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Effects of the Nuss procedure on chest wall kinematics in adolescents with pectus excavatum☆

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ABSTRACT

No data are available on the effects of the Nuss procedure on volumes of chest wall compartments (the upper rib cage, lower rib cage and abdomen) in adolescents with pectus excavatum. We used opto-electronic plethysmography to provide a quantitative description of chest wall kinematics before and 6 months after the Nuss procedure at rest and during maximal voluntary ventilation in 13 subjects with pectus excavatum. An average 11% increase in chest wall volume was accommodated within the upper rib cage ($p = 0.0001$) and to a lesser extent within the abdomen and lower rib cage. Tidal volumes did not significantly change during the study. The repair effect on chest wall kinematics did not correlate with the Haller index of deformity at baseline. Six months of the Nuss procedure do increase chest wall volume without affecting chest wall displacement and rib cage configuration.

1. Introduction

Pectus excavatum (PE), the most common congenital chest wall deformity, is characterized by a depression of the anterior chest wall and sternum. PE occurs in more than 1 of every 1000 births with a 3–4:1 male predominance and deformity progression during the period of rapid skeletal growth in early adolescence (Fonkalsrud, 1995; Molik et al., 2001).

Despite well-documented reports to the contrary, the long standing misconception persists that PE is a cosmetic defect with no physiological consequences. Some patients, however, will develop cardiopulmonary symptoms for the first time as adolescents while others will experience a worsening of the symptoms they have endured for years. Surgical repair of PE has evolved significantly during the past 50 years. A minimally invasive technique for repair was described by Nuss et al. (1998) and Nuss (2005, 2008). The method involves the placement of substernal concave bar(s) that will be rotated to elevate the sternum outward. The bar is left in place for 2–3 years while the anterior chest wall remodels. Cosmetic results are reported as good to excellent in 85% of patients. Although the effects of repair on symptoms, lung volumes and cardiopulmonary function have long been described in PE patients (Aronson et al., 2007; Borowitz et al., 2003; Derveaux et al., 1989; Kelly et al., 2007; Kinuya et al., 2005; Lawson et al., 2005; Malek et al., 2006; Morshuis et al., 1994; Nuss, 2008; Sigalet et al., 2007), no data are available on the effects of the Nuss procedure on kinematics of the chest wall compartments: the upper rib cage (RC,p), lower rib cage (RC,a) and abdomen (AB) (Ward et al., 1992). It should be remembered that in case of respiratory abnormalities the chest wall is primarily involved, so that the effect of repair should be assessed mainly by observing chest wall kinematics and possibly chest wall mechanics in PE patients.

The technique for quantifying the magnitude of PE defects has been developed based on measurements taken from chest X-ray, photographs of the body surface, and computed tomography. Discrepancies and controversies on the effects of corrective surgery in PE patients are at least in part attributable to the frequent lack of accurate evaluation of the severity of the deformation (e.g., by radiological indices) and even more by the lack of estimations of relationships between functional variables and the degree of chest deformity. A previous study carried out in adolescents with mild restrictive defect has shown that abnormalities in chest wall kinematics during maximal voluntary ventilation (MVV) were not correlated with the Haller computed tomography (CT) scan severity index (Haller et al., 1987), indicating the contribution of chest wall kinematics to clinical evaluation of PE patients (Binazzi et al., 2012).

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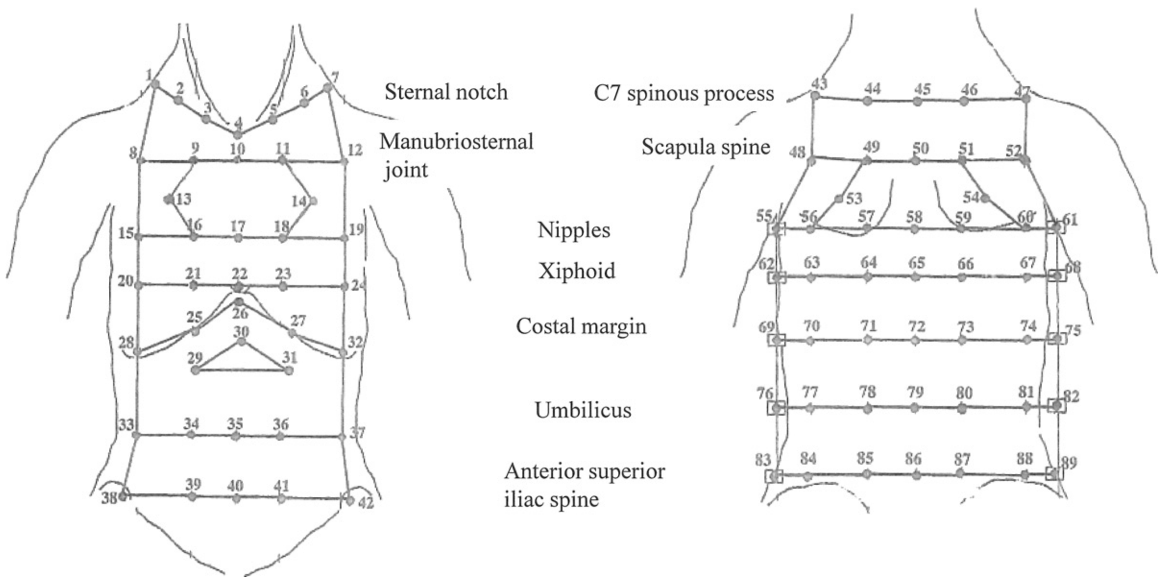

Fig. 1. Position of the 89 reflecting markers. The upper rib cage is separated from the lower rib cage (RC,a) at the level of the xifisternum; the caudal border of RC,a is at the costal margin anteriorly down from the xifisternum and straight across posteriorly at the most caudal level of the lower rib cage.

Questions now arise. Should we wait 2–3 years before assessing repair effects (if any) on chest wall kinematics? Can the Nuss procedure influence timing, and kinematics of the chest wall and rib cage configuration in otherwise healthy subjects? We postulate that the repair effect based on increased chest wall end-expiratory volume, as shown in humans (Jubran and Tobin, 1992; Gorini et al., 1999; Binazzi et al., 2008), does not affect chest wall displacement and dynamic configuration of the rib cage. These questions and the hypothesis have yet to be answered and proved.

Optoelectronic plethysmography (OEP) provides an accurate evaluation of the kinematics of the three chest wall compartments during quiet breathing and hyperventilation in healthy and diseased individuals (Binazzi et al., 2008, 2012; Lanini et al., 2003; Romagnoli et al., 2011). We decided to use OEP to provide a quantitative description of chest wall kinematics before and after the Nuss procedure in young PE subjects.

2. Patients and methods

2.1. Subjects

We studied 13 young male PE patients before and 6 months after the Nuss procedure. None of the patients had comorbidities. The study was approved by the local ethics committee and subjects gave their informed consent.

2.2. Protocol

All patients were well acquainted with the experimental protocol and equipment used. After baseline lung function measurements, optoelectronic measurements of chest wall motion were made with subjects sitting upright in a comfortable armchair. The measurements were made at rest during quiet breathing (QB), which was defined as habitual comfortable breathing, and during MVV. The evaluation was repeated 6 months after implantation of the Nuss bar.

2.3. Pulmonary function tests

Routine spirometry was measured according to ATS/ERS guidelines (Miller et al., 2005). Functional residual capacity (FRC) was measured with a body plethysmograph (Autobox DL, 6200; SensorMedics; Yorba Linda, CA) according to a standardized procedure (Wanger et al., 2005). The normal values for lung function were those of Zapletal et al. (1972). Volume and time components of the breathing pattern were assessed as previously described (Binazzi et al., 2012).

2.4. Maximal voluntary ventilation (MVV)

12 s MVV was performed according to Miller et al. (2005). Tidal volume was set at 40% of vital capacity (VC), with a breathing frequency of ~90 breaths/min. Details of this test have been previously described (Binazzi et al., 2012).

2.5. Optoelectronic measurements

Chest wall volume changes during QB and MVV were assessed by optoelectronic plethysmography (OEP). Details of this technique have been thoroughly described previously (Binazzi et al., 2008, 2012; Lanini et al., 2003; Romagnoli et al., 2011). In brief, six television cameras (three placed 4 m in front and three placed 4 m behind the subjects) tracked the three-dimensional movements of 89 small surface markers attached to the skin of the trunk with double-sided adhesive tape and lit by infrared light-emitting diodes coaxial with lenses of the cameras (Fig. 1).

2.6. Radiographic evaluation

CT of the chest was used to provide an objective assessment of the severity of the deformity. The severity was assessed on the basis of the Haller index which is the ratio of the transverse diameter of the thorax to the antero-posterior diameter at its narrowest point (Haller et al., 1987).

2.7. Analysis of the data

The OEP calculates absolute volumes, and the absolute volume of each compartment at FRC in control conditions was considered as the reference volume. Volumes are reported as absolute values and as changes from the volume at FRC in control conditions. OEP

Table 1
Anthropometric and lung function data of the 13 patients before and after Nuss procedure.

Patients		Age (year)	BMI (Kg/m^2)	TLC L (% pred)	FRC L (% pred)	FEV1 L (% pred)	FEV1/SVC (%)	MVV L (% pred)
Before	Mean ± SD	14 ± 1	19 ± 3	4.78 ± 1.74 (96 ± 17)	2.69 ± 0.75 (97 ± 19)	3.36 ± 0.78 (88 ± 15)	85 ± 7	92 ± 14 (118 ± 18)
After	Mean ± SD	15 ± 1	20 ± 3	5.14 ± 1.87 $p = 0.04$	3.08 ± 0.9 $p = 0.009$	3.32 ± 0.86	88 ± 7	109 ± 26

BMI, body mass index; TLC, total lung capacity; FRC, functional residual capacity; FEV1, forced expiratory volume in 1 s; SVC, slow vital capacity; MVV, maximal voluntary ventilation.

data were averaged over 3 min of quiet breathing and over MVV at different times (start, 4 s, 8 s and end).

The presence of rib cage distortion was established (1) by comparing the time courses in volume of the RC,p (Vrc,p) vs. volume of the RC,a (Vrc,a) and (2) by the phase shift between Vrc,a and Vrc,p when these two volumes were plotted against each other. This was measured as previously adopted and described (Agostoni and Mognoni, 1966; Romagnoli et al., 2011). In this system a phase angle of zero represents a completely synchronous movement of the compartments and 180° total asynchrony. Values are the mean ± SD. Significance of changes in variables during maximal voluntary ventilation was evaluated by two-way analysis of the variance (ANOVA). Com was made by tests for paired data with a 5% significance level. SEM interval confidence was assessed at 95% of significance. All statistical procedures were carried out using the PASW Statistics 18.0 (SPSS inc, Chicago IL, USA).

3. Results

3.1. Changes at baseline

The Nuss procedure resulted in a total lung capacity (TLC) absolute volume increase and an average 16% FRC increase (Table 1). An average 11% ± 11 (from 16.1 L ± 3.9 to 17.9 L ± 5.1; $p = 0.008$) increase in end-expiratory-lung-volume of the chest wall (Vcw,ee) (for individual data see Table E1 in the supplementary material) was mostly accommodated within the RC,p and to a lesser extent within the AB and RC,a; the breathing pattern (VT, Ti, Te, Ttot, VT/Ti and Ti/Ttot) lay unchanged across compartments (Fig. 2).

3.2. Ventilatory response

The increment in end-expiratory volume of the upper rib cage (Vrcp,ee, $p = 0.0001$) along with a lack of changes in Vrca,ee and Vab,ee shifted the chest wall operational volumes upward (Fig. 3) which remained unchanged during MVV before and after the Nuss procedure. As a result, neither VTcw (0.76 L ± 0.26 vs. 0.67 L ± 0.23) nor VTrc,p (%VTcw) (42.6 ± 7.7 vs. 37.9 ± 6.6) changed; and changes were even smaller for VTrc,a and VTab.

The normal values of phase angle degree (FAD) of our lab range from 0 to 14. One patient (#13) exhibited rib cage distortion during MVV before and after the Nuss procedure (see Table E1 in the supplementary material). Two demonstrative cases are shown in Fig. 4: in patient #4 the Nuss procedure did not modify chest wall kinematics at rest and during MVV, whereas in patient #13 a mild rib cage distortion was not affected by the Nuss procedure.

3.3. Relationships

Changes in Vcw,ee or Vrcp,ee did not correlate with baseline lung function or the Haller index.

4. Discussion

This is the first study assessing chest wall kinematics during ventilatory efforts in patients with PE after having undergone the Nuss procedure. The novel findings of this study include the following: (i) six-months after bar insertion, the volume of the upper rib cage (Vrcp,ee) increases, without modifying the breathing pattern, chest wall displacement (VTcw), rib cage configuration and routine spirometry; (ii) the repair effect is not correlated with baseline lung function or with anatomical defect radiological indexes.

4.1. Criticism

The reasons for choosing MVV rather than the cardiopulmonary cycle exercise test (CPET) to assess chest wall kinematics in patients with PE have been provided elsewhere (Binazzi et al., 2012). Briefly, the maximal ventilation occurring during exercise is only about two-thirds of the maximal ventilation that can be achieved for brief periods of voluntary effort (Olafsson and Hyatt, 1969). Also, during maximal CPET considerable negative pleural pressure can occur during inspiration, but rarely, if ever, during expiration does expiratory pleural pressure occur in excess of that required for maximal expiratory flow (Olafsson and Hyatt, 1969). This avoids waste of muscular effort that would occur if expiratory flow were being limited. In contrast, minimal pressure required to achieve maximal flow at a given lung volume (effort independent expiratory flow) is exceeded during MVV.

4.2. Comments on results

Few detailed physiological studies have been carried out in adolescent PE subjects either preoperatively or postoperatively (Binazzi et al., 2012; Mead et al., 1985; Redlinger et al., 2012). This is the first study which investigates chest wall kinematics during ventilatory efforts before and 6 months after the Nuss procedure, with the aim of quantifying the effect of early repair on the dimension of rib cage deformity in young people with PE. OEP provides a detailed description of chest wall kinematics, i.e., the volume at end-expiration and volume displacement (VT) of the pulmonary rib cage (upper rib cage), abdominal rib cage (lower rib cage) and abdomen.

Some conflicting data acquired before the adoption of the Nuss procedure indicate that PE surgery may modify lung function over a long period. A decrease in static and dynamic lung volumes (Cahill et al., 1984; Derveaux et al., 1988), further impairment of restricted lung function despite improvement in symptoms, and a significant increase in the antero-posterior diameter of the chest have long been demonstrated (Derveaux et al., 1988; Morshuis et al., 1994). Conversely, lung function improvements have been shown in some studies (Cahill et al., 1984), but have remained largely unchanged or worsened in others (Gimeno et al., 1984). These discrepancies and controversies were at least in part attributable to the frequent lack of accurate evaluation of the severity of the deformation (e.g., by radiological indices) and even more by the lack of estimation of relationships between lung functional variables and degree of chest deformity (Shamberger and Welch, 1988).

Improvement (Borowitz et al., 2003; Kelly et al., 2007; Kinuya et al., 2005; Lawson et al., 2005; Sigalet et al., 2007) or no change (Aronson et al., 2007) in pulmonary function, and increase in cardiac filling and stroke volume (Kelly et al., 2007; Malek et al., 2006;

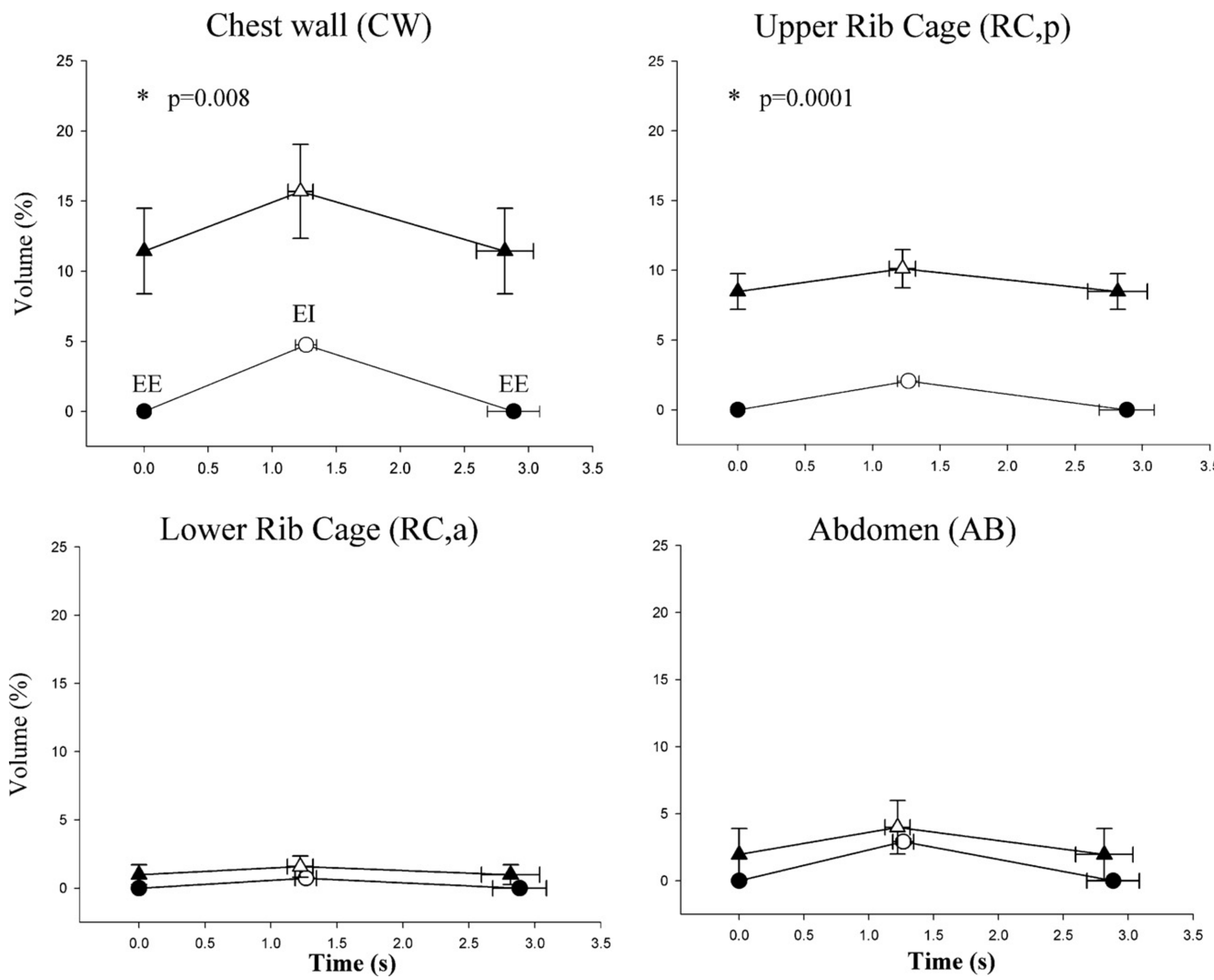

Fig. 2. Percent changes from control in volumes and timing components of the chest wall compartments during quiet breathing. Circles: values before the Nuss procedure; triangles: values after the Nuss procedure. Closed symbols: end expiration (EE); open symbols: end inspiration (EI). Values are means and SE. *Compare EE and EI before and after the Nuss procedure.

Sigalet et al., 2007) have been reported after the Nuss procedure. According to Nuss (2008), the reasons for these discrepancies are multifactorial and include the size of the cohort being studied, the duration of the study and the severity of PE. In the Nuss series of more than 900 cases the preoperative resting pulmonary studies showed a distribution of forced vital capacity markedly shifted to the left, and a significant postoperative correction (Kelly et al., 2007; Lawson et al., 2005), with the distribution being shifted toward normal. Yet the value of routine testing of pre- and postoperative lung function in PE patients has been questioned (Kubiak et al., 2007).

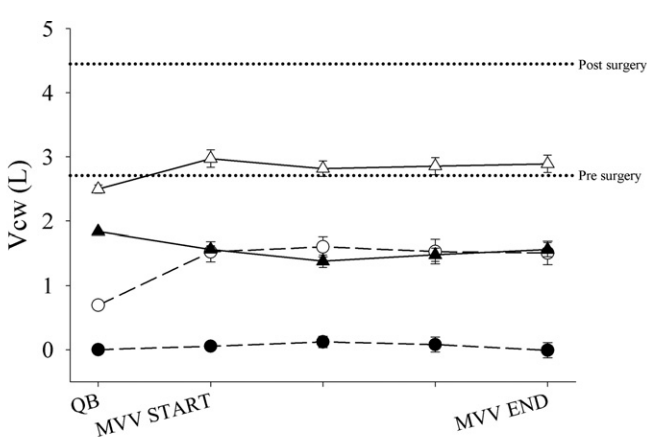

Fig. 3. Changes in chest wall volumes from quiet breathing (QB) to end of maximal voluntary ventilation (MVV). Circles: values before the Nuss procedure; triangles: values after the Nuss procedure. Closed symbols: end-expiratory-volumes; open symbols: end-inspiratory-volumes. Dotted line: average total lung capacity.

The new and important data collected by using OEP and functional consequences of the Nuss procedure are as follows. End-expiratory volume of the chest wall (Vcw,ee) increased by 11% and was allocated mostly to the upper rib cage and to a lesser extent to the abdomen and lower rib cage. Based on increased Vcw,ee the repair effect did not affect chest wall displacement and dynamic configuration of the rib cage in PE patients without rib cage distortion. These findings require two major comments. On the one hand, unchanged inspiratory reserve volume associated with an increase in end-expiratory volume indicates the need to keep end-inspiratory volume far from the increased total lung capacity (Fig. 3) with the aim of increasing inspiratory reserve volume and lower inspiratory muscle activation. Such a kinematic pattern which leaves driving and timing components of the breathing pattern unmodified (Fig. 2) prevents dynamic volume constraints and effort-related sensory consequences (breathlessness) during hyperventilation. On the other hand, the size of abdominal volume (Vab) provides information on rib cage configuration. A marked

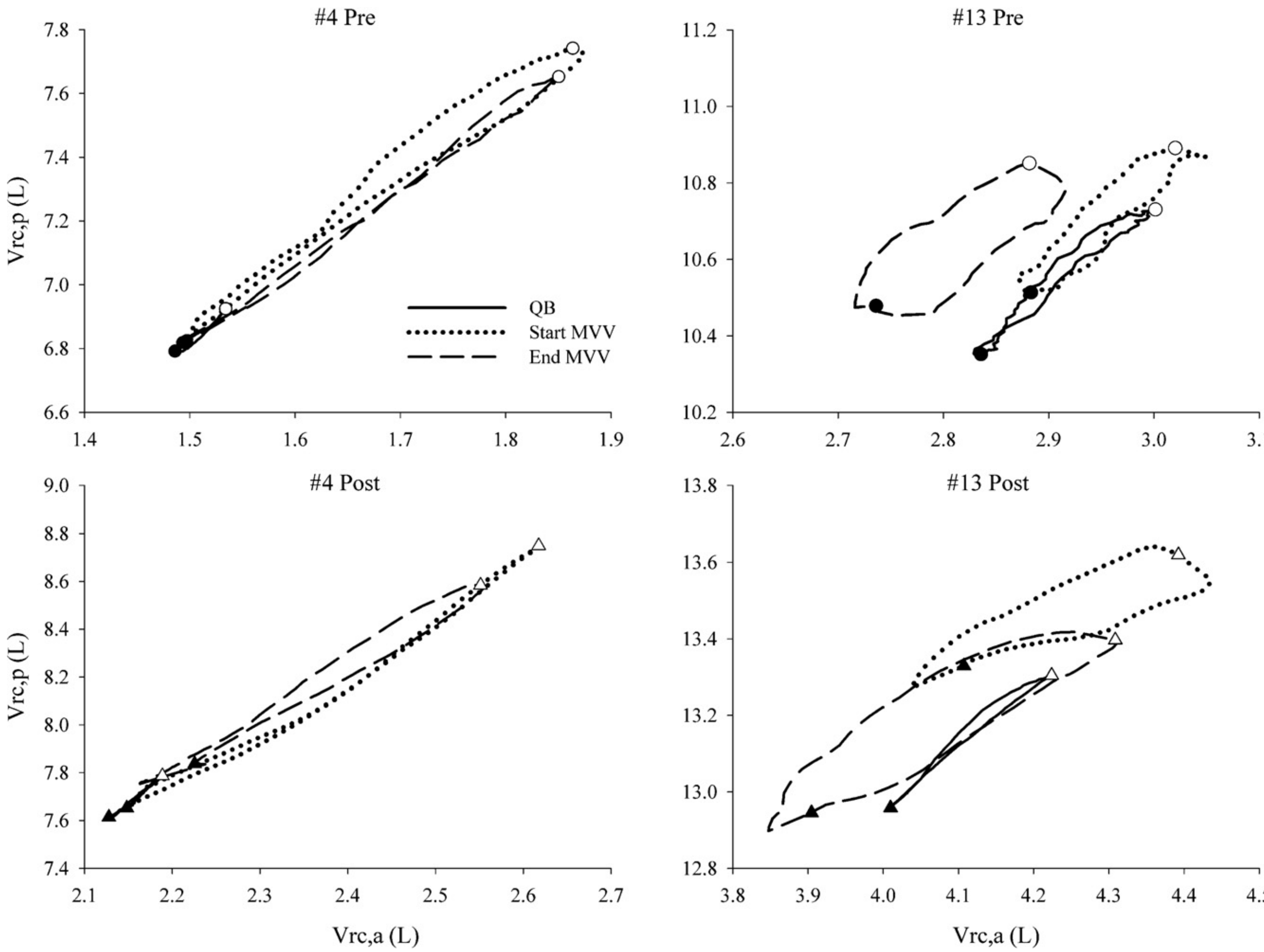

Fig. 4. In two PE patients rib cage distortion is calculated by plotting change in Vrc,p to change in Vrc,a during quiet breathing (QB) and during maximal voluntary ventilation (MVV). Vrc,p: volume of the upper rib cage; Vrc,a: volume of the lower rib cage. Closed symbols: end expiration; open symbols: end inspiration. For explanation see Section 2.

increase in Vab has been recently reported to be associated with increased chest wall motion in the area of pectus defect after repair in PE patients during spirometry (Redlinger et al., 2012). By contrast, no changes in Vab were found in our study and in a former one before repair in patients without RC distortion (Binazzi et al., 2012).

Binazzi et al. (2012) have recently shown no correlation between the Haller index and dynamic assessment of preoperative deformity, and indicate that chest wall kinematics contribute to clinical evaluation of PE patients. Likewise, in this study we found no correlation between the Haller index and repair effects on chest wall kinematics. An increase in chest wall (Vcw,ee) or upper rib cage (Vrcp,ee) volumes did not correlate with lung volumes at baseline. These results, however, should be interpreted in light of the limitations imposed by the mild level of the functional abnormalities found preoperatively in our patients.

OEP allows both the clinician and surgeon to assess the effects of the procedure longitudinally over a 3-year period.

5. Conclusion

Six months following the Nuss procedure in adolescents with PE, end-expiratory volume of the upper rib cage increases without affecting chest wall displacement (VTcw) or rib cage configuration. Neither routine lung function nor the radiological deformity index at baseline are able to predict the repair effect of the Nuss procedure.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.resp.2012.05.015.

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