Healthcare Improvement Scotland | SHTG Advice on health technologies

SHTG Assessment
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In response to an enquiry from the Scottish National Chest Wall Service

Chest wall bracing for children and young people with pectus carinatum

What were we asked to look at?

The Scottish Health Technologies Group (SHTG) was asked to evaluate the clinical effectiveness, cost effectiveness, safety and patient experience of orthotic bracing in people who have pectus carinatum (sometimes known as pigeon chest).

Why is this important?

The primary concern associated with pectus carinatum often relates to its physical appearance, and the potential impact of this on a child or young person's quality of life and mental wellbeing. Less commonly, physical health issues have been reported in the literature.

The Scottish National Chest Wall Service, based in the Royal Hospital for Children Glasgow, offers conservative treatment for children and adolescents (up to 16 years old) in Scotland with pectus carinatum. They requested this work to inform treatment plans and enhance patient care, and to inform National Services Division (NSD) who fund the service.

What was our approach?

A comprehensive literature search was conducted to evaluate clinical effectiveness, cost effectiveness, patient issues and safety of chest wall bracing for people with pectus carinatum. All outcomes reported in the literature were included. A draft of this SHTG Assessment was peer reviewed by topic experts.

What next?

This work will be shared with colleagues at the Scottish National Chest Wall Service and NSD.

Key findings

- The evidence base for chest wall bracing in people with pectus carinatum consists largely of lower-level observational studies (case series). These studies consistently report that chest wall bracing in people with pectus carinatum is a safe, and clinically effective alternative to surgery, which is acceptable to most patients. Some people may still require surgery if, for example, chest wall bracing fails or is not appropriate in practice.
- The evidence base is heterogeneous, particularly with regard to the bracing protocol used (including wear time and duration of treatment), the selection of patients and how clinical outcomes are measured.
- People with pectus carinatum may have reduced self-esteem and body image. Successful treatment is associated with improved patient outcomes such as body image and quality of life.
- Accurate selection of patients based on age, pressure of initial correction and motivation appears to be important. High attrition rates were reported in several studies, where the treatment was abandoned for reasons including skin issues, discomfort, lack of motivation, slow/no improvement in the pectus carinatum and lengthy treatment durations. Protocols may be improved by addressing pain reduction, skin problems and discomfort. Family support was also associated with improved adherence. Rapid or immediate improvements in the pectus carinatum may increase patient motivation to continue with treatment.
- The existing literature suggests that patients are suitable for dynamic chest wall bracing if the pressure of initial correction does not exceed 7.5 to 10 pounds per square inch (PSI). The literature also suggests that, to avoid skin ulcerations, the pressure of treatment should not exceed 2.5 to 3 PSI.

Contents

Literature search

A systematic search of the secondary literature was carried out between 20–25 October 2021 to identify systematic reviews, health technology assessments and other evidence based reports. Medline, Medline in process, Embase and the Cochrane Database of Systematic Reviews were also searched for systematic reviews and meta-analyses. No date restrictions were applied.

The primary literature was systematically searched between 20–25 October 2021 using the following databases: Medline, Medline in process and Embase.

Results were limited to English language.

Key websites were searched for guidelines, policy documents, clinical summaries, economic studies and ongoing trials.

Concepts used in all searches included: pectus carinatum, funnel chest, pigeon chest, orthotic compression bracing, chest compression, chest bracing, orthotic bracing. A full list of resources searched and terms used is available on request.

Research question

The main research question was:

- What is the clinical effectiveness, cost effectiveness, safety and patient experience of orthotic bracing for pectus carinatum?

The following supplementary questions were also considered:

- What is the prevalence of pectus carinatum in the general population aged 0 to 16 years and is there any deprivation effect?
- What is the evidence on wear time, including the use of technology for recording/assessing wear time in pectus carinatum chest wall bracing?
- What is the evidence around patient adherence to recommended brace wear time?
- What is the evidence on pressure for initial correction (PIC) and pressure of treatment (POT) used to guide initial patient selection and treatment while in brace?

Introduction

Pectus carinatum is characterised by an anterior protrusion of the sternum and the adjacent costal cartilages. There are two subtypes:

- The chondrogladiolar subtype is most common (92–95 % of cases), and involves a protrusion of the body of the sternum (gladiolus).[1]
- The chondromanubrial subtype involves a protrusion of the superior part of the sternum (manubrium) and can occur as an isolated defect or combined with a gladiolar depression (pectus excavatum).[1]

Pectus carinatum normally occurs following a growth spurt, typically in children between 9 and 14 years old, although, it can occur in younger children. It is caused by abnormal growth of the cartilage ribs that attach to the sternum (breast bone).[2]

The primary concerns with pectus carinatum are usually related to the physical appearance of the chest wall, and the potential impact this may have on a child or young person's mental wellbeing and quality of life.[1, 2] Pectus carinatum may also present with physical symptoms such as exercise intolerance, chest pain, chest wall tenderness, shortness of breath, palpitations or wheezing.[3] It may also make wearing a bra more difficult for some people.[4]

Compressive orthotic bracing is a time sensitive therapy. A chest wall becomes more rigid (less compliant) with age, and the optimal age for bracing is between 10 and 15 years. Compressive orthotic bracing is the first line therapy for most children with a compliant pectus carinatum, but may be less effective in patients with a non-compliant chest wall or asymmetry.[1] Conservative treatment following ossification of the costal cartilages (which occurs with age) is not possible, leaving surgery as the only treatment option for correction.[1]

The Scottish National Chest Wall Service, based in the Royal Hospital for Children Glasgow, offers conservative (non-surgical) treatment for children and adolescents (up to 16 years old) in Scotland with pectus carinatum.

In line with the current published evidence, The Scottish National Chest Wall Service offers three treatment options, listed below:

1. For the mildest cases, physiotherapy assessment and advice, aimed at improving posture, core strength and breathing are usually adequate.
2. Most people will be offered dynamic bracing.
3. Surgery is normally reserved for people who are severely affected and typically follows an unsuccessful trial of bracing.[2]

Epidemiology

The reported prevalence of pectus carinatum ranges from 0.1 % to approximately 1 %.[2, 5, 6] It affects males about four times more frequently than females.[6] The cause is unknown, but

25 % of patients have a family history, and it is a common feature in people with Marfan syndrome or Noonan syndrome.[1] Scoliosis is an associated condition in about 10 % of people with pectus carinatum.[2]

Health technology description

The pectus carinatum orthoses used in NHSScotland are typically manufactured in carbon fibre.[2] Custom-designed aluminium braces are also described in the literature.

Bracing protocols vary. Daily use generally ranges from 8 to 24 hours per day, although, longer wear times (≥20 hours) are more commonly reported. Some protocols start with high daily use (up to 23 hours per day) until the pectus carinatum resolves, followed by a maintenance phase (for example overnight use only). Duration of treatment generally ranges from 6 months until completion of linear growth, although, shorter durations of treatment are described in the literature.[1]

Patients in NHSScotland are generally advised to wear the braces for between 16 and 20 hours a day.[2] Total wear time (until treatment completion) varies, and will be individual to each child or young person.

The braces described in the literature appear to be either:

- standard orthoses that do not allow measurement of pressure over the thoracic wall, or
- dynamic compression braces that allow measurement and adjustment of the brace's pressure over the thoracic wall, leading to a controlled correction.

The type of brace being used was not always clear in the studies reviewed. In addition, the definition of 'dynamic' bracing varies and includes the use of braces that do not have a pressure measurement system, but allow for the pressure to be adjusted. The evidence relating to any type of chest bracing (and bracing protocol) are included in this assessment.

In NHSScotland, patients are offered customised braces that are fitted to the shape of their chest. The braces are described as 'dynamic', although, they do not have a pressure measurement system.

Clinical effectiveness

A description of the quality and quantity of the evidence

The main evidence on clinical effectiveness comes from two systematic reviews.[6, 7] Both report favourable conclusions for chest wall bracing in young people with pectus carinatum, but also note the need for more high quality research.

Hunt and Patel (2020) published a moderate quality systematic review on external bracing in patients with flexible (compliant) pectus carinatum.[7] Their literature search was limited, and the included studies were all case series. Their results are presented as a narrative summary. The authors acknowledge and discuss the limitations of the included evidence and their conclusions are reasonable based on the evidence reviewed.

Hunt and Patel stated that they included 16 case series, although, 17 are described within the main text. The included case series had a study period of between 4 and 24 months. The authors do not report the total number of patients included in the studies, but note that ten studies included less than 100 patients, and two included more than 250. In most of the studies, treatment commenced in adolescents around 12 to 14 years of age, and the median age of the included patients was 14 years (range 5–28 years). The authors do not report the percentage of males and females in the studies. The assessments varied between the studies and included clinical outcomes that were subjective, objective or a combination of the two.

The systematic review by de Beer et al. (2018) included a more extensive literature search, and had a more focused scope on studies measuring dynamic compression.[6] It included eight studies (encompassing 1,185 patients), seven of which were included in the Hunt and Patel review. The median age of the included patients was 14 years (range 2–28 years) and 87 % were male. The mean study follow-up period was 16 months. This systematic review was well-conducted and, like Hunt and Patel, the authors acknowledged that the evidence base tends to consist of lower-level observational studies. The authors' conclusions are reasonable based on the evidence presented.

This assessment identified seventeen additional studies. These were mostly retrospective observational studies which had collected data prospectively (n=14), generally describing a series of patients treated at a particular centre (case series). Two were small randomised controlled trials and one was a small pilot study.

Bracing protocols

Systematic reviews

The systematic review by Hunt and Patel stated that all of the included studies (n=17) described the bracing protocol they used.[7] In five studies, patients were advised to wear the brace 'as often as possible' or for most of the day (wear times used ranged between 20 and 23 hours per day). Four of the studies included in the review reported shorter wear times (ranging between 8 and 12 hours per day). One study described starting bracing slowly (for 6 hours per day), increasing it by 1 hour every week until reaching a wear time of 16 hours per day.

The remaining seven studies described a protocol that included the custom fitting of an external orthotic brace, a period of active treatment, followed by a period of reduced or maintenance wear. The active treatment phase tended to consist of continuous or near-continuous wear for a period 2 to 8 months (although, one study described an active treatment phase of only 2 weeks). Maintenance wear times varied again, but were mostly between 8 and 12 hours per day, for a period of 3 to 6 months (or until the patient reached skeletal maturation).

Most studies included in the review reported some form of ongoing assessment and brace tightening over the bracing period with five of the 16 studies using a pressure assessment to guide degree of tightening (as described in the next section 'Dynamic compression').

The review authors reported that treatment failure (including non-adherence) rates varied between very low to as high as 43 % in one study. Complications were generally low (<10 %), and mainly involved skin issues. Reasons for treatment abandonment included skin issues, discomfort, lack of motivation, persistent sternal protrusion and rib flaring (when the lower portion at the front of the rib cage protrudes forwards and out). Some patients were also lost to follow up.

The review authors concluded that:

> Compressive external bracing appears to be a safe and well tolerated nonsurgical treatment option for young patients with flexible pectus carinatum deformities. However, there is still a need for robust level I randomised data from multiple centers with a clearly standardized bracing protocol, objective measurement of outcomes, and recording of results at the end of the bracing treatment program in sufficiently powered sample sizes over a significant follow-up period.

Additional studies

The search for this assessment identified nine additional studies (in ten publications) which evaluated various chest wall bracing protocols.

Five did not add significantly to the conclusions of the Hunt and Patel review, and so have not been described in detail in this assessment. Four of the five were retrospective observational studies describing bracing protocols similar to those already discussed.[8-11]

Two were from South Korea (n=119 and 320), one from China (n=767), and one from America (n=30). These described various bracing protocols, but generally proposed longer wear times (>20 hours per day). Overall they are in agreement with the Hunt and Patel review, that compressive orthotic bracing is a safe and effective alternative to surgical correction, but that patient adherence to treatment is critical to success. The fifth study, from China (n=125), measured the force required for correcting pectus carinatum, and noted that this correlated positively with age and body mass index (BMI) and negatively with Haller index (a ratio of thoracic width and height).[12]

The remaining four studies (in five publications) are described below.

Less intensive bracing protocols

Two of the studies, a case series (with a literature review and pooled analyses)[13] and a small randomised controlled trial[14], evaluated the use of less intensive bracing protocols (in other words, shorter wear times). The results of these are conflicting, with one endorsing shorter wear times (≥12 hours per day),[13] and the other recommending longer wear times (23 hours per day).[14]

Wahba et al. (2017) described the treatment of a series of patients in their practice (Canada) for pectus carinatum using a less intensive bracing protocol (wear time 8–12 hours per day).[13] Of the patients who were offered bracing and had adequate follow up (n=32), the success rate (full correction or improvement) was 90.6 %. The adherence rate was 93.8 %. The authors pooled their results with the findings from 15 other studies (11 of which were included in the Hunt and Patel review), and reported that less intensive brace usage (<12 hours per day), when compared to more intensive usage (≥12 hours per day), was associated with higher patient adherence (89.6 % versus 81.1 %) with a similar time to correction (7.3 versus 7.1 months) and success rate (85.3 % versus 83.5 %). The included studies were all lower-level observational studies with no control group, and to pool the results of such a heterogeneous group of studies is questionable.

A small randomised pilot study from Turkey by Giray et al. concluded that longer brace wear times (23 hours per day) should be recommended over shorter wear times (8 hours per day).[14] In this study, male patients with pectus carinatum aged 7 to 17 years (n=27) were randomised into three groups: compression orthosis 23 hours per day, compression orthosis 8 hours per day, and no orthoses. Dynamic compression orthoses were used. All groups received exercises for 1 hour a day for 3 weeks. The authors reported that the group that received compression orthosis for 23 hours showed greater improvements than the other groups. After treatment, all groups showed significant changes in protrusion, pressure of correction, and external chest wall measurements, and adverse events occurred with similar frequency across groups. Retention percentages did not differ among groups. The short-term nature of this trial (outcomes were assessed after 3 weeks of treatment) is a significant limitation.

The results of both the Wahba et al.[13] and Giray et al.[14] studies need to be interpreted with a degree of caution and further research would be needed before confident conclusions could be drawn on the effectiveness of shorter brace wear times.

Physiotherapy as an adjunct to bracing

A small randomised trial (n=30) from Turkey evaluated the effect of a physiotherapy protocol applied as an adjunct to compression brace treatment in patients with pectus carinatum.[4] The included patients were all male (age range 11 to 18 years) and were randomised either to brace treatment alone (23 hours per day for 12 weeks) or brace treatment (23 hours per day for 12 weeks) with physiotherapy. The authors reported that both groups showed improvements based on external chest measurements related to pectus carinatum protrusion following treatment. The physiotherapy group displayed greater improvement in maximum protrusion degree and lateral length values ($p<0.05$). The authors reported that patient perception of the pectus carinatum, posture, psychological life quality and treatment satisfaction scores were significantly better in the physiotherapy group ($p<0.05$). The statistical analyses were not intention to treat. This is a small study with a relatively short treatment duration, which was limited to males, and further studies would be needed to confirm these results.

Prior manipulation to correct pectus carinatum

A retrospective observational study from the United Kingdom, published in 2020, described a novel bracing protocol.[15] The authors hypothesised that the reasons for treatment failure often relate either to prolonged bracing protocols (up to 2 years in some cases) and the slow improvement in the correction of the pectus carinatum. They developed a protocol that involved immediate correction of the pectus carinatum within the first patient consultation using a physical therapy technique ('soft tissue release') followed by application of a custom fitted non-compressive external brace together with prescribed schedule of brace wear. The protocols were tailored to each patient, but seemed to consist of longer wear times (>20 hours per day). Bracing was considered complete any time after 32 weeks if the patient had fully weaned from routine bracing and was satisfied with the cosmetic result. The authors reviewed 249 consecutive patients. Thirty-four patients had pectus carinatum that were too stiff for immediate correction and so underwent bracing with traditional progressive tightening. The remaining 215 underwent successful reduction of their pectus carinatum at their initial consultation, and were followed for a mean of 32.6 weeks (range 8–83).

There was successful sustained reduction of the pectus carinatum in 244/249 patients with high reported rates of concordance (98 %) and satisfaction (94 %). Patients experienced a reduction in symptoms of anxiety and depression ($p<0.001$) and had improved body satisfaction ($p<0.001$). Mild skin irritation occurred in 18 % of patients (n=44) and there were two severe cases of skin irritation, one of which resulted in abandonment of bracing.

Another paper by the same group of authors was also identified.[16] The full text was not available, but the abstract describes the same bracing protocol. It is not clear from the

abstract whether it is the same group of patients, but it seems likely that this is an interim analysis of the study already described. The conclusions are the same.

Dynamic compression

Dynamic compression braces (as described in the review by de Beer et al.[6]) allow measurement and adjustment of the brace's pressure over the thoracic wall, leading to a controlled correction. These braces have been proposed as an alternative to other orthotic devices and proponents suggest that they are more comfortable and less likely to cause skin lesions.[6] As already noted, it was not always clear if the studies were evaluating dynamic or non-dynamic bracing, and the terms are used inconsistently, which made describing the literature under two discrete headings challenging. It was felt that there would still be value in considering the evidence for dynamic bracing separately, although, it is acknowledged that there will be overlap with the literature already described.

This use of dynamic compression braces (with a pressure measurement system) was first described in a case series by Martinez-Ferro et al. in 2008.[17] In this series, the PIC was obtained at the first patient consultation, using a measuring device which was applied over the pectus carinatum until a normal thoracic shape was obtained. PIC is measured in PSI. Martinez-Ferro et al. stated that patients who required a pressure higher than 7.5 PSI should not be treated with bracing.[17] In contrast, de Beer et al. argue that people are considered suitable for this treatment if their PIC does not exceed 7.5 to 10 PSI.[6] The electronic pressure measuring device can be added to the brace to adjust the correction pressure to the desired level for the POT. A pressure that is too high can increase complications (such as skin lesions) and/or abandonment of treatment. In order to avoid skin ulcerations, the literature reports that POT should not exceed 2.5 to 3 PSI.[6, 17] With this approach, patients are generally assessed every 4 to 6 weeks, with the POT measured and readjusted to 2.5 to 3 PSI. When correction of the pectus carinatum is achieved (PIC around 0 PSI), treatment moves to a maintenance phase, with wear time gradually decreased over a period of 2 to 6 months.

The systematic review by de Beer et al. (2018) provides a comprehensive summary of the literature on dynamic compression bracing in pectus carinatum.[6] It includes eight studies (n=1,185), seven of which were included in the review by Hunt and Patel. All of the included studies used the FMF® Dynamic Compressor System (Pampamed, Buenos Aires, Argentina). The authors reported that individually and combined, the eight studies showed favourable outcomes for measured dynamic compression bracing. The results of the eight studies are summarised in *Table 1*. Of the 1,185 patients, 347 (29.3 %) successfully completed treatment after a mean follow-up time of 16 months; 248 (20.9 %) were lost to follow up and 71 (6 %) patients dropped out (main reasons being social discomfort and failure of treatment). The remaining 519 (48 %) of patients were still under treatment. Recurrence of pectus carinatum was described in 2.6 % of patients, and secondary bracing of these patients showed good results.[6]

One additional observational study from America (published after the systematic review by de Beer et al.) was identified.[18] The authors reviewed prospective institutional data for patients undergoing dynamic chest bracing over a 7-year period (n=460). The authors reported that 57 % of patients adhered to recommended brace wear times, and that the median time to 'retainer mode' in this subset was significantly shorter than in patients who did not adhere to recommended wear times (3.5 months versus 10 months, $p<0.001$). The main barrier to adherence with wearing the brace was discomfort (37 %), while the main motivation for wearing the brace was appearance (58 %). Patients were surveyed via the telephone, and all endorsed bracing as worthwhile, with 94 % reporting a satisfaction rating of eight or greater (on a scale on one to 10) for the correction outcome.

Table 1: Dynamic bracing – study details taken from Hunt and Patel and de Beer reviews, or from study abstract

Reference	Patients	Patient selection criteria	Results
Martinez-Ferro et al. (2008)[17]	n=208 Argentina 74 % male Mean age started bracing: 12.5 years (range 3–18)	PIC<7.5 PSI	Mean utilisation time=7.2 hours daily for 7 months (range 3 to 20 months). Twenty-eight (13.4 %) patients abandoned treatment and were not evaluated for final results. Of the remaining 180, 112 completed treatment (68 were still receiving treatment) and 99 (88.4 %) had good to excellent results scoring between 7 and 10 points (out of 10). Thirteen (11.6 %) patients scored 1 to 6 points and were judged as poor or failed results.
Cohee et al. (2013)[19]	n=122 USA Median age started bracing: 14 years (range 10–28)	This detail was not given in either systematic reviews, or in the study abstract (full text not freely available)	Sixty-seven (55 %) were still progressing under active treatment at the end of the study. Five patients (4 %) were lost to follow up, and thirteen (11 %) failed treatment. Thirty-seven patients (30 %) exhibited flattening of the sternum after 6 (range 1–24) months without surgery. After flattening, patients then wore the brace for progressively fewer hours each day as a "retainer" for 5 (range 3–19) months. Five patients (4 %) experienced recurrence 5 (range 3–7) months after brace treatment was discontinued.
Lopez et al. (2013)[20]	n=61 France 97 % male Mean age started bracing: 13.5 years (range 5–25)	typical chondrogladiolar PC PIC<9 PSI	All patients and parents considered the results as excellent. Fourteen patients with a PIC>7.5 PSI were included. Even though treatment was still in progress at the end of the study period, they noted that despite the higher PIC, eight of the 14 had experienced some correction of the sternum.

Sesia et al. (2016)[21]	n=36 Switzerland Mean age started bracing 14.4 years (2–25)	chondrogladiolar PC No information given in either systematic reviews, or in the study abstract (full text not freely available) – apart from that some of the included patients had a PIC>7.5	Seventeen completed treatment by the end of the study period, and the remainder were still receiving treatment. Seventeen (47 %) were cured with good (n=7) to excellent (n=10) cosmetic results after a median treatment period of 9 months (range 2.5–16 months). The mean daily time of wearing of the device in the 17 patients was 9 hours (range 5–18). None abandoned the treatment and there were almost no complications.
Al Githmi et al. (2016)[22]	n=18 Saudi Arabia 94 % male Mean age started bracing 15.5 years (10–23)	Chondrogladiolar PC Mean PIC of included patients given in results, but not as inclusion criteria to study.	Mean PIC was 4.5 PSI (range 2.2–7.3 PSI). Bracing time was 12.8 hours per day (range 8–24), and satisfaction score was 3 (scale: no correction, 0; complete correction, 4). There was complete correction in seven patients (39 %), remarkable improvement in five patients (28 %), minimal improvement in three patients (17 %), and no correction in three patients (17 %). There were no major complications.
de Beer SA, et al. (2017)[23]	n=286 The Netherlands 91 % male Mean age started bracing: 15 years (range 4–21)	PIC≤10 PSI	At the end of the study: - 27 % (n=78) completed treatment by the end of the study (mean treatment time 14 months) - 10 % (n=27) abandoned treatment (reasons include of lack of motivation, loss to follow up, persistent protrusion of the sternal bone or flaring that required surgical correction and delayed correction), and - 63 % (n=181) were still under treatment.

Emil et al. (2017)[24]	n=114 Canada 92 % male Mean age started bracing: 14 years (7.5–17)	This detail was not given in either systematic reviews, or in the study abstract (full text not freely available)	Sixty-four (56 %) were successfully treated, 33 (29 %) were still in active bracing, and 17 (15 %) failed or were lost to follow up. In successful patients, active and maintenance bracing was 5.66±3.81 and 8.80±3.94 months, respectively. Asymmetry and older age were significantly associated with failure. Multivariable Cox proportional hazard analysis of time-to-maintenance showed that asymmetry (p=0.01) and smaller first drop in POT (p=0.02) were associated with longer time to reach maintenance.
Poola et al. (2018)[25]	n=340 USA 81 % male Mean age started bracing 14 years ±2 standard deviations	No information given in either systematic reviews, or in the study abstract (full text not freely available) – apart from that some of the included patients had a PIC>7.5	High loss to follow up (n=227, 67.1 %), reasons not clearly specified. Of those lost to follow up, 123 had less than two visits over the 5-year study period and 105 patients were lost after an average of seven brace adjustment visits after initial fitting. A low PIC and an increased duration of bracing were predictive factors for successful resolution of the pectus carinatum. 217 patients had two or more visits after the patient was fitted for the brace. The mean PIC in this cohort was 4 PSI (range: 1.5–7.8), and the median duration of bracing in this group was 16 months (IQR: 7–23 months). 103 (47 %) achieved complete correction after an average bracing time of 7.5 months and were then placed in the retainer mode. 30 patients successfully completed bracing therapy and required an average of 23 months of therapy (2 months to 4 years). No patient recurred after bracing was completed, but one failed bracing and required operative correction.

Patient selection protocol

The systematic review by Hunt and Patel stated that the method of patient selection was poorly reported by the included studies.[7]

- Five of the 17 studies included in the review reported the use of pressure monitoring as an assessment tool (see patient selection criteria in *Table 1*).
- In five of the 17 studies, the type of pectus carinatum was characterised and only patients with a lower sternal or chondrogladiolar type were considered candidates for dynamic chest bracing. This may be explained by the fact that the chest wall in people with chondrogladiolar pectus carinatum tends to be more flexible than in patients with chondromanubrial pectus carinatum.[6]

Ease of use for patients

As already discussed, many of the bracing protocols described in the literature (particularly those that involve a gradual reduction of the pectus carinatum and long periods of bracing) had high attrition rates. Various approaches to make it easier for patients to persist with chest wall bracing have been proposed, and some have been described above, for example immediate correction of the pectus carinatum,[15, 16] or reducing discomfort (for example, with dynamic bracing).[6]

Two additional retrospective observational studies (not included in the systematic reviews described above) were identified that evaluated the factors impacting on patient adherence to treatment.[26, 27]

A study from South Korea (Kang et al. 2014) included 86 patients (seven female and 79 male) who were instructed to wear the brace over a t-shirt for 20 hours per day during the active treatment period (2–4 weeks) and 10 hours per day during the maintenance period (6 months).[27] For the analysis, the authors split the patients into a 'compliance' group (patients who wore the brace for ≥6 months) and a 'non-compliance' group (patients who wore the brace for <6 months). Factors affecting patient adherence with treatment were assessed at the last day of follow up with a multiple-choice questionnaire. The questionnaire comprised seven items: pain at compression site, skin problems on compression area, confidence in brace treatment, shame, discomfort, initial result of bracing treatment and total number of factors affecting patient adherence. The authors reported that the initial result of the treatment period ($p<0.001$) and total number of factors affecting patient adherence ($p<0.05$) were significant predictors of patient adherence. Mean reported values for pain on the compression area, skin problems on the compression area, confidence in brace treatment, shame and discomfort were not significantly different between the two groups. They concluded that an initial successful result of the active treatment period may increase patient adherence during treatment for pectus carinatum. In addition 'efforts to decrease

pain, skin problems, shame and discomfort, and to give confidence may be beneficial in increasing compliance with bracing treatment.'

A study from the USA (Thaker et al. 2017) aimed to identify factors that predicted successful treatment with chest wall bracing in patients with pectus carinatum.[26] The authors used a mixed methodology approach, encompassing an online survey of patients and a retrospective chart review on patients who participated in the survey. The authors contacted 176 patients via phone, asking them to participate, but only 25 completed the survey. The patients had been treated with chest wall bracing up to 10 years previously, increasing the risk of recall bias. It is possible that patients who had a more positive experience may have been more inclined to take part in the survey (participation bias). The authors reported that subjects rated themselves as more confident after bracing (p=0.002). Patients who had family support and no documented complaints (p=0.024) and (p=0.009) respectively, were more likely to say they had made the right choice to wear the brace (results taken from abstract – full text not obtained).

With regard to technology for monitoring patient adherence to wear times, a small 4-week pilot study (n=8, Argentina) assessing a novel, wireless, real-time monitoring system (MyPectus) was identified.[28] The components of the MyPectus system included: a data logger, smartphone app, web-based platform and cloud-based storage system. The data logger was inserted into the chest brace. All patients were fitted with the FMF® Dynamic Compressor System (Pampamed, Buenos Aires, Argentina). The authors concluded that the components of the MyPectus system recorded, stored and provided data to patients and clinicians and may be useful for measuring patient wear times. They noted that this was a preliminary study, and that further refinements to the system were required, along with additional research.

Other issues identified in literature

Patient-applied force

One small observational study from Canada (n=21, all male) evaluated the differences between prescribed force and patient-applied force.[29] In this study the force applied by the brace was manually adjusted by the patient over the course of treatment. The authors aimed to determine the accuracy of patients in applying their prescribed force over time, and to determine whether the protrusion stiffness influenced the patient-applied forces and the protrusion correction rate. The brace used by all participants was produced by Braceworks Custom Orthotics (Calgary, Alberta, Canada), and participants were evaluated on three visits: at the first fitting, 1 month post-fitting and 2 months post-fitting.

The majority of individuals followed for 2 months (75 %) had a significantly different patient-applied force ($p<0.05$) from their prescribed force. The magnitudes of some of these differences were minimal and so the clinical significance of the differences is not clear. Protrusion stiffness had a positive relationship with patient-applied force, but no relationship with correction rate.

This was a very small study and it only measured forces as they were applied in a clinical setting (not in patients' home environments).

Overcorrection of pectus carinatum

One study included 17 patients (Brazil) who experienced overcorrection during orthotic treatment of pectus deformities (15 had pectus carinatum and two had pectus excavatum).[30] The authors described the use of orthoses and exercises to reverse this complication. The procedures varied according to each patient's needs and included: decreasing the time of orthosis wear, introducing a second orthosis, improving the prescribed exercises and/or encouraging the patient to perform them more intensively. Overcorrection was reversed in all but one patient with pectus excavatum.

Use of pressure of correction for predicting brace failures

A paper from a centre in America described a team's 10-year experience of using a staged management protocol for pectus carinatum.[31] The protocol consisted of bracing initially, with surgical repair reserved for patients who failed bracing, those with stiff or very asymmetric chest walls and patients for whom bracing therapy was unlikely to be workable. The main results do not add significantly to the literature already described - the authors reported that of 364 patients who had bracing, 144 (40 %) were successful, 77 (21 %) were still being treated, the treatment failed in 25 (7 %), and 118 (32 %) dropped out of the study. The authors reported that patients for whom the treatment was successful experienced a 50 % decrease in pressure of correction beginning 1 month after starting treatment. Based on this, the authors concluded that a considerable decline in pressure of correction during the early months of bracing therapy predicted eventual success.

Safety, complications and drop-out rates

No significant safety concerns were identified from the literature.

The review by Hunt and Patel stated that all 17 included studies recorded treatment failures, with failure rates varying from very low in some studies to very high in others (one study reported a failure rate of 43 %). This included people who abandoned treatment, or were lost to follow up. Most complications recorded were skin issues, but mechanical problems, vasovagal reactions, as well as complaints of tightness and pain were also recorded.[7]

Another general review article noted that complications that have been recorded may include:

- skin discolouration, rash, or ulceration
- back pain
- haematoma, and
- overcorrection leading to iatrogenic pectus excavatum.[32]

Two studies included in the clinical effectiveness section stated that they only included male patients because of concerns around the impact of bracing on breast development or the impact of breasts on chest wall bracing effectiveness.[4, 14] This was not discussed in any great detail, or mentioned in any of the other studies.

Patient and social aspects

This section focuses on the psychosocial issues that may affect patients with pectus carinatum. Issues around patient experience of chest wall bracing and patient adherence to treatment are covered in the clinical effectiveness section.

The British Lung Foundation includes some patient and carer information on pectus carinatum.[5] This states that:

> Because it affects the shape of their chest, some children and adults with pigeon chest may be unhappy with their body. This can have a big impact on their lives. Even if pigeon chest isn't causing any physical problems, treatment may be recommended to improve your child's self-esteem and quality of life.

The literature search for this assessment identified two studies which highlighted the potential impact of pectus carinatum on body image and quality of life.

A prospective study from Turkey (2021) compared physical and psychosocial parameters in adolescents with pectus excavatum (n=90) and pectus carinatum (n=90) with healthy controls (n=90).[33] The participants had mild, moderate and severe pectus deformities. For most of the psychosocial parameters, no statistically significant differences were reported between patients with pectus carinatum and healthy controls. Perceived appearance of the chest area correlated with some physical functions and many parameters of psychosocial status, for example perception of social appearance, the SAD-New (Social Avoidance and Distress-New) score and subscales of the brief symptom inventory (excluding hostility). Based on these findings, the authors concluded that all adolescents with mild, moderate or severe pectus carinatum should undergo a biopsychosocial evaluation and receive psychosocial support.

A case-control study from Germany (2011) evaluated the effects of having anterior chest wall deformities on disease-specific and health-related quality of life, body image and psychiatric comorbidity prior to surgical correction.[34] It included 90 male patients with severe pectus deformities (71 pectus excavatum and 19 pectus carinatum) scheduled for surgical correction, who were assessed by questionnaire and compared with 81 controls.

Compared to the control group, the authors reported that having severe pectus carinatum was associated with:

- poorer disease-specific quality of life ($p<0.001$)
- increased dysmorphic concerns ($p<0.001$)
- increased body image impairment ($p<0.001$), and
- lower self-esteem ($p<0.01$).

Cost effectiveness

No literature relating to cost effectiveness was identified.

Conclusion

The conclusions are presented under each question asked by the topic referrer. The evidence base consists largely of lower-level observational studies and so a degree of caution is required when considering the results.

What is the clinical effectiveness, cost effectiveness, safety and patient experience of orthotic bracing for pectus carinatum?

Clinical effectiveness

The evidence base for chest wall bracing in patients with pectus carinatum consists largely of lower-level observational studies (case series). These studies suggest that this treatment modality is safe, acceptable to most patients, and clinically effective.

The identified studies were extremely variable with regard to:

- the bracing protocol used, including wear times and the duration of treatment
- the selection of patients, including the use of pressure monitoring as an assessment tool, and
- how clinical outcomes are measured.

Individually, the studies reported positive results for chest wall bracing in patients with pectus carinatum. The heterogeneity between the studies means that drawing conclusions on the optimal treatment approach is not possible.

The use of 'dynamic bracing' was described in several studies. The term 'dynamic' is used inconsistently, making it difficult to accurately categorise studies under discrete headings. One systematic review provided a comprehensive summary of the literature on 'dynamic

bracing', defined by the authors as a system which allows measurement and adjustment of the brace's pressure over the thoracic wall. This concluded that patients may comply more with this bracing approach as it is more comfortable. The authors note that accurate selection of patients based on age, pressure of initial correction and motivation is important.

Cost effectiveness

No evidence was identified relating to cost effectiveness.

Safety

No significant safety concerns were identified from the literature.

Patient issues

Compared to a control group, people with pectus carinatum may have reduced quality of life and poorer body image. Treatment of the pectus carinatum might help improve self-esteem, and some authors noted that psychosocial support may also be beneficial.

What is the prevalence of pectus carinatum in the general population aged 0–16 years and is there any deprivation effect?

Pectus carinatum is the second most common chest wall deformity in children, affecting males about four times more frequently than females. The reported prevalence of pectus carinatum varies in the literature, ranging from 0.1 % to approximately 1 %.

The association between incidence of pectus carinatum and levels of deprivation was not explored in the published literature, or raised by any authors as a potential area for research.

What is the evidence on wear time, including the use of technology for recording and/or assessing wear time in pectus carinatum chest wall bracing?

With regard to wear time, there was considerable variability in the published studies, though they mostly reported longer wear times (≥20 hours per day). Of the 17 studies included in the systematic review by Hunt and Patel.[7]

- Five reported that patients were advised to wear the brace 'as often as possible' or for most of the day (wear times used ranged between 20 and 23 hours per day).
- Four reported shorter wear times (ranging between 8 and 12 hours per day).
- One study described starting bracing slowly (for 6 hours per day), increasing it by 1 hour every week until an optimal wear time of 16 hours per day.

- Seven studies described a protocol that included a period of active treatment, followed by a period of reduced or maintenance wear. The active treatment phase tended to consist of continuous or near-continuous wear for a period 2 to 8 months (although, one study described an active treatment phase of only 2 weeks). Maintenance wear times varied but were mostly between 8 and 12 hours per day, for a period of 3 to 6 months (or until the patient reached skeletal maturation).

Two additional studies were identified which evaluated shorter wear times and reported conflicting results.[13, 14] Additional research would be required before confident conclusions could be drawn around shorter wear times.

With regard to technology for monitoring patient adherence to wear times, only one small 4-week pilot study (n=8, Argentina) assessing a novel, wireless, real-time monitoring system (MyPectus) was identified.[28] The authors concluded that the components of the MyPectus system recorded, stored, and provided data to patients and clinicians, and may be useful for measuring patient wear times. This was a preliminary study, and further refinements to the system are required along with additional research.

What is the evidence around patient adherence to recommended brace wear time?

While the literature collectively suggests that orthotic bracing is a safe and effective alternative to surgical correction, the factors that allow patients to persist with treatment are critical to success.

Many of the bracing protocols described in the literature (particularly those that involve gradual reduction of the pectus carinatum and long periods of bracing) reported high attrition rates. Careful selection of patients based on age, pressure of initial correction and motivation appears to be important.

The systematic review by Hunt and Patel reported that treatment failure (including non-adherence) rates varied between very low in some studies to 43 % in on study.[7] Reasons for treatment abandonment included skin issues, discomfort, lack of motivation, persistent sternal protrusion and rib flaring. Some patients were also lost to follow up.

Various factors associated with improved adherence to treatment were reported, including family support and efforts to decrease pain, skin problems, shame and discomfort. de Beer et al. proposed that dynamic bracing protocols (with pressure measurement systems) were associated with less discomfort and improved tolerability of treatment.[6]

Some studies reported that rapid or immediate improvements in the pectus carinatum seemed to increase patient motivation to continue with treatment. One study noted that an initial successful result during the active treatment period may increase patient adherence.[27] Similarly, another study described a novel protocol that involved immediate

correction of the pectus carinatum using a physical therapy technique ('soft tissue release').[15] The authors reported that this approach was associated with high rates of treatment completion, although, further research would be needed to corroborate these findings.

What is the evidence on PIC and POT used to guide initial patient selection and treatment while in brace?

de Beer et al. suggest that dynamic bracing is suitable for patients whose PIC does not exceed 7.5 to 10PSI.[6] The literature also suggests that in order to avoid skin ulcerations, the POT should not exceed 2.5 to 3 PSI.[6, 17] Using the dynamic bracing approach, patients are generally assessed every 4 to 6 weeks, with the POT measured and readjusted to 2.5 to 3 PSI. When correction is achieved (PIC around 0 PSI), treatment moves to a maintenance phase, with wear time gradually decreased over a period of 2 to 6 months.

One case series proposed that treatment could be guided (and treatment failure predicted) by measuring falls in PIC.[31] The authors reported that patients for whom bracing was successful experienced a 50 % decrease in PIC beginning 1 month after starting treatment. They concluded that a considerable decline in PIC during the early months of bracing therapy predicted eventual success.

Identified research gaps

- There is a need for well-conducted, adequately-powered trials on the clinical effectiveness of chest wall bracing in people with pectus carinatum. These should clearly describe the chest wall bracing protocol, and should include objective measures of clinical outcomes, and a reasonable follow-up period.
- The impact of chest bracing on breast development, and the impact of breast development on the effectiveness of chest wall bracing, was mentioned in two studies. It may be useful to explore this further in future studies, to get clarity on whether this is an issue that needs to be considered during patient selection and treatment.
- Studies are needed to evaluate the management of patients with more complex pectus deformities, such as mixed deformities, and the role of combined bracing and vacuum bell therapy (a treatment pectus excavatum) or the role of multiple braces to treat patients with pectus carinatum with significant rib flare.[7]
- Investigating how to make treatment easier to persist with is something that should be highlighted in future studies.[7]
- A patient organisation who peer reviewed the SHTG Assessment noted that they would like to see greater emphasis on how patients and parents are supported to manage the side effects of chest wall bracing to reduce drop out from the procedure.

Equality and diversity

Healthcare Improvement Scotland is committed to equality and diversity in respect of the nine equality groups defined by age, disability, gender reassignment, marriage and civil partnership, pregnancy and maternity, race, religion, sex, and sexual orientation.

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