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ORIGINAL ARTICLE

Outcomes of the Nuss procedure in children with pectus excavatum: 14 years of experience

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Abstract

Background: We aimed to assess the effectiveness of the Nuss procedure for pectus excavatum (PE) and explore the impacts of sex and age on outcomes.

Methods: We retrospectively reviewed 594 consecutive children ≤18 years of age who underwent the thoracoscopy-assisted Nuss technique between January 2006 and July 2019. The severity of pectus deformity was calculated according to the Haller index (HI). The classification of PE and clinical data including complications was analyzed.

Results: Of the 594 patients, 456 (76.8%) were boys and 138 (23.2%) were girls. The mean age at surgery was 10.0 ± 5.0 years. The most common types of PE were 1A and 2A2 according to Park classification. Intraoperative and postoperative complication rates were 2/594 (0.3%) and 74/594 (12.5%), respectively. The most common complication was bar displacement. The bar was removed in 414 patients 3.5 ± 0.8 years later. The mean preoperative HI, postoperative HI with bar, and HI after bar removal were 4.2 ± 1.7, 2.4 ± 0.3, and 2.7 ± 0.5, respectively. Compared to the preoperative HI, both the postoperative HI with bar and HI after bar removal were significantly lower ($p < 0.001$). For preschool-age children, the preoperative HI was significantly higher ($p = 0.027$) and the change in HI significantly improved compared to school-age children ($p = 0.004$). Boys and adolescents needed significantly more bars and stabilizers.

Conclusion: Surgical correction of PE using the Nuss procedure is a safe procedure and improves the HI in children of different ages, even in those younger than 6 years of age.

Keywords: Haller index; Nuss procedure; Pectus excavatum; Pediatric

1. INTRODUCTION

In 1998, a minimally invasive technique was introduced by Donald Nuss[1] to remodel the anterior chest wall deformity in pectus excavatum (PE) by inserting a retrosternal metal bar. Because the Nuss procedure offers technical simplicity and better cosmetic outcomes, it has become widely accepted as the preferred surgical technique to treat PE. Although several studies have indicated its efficacy and safety in children and adults, an overall complication rate of 2% to 43%[2,3] and a variety of life-threatening complications have also been reported.[3] Modifications of the technique such as using a stabilizer and more bars at different levels of the chest have significantly reduced the complication rate.[4,5]

PE often worsens at about the time a child starts puberty. However, the ideal age for elective repair of PE is still controversial. The timing of surgery is usually based on clinical symptoms, psychosocial disposition of the patients and parents, and expected surgical outcomes. Repair of PE is generally undertaken in children at the beginning of puberty,[6,7] however the early repair of PE in children older than 3 years of age has also been reported to be safe and effective.[8]

PE is more common in males than in females, however, data regarding the relationship between sex and outcomes in children undergoing surgical repair of PE are limited. Therefore, the purpose of this study was to explore the surgical results and postoperative complications of the thoracoscopy-assisted Nuss procedure to treat various types of PE among different age groups of children. Furthermore, we explored the impact of sex on clinical characteristics and outcomes.

2. METHODS

2.1. Patients and preoperative evaluation

We analyzed the demographics, comorbidities, complications, and outcomes of 594 consecutive patients ≤18 years of age with PE who underwent the Nuss procedure between January 2006 and July 2019. All patients underwent a complete evaluation including history taking, echocardiogram, electrocardiogram, chest roentgenogram, and non-contrast chest computed tomography. Computed tomography was used to determine the type and severity of thoracic cage deformity according to Park

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classification, heart deviation, and Haller index (HI).[9] HI scores of 2.0 to 3.2, 3.2 to 3.5, and ≥3.5 were considered to indicate mild, moderate, and severe PE, respectively. The criteria for surgery were severe body image disturbances, symptoms of cardiopulmonary limitation, and recurrent PE from previous surgery. The study population was divided into three subgroups: <6 years (preschool-age), 6 to 12 years (school-age), and 12 to 18 years (adolescent). The research was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This study was approved by the Institutional Review Board of Taipei Veterans General Hospital (NO.: 2022-05-009BC) and the requirement for individual consent for this retrospective analysis was waived.

2.2. The surgical technique

Under general anesthesia, the patient was intubated with a single-lumen endotracheal tube. The patient was placed in the supine position with both arms extended above their head with a 90° angle at the shoulders as well as the elbows, and the forearms were wrapped with towels and hung on the screen frame to ensure an adequate operative field. The deepest point of the sternum was identified and the hinge points, bar entry/exit points, and incision lines were marked on the skin. An optimal length of template was then used to design the correction, and we choose bars 1 or 2 cm longer than the template. The crane technique for sternal elevation was routinely used, with wire sutures passed through the sternum and then hooked onto a rib approximator for elevation of the sternum. A thoracoscope was inserted via a trocar from the right side through the whole procedure to guide the tunnel dissection and to monitor bar placement. A water seal with positive pressure ventilation was used to evacuate the pneumothorax, and a chest tube was rarely required. The typical length of hospital stay was 6 days, including surgery, postoperative intensive care unit stay for 1 day, respiratory training, rehabilitation prescriptions, and discharge preparations. The numbers of bars and stabilizers, surgical complications, and hospital courses were recorded and analyzed.

2.3. Postoperative evaluation and bar removal

To protect the patient against radiation overexposure, we estimated the HI by measuring the minimum anteroposterior chest dimensions and maximum transverse diameter in two-view chest radiographs postoperatively and after the bar had been removed.[10] The criteria for bar removal were (1) the size of the bar was smaller than the adjacent ribs in serial follow-up; (2) surgical complications occurred due to bar displacement, intractable implant infections, unbearable pain or thoracic trauma; and (3) the bar had been placed for more than 3 years.

2.4. Statistical analysis

Continuous variables are expressed as the mean ± SD. Paired *t* tests were used for preoperative and postoperative comparisons of the HI in each subject. Continuous variables were compared between groups using the independent *t* test and one-way analysis of variance (ANOVA), followed by Tukey post hoc test. Categorical variables were analyzed using Fisher exact test or the chi-square test. All statistical analyses were performed using SPSS (version 18.0; SPSS Inc., Chicago, IL). Results were considered to be statistically significant at $p < 0.05$.

3. RESULTS

3.1. Demographics and clinical characteristics

A total of 594 patients who had undergone the Nuss procedure for PE were enrolled, of whom 456 (76.8%) were boys and 138 (23.2%) were girls (male to female ratio, 3.3:1). The mean age at surgery was 10.0 ± 5.0 (range, 1.7-18) years. The distribution of age at the time of surgery is shown in Fig. 1. The most common elective age for repair of PE was 12 to 18 years (n = 252,

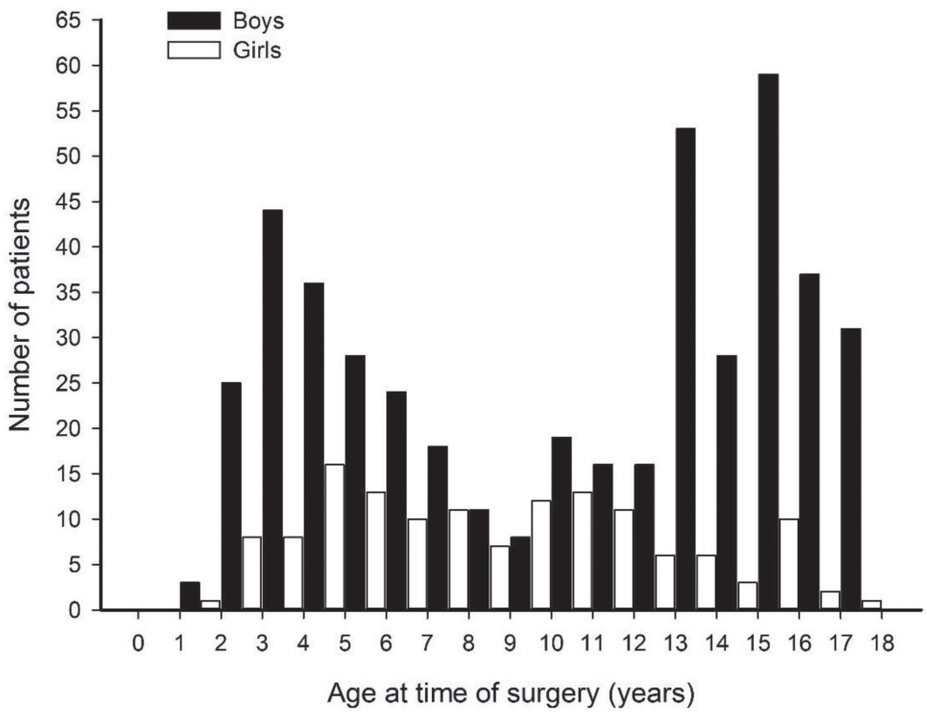

Fig. 1 Age distribution at time of surgery for pectus excavatum.

42.4%), followed by under 6 years (n = 182, 30.6%) and 6 to 12 years (n = 160, 26.9%). Other associated abnormalities were found in 106 (17.8%) patients, including 54 (9.1%) with scoliosis, 46 (7.7%) with straight back syndrome, 6 (1.0%) with Marfan syndrome, 4 (0.7%) with congenital heart diseases, 4 (0.7%) with chromosome abnormalities, 2 (0.3%) with congenital emphysema, 1 (0.2%) with pulmonary sequestration, 2 (0.3%) with diaphragmatic hernia, 2 (0.3%) with neurofibromatosis, 2 (0.3%) with Poland syndrome, 1 (0.2%) with pyloric stenosis, and 1 (0.2%) with bilateral hearing loss. The most common types of PE were 1A (n = 203, 34.2%) and 2A2 (n = 140, 23.6%) followed by 1B (n = 108, 18.2%) and 2B (n = 42, 7.1%) according to Park classification.

3.2. Comparison of preoperative and postoperative HI and clinical features

The HI and grade of PE preoperatively, postoperatively, and after bar removal are shown in Table 1. The preoperative HI was 4.2 ± 1.7, and 98 (16.5%) of the patients had mild PE, 112 (18.9%) had moderate PE, and 384 (64.6%) had severe PE. A total of 419 patients (70.5%) required 1 bar insertion, 161 patients (27.1%) required 2 bars, and 14 (2.4%) patients required 3 bars at the time of surgery. Overall, the mean numbers of implanted bars and bar stabilizers were 1.3 ± 0.5 and 0.9 ± 1.0, respectively. The postoperative mean HI was 2.4 ± 0.3, showing a significant improvement compared with the preoperative HI ($p < 0.001$). The results showed significant postoperative improvements in the grade of PE, including 36 (6.1%) with normal HI, 549 (92.4%) with mild PE, 7 (1.2%) with moderate PE, and 2 (0.3%) with severe PE.

3.3. Complications

The intraoperative complication rate was 2/594 (0.3%), including one patient with a pericardial injury and one with a diaphragm/liver injury. There were no cases of mortality. Postoperative complications occurred in 74 patients (12.5%).

Table 1

HI and grade of PE before and after the Nuss procedure and removal of the bar according to age at time of surgery

	Total	Age subgroup			*p*
	n = 594	<6 y n = 182 (Preschool-age)	6-12 y n =160 (School-age)	12-18 y n = 252 (Adolescent)	
Sex					<0.001[a]
Male	456 (76.8%)	136 (74.7%)	96 (60.0%)	224 (88.9%)	
Female	138 (23.2%)	46 (25.3%)	64 (40.0%)	28 (11.1%)	
HI					
Preoperative	4.2 ± 1.7	4.5 ± 2.0	4.0 ± 1.9*	4.1 ± 1.2	0.027[b]
Postoperative	2.4 ± 0.3	2.3 ± 0.3	2.4 ± 0.3*	2.5 ± 0.3*,**	<0.001[b]
After removal of the bar	2.7 ± 0.5	2.7 ± 0.5	2.6 ± 0.3	2.8 ± 0.5	0.415[b]
Difference in HI					
Postoperative minus preoperative HI	−1.8 ± 1.6	−2.2 ± 1.9	−1.6 ± 1.9*	−1.6 ± 1.1*	0.001[b]
After removal of the bar minus preoperative HI	−1.9 ± 2.7	−2.3 ± 2.6	−1.9 ± 3.9	−1.2 ± 1.2	0.264[b]
Number of bars implanted					<0.001[a]
One bar	419 (70.5%)	182 (100%)	150 (93.8%)	87 (34.5%)	
Two bars	161 (27.1%)	0 (0.0%)	10 (6.3%)	151 (59.9%)	
Three bars	14 (2.4%)	0 (0.0%)	0 (0.0%)	14 (5.6%)	
Mean number of bars implanted	1.3 ± 0.5	1.0 ± 0.0	1.1 ± 0.2	1.7 ± 0.6*,**	<0.001[b]
Mean number of bar stabilizers used	0.9 ± 1.0	0.1 ± 0.2	0.3 ± 0.6*	1.9 ± 0.7*,**	<0.001[b]
Grade of PE preoperatively					0.039[a]
Mild (2.0-3.2)	98 (16.5%)	29 (15.9%)	35 (21.9%)	34 (13.5%)	
Moderate (3.2-3.5)	112 (18.9%)	28 (15.4%)	37 (23.1%)	47 (18.7%)	
Severe (≥3.5)	384 (64.6%)	125 (68.7%)	88 (55.0%)	171 (67.9%)	
Grade of PE postoperatively					0.031[a]
Normal (<2.0)	36 (6.1%)	18 (9.9%)	8 (5.0%)	10 (4.0%)	
Mild (2.0-3.2)	549 (92.4%)	163 (89.6%)	150 (93.8%)	236 (93.7%)	
Moderate (3.2-3.5)	7 (1.2%)	0 (0.0%)	1 (0.6%)	6 (2.4%)	
Severe (≥3.5)	2 (0.3%)	1 (0.5%)	1 (0.6%)	0 (0.0%)	
Grade of PE after removal of the bar					0.342[a]
Normal (<2.0)	2 (1.9%)	2 (4.4%)	0 (0.0%)	0 (0.0%)	
Mild (2.0-3.2)	85 (82.5%)	34 (75.6%)	25 (92.6%)	26 (83.9%)	
Moderate (3.2-3.5)	9 (8.7%)	4 (8.9%)	1 (3.7%)	4 (12.9%)	
Severe (≥3.5)	7 (6.8%)	5 (11.1%)	1 (3.7%)	1 (3.2%)	
Duration of bar placement for those removed	3.5 ± 0.8	3.8 ± 0.9	3.6 ± 0.9	3.3 ± 0.6*,**	<0.001[b]
Postoperative complications	74 (12.5%)	26 (14.3%)	19 (11.9%)	29 (11.5%)	0.665[a]

The data are expressed as the mean ± SD or number (%).

ANOVE = analysis of variance; HI = Haller index; PE = pectus excavatum.

[a]Fisher exact test or chi-square test for categorical variables.

[b]ANOVA for continuous variables between groups.

*$p < 0.05$ compared with preschool-age group.

**$p < 0.05$ compared with school-age group.

The most common postoperative complication was bar displacement (n = 41, 6.9%), followed by implant infection (n = 12, 2.0%), wound infection (n = 10, 1.7%), wire disruption (n = 10, 1.7%), pneumothorax (n = 6, 1.0%), hemothorax (n = 3, 0.5%), and pleural effusion (n = 2, 0.3%).

3.4. Outcome of bar removal and recurrence rate

The bar was removed in 414 patients, with a mean time from implantation to removal of 3.5 ± 0.8 years. The mean HI after bar removal was 2.7 ± 0.5. Compared to the preoperative HI, the postoperative HI after bar removal was significantly lower ($p < 0.001$). The recurrence rate after bar removal was 4.1% (n = 17).

3.5. Clinical features according to age group

Clinical information of the patients according to age at the time of surgery is shown in Table 1. Of the 182 preschool-age children, 160 school-age children and 252 adolescents, 136 (74.7%), 96 (60.0%), and 224 (88.9%) were boys, respectively. There was significant male predominance in all three groups. Regarding the grades of HI, the preschool-age group had the highest preoperative HI (4.5 ± 2.0), lowest postoperative HI (2.3 ± 0.3), and greatest difference in the change in HI postoperatively (–2.2 ± 1.9). Significant differences in preoperative and postoperative HI were found between the three age groups ($p = 0.027$ and $p < 0.001$, respectively). A post hoc Tukey test showed that the preoperative HI was significantly higher in the preschool-age group compared to the school-age group ($p = 0.027$), while there was no significant difference between the adolescent group and school-age group. Compared to the preschool and school-age groups, the adolescent group had a significantly higher postoperative HI ($p < 0.001$ and $p = 0.001$, respectively). The school-age group also had a significantly higher postoperative HI compared to the preschool-age group ($p = 0.026$). In addition, there were significantly greater changes in postoperative HI between the preschool-age and school-age (–2.2 ± 1.9 vs –1.6 ± 1.9, $p = 0.004$) and adolescent (–2.2 ± 1.9 vs –1.6 ± 1.1, $p = 0.002$) groups. The mean numbers of bar implanted were 1.0 ± 0.0, 1.1 ± 0.2, and 1.7 ± 0.6 for the preschool-age, school-age, and adolescent groups, respectively. A post hoc Tukey test showed that the adolescent group needed significantly more bars for PE surgery than the other two groups (adolescent vs preschool-age group, $p < 0.001$; adolescent vs school-age group, $p < 0.001$). The mean numbers of stabilizers used were 0.1 ± 0.2, 0.3 ± 0.6, and 1.9 ± 0.7 for the preschool-age, school-age, and adolescent groups, respectively. The older children needed significantly more stabilizers (adolescent vs preschool-age group, $p < 0.001$; adolescent vs school-age group, $p < 0.001$; school-age vs preschool-age group, $p < 0.001$). Among the patients who underwent bar removal, the bars were removed at a mean of 3.3 ± 0.6 years after the Nuss procedure in the adolescent group, which was significantly shorter than that in the preschool-age ($p < 0.001$) and school-age groups ($p = 0.016$). Differences in HI and grade of PE after removing the bar among the three age groups did not reach statistical significance. Postoperative complications were most frequently observed in the preschool-age group (14.3%), followed by the school-age group (11.9%) and adolescent group (11.5%). However, there was no significant difference in postoperative complication rate among the three groups ($p = 0.665$).

3.6. Clinical features according to sex

The clinical information of the patients according to sex is shown in Table 2. The mean age at time of surgery was significantly older in the boys than in the girls (10.4 ± 5.1 vs 8.5 ± 4.0 years, $p < 0.001$). Boys needed more bars for PE repair (1.4 ± 0.5 vs 1.1 ± 0.3, $p < 0.001$), and more stabilizers were used in the boys than in the girls (1.1 ± 1.1 vs 0.5 ± 0.8, $p < 0.001$). However, differences between the boys and girls in HI and grade of PE preoperatively, postoperatively, and after removal of the bar did not reach statistical significance. Both boys and girls had a lower HI with the bar and a lower HI after bar removal, and the changes in HI were similar.

Table 2

HI and grade of PE preoperatively, postoperatively, and after removal of the bar by sex

	Boys n = 456	Girls n =138	*p*
Mean age at time of surgery	10.4 ± 5.1	8.5 ± 4.0	<0.001
HI			
Preoperative	4.2 ± 1.5	4.2 ± 2.2	0.826
Postoperative	2.4 ± 0.3	2.4 ± 0.3	0.187
After removal of bar	2.7 ± 0.4	2.7 ± 0.7	0.878
Difference in HI			
Postoperative minus preoperative HI	–1.8 ± 1.5	–1.8 ± 2.1	0.986
After removal of the bar minus preoperative HI	–1.7 ± 2.2	–2.4 ± 4.4	0.342
Mean number of bars implanted	1.4 ± 0.5	1.1 ± 0.3	<0.001
Mean number of bar stabilizers used	1.1 ± 1.1	0.5 ± 0.8	<0.001
Grade of PE preoperatively			0.979
Mild (2.0-3.2)	76 (16.7%)	22 (15.9%)	
Moderate (3.2-3.5)	86 (18.9%)	26 (18.8%)	
Severe (≥3.5)	294 (64.5%)	90 (65.2%)	
Grade of PE postoperatively			0.816
Normal (<2.0)	28 (6.1%)	8 (5.8%)	
Mild (2.0-3.2)	422 (92.5%)	127 (92.0%)	
Moderate (3.2-3.5)	5 (1.1%)	2 (1.4%)	
Severe (≥3.5)	1 (0.2%)	1 (0.7%)	
Grade of PE after removal of bar			0.736
Normal (<2.0)	2 (2.4%)	0 (0.0%)	
Mild (2.0-3.2)	68 (81.9%)	17 (85.0%)	
Moderate (3.2-3.5)	8 (9.6%)	1 (5.0%)	
Severe (≥3.5)	5 (6.0%)	2 (10.0%)	

The data are expressed as the mean ± SD or number (%).
HI = Haller index; PE = pectus excavatum.

4. DISCUSSION

PE is the most common morphological chest wall abnormality. Although most cases of PE are sporadic, a familial predisposition has been reported.[11] The reported pathophysiology of PE includes overgrowth of costal cartilage, abnormal flexibility of the sternum, overgrowth of the ribs, defective metabolism in the sternocostal cartilage, or an imbalance between the respiratory muscles.[12] PE may be associated with monogenic syndromes such as Marfan syndrome,[13] Noonan syndrome, and other connective tissue diseases.[14] Cases of non-syndromal PE can be considered to be an isolated anomaly, however it is usually accompanied by other thoracic cage deformities, congenital heart diseases, respiratory system defects, and underdeveloped chest muscles, that is, Poland syndrome. Like previous studies,[15,16] in our cohort, there were high rates of scoliosis (9.1%) and straight back syndrome (7.7%) associated with PE. The high prevalence (17.8%) of comorbidities supports the need for screening strategies for associated conditions in patients with PE.

Precisely assessing the dysmorphology of PE may help determine treatment options. Several classifications of PE subtype have been reported in prior studies. Both the Park et al[9] and Cartoski et al[17] classification systems are based on subjective

morphological findings such as localized/diffuse or symmetric/asymmetric depression, and sternal torsion may result in inter-observer variability. Choi et al[18] used computed tomography to obtain objective parameters such as asymmetry index, angle of Louis, flatness index, pectus index, and sternal torsion angle. In this study, 1A (34.2%) and 2A2 (23.6%) were the most common types of PE according to Park classification.

The incidence of PE is more common in males, as also shown in this study with a male to female ratio of 3.3:1. PE tends to progress and often worsens as a child starts puberty. Untreated PE has cosmetic as well as progressive cardiopulmonary impacts on the patients. The optimal timing of the Nuss procedure is based on the severity of deformity, clinical symptoms, the anticipated end result, potential complications, and recurrence rate. There is no absolute consensus on the optimal age for surgical correction.[19,20] Some experts recommended that the optimum age for PE repair is at 6 to 12 years,[21,22] while others advise that the surgical intervention should be performed at around 10 to 14 years of age to decrease growth-related recurrence.[19,23] The biggest concern with young children undergoing surgery is over iatrogenic complications influencing thoracic growth. Unlike the more invasive Ravitch procedure,[24] the Nuss procedure does not interfere with the growth plate. Therefore, children of preschool age are not currently contraindicated for the Nuss procedure. Some surgeons prefer to perform the surgery earlier in symptomatic children older than 3 years of age.[8,25] The sternum in younger children is more flexible than that in adolescents, which can contribute to a better postoperative effect, less postoperative pain, and early relief of cardiopulmonary compression. In the present study, 73.1% and 26.9% of the patients were 6 to 18 and <6 years of age, respectively. We found that the children who underwent surgery before school age had more severe PE, as reflected by a higher HI. In this group, we choose a shorter bar length of about 3/8 of the thoracic circumference instead of 1/2 which was used in the others, to achieve the best stability and thoracic contours. All of the children under 6 years of age only required one bar, and the number of bars implanted increased with age, which is consistent with other studies. Furthermore, the bars were placed for a longer period of time in the preschool-age and school-age groups to avoid recurrence after removal. Postoperatively, the HI in the preschool-age children was lower than that in the school-age children and adolescents. The results of our study are consistent with previous studies showing that the costal cartilage is more compliant. Furthermore, the postoperative complications were similar among the three age groups. Taken together, these findings suggest that the Nuss procedure is safe and effective for symptomatic children <6 years of age.

In terms of the age at surgery, we found that the girls received surgical correction earlier than the boys. The possible reasons are that girls' body weight is relatively lighter, thoracic deformation is more obvious, and puberty occurs earlier, so that a rapid deterioration has a greater negative impact on self-esteem and they seek medical treatment. In contrast, girls tend to be more reluctant to talk about problems such as concave contours as they become older. In addition, the progression of PE is not easily discovered by parents in older girls, so the number of corrections is reduced. Most boys receive surgery during adolescence when their height and chest size are significantly larger, and hence more bars and stabilizers are required to achieve the same efficacy of correction.

The traditional supine position with both arms extended horizontally is prone to cause brachial plexus injuries after surgery.[26,27] We placed the patient's arms with elbow flexion in front of their heads, which greatly reduced the complication of hand numbness. Thoracoscopy is necessary throughout the operation to determine the path of the dissection, to minimize injury to nearby tissue and organs, and even to avoid pulmonary entrapment caused by bar flipping. Postoperative regular follow-up is mandatory to identify and deal with surgery-related complications. From an imaging point of view, bar displacement is the most common complication,[28] and reoperation is considered if the displacement compromises the corrections. Unexpected and violent impacts to the chest can lead to hemopneumothorax requiring urgent treatment. Rarely, patients may be allergic to the implanted metal materials, resulting in poor wound healing, skin erosion and infections. Most complications were resolved by conservative treatment without having to remove the implants. All of the subjects with recurrence in our study kept their bars in place for more than 3 years. Further studies are needed to elucidate the factors associated with recurrence and the optimal time of bar removal.

In conclusion, the Nuss procedure is always challenging and needs experience and continuous modification of surgical skills to ensure safety. Many experts in this area have invented various kinds of new implants and modalities to improve the existing deficiencies and shortcomings. The Nuss procedure continues to have excellent results for pediatric PE correction. Surgical plans and expected goals need to be tailored to each patient, especially when the patient also has other musculoskeletal abnormalities.

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