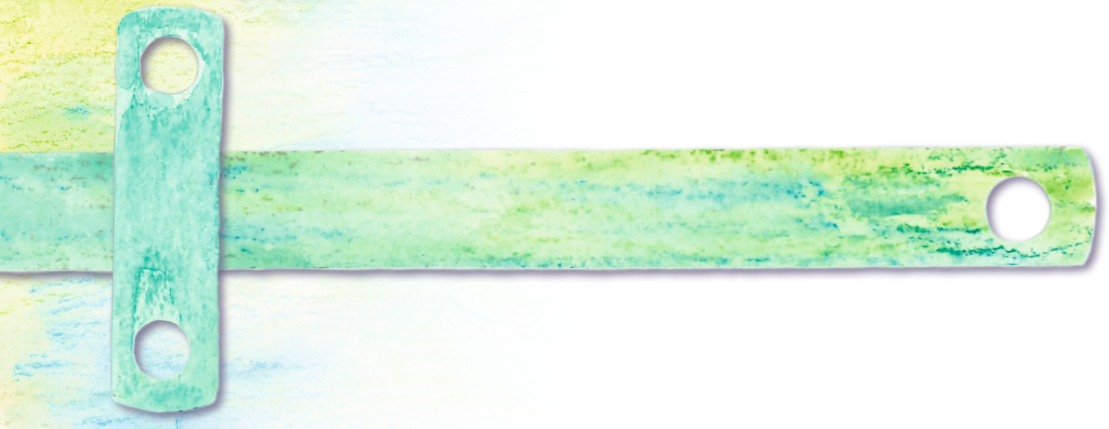
Advancing pectus deformity care: evaluation of current treatments, complications and future innovations

Braak, H. van

Part I

Pectus carinatum



Chapter 2

Ravitch surgery or dynamic compression bracing for pectus carinatum: a retrospective cohort study

Hendrik van Braak, Sjoerd A. de Beer, Sander Zwaveling, Matthijs W.N. Oomen, Justin R. de Jong

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ABSTRACT

Background

Pectus carinatum is a pediatric condition which can be treated by dynamic compression system (DCS) bracing or surgery. Several publications concerning DCS-bracing or surgery are published, however without comparing both treatments.

Methods

Over a 10 year period 738 patients with pectus carinatum were treated. We describe our 10-years' experience and results of both treatments.

Results

Of the 631 patients who received DCS-bracing treatment, 553 finished treatment and 78 patients are still under treatment. Seventy-three point eight percent (408/553) of patients finished treatment successfully, 13.6% (75/553) failed treatment and 12.7% (70/553) were lost to follow-up. Success rate decreased with an increasing pressure of initial compression (84.2%-67.3%). Marfan and Poland syndrome were related to bad results.

Ravitch surgery was performed in 105 patients with a success rate of 92.4%. Complications occurred in 32.4% of patients and 6.7% of patients had complications for which surgery was needed. No relationship was found between osteotomy or sternal fixation and outcomes or complications. Abramson procedure was performed in two patients.

Conclusions

DCS-bracing should be the treatment of choice in patients with pectus carinatum because its non-invasiveness, good results and a lower complication rate compared to surgery. Besides pressure of initial correction, motivation is an important factor influencing outcomes and compliance remains a major challenge in treating pectus carinatum using DCS-bracing. Bracing patients before their growth spurt should be discouraged. Patients with a higher pressure of initial compression (>8.0-8.5) and Marfan and Poland syndrome have poorer outcomes. In those cases surgery may be considered.

INTRODUCTION

Pectus carinatum (PC) is a pediatric condition characterized by overgrowth of costal cartilage which causes protrusion of the sternum and adjacent cartilage. PC has an incidence of 0.3-0.7% and affects males four times more frequently than females.[1,2] PC is highly associated with a disturbed body image, embarrassment and a reduced quality of life.[3] Patients are mostly asymptomatic and treatment is started mainly for aesthetic reasons. Patients that are symptomatic complain of pain, dyspnea, tachypnea with exertion and reduced endurance.[4]

Traditionally PC patients were treated surgically using the Ravitch procedure which was first introduced in 1960 and modified during later years.[5, 6] Another surgical technique is the Abramson procedure, which is a 'reverse' Nuss procedure.[7] From the seventies onwards orthotic treatment was introduced as a non-operative treatment with a success rate ranging from 40-90%. Orthotic treatment was complicated by long treatment time (up to 30 months) and complications such as discomfort, pain, skin rash or discoloration and skin lesions.[8-12] In 2008 Martinez-Ferro et al. introduced the dynamic compression system (DCS) for treating pectus carinatum and claimed a success rate up to 90%.[13, 14] This system was adopted in the Amsterdam Pectus Center in 2013. Several publications concerning the outcomes, complications and improvement of quality of life after DCS-bracing or Ravitch surgery were published, however most studies focused on either Ravitch [6, 15-18] or DCS treatment[1, 2, 19-23] without comparing both treatments.

Recently the first comparison of both treatments in one medical center was published.[24] In this study we describe the results of both the surgical procedures and DCS-bracing for patients with PC in our center.

PATIENTS AND METHODS

Study design

We conducted a retrospective cohort study including all patients with PC, aged 0-18 years (y), that were treated from 2009 to 2019 with either surgery (Ravitch or Abramson) or DCS-bracing. A Medical Research Ethics Committee (METC) official waiver of ethical approval was granted from the METC of the Amsterdam Medical Center. Informed consent for this specific study was prospectively obtained from all individual patients (or their parents) included in this study. Patients who were retrospectively included were contacted by their treating clinician to receive informed consent. This was recorded in their electronical medical record system. No patient opted out of providing consent.

Data collection

Patients were prospectively included, data were retrospectively complemented and reviewed using the patients electronical medical record system. Sixteen patients, who underwent Ravitch surgery before we started including patients prospectively for this study, were retrospectively included. Baseline characteristics were age, gender, breast growth, underlying diseases, symmetry and degree of the deformity, flaring, pressure of initial compression (PIC), symptoms, follow-up (months (m)), hospitalization (days (d)), details about the surgical procedure (osteotomy, fixation methods), length of treatment and prior surgeries. DCS-bracing patients were distributed into two age groups (<10y & 10-18y) and three groups based on PIC (<5.0 PSI (pounds per square inch), 5.0-7.5 PSI & >7.5 PSI). Patients were compared on different variables (PIC, age) to identify possible factors influencing the outcome of treatment.

Indications

After anamnesis and physical examination the type of treatment was determined using shared decision making. DCS-bracing was advised for all patients except for patients with pectus arcuatum (PA), an asymmetrical deformity, a high PIC (>10 PSI) or a history of earlier DCS-bracing. They were advised to choose Abramson or Ravitch surgery, but they could still choose DCS-bracing. Patients who were not eligible for DCS-bracing usually had a rigid, stiff thorax with a high PIC, a asymmetric deformity or PA. In these cases Abramson surgery is unfit because of more complications and the risk of plates breaking out.[25] Another reason for choosing Ravitch over Abramson was the fact that Ravitch procedure required only one surgery.

Techniques

For DCS-bracing the FMF Dynamic Compression System (FMF-DCS) was used (Figure 1). First we measure the PIC using a Pressure Measuring Device which is pressed on the chest and measures the average applied pressure until the deformity is corrected. Thereafter the thorax is measured, the brace is ordered, fitted and the patient receives training on how to use it. The brace is typically worn daily for 12-24 months, 12 hours a day, until the deformity is corrected. More about the procedures and technique can be found in our earlier manuscript[17] and a step-by-step plan on how to fit the brace is described by Martinez-Ferro et al.[14] Ravitch surgery was performed using the modified Ravitch technique as described by Welch et al.[6] Abramson surgery was performed using the technique as described by Abramson et al.[7]

Statistical analysis

Due to the nature of this study only descriptive measurements were used. Probability values of less than 0.05 were considered statistically significant. All data were analyzed using IBM SPSS Statistics 24.

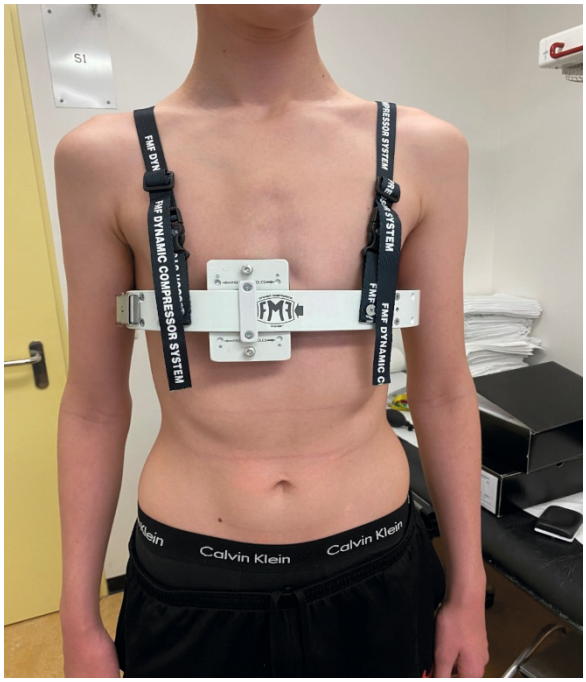

Figure 1 Patient wearing a Dynamic Compression System Brace

Outcomes

Primary outcomes were success or failure and complications (number of patients with complications and severity of complications). Active treatment as outcome was only possible for patients who received DCS-bracing. Treatment was regarded successful if patients achieved a satisfactory result, in consultation with the clinician. Pictures of the thorax were taken before and after treatment by a medical photographer to give patients and clinicians a reference. Failure for DCS-bracing was defined as: patients who started brace therapy but did not accomplish a sufficient result or could not endure bracing therapy. Patients were regarded lost to follow-up if they were not seen for two years in the outpatient clinic before ending treatment. Relapse (or failure) in patients who underwent surgery was defined as a disturbing comeback of PC after initial correction. Complications (wound infection, wound dehiscence, bleeding, pneumonia, pneumothorax, atelectasis, pseudoarthrosis, malunion, pectus excavatum (PE), painful/

numb/hypertrophic scar, pressure ulcers and dermatosis) were ranked on severity and impact using the Clavien-Dindo Classification.[26]

RESULTS

Baseline characteristics of all patients

Baseline characteristics can be found in Table 1. Between April 2009 and November 2019 738 patients were treated at the Amsterdam Pectus Center. Two patients deliberately opted for the Abramson procedure. Six hundred thirty-one patients started with DCS-bracing. One hundred and five patients applied for Ravitch surgery. Seven patients had a pectus deformity of the chondromanubrial (PA) type. Asymmetrical deformities, scoliosis, PA and deformities accompanied by costal flaring were found relatively more often in patients receiving Ravitch surgery. A little less than 50% of the patients was bullied or felt embarrassed about their chest (361/738).

Table 1 Patient characteristics and treatment groups

	n	%	Age [a] (y)	Follow-up [a] (m)
Total treated patients	738	100.0	14.0 (13.0-16.0)	53.0 (35.0-74.0)
Male	683	92.5		
Female	55	7.5		
Treatment type				
DCS	631	85.5	14.0 (13.0-15.0)	53.0 (35.0-71.0)
Ravitch	105	14.2	15.0 (14.0-16.0)	54.0 (34.0-79.0)
Abramson	2	0.3	15.5 (15.0-16.0)	85.0 (85.0-85.0)
Underlying conditions				
Marfan syndrome	10	1.4		
Poland syndrome	2	0.3		
Osteogenesis imperfecta type 3	1	0.1		
Scheuermann's disease	1	0.1		

Characteristics of deformities			**%Ravitch**	**%DCS**	**%Abramson**
Asymmetric	277	37.5	*65.7*	*32.6*	*100.0*
Symptomatic	276	37.4	*33.0*	*38.0*	*50.0*
Flaring	134	18.2	*21.7*	*17.4*	*50.0*
Scoliosis	92	12.5	*17.9*	*11.5*	*0.0*
Chondromanubrial (PA)	7	0.9	*6.6*	*0.0*	*0.0*

DCS = dynamic compression bracing, IQR = interquartile range, m = months, n = number of patients, PA = pectus arcuatum, PC = pectus carinatum, y = years
[a] Continuous variables expressed as median (IQR)

Outcomes of DCS-bracing

Figure 2 summarizes the DCS-bracing treatment group. Five hundred fifty three (87.6%) of patients had completed treatment. From those patients 73.8% (408/553) finished treatment successfully, 13.6% (75/553) failed and 12.7% (70/553) were lost to follow-up. The combination of not wearing the brace enough and a lack of motivation was (nearly) always the reason for failing treatment. Female patients (44/553) had a success percentage of 81.8% compared to 73.1% in male patients (509/553). No significant relationship was found between breast growth and outcome of DCS-bracing (P=0.64).

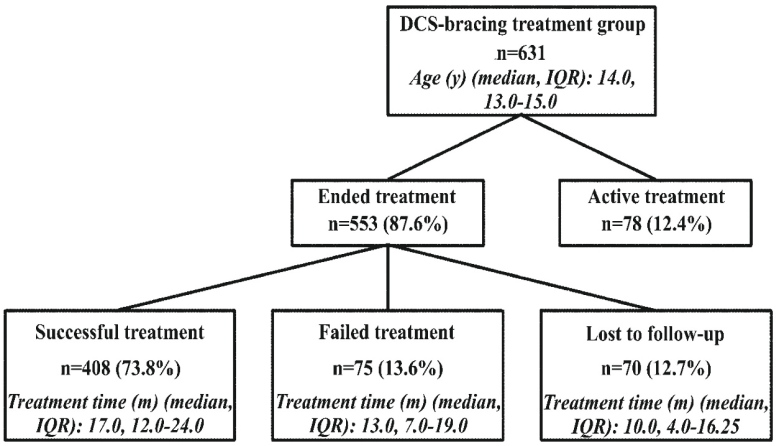

Figure 2 DCS-bracing group details (DCS = dynamic compression system, IQR = interquartile range, m = months, n = number of patients, y = years)

As earlier research suggested that in patients with a higher PIC treatment is less successful,[13] patients were divided into three PIC-groups. Results are shown in Table 2. The PIC varied from 1.9 to 13.3 PSI. Patients with a PIC less than 5.0 PSI had the best results with 84.2% finishing treatment successfully. Higher PIC decreased the rate of success, with success percentages decreasing to 60.0% when the PIC exceeded 10.0 PSI (Figure 3). A statistically significant (P=0.01) inversely proportional relation was found between success of DCS-bracing and PIC. A higher PIC was also related with longer treatment duration and higher age (respectively P<.001 & P<.001). No relevant complications of DCS-bracing were noted. Younger patients (<10y) had initially better results (83.9% treated successfully) compared to other age groups (10-18y: 73.2% treated successfully), but no statistical relation was found between age and outcomes (P=0.59), recurrence (P=0.74) or length of retainer mode trajectory (P=0.48). Four percent of the patients suffered from minor skin lesions during the treatment. One patient (0.2%) had overcorrection and developed PE for which the patient was unsuccessfully treated with vacuum bell therapy. The patient subsequently received a

Nuss-bar. Twenty-three patients (3.6%) decided to quit bracing therapy and received a Ravitch procedure.

Table 2 Pressure of initial correction (PIC) & results

	n	%	Treatment time [a] (m)
Total treated	631	100.0	
PIC < 5.0	116	18.4	
PIC 5.0-7.5	275	43.6	
PIC > 7.5	240	38.0	
PIC < 5.0 (active patients left out)			
Successful	80	84.2	15.0 (11.0-19.0)
Failed	7	7.4	
Lost to follow-up	8	8.4	
PIC 5.0-7.5 (active patients left out)			
Successful	184	75.4	16.0 (12.0-23.0)
Failed	29	11.9	
Lost to follow-up	31	12.7	
PIC > 7.5 (active patients left out)			
Successful	144	67.3	19.5 (14.5-25.0)
Failed	39	18.2	
Lost to follow-up	31	14.5	

IQR = interquartile range, m = months, n = number of patients, PIC = pressure of initial correction
[a] Continuous variables expressed as median (IQR)

Table 3 Ravitch treatment group details

	n	%
Ravitch patients never braced	94	89.5
Ravitch after DCS failure	9	8.6
Ravitch after Nuss	2	1.9
Sternal fixation after osteotomy		
None	47	61.0
Soluble stitches	15	19.5
Non-soluble stitches	4	5.2
LCP-plate	11	14.3
Relapse PC	8	7.6

DCS = dynamic compression bracing, n = number of patients, PC = pectus carinatum

Outcomes of Ravitch/Abramson surgery

Table 3 summarizes the Ravitch treatment bracing group. Mean hospitalization time was 6.4 days (SD=1.3). Surgery was performed on patients who did not receive prior brace therapy (94/105, 89.5%), failed DCS-bracing (9/105, 8.6%), or were treated with a Nuss-bar before and needed treatment for overcorrection (2/105, 1.9%). The PIC of patients who failed DCS-bracing was high with an median of 9.0 PSI (IQR 7.8-9.2), compared to 7.0 PSI (IQR 5.6-8.0) in the total DCS-bracing population. Ninety-seven (92.4%) of the patients finished treatment with satisfying results. Relapse occurred in eight patients (7.6%). Sixteen patients received Ravitch surgery before the introduction of DCS-bracing. The success rate of surgery was lower before (77.5%) than after the introduction (93.3%) with a comparable percentage of complications (respectively 31.3% and 32.6%).

Complications of Ravitch surgery are summarized in Table 4. Thirty-four patients (32.4%) had complications. There were seventeen patients with one complication, thirteen patients with two complications and four patients with three complications or more. Patients with complications spent on average 1.0 days longer in the hospital compared to patients without complications (P=0.01). Grade IIIb complications, for which an intervention under general anesthesia is needed, were pseudoarthrosis (5), wound dehiscence (1) and hypertrophic scars (1). Eight patients developed PE, for which three patients were successfully treated with a vacuum bell. No statistically relevant relations were found between osteotomy and the amount of complications (P= 0.92) nor the severity of the complications (P=0.67). No statistical relevant relation was found between PE and sternal fixation method (P=0.11). Relapse was not related to osteotomy nor complications (respectively P=0.91 & P=0.38). Two patients successfully underwent Abramson surgery, one patient needed an additional procedure for resection of flaring. No complications were noted.

DISCUSSION

In general, both DCS-bracing and surgery obtained good results. In the DCS-bracing group the success rate decreased with an increasing PIC, with a steep decrease in success percentage when the PIC exceeds 8.0-8.5. This, in combination with the high median PIC (9.0 PSI) in patients who underwent Ravitch surgery after failing DCS-bracing, makes DCS-bracing in patients with a high PIC debatable. Figure 3 however shows that the success rate increases slightly after reaching its ultimate low at a PIC of 9.0-9.5. A reason for this could be that the patients with a high PIC deliberately made a highly motivated choice for DCS-bracing, despite being well informed about the difficulty of DCS-bracing in their situation by their attending surgeon. This, combined

with the fact that overall a lack of motivation is the most important reason for DCS-bracing failure, might prove that not primarily the PIC, but also motivation is one of the most important factors determining the success of DCS-bracing, which might prove to be clinical useful.

Table 4 Complications of Ravitch repair

	n	**%**
Complications	56	100.0
Hypertrophic scar	14	25.0
Pectus excavatum	8	14.3
Painful scar	7	12.5
Pseudoarthrosis	5	8.9
Wound infection	5	8.9
Numb scar	3	5.4
Pneumothorax	3	5.4
Wound dehiscence	3	5.4
Malunion	2	3.6
Bleeding	2	3.6
Atelectasis	1	1.8
Dermatosis	1	1.8
Pneumothorax	1	1.8
Pressure ulcers	1	1.8
Clavien-Dindo Classification		
I	37	66.1
II	12	21.4
IIIb	7	12.5

n = number of patients

Bracing in the young patients

It was expected that young patients (36) would struggle with a prolonged time of wearing the brace and relapse. Yet within the study period no recurrence occurred and no differences in retainer mode time were found between age groups. However secondary evaluation showed that, several years after the study period, eight (22.2%) patients contacted the clinic again because of relapse. Those patients restarted treatment. More than half of the other patients wear their brace occasionally at night to maintain the obtained results. Relapse occurred nearly always in their growth spurt. DCS-bracing should probably be delayed for several years until children have had their growth spurt.

DCS-bracing in the literature

When DCS-bracing was first adopted great results were expected. In the original article of Martinez-Ferro et al. a success rate of 88.4% was mentioned[13] while others noted a success rate of 100%.[19, 23] Those studies were, however, a lot smaller and Martinez-Ferro et al. excluded patients with a PIC above 10.0 PSI. Our study included 38 patients with a PIC of 10.0 PSI or higher. Their results were also influenced by the exclusion of 24 (10.3% of the total population) patients with Marfan or Poland syndrome. Both syndromes seem to be related to bad outcomes, which could have raised the success rate in their study. This is confirmed by our results. In our study two (33.3%) patients with either Marfan or Poland syndrome finished DCS-bracing successfully, three were lost to follow-up and one failed treatment. In patients with Marfan or Poland syndrome surgery could be considered the treatment of first choice.

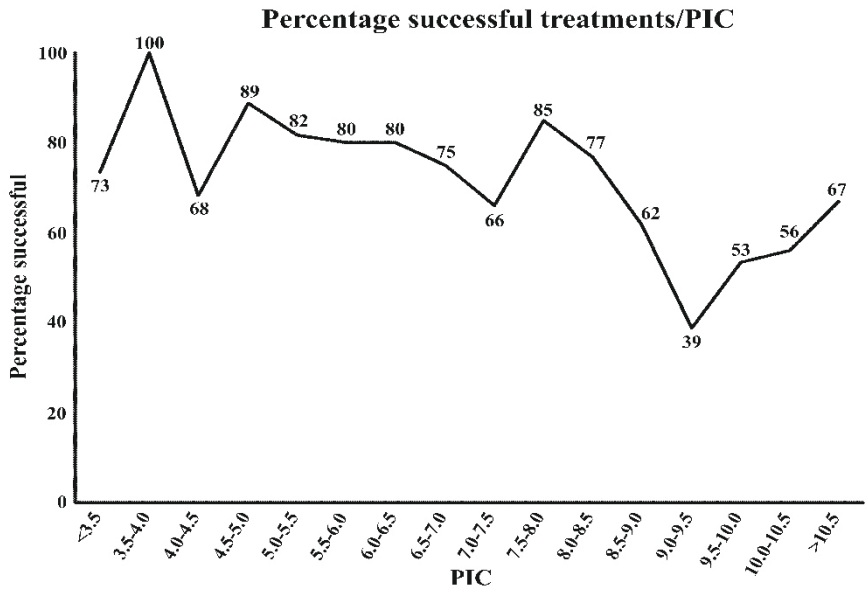

Figure 3 Percentage of successful DCS-treatment per PIC (DCS = dynamic compression system, PIC = pressure of initial correction)

In recent literature Kelly et al. et al. also evaluated the outcomes of DCS-bracing of the last ten years. They showed a much lower success percentage of 50.2% with a loss to follow-up of 41.1%.[24] In our study 12.7% of the patients were lost to follow-up, which is in accordance with the 13.4% noted by Martinez-Ferro et al. Despite this, compliance remains a major problem of the DCS-bracing. There is room for improvement if patients could stay motivated. Earlier studies stated social discomfort and lack of motivation as major contributors to decreased compliance and loss of follow-up.[2, 13, 21] Monitoring (non-)compliance, for example with a real-time monitoring system such as MyPectus[27], could be useful for early detection of non-compliance and could give more insight in the reasons behind it. Kelly et al. experimented with an optical scanning device to monitor the progress of the chest deformity. This could be shown to patients to prove the efficacy of the therapy and encourage them to continue wearing it.[28] Another study did the same with a white light scanner.[29] The use of the Pectus Carinatum Body Image Quality of Life questionnaire (PeCBI-QOL) could also help improve and monitor the body image of patients.[30]

Ravitch and Abramson surgery

Ravitch surgery received excellent outcomes with 92.4% of the patients successfully treated compared to 73.8% in the DCS-bracing group. Despite this, surgery leads to more complications. While no major complications were observed in the bracing group, one in three surgical patients had postoperative complications. Most complications were minor and only 6.7% of the patients with complications needed surgery under general anesthesia. This corresponds with other studies on Ravitch surgery.[15, 18] The results are also similar to recent figures on the Abramson procedure: satisfactory results in 91.0% of patients and complications in 26.5% of patients (with 6.6% of patients needing surgery because of complications)[25]. Despite the Abramson procedure being a minimal invasive technique with only little scars, patients in our study who were suitable for this procedure all received a DCS-brace, except for one patient who preferred the Abramson procedure over DCS-bracing. Patients who were not eligible for bracing were also unfit for Abramson (asymmetric, flaring, high PIC) and therefore underwent Ravitch surgery except for one patient. This patient, despite having a asymmetrical thorax with costal flaring, deliberately opted for Abramson surgery. This patient needed additional surgery to remove the costal flaring which could not be corrected using the Abramson procedure.

Conclusion

In our opinion, DCS-bracing should be the treatment of first choice in patients with PC. With a success rate varying from 67.3-84.2% results are not as high as the percentages achieved by surgery, but the risks are much lower and if conservative therapy fails there is still the possibility to opt for surgery. Bracing patients before their growth spurt should be discouraged, unless they are willing and motivated to wear the brace in retainer mode for many years. Besides PIC, motivation seems to be an important factor influencing the outcomes. If compliance is suspected to be a problem or patients have complex deformities (PA, underlying syndromes like Marfan or Poland, asymmetry, and flaring) primary surgery should be considered.

Limitations

This study might seem a two-era study, before and after the introduction of DCS-bracing in 2013. Before 2013 only patients with major deformities applied for treatment. While the indication for surgery remained the same after 2013, some patients with less severe deformities, despite being advised to choose DCS-bracing, wanted surgery too. Perhaps this explains the differences in success rate of Ravitch surgery before and after 2013. Although only 16 patients were treated before 2013, this could bias the results.

It is difficult to quantify both degree of deformity before treatment and outcomes after treatment in Ravitch and DCS-bracing patients since PC is mainly a cosmetic problem. Although some clinics use 3D imaging to map the deformities this might not be the ultimate solution, since the appearance of the deformity also depends on muscle mass and BMI. The quantification of initial deformities and outcomes remains a hurdle which is yet to be taken. Further investigation into the reasons for discontinuing DCS-bracing and loss of follow-up is needed to optimize DCS-bracing. No evaluation of resolution of symptoms in symptomatic patients was performed in this study because we lack data about the resolution of symptoms after treatment.

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