of the treatment according to a scale based on the resultant deformity. A score of 1 was defined when the deformity was judged as worsened, 2 when it was judged as being the same, 3 when judged as partial improvement and 4 when judged as total improvement.



Figure 1: A custom-made orthotic brace.



Figure 2: The belt markings showing neutral, initial correction pressure and full correction pressure positions.

Echocardiography, pulmonary function tests and thorax computed tomography (CT) were obtained at the beginning of the brace treatment, and they were repeated after completion of the brace treatment. Pulmonary function tests were evaluated according to the spirometric reference values for children as described by Zapletal *et al*. [8]. Forced vital capacity (FVC) and forced expiratory volume in 1 second (FEV1) values are expressed as percent predicted values for age according to Zapletal *et al.* The pectus severity index (Haller index) and the angle of sternal rotation were measured using CT (Fig. 5). The Haller index is described as the transverse inner diameter of the chest divided by the anteroposterior diameter between the sternum and the vertebral bodies. At the point of maximum sternal protrusion on CT, a line is drawn tangential to the posterior lamina of the rotated sternum (Fig. 5, line A). The angle between the horizontal axis of the thorax (Fig. 5, line B) and aforementioned retrosternal line was measured as sternal rotation angle.

Values were expressed as mean ± 1 SD. The results were analysed using the paired *t*-test.

RESULTS

There were 43 boys and 10 girls in the age range from 5 to 18 years (mean ± 1 SD = 12.3 ± 6). While 36 patients (26 boys and 10 girls) had a symmetric deformity, 17 were asymmetric. All the asymmetric cases were male. Family history for PC was positive in 11 patients with symmetrical deformity and in 4 with asymmetrical deformity. One female patient could not tolerate bracing and treatment was stopped. Before bracing, 8 patients had pulmonary symptoms such as frequent respiratory infections ($n = 4$), hyperpnoea ($n = 2$) and exercise intolerance ($n = 2$).

Echocardiography showed minimal mitral valve insufficiency ($n = 5$), mitral valve prolapse ($n = 4$) and juxtaductal shelf ($n = 1$). Echocardiography findings persisted after the brace treatment.

The mean distance between the first marking on the belt and the second marking was 1.9 (range 1.2–2.4) cm. While the mean pretreatment Haller index was 1.96 ± 0.24, the mean post-treatment index was 2.26 ± 0.32. While the mean pretreatment sternal rotation angle was 14.3 ± 5.4°, post-treatment rotation angle was 6.8 ± 0.9°. The angle of rotation was improved by 47.5% (Fig. 6).

Four patients were non-compliant during the pulmonary function tests and their data were excluded. Pulmonary function tests are summarized in Table 1. FVC and FEV1 were correlated with the predicted values for age. There was no statistically significant difference between pre- and post-treatment values.

The average duration of the brace treatment was 21.3 ± 3.4 months. The average follow-up period was 24.1 ± 13.5 months. None of the patients required analgesics during bracing. No skin breakdown or bruising was encountered, but a mild erythema was usually observed at the point of pressure on PC. Satisfaction was scored as 4 by 49 patients and 3 by 4 patients. Thus, the overall average satisfaction score was 3.92 ± 0.27.

DISCUSSION

Chest wall deformities have been traditionally repaired with operative procedures. Haje and Bowen [7] have defined orthotic

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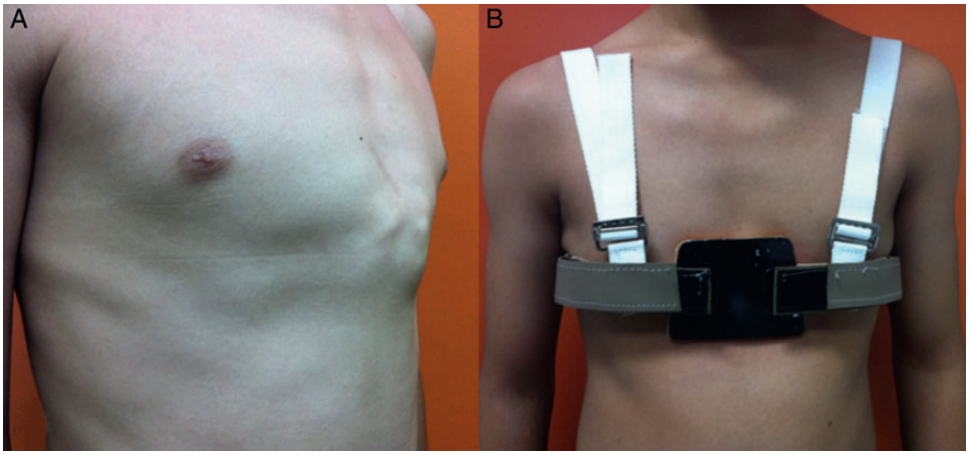


Figure 3: (**A**) A 12-year old boy with a symmetrical deformity, (**B**) a custom-fitted orthotic brace worn by the patient.



Figure 4: The appearance of the chest after treatment.

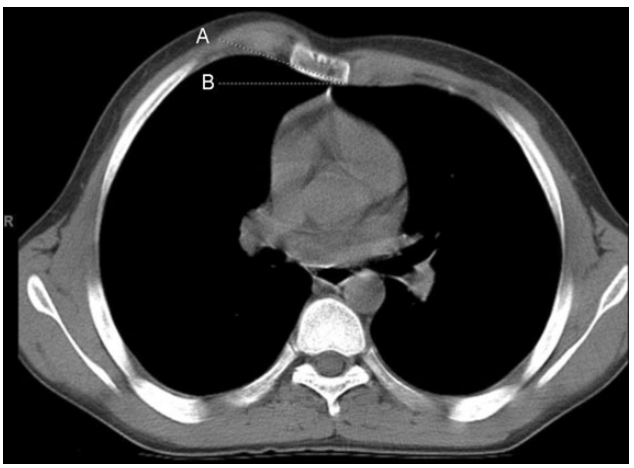


Figure 5: CT showing the angle of sternal rotation as measurement before treatment (the angle of sternal rotation is between **A** and **B**).

brace treatment for PC. Bracing has been gradually accepted by surgeons [9–15].

Two subtypes of PC exist, chondrogladiolar and chondromanubrial [16]. In chondrogladiolar PC, the chest cage is flexible and highly compliant, but in chondromanubrial PC, the chest is rigid and non-compliant. Unilateral overgrowth of the costal cartilages due to sternal rotation causes asymmetrical PC that is a variant of chondrogladiolar type (Fig. 7). While patients with chondrogladiolar PC are suitable for orthotic brace treatment, patients with chondromanubrial PC do not respond to orthotic brace treatment [7, 9]. Therefore, bracing was applied to chondrogladiolar PC cases in the present series.

Most authors think that cardiorespiratory function is less affected in patients with PC compared with pectus excavatum [1, 2]. However, some authors have observed shortness and tachypnoea, exercise intolerance and asthmatic symptoms in patients with PC, especially during the adolescent period [2]. They have suggested that PC leads to increased residual volume, decreased vital capacity and reduction in chest movements during respiration. They also think that chest flexibility in PC is decreased due to protrusion. Despite this restrictive defect, the majority of patients report improved exercise tolerance. Impairment of pulmonary functions after the surgical treatment of PC has been reported [1, 6]. Extensive surgical dissection of costal cartilages decreases chest flexibility by increasing chest rigidity due to fibrosis of costal cartilage [5]. Derveaux *et al*. have compared the preoperative and postoperative abnormalities in chest X-ray indices and in lung function in pectus deformities [6]. They have shown that pulmonary function in all deformities was deteriorated after surgery, although radiological parameters were improved. They have stated that resection of several costal cartilages, sternophrenolysis and incision of the parasternal muscles during surgery decrease chest wall compliance and diminish inspiratory muscle efficiency.

Bracing (∼2 years) limits chest expansion and compliance, and in consequence, lung expansion restriction may take place as effective pressure is applied to the chest, which was determined to be 3.7 psi average (range 0.4–9.5 psi) [10]. Our rationale for evaluating the effects of the bracing on pulmonary functions is based on this fact. In the present study, pretreatment FVC and FEV1 of the PC patients including those with pulmonary symptoms were concordant with age-predicted values. Pre- and post-treatment FVC and FEV1 were similar. Postoperative pulmonary functions are worsened after operative treatment [1, 6]. Present study has shown that pulmonary functions are not affected by non-operative treatment. Thus another advantage of non-operative treatment is that pulmonary functions remain unaffected after completion of bracing.

Major problems encountered during brace treatment are patient compliance and formation of skin lesions [9–14]. Martinez-Ferro

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Figure 6: CT showing improvement in the angle of sternal rotation.

Table 1: Lung functions in patients with PC deformity

	Pretreatment		Post-treatment	
	FVC	FEV 1	FVC	FEV 1
Patient values	2.97 ± 1.16	2.76 ± 0.99	2.95 ± 1.08	2.75 ± 0.92
Predicted values	3.48 ± 1.18	2.90 ± 0.95	3.46 ± 1.13	2.95 ± 0.92
% Predicted	85 ± 13	95 ± 14	85 ± 12	93 ± 8



Figure 7: Application of a custom-fitted orthotic brace to an asymmetrical deformity.

et al. have shown that patients cannot tolerate brace treatment and skin breakdown is encountered if the bracing pressure >2.5 psi [10]. Early in our experience, we realized that application of full correction brace pressure at the beginning of treatment leads to patient discomfort and thus to non-compliance. Similarly, skin lesions including hyperaemia, abrasion and breakdown may occur with application of full correction brace pressure at the beginning of treatment. Thus, brace treatment was started with low initial correction pressure for the first month of bracing, and bracing pressure was increased afterwards in the present study. On the other hand, daily bracing time was gradually increased in the present study. This approach increased patient compliance and prevented the occurrence of skin lesions. Brace treatment was not ceased in any of the patients excluding the single patient living in a hard-to-reach rural area in the present series.

In more than half of the children, protrusion was indeed noticed after 11 years of age. Deformity in the remainder of children emerges during early childhood. The possibility of recurrence is one of the reasons that surgical therapy in early childhood is not be recommended. Also, costal cartilage injury after surgical repair of PC is more frequent during early childhood compared with the adolescence and post-adolescence periods. There are no surgical risks and possibility of costal cartilage injury during brace treatment. Recurrence is a possibility after brace treatment; therefore, we have continued brace treatment until linear growth of the patient has been completed. In our series, there was no recurrence after follow-up in the adolescence and preadolescence periods. However, long-term results of bracing for PC are still unknown despite good short-term results. The present bracing protocol yielded successful results as evidenced by visual evaluation, Haller index and improved sternal rotation angle. Nevertheless, we think that long-term follow-up is necessary for detection of possible recurrence.

We conclude that pulmonary function tests are not affected after brace treatment and gradual progression of bracing increases the patient's compliance.

Conflict of interest: none declared.

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