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LOCALIZED PECTUS EXCAVATUM TREATED WITH BRACE AND EXERCISE: LONG TERM RESULTS OF A BRAZILIAN TECHNIQUE

TRATAMENTO DO PECTUS EXCAVATUM LOCALIZADO COM ÓRTESE E EXERCÍCIOS: RESULTADOS DE LONGO PRAZO DE UMA TÉCNICA BRASILEIRA

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ABSTRACT

Objective: Pectus excavatum is a deformity that affects aesthetics and causes emotional disorders. Surgical correction is well established, but conservative treatment is less common. We investigated the long-term results of using a brace and performing specific physical exercises to treat localized pectus excavatum, a type of deformity in which the depressed area is restricted to the midline region along the nipple line. Methods: We selected 115 patients (mean age 12.8 years), with a minimum follow-up of 36 months, who were evaluated more than one year after the end of treatment and skeletal maturity. Results were correlated with deformity flexibility, severity, regular use of the device, and performance of specific exercises. The chi-square (χ^2) and the Cochran-Mantel-Haenszel tests were used for statistical analysis. Results: Treatment was successful in 58% of patients, however, when exercises were performed and the brace was used regularly by patients with flexible deformities, the rate increased to 83% ($p = 0.005$). Severity and adherence to treatment greatly impacted successful treatment ($p = 0.009$ and < 0.001, respectively). Conclusion: The proposed treatment method was effective for correction or partial correction of the deformity in motivated patients followed up until skeletal maturity, especially when started early in milder and more flexible deformities. ***Level of Evidence V, Expert opinion.***

Keywords: Pectus Carinatum. Funnel Chest. Orthotic Devices. Exercise Therapy. Conservative Treatment.

RESUMO

Objetivo: O pectus excavatum é uma deformidade importante por comprometer a estética e causar distúrbios emocionais. A sua correção cirúrgica é bem estabelecida, mas o tratamento conservador é menos familiar. Investigamos os resultados de longo prazo do tratamento do pectus excavatum localizado (deformidade restrita a linha média e na linha mamilar) com uso de órtese e exercícios físicos específicos. Métodos: Selecionamos 115 pacientes (média de 12,8 anos) com seguimento mínimo de 36 meses, sendo avaliados mais de um ano após o término do tratamento e maturidade esquelética. Os resultados foram relacionados estatisticamente (qui-quadrado e Cochran-Mantel-Haenszel) com a flexibilidade da deformidade, a gravidade, o uso regular da órtese e a realização de exercícios específicos. Resultados: O tratamento foi bem-sucedido em 58% dos pacientes, mas quando o uso da órtese e os exercícios foram regulares em pacientes com deformidades flexíveis, essa taxa aumentou para 83% ($p = 0,005$). A gravidade e a adesão ao tratamento tiveram grande impacto no sucesso do tratamento ($p = 0,009$ e $<0,001$, respectivamente). Conclusão: O método de tratamento proposto foi eficaz para correção total ou parcial da deformidade em pacientes motivados acompanhados até a maturidade esquelética, principalmente quando a terapêutica foi iniciada precocemente em deformidades mais leves e flexíveis. ***Nível de Evidência V, Opinião do especialista.***

Descritores*: Pectus Carinatum. Tórax em Funil. Aparelhos Ortopédicos. Terapia por Exercício. Tratamento Conservador.*

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INTRODUCTION

Pectus excavatum (PE) and pectus carinatum (PC) account for 90% of all anterior chest wall deformities, with one case per 100-300 live births.[1] Both conditions can be treated with a brace that applies gradual compressive force on the anterior chest wall and via specific physical exercises. This type of treatment is more accepted for carinatum deformities.[2,3] However, conservative treatment of PE in which a brace applies compressive force below the depressed

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The study was conducted at the Centro Clínico Orthopectus.
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area along with specific exercises, is less common.[4,5] Previous reports have shown that flexibility and application of a standard treatment protocol are important factors to achieve good results.[3,6,7] There are no publications on PE treatment with braces and exercises apart from those by Haje et al.,[4-8] which describe short term results. Most of the musculoskeletal system deformities (like clubfoot, teeth deformities, scoliosis, pectus, etc.) can relapse after a short period or during growth when treated by conservative or surgical methods.[9-12] This study aimed to evaluate the long-term results after skeletal maturity of conservative treatment of localized PE, using a dynamic chest reshaping method that consists of the use of a custom-made bespoke brace[5] and specific exercises.

MATERIALS AND METHODS

The evaluation protocol was approved by the Institutional Ethics Committee (58417516.3.0000.5553). Written consent was obtained from all patients. In total, 6072 medical records of individuals suffering from pectus carinatum and pectus excavatum deformities were considered to select patients. All patients were treated according to a pre-established protocol, from 1977 to 2017, by the same group of physicians. All patients were supervised by one of the authors. The study only evaluated the treatment of localized PE patients with a depressed area restricted to the midline region along the nipple line.[7,8] Patients with the broad type of PE, in which the depression was located above and below the nipple line, were excluded, whether the nipple area was affected or not,[7,8] and their treatment results will be shown in another study.

Deformity severity was classified subjectively, based on aesthetics, by the first or second authors as mild, moderate, or severe, as shown in Figure 1. Flexibility was assessed by the first or second author, by manually compressing the lower rib cage and using the Valsalva maneuver, associated with upper limb adduction against resistance. Younger patients who were unable to execute the Valsalva maneuver were asked to blow a balloon, while a manual compression of the flaring ribs was performed. Each deformity was classified as "flexible" when it was completely reversed by maneuvers; "rigid" or "poorly flexible" when no or little reduction of the deformity occurred, respectively; or "moderately flexible" in intermediate situations.[5] For result analyses, the "flexible" and "moderately flexible" deformities were grouped, as the "rigid" and "poorly flexible" pectus.

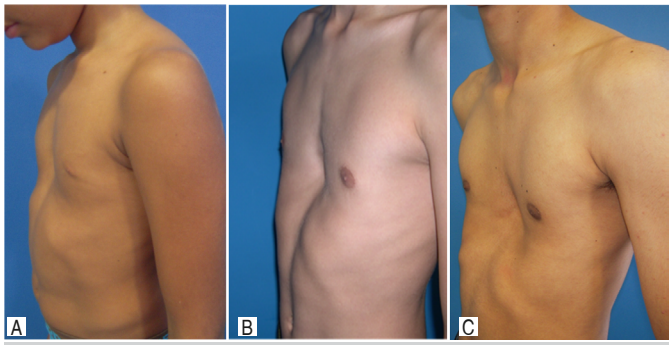

Figure 1. Classification of localized pectus excavatum severity according to esthetic compromise: A: mild; B: moderate; and C: severe.

The treatment indication criteria were patients between 2 and 19 years of age, with localized PE, and motivation to adhere to the treatment protocol by the patients and/or family members. Most deformities were moderate or severe, but some adolescent patients with mild deformities were also treated when it was important for the patient or the family. If the depression component of the localized pectus excavatum started worsening before puberty, treatment would begin in this age group. Adolescent patients with postural problems (like postural kyphosis) were encouraged to start treatment. Despite severity, treatment was indicated to all patients whose conditions were associated with mild scoliosis (less than 20° Cobb angle, measured in standing position). The treatment comprised of wearing a brace developed by Haje and Bowen[5] and specific exercises to strengthen trunk muscles. Figure 2 shows the patient selection flowchart. Only patients with follow-up periods longer than 36 months and at least one-year after they stopped growing were included (n = 115). Patients with iatrogenic deformities, associated with scoliosis greater than 20° or patients treated with a vacuum bell were excluded. Patients with deformities that did not bother them or cases where treatment was rejected by the family were also not included. The comparison group was composed of patients who remained untreated, although treatment was indicated, and who subsequently returned for a new evaluation.

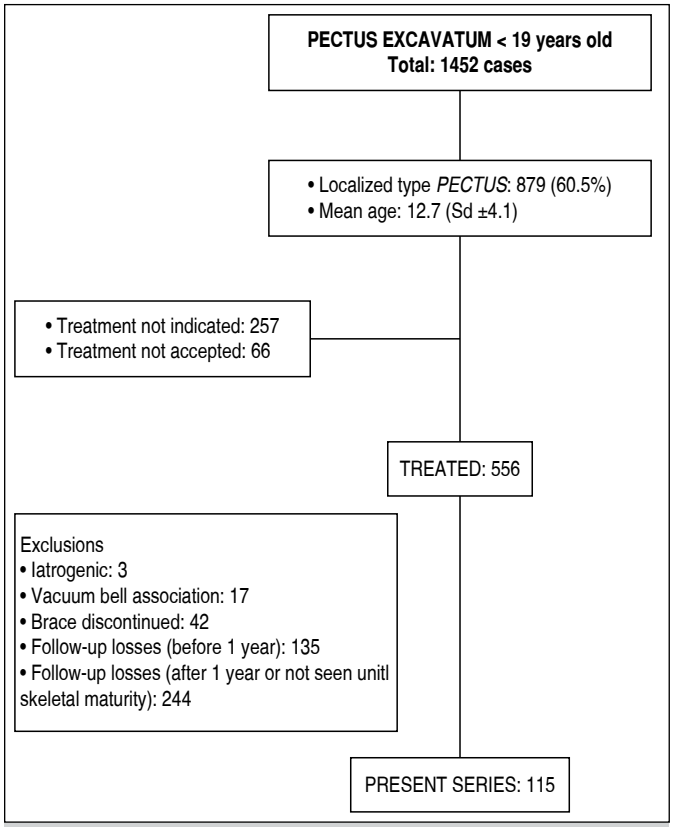

Figure 2. Flowchart of the treatment and monitoring until skeletal maturity of patients with localized pectus excavatum.

All patients were treated with the device shown in Figure 3. They were instructed to wear the brace until maximum correction for 23 hours a day, with a minimum of 18 hours. It could be removed for aquatic activities and contact sports. The patients themselves controlled the compression force at a comfortable level. This adjustment was discontinued only when the costal arch flare had disappeared. It was recommended that the specific exercises to strengthen the anterior chest wall muscles were performed while wearing the device, at least five times a week, maintaining maximum inspiration during concentric contraction. These exercises were initially taught by a physical therapist and adapted according to the patient's age and acceptance. The main exercises for older children and adolescents were the following: abduction and simultaneous counter-resistant extension of the upper limbs, trunk extension in the prone position, push-ups, sit-ups, crucifix, and blowing a balloon for 10 minutes.

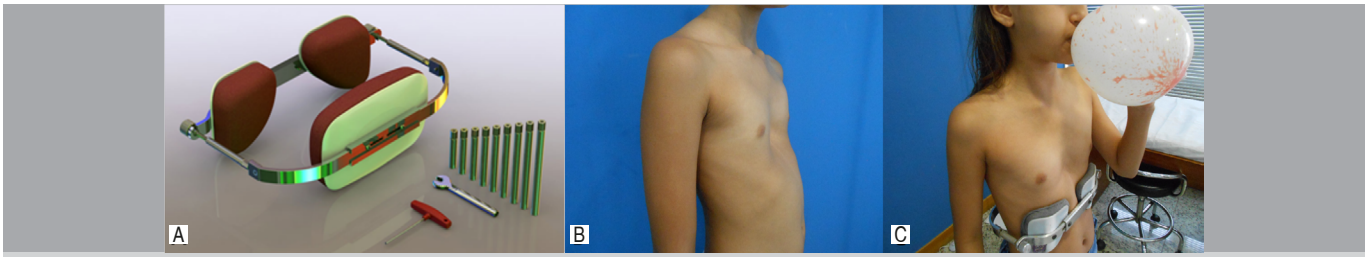

Figure 3. Illustration of the dynamic chest compression brace used for the treatment of all localized pectus excavatum cases. The brace is made up of two anterior padded plates resting on the lower costal margins and another padded plate on the dorsal region. A: compression between the anterior and posterior plates is controlled by the patient and it is applied by lateral screw threaded rods; B: in the picture, the patient with a moderate deformity performs an exercise to increase intrathoracic pressure by inflating a balloon; C: the pectus was classified as moderately "flexible".

After correction, patients were gradually weaned off the brace after completing at least 24 months of treatment. Treatment was discontinued after skeletal maturity, and additional follow-up was extended for at least another 12 months. Treatment adherence was classified as inadequate when the brace was not worn for the prescribed time or when the physical exercises were not performed regularly. Patients who stopped using the device before discharge were excluded, but those who discontinued the exercises before the recommended period were maintained in the final analysis, being classified as "irregular exercise adherence". Patients who used the brace for fewer hours than the indicated were classified as "irregular adhesion to the brace". Figure 3 shows the Dynamic Chest Compression Brace 2 (DCC 2) used for all patients, manufactured after receiving patient measurements and specifications from the doctor.

Results were considered "poor" when the deformity was aggravated or not corrected; "average" when it was under-corrected; and "good" when it was significantly improved (determined by one of the first two authors). Patient satisfaction with the treatment was defined as "satisfied" or "not satisfied". Successful treatments were defined as a "good" result and a "satisfied" patient. Photos of all patients were taken before and after treatment, and the clinical pictures were used for evaluation.

The Statistical Package for the Social Sciences (SPSS) software v. 22.0 (IBM, USA) was used for statistical analysis. The level of significance was set at $p < 0.05$. The chi-square (χ^2) test was used to verify associations between categorical variables. The Cochran-Mantel-Haenszel test was used to verify if the odds ratio between two variables remained the same for the categories of a third variable.

RESULTS

The mean age at the beginning of treatment was 12.8 years (SD = 4.0; median 13.7 years, variation 5.4 to 17.2; 24% were less than 10 years old, n = 28). Figure 4 shows age histogram of treated patients, with a mean follow-up duration of 23.9 months (S.D = 7.6; median 24.7) after the end of the treatment. Of the treated patients, 85 (74%) were male and 30 (26%) were female.

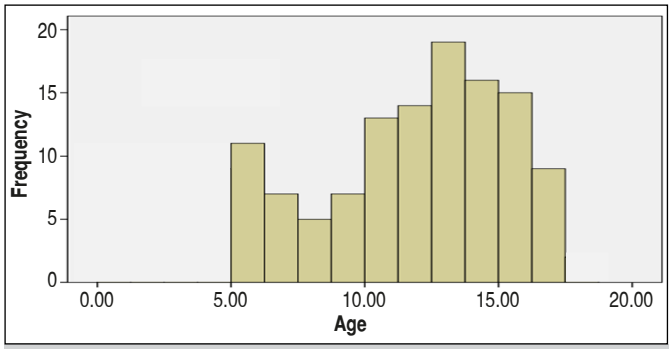

Figure 4. Age histogram of treated patients.

Thirty-four patients (30%) showed mild deformity, 67 (58%) showed moderate deformity, and 14 (12%) showed severe deformity. The deformity was very flexible or moderately flexible in 64 patients (56%) and rigid or poorly flexible in 51 patients (44%). The results were considered good in 66 patients (58%), average in 36 (31%), and poor in 13 (11%). Patients were satisfied with their treatment in all cases with good results, in 25%(n = 9) of average outcomes, and in none with poor results.

Good results were obtained in 57% (n = 20) of patients with mild deformities, 61% (n = 41) with moderate deformities, and 36% (n = 5) with severe deformities. In terms of flexibility, the results were good in 55% (n = 35) of patients with flexible or moderately flexible deformities and in 61% (n = 31) of patients with rigid or poorly flexible deformities (Figure 5).

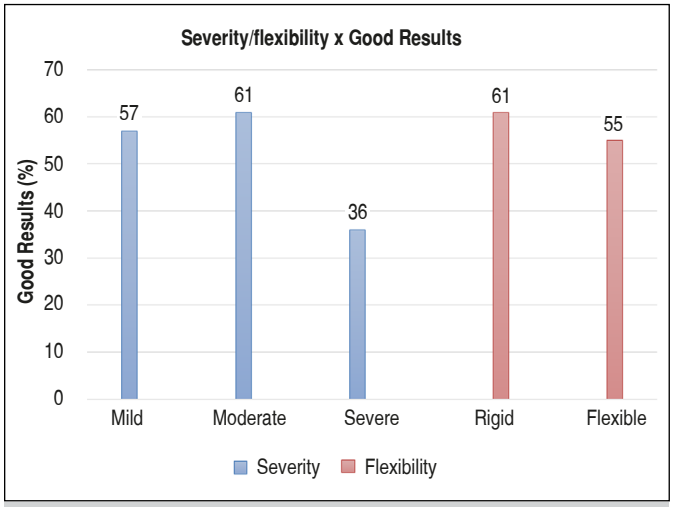

Figure 5. Results according to severity and flexibility.

Brace use was regular in 63% (n = 72) and irregular in 37% (n = 43). The specific exercises were regular in 49% (n = 57) and irregular in 51% (n = 58).

When flexibility was analyzed alone, no statistical difference between the results (p = 0.27) was observed, but when analyzed together with the regular use of braces, a significantly positive response to treatment was observed, with 83% of patients showing good results (p = 0.005). On the other hand, when the use of the brace was irregular, even with regular exercise performance, only 38% of the results were good (p = 0.167). In patients with regular orthosis use and exercise performance, good results were observed in 62% of patients (p < 0.001) (Figure 6). Table 1 shows the comparisons made. Figures 7 illustrates the final treatment results.

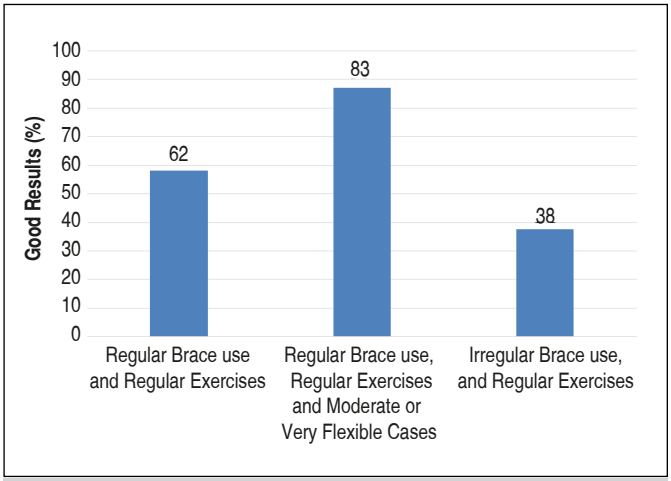

Figure 6. Results according to treatment regularity (brace use and specific exercises) associated or not with chest flexibility.

Table 1. Comparisons and interpretation of results.

Comparison	χ^2 value / p / odds ratio and 95%CI	Association between variables
Good results vs. pectus severity	17.09 / p = 0.009 / 3.8 (1.2–15.4)	negative
Good results vs. pectus flexibility	3.95 / p = 0.27/ 0.90 (0.38–2.13)	no correlation
Good results vs. regular brace use, and flexibility	23.84 / p = 0.005 / 6.45 (1.85–9.45)	positive
Good results vs. regular brace use	15.39 / p < 0.001 / 8.67 (2.74–27.4)	positive
Good results vs. regular physical exercises	7.40 / p = 0.007 / 4.05 (1.45–11.34)	positive
Good results vs. regular brace use and regular physical exercises	14.90 / p < 0.001 / 14.17 (3.23–62.11)	positive
Good results vs. irregular brace use and regular physical exercises	5.09 / p = 0.167/ 0.91 (0.27–1.28)	no correlation

Complications were not significant, but skin rashes, superficial skin injuries, and transient hyperpigmentation appeared in pressure areas (Figure 7B) in approximately 5% of the patients. In 15% of patients, discomfort or transient pain was experienced in the areas supported by braces. This was resolved by partially releasing the pressure. These complications did not cause discontinuance of device use. In three patients, there was mild overcorrection, which was managed by reducing the device use time or starting a second brace to treat the iatrogenic pectus carinatum (n = 2) (Figure 8). There were no relapses for those who completed follow-up.

The main reasons of the 66 patients who did not accept treatment were the following: surgery was selected as the primary treatment; psychological issues; or socioeconomic conditions. Twenty-eight returned in a mean follow-up of 18 months. The deformity had worsened in 20 patients (an example is in Figure 9), while it remained stable in eight. Among those who had worsened deformities, all showed moderate or accentuated deformities, which were very or moderately flexible in 11 (55%) and rigid in 9 (45%) in the last evaluation (at approximately 14.6 years of age).

Figure 7. Localized pectus excavatum patients treated with braces and exercises with good final long-term results. A: 13-year-old male patient, showing moderate severity and flexibility of the deformity at the beginning of treatment; B: with transient hyperpigmentation in pressure areas and overcorrection of flaring ribs; and C: final result at the age of 17; D: 10-year-old female patient with good adherence to treatment; E: Correction was achieved after 12 months of treatment; F: Final correction at 15 years and 5 months of age; G: 5-year-old female patient before treatment; H: Complete correction showed when she was 8 years old; and I: 12 years old.

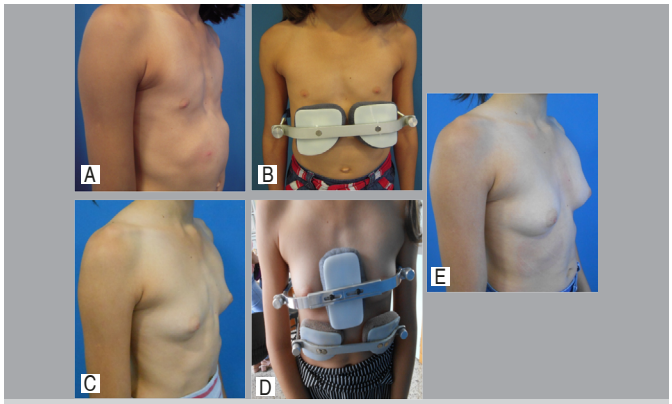

Figure 8. A: 9-year-old female patient with mild localized pectus excavatum and good adherence to the DCC 2 device and exercises; B: first treatment day with the DCC 2 device; C: overcorrection started after 2 years of follow-up and became more evident after 3 years of follow-up, when she demonstrated aesthetic complaints; D: a second DCC 1 device was associated to treat the mild reactive pectus carinatum; E: a good result was observed when she was 14 years old and discharged from using the brace.

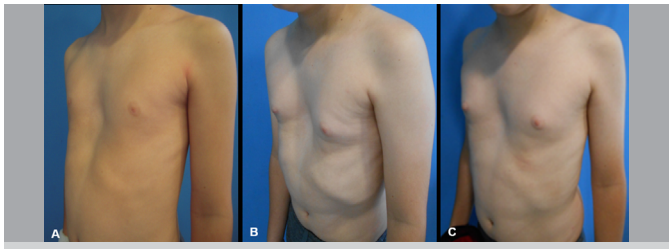

Figure 9. A: 8-year-old male patient with a moderate deformity whose family refused the indicated treatment; B: two years later, the deformity had worsened, and treatment was initiated; C: after nine months, there was an improvement with regular treatment.

DISCUSSION

Our method of treating localized PE using a brace and exercises was effective for the correction or partial correction of the deformity in motivated patients. It had no recurrence after finishing treatment in patients who continued treatment until skeletal maturity. Moreover, it was a low-risk treatment and that offers the possibility of discontinuance in the event of intolerance.

In terms of treatment, surgical correction of the pectus is quite effective.[10] However, with surgery, complications may be devastating and may occur even when patients are in the hands of experienced surgeons.[13] Other less invasive options include procedures that fill the cavity with substances, such as silicone.[14]

There are reports on the use of a device called a vacuum bell (Eckart Klobe, Germany) that applies negative pressure to the depressed area of the PE, resulting in complete improvement in 15 to 31% of patients.[15,16] The method does not reposition the ribs efficiently or correct asymmetrical pectus. When vacuum bell was combined with a braces, additional corrective improvements were observed.[4,17]

The non-invasive alternative treatment for pectus using an orthotic device and thoracic muscle exercises was initially developed by Haje and Raymundo[3] to treat pectus carinatum, and, later, a new device to treat PE was created.[5] In carinatum patients, the principle of correction is quite intuitive, as it applies pressure to the deformity apex. However, in excavatum patients, the action on the sternum and the chondrosternal joints results from the pressure applied to the anterior costal arches at a distance. Furthermore, we believe that this pressure results in a mechanical effect that acts from the mediastinum to the surface, promoting regional thoracic expansion, changing the forces that act on several growth plates, which contributes to reshaping (Wolff's law). These ideas are supported by Wong and Carter, who reported that mechanical forces on the sternum can influence skeletal morphogenesis.[18] Another mechanism that may play a role is the diaphragm, whereby the lowering of the last ribs starts to affect the chest expansion mechanism more efficiently. The secondary actions of the brace may contribute to improved posture. The performance of specific and repetitive exercises, with the patient maintaining inspiration at its maximum volume during muscle contraction, works actively to correct the depressed area. Data from the literature[5,10] show that pectus deformity is progressive. The patients in this study who initially refused treatment and subsequently returned reinforce this idea. Nuss et al.[10] do not recommend surgery in very young patients due to the risk of relapse. In our experience, the conservative method can be used at very early ages, but it should be maintained until skeletal maturity to avoid relapses.

Previous studies reporting results on brace treatment for pectus excavatum or pectus carinatum had short follow-ups, and we believe that Moon et al.[19] overstated when they claimed that their study reported long-term results, as their follow-up period lasted only 13 months. The need for a long treatment period may lead to treatment discontinuation and can ultimately be regarded as a shortcoming of the non-surgical methods of PE treatment.

One limitation of this study is that subjective criteria were used to evaluate the initial flexibility and severity. This is common in publications because imaging methods mentioned in the literature pose limitations. There is no previous description of an objective flexibility measurement for pectus excavatum. Using a CT before and after treatment implies radiation concerns, it does not alter the pectus treatment methodology when using braces and exercises, and there is no correlation between CT indexes and clinical aspect. More recently, an imaging evaluation technique using a structured-light 3D scanner has been introduced to perform more accurate evaluations;[20] this technique was not available when our patients were treated, and nowadays it is still expensive for our center. Also, we believe that it is enough to compare results using photos taken in the same position before and after treatment.

Our results clearly show an association between brace -use and physical exercise and positive results. This information is important to consider when advising families. Treatment regularity was quite important, since most patients who completed the protocol showed good results. Furthermore, when flexibility was analyzed together with regular treatment, our results improved. Haje et al.[4] described flexibility as a key prognostic factor in the treatment of pectus deformities using braces and specific exercises.

The most frequent complication in this study was skin irritation. Wearing a t-shirt under the brace and adequate body hygiene,[4,5] device pressure, size of the anterior and posterior padded plates, and brace width are crucial to minimize discomfort and skin problems. The overcorrection reported in our study, and in previous studies,[4,6] certifies the effectiveness of the treatment method for correction of the chest depressed area.

One expected result was that more severe deformities were related to worse results, which corroborates the idea that early treatment is crucial when using braces and specific exercises.

CONCLUSION

The non-invasive method for correcting PE resulted in positive outcomes in the patients that were adherent to the treatment protocol and continued the treatment until skeletal maturity, especially in flexible and mild deformities.

AUTHORS' CONTRIBUTIONS: Each author contributed individually and significantly to the development of this article. DPH: conceived and planned the activities that led to the study, participated in the review process, performed the treatment together with Sydney Haje from 1999 to July 22nd, 2011, and by himself since this last date, interpreted the results of the study and approved the final version; SAH (*in memorian*): performed the treatment from 1988 to July 22nd, 2011 (3 days before he passed away); JBV: manuscript preparation, participated in the review process and approved the final version; ACOS: data collection and approved the final version; LFBL: data collection and approved the final version; WH data collection and approved the final version.

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