Journal of Pediatric Surgery 55 (2020) 1347–1350

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Other Conditions

Initial reduction of flexible pectus carinatum with outpatient manipulation as an adjunct to external compressive bracing: technique and early outcomes at 12 weeks

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ARTICLE INFO

Article history:
Received 15 March 2019
Received in revised form 10 June 2019
Accepted 1 September 2019

Key words:
Pectus carinatum (PC)
Haller index (HI)
External compressive bracing (ECB)
Manipulation

ABSTRACT

Introduction: Our aim was to assess whether initial reduction with outpatient soft-tissue manipulation of flexible pectus carinatum deformity prior to external compressive bracing was associated with improved compliance and patient satisfaction compared to reported outcomes of external brace with progressive tightening.
Materials and methods: From our observational cohort of 227 patients, 177 were felt appropriate to undergo initial reduction and soft tissue manipulation prior to immediate custom fitting of an external compressive brace. These patients then followed a prescriptive schedule of 12 weeks of continuous external bracing with subsequent follow-up in clinic.
Results: The reduction in Haller Index was maintained throughout the period of external bracing without the need for progressive tightening of the external brace. The treatment was associated with high levels of patient satisfaction and high patient concordance compared to other protocols. There were no major complications and minor complications included only skin irritation.
Conclusions: Out-patient initial reduction with manipulation prior to external compressive bracing is a novel technique which resulted in excellent concordance and high rates of patient satisfaction and should be considered as an adjunct to standard external bracing techniques.
Type of Study: Treatment Study.
Level of Evidence: Level II.

Pectus carinatum (PC) is a common chest wall deformity presenting in childhood with progression during growth periods. Abnormal cartilage growth causes outwards protrusion of the anterior chest wall which is often referred to as 'pigeon chest'. Historically, surgery in the form of an open operation to resect the deformed cartilage and realign the sternum with an osteotomy was the mainstay of treatment (modified Ravitch procedure). However, this was associated with significant operative risk as well as post-operative pain, wound infection, pneumothoraces and scarring [1].

Compressive external bracing is increasingly considered as a first-line treatment for flexible PC due to the growing body of evidence demonstrating safety and efficacy [2]. There have been few complications reported with bracing. The main limitations therefore are the tolerability and compliance of the treatment in young patients allied with the need to brace for a prolonged period. A recent literature review of external compressive bracing highlights the high levels of treatment abandonment and low levels of successful bracing (Hunt et al., 2018 submitted).

Much of the current literature mentions relatively high rates of poor compliance and failure with complete external bracing [3,4]. This is likely due to pain relating to progressive tightening of the brace allied with the protracted duration of bracing and loss of motivation secondary to the patient's perceived failure to improve their deformity. Compliance with bracing, as with any medical intervention, is therefore critical in order to achieve a successful outcome.

Here, we present a novel method of initial reduction of PC through out-patient soft tissue manipulation prior to custom fitting of an external compressive brace. This technique avoids the need for significant adjustments during the bracing period, including routine tightening or pressure monitoring. In addition, it allows for a more prescriptive wearing schedule with fewer follow-up appointments as brace alterations and adjustments are rarely required.

1. Materials and methods

We conducted a retrospective review of 192 consecutive patients presenting to our centre between 1 January 2015 and 8 February 2018. Four patients were not considered candidates for bracing due to stiff, inflexible deformities. A further 5 patients chose not to wear the

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https://doi.org/10.1016/j.jpedsurg.2019.09.024
0022-3468/

brace and opted for other treatments and 6 patients were lost to follow-up (4 of whom lived abroad). The remaining 177 patients were all included in this analysis with no other exclusion criteria. Ethical approval was obtained accordingly. Statistical analysis was performed using STATA 12.0.

Data collected included the patient's age, height, weight, chest wall measurements, associated features, associated symptoms and medical history. The measurements taken to describe the morphology of the deformity are described elsewhere and included an anteroposterior/lateral chest wall calculation (Pectus Index) (Hunt et al., SCTS abstract presentation, Glasgow 2018). All measurements were repeated at each follow-up appointment and patients were assessed for any complications of bracing. Information around appropriate wearing of the brace, including a wearing schedule was provided and patients were encouraged to follow the schedule.

Prior to starting and during the bracing programme, a patient satisfaction questionnaire was completed. This survey asked patients to report their compliance with bracing, any associated symptoms and the psychological impact of their deformity. Patient's responses were either descriptive or rated on a scale of 1 to 10.

1.1. Procedure

Once consent had been obtained and the deformity examined and measured including photographic records, a custom brace was manufactured on-site and an initial reduction with soft tissue manipulation was performed during the patient's consultation. The patient was placed in a supine position and the flexibility of the PC assessed. Since April 2018 this includes an additional pressure measurement (in Pressure per Square inch, PSI) to correct deformity (POC). This has been employed for all patients attending clinic for assessment of external compressive bracing. In a similar series of patients, pressures of correction measurements (PSI) are taken at regular intervals during the bracing treatment in order to accurately ascertain compliance. Initial results suggest a PSI > 14 is unsuitable for manipulation.

Once assessed, approximately 1 g (equals 3.5 cm coverage) to 5 g based on age and severity (and according to manufacturer's instructions) of topical local anesthetic cream (EMLA® (lidocaine/prilocaine) cream 5%) was applied to the skin over the deformity and covered with plastic wrap (cling film) to aid speed of absorption. Thereafter, the anterior chest is warmed using a heating pad and gentle pressure applied to the deformity. After 10 min, an infrared vibrating massager (Beurer®) was applied for a further 10 min with gentle 'massage' over the cartilaginous component of the deformity.

Entonox® (BOC Healthcare) a medical gas mixture consisting of 50% nitrous oxide and 50% oxygen was offered and self-administered as required, this was carried out according to the manufacturer's instructions regarding procedural analgesia. This technique is usually employed in all situations where analgesia and sedation with rapid onset and offset is required and is particularly popular in children. The majority of patients did not require Entonox®. We found that by allowing more time during the warming phase pre-manipulation and by performing the manipulation in a slower and steadier manner over a longer time period, the analgesia requirements were much lower thus precluding the need for Entonox in most cases. No immediate complications were noted, or side effects reported.

The initial reduction and manipulation was performed by a thoracic surgeon and supported by a specially trained physiotherapist and pediatric nurse (if necessary). The anterior chest wall and specifically the cartilaginous component are reduced using a well described physical therapy technique of manual manipulation or 'soft tissue release'. This is similar to deep tissue massage and the manipulation involved 'probing', 'shearing' and 'rolling' techniques to help break down fibers [5]. Gentle sustained pressure was applied directly to the abnormally elongated and protruding costal cartilage to allow 'mobilization' of the pliable cartilage into a more natural alignment. The key feature of this procedure was controlled and consistent movements with progressive and targeted pressure over the peak of the pectus deformity for around 10 to 15 min. This mobilized the deformity into a flatter anatomical or near-anatomical position. Care was taken not to apply pressure directly over the ribs or sternum but only to the flexible costal cartilage.

Through consistent and maintained pressure of the pectus deformity and once a desirable position had been achieved, the flattened cartilages were held in place while the custom-made brace was fitted. It was essential that the reduced deformity was held in place while the brace was applied and tightened enough to maintain the reduction. No additional pressure was applied or required. The technique on average took 15 to 20 min to achieve reduction or near reduction of the deformity. POCs were recorded in a small cohort of patients, at two time points; initially and immediately following the reduction (since April 2018).

Following immediate bracing, the patient was advised not to tighten the brace which simply maintains the reduction. However, they were required to wear the brace for 5 days continuously; a wearing schedule was prescribed thereafter. Anti-inflammatory drugs were prescribed as required, along with advice on sleeping positions and suitable levels of activity in the short and long term. The patient attended clinic 24–48 h later for clinical review. During the study period, and in response to the success of the initial reduction and manipulation adjunct to external compression bracing, the program was altered with the brace weaning schedule reduced by 2 weeks. The current wearing schedule is described in Fig. 1.

2. Results

The average age of patients undergoing initial reduction with manipulation and external bracing was 15.04 +/− 3.07 years (range 8.3 to 30.5). The population group was 91% Caucasian (n = 157) and 95% male (n = 165). Many patients had a family history of pectus deformity and Marfans syndrome and several patients had co-morbidities including neurofibromatosis and cystic fibrosis.

All 177 patients included in this analysis underwent successful reduction of their deformity at the initial consultation without immediate complication. One hundred seventy-three patients had attended 12 weeks of follow-up (mean 11.5 weeks; range 5–40 weeks). The remaining four patients are included in the results below under 'failed bracing'.

During the bracing period, the average number of hours of self-reported bracing wearing was 22.4 h per day (median 24; range 0–24). Of the 46 patients who were bracing for less than 24 h per day, 17 patients attended their first follow-up after 12 weeks, meaning that they had already entered the weaning phase and were bracing according to the above protocol. Two patients had overcorrected and were advised to reduce the number of hours spent bracing per day. The remaining 27 patients reflect a recent change in the protocol whereby younger patients with flexible deformities were given tailored programmes of reduced bracing (i.e. 16 h on during the day, 8 h off overnight) or had started weaning due to early correction of the deformity. Overall, the compliance rate with the recommended hours of bracing was 100% on self-reporting (see Table 1).

The measurements performed at the initial consultation enabled comparison of the morphology of the deformity throughout the period of bracing. The apex of the pectus deformity was measured with calipers and the average height from the patient's back to the highest point of the deformity was 21.6 cm (range 15.5–33.0 cm) prior to manipulation and external bracing. At 12 weeks, the apex of the deformity was on average 18.1 cm (range 14.0–28.0). The average reduction in the apex was 3.5 cm (range 0.0–12.0 cm) following initial reduction and manipulation and 12 weeks of bracing which was statistically significant ($p < 0.0001$). The average POC recorded prior to manipulation was 9.43PSI (n = 39). In 20 patients from this subset, the POC was also measured post initial manipulation, average POC was 9.02 kg, the matched pre-manipulation POC was 9.2 kg ($p = 0.796$).

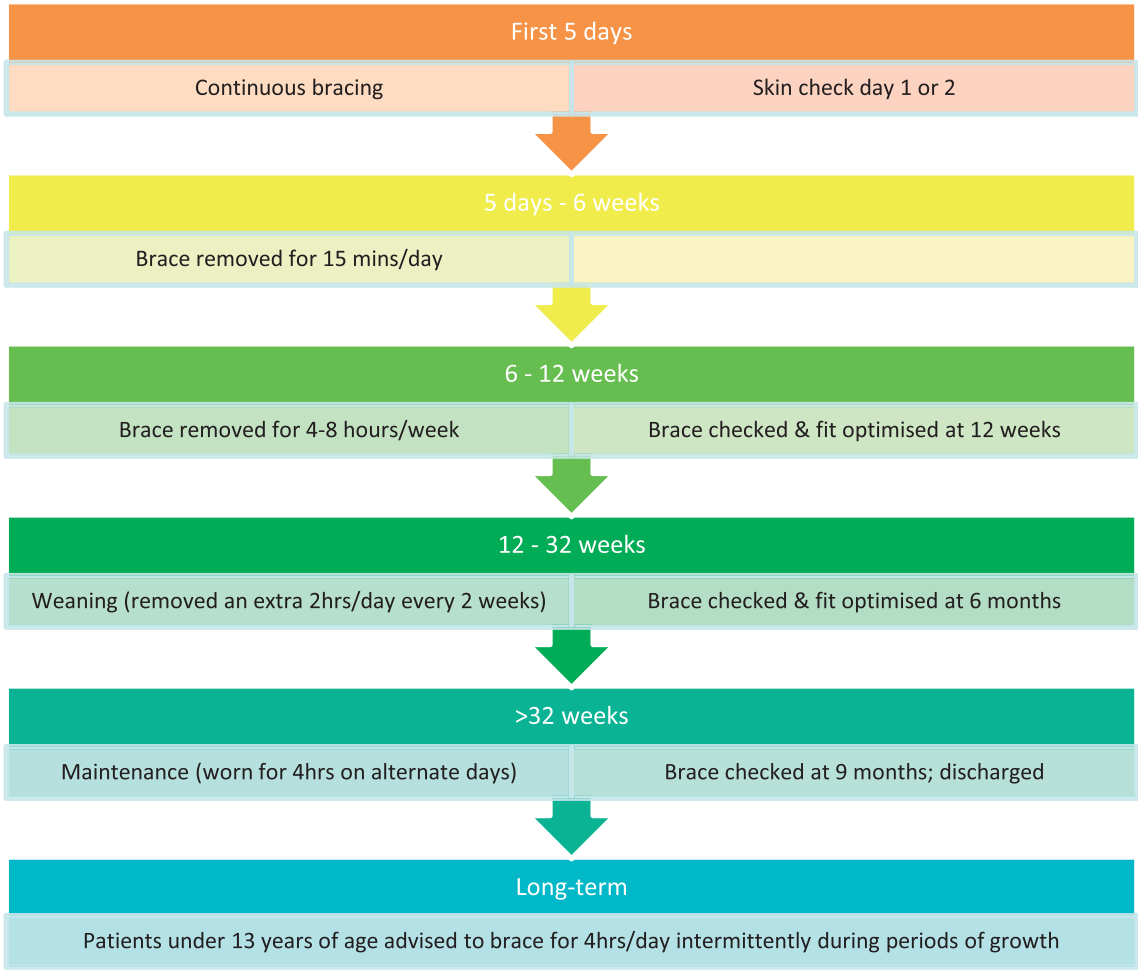

Fig. 1. Bracing schedule.

Patients self-reported feeling that the results were as good as they hoped for with an average score of 8 out of 10 (median 9; range 1–10). 88% of patients stated that they would strongly recommend this technique (n = 152) while only 2% (n = 3) said that they would not recommend the procedure in the future with 10% undecided (n = 18).

A minority of patients experienced shortness of breath on exertion or pain relating to their deformity both prior to bracing and while wearing their brace. Prior to bracing, 14% (n = 25) reported significant breathlessness as assessed by a score of >5/10 compared to 29% (n = 48) following initial reduction with manipulation and external bracing. There was a small but statistically significant increase in the score for breathlessness from 2.7 to 3.9 ($p < 0.0001$; 95% CI -1.64 to −0.59). This compared to a non-statistically significant reduction in pain scores from an average of 2.5–2.4 following initial reduction with manipulation and external bracing ($p = 0.6532$; 95% CI −0.29 to 0.46). Of those that described pain following treatment, this was largely described as mild discomfort, with no patients reporting severe pain or scoring >7 out of 10 on their questionnaire. The reporting of pain or breathing difficulty did not lead any patients to stop wearing their brace at 12 weeks and this was therefore not a negative predictor of compliance.

Table 1
Patient compliance.

Number of weeks	n	Mean number of hours worn per day	S.D.	Min	Max
0–6	22	23.5	1.74	16	24
6–12	78	23.1	3.30	0	24
12–24	67	22.0	3.79	8	24
>24	6	13.3	7.97	2	24

2.1. Complications

Skin complications were encountered in 39 patients. The majority of these (n = 38) were mild to moderate skin reactions which required application of either a simple water-based emollient or a zinc-oxide based barrier cream. This was applied in the first 12 weeks and all cases had resolved by 24 weeks. Only one severe case of skin irritation was noted, which was subsequently confirmed as ring worm following dermatology review. This required treatment, but the patient continued bracing and the symptoms resolved. There were no other complications or mechanical failures relating to the external brace or the manipulation technique.

2.2. Failed bracing

Of the four patients (2.2%; n = 177) who failed to complete the above protocol for bracing, three patients chose to stop wearing their brace, and one patient developed severe skin ulceration 5 days after initial reduction with manipulation and external bracing. Bracing was terminated to allow the skin to heal without subsequent complication.

3. Discussion

Initial reduction of the PC using the deep tissue massage of manipulation, is a well described technique in sports physiotherapy involving a graduated deep massage, which has been applied to the chest,

combined with sustained pressure to the deformed costal cartilages, followed by immediate application of the custom brace to the reduced deformity. The technique was uniquely employed as an adjunct to compressive external bracing for flexible PC deformities and remained constant throughout the study period. It is the first time such a technique has been described in relation to pectus deformities.

We found that the most important factor following reduction of the deformity with controlled pressure was holding the deformity in place while the brace was applied followed by 5 days of continuous bracing without a break. This maintained the reduction for an adequate period of time to reduce the likelihood of recurrence and once active external bracing began, avoided need for constant tightening. With regards to the clinic set-up, the most useful adjuncts were warming pads, infrared massager and topical local anesthetic. Although inhaled analgesia (Entonox) was offered during all initial reduction and manipulation cases, this was required less frequently with adequate preparation including warming and topical anesthetic and as the senior author became more experienced with the technique. This reduced the likelihood of anesthetic-related complications such as lightheadedness and did not increase the related pain scores.

Patient selection changed throughout our practice. As a result of the skin complication mentioned above which required termination of bracing, older patients over the age of 18 years of age with stiff deformities are now offered traditional progressive tightening of their brace without initial reduction with manipulation. In later cases, where there was evidence of skeletal maturity, the patients were additionally assessed by the pressure gauge measurements described elsewhere [6]. By avoiding initial reduction and manipulation, there was reduced tension at the peak of the deformity thereby reducing the pressure on the overlying skin. The external brace was then worn typically for 16–20 h for approximately 6 months with progressive tightening over the time period followed by a period of progressive weaning over a further period of 6–12 months.

Patients with stiff deformities which were not felt to be amenable to external compressive bracing regardless of the bracing schedule (typically older patients with a PSI > 14) were counseled appropriately and not offered external bracing.

For young patients under 11 years of age with flexible deformities, a shortened protocol for external bracing has been subsequently adopted. Once correction of the deformity has been achieved, active intense wear in the first 12 weeks may not be necessary, and typically the brace would be worn for 16 h, with 8 h off (usually coinciding with the school day), and an early weaning period This is caveated by a longer period of sporadic external bracing for maintenance during period of growth typically where the brace is only worn 6–8 h every other day or less.

Early involvement of physiotherapists for education and treatment of associated musculoskeletal abnormalities is also important in optimizing results and now forms part of our initial assessment. This is consistent with units outside of the UK where teams including surgeons and therapists assess the patients to determine the best course of treatment [7].

Regarding the technique as a whole, there were no significant complications apart for skin discomfort. The patient questionnaire highlighted a statistically significant increase in the level of self-reported breathlessness, however, by 9 months; this was no longer statistically significant. It also did not correlate with concordance with bracing or the patient's perception of physical fitness as ascertained in their questionnaire. The low level of morbidity described is consistent with the APSA's assessment of traditional compressive bracing as a safe technique in a pediatric population.

Pre-bracing, in young patients with PC, there are high levels of anxiety and depression. As such, there is a need for prompt treatment, counseling and education of this condition. With compliance being such a vital cornerstone to the success of this safe and effective therapy; a fast-tracked protocol such as the one we have described provides a less intensive and more flexible brace schedule which suits the patient population and is thus likely to yield a better quality of life and reduced burden of morbidity.

Overall, there were high levels of patient concordance with initial reduction with manipulation with external compressive bracing and a low failure rate (2.2%). This compares very favorably with previously published reports which documented longer periods of bracing and attrition rates of up to 50% [3,4]. We postulate that compared to other protocols, immediate correction of the deformity through initial reduction motivates the patient to comply with the treatment, improving the end results. This correlates with other research which suggested that the greater the immediate correction with dynamic external bracing, the more likely the patient was to continue with bracing [8]. Early correction may also reduce the more protracted discomfort typically associated with traditional techniques of progressive brace tightening, thereby improving concordance. In this study, low levels of pain were reported with no patients experiencing severe pain during the 12 weeks of brace treatment despite the near continuous wear.

3.1. Conclusions

Initial reduction with soft tissue manipulation of flexible PC as an adjunct to external compressive bracing was associated with excellent short outcomes at 12 weeks with high patient concordance, high patient reported satisfaction and low complication rates, when compared to other more traditional bracing programmes.

Acknowledgments

We acknowledge the invaluable assistance of Pectus Services LLC in developing and applying this technique in clinical practice.

Funding statement

This research received no specific funding.

Conflict of interest statement

All authors disclose no conflict of interest.

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