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Musculoskeletal deformities after thoracic surgery in children: An observational long-term follow-up study☆,☆☆

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ABSTRACT

Purpose: This study reports the incidence, severity, and predictors of musculoskeletal deformities (MD), including scoliosis and chest wall anomalies, following thoracic procedures in children.
Methods: Children younger than 14 years who had thoracic surgery between 1997 and 2012 and had no other predispositions to MD, underwent longitudinal follow-ups with dedicated musculoskeletal examination performed in an esophageal atresia, orthopedic, or research clinic. Incidence of MD was calculated, and logistic regression methods were used to determine independent predictors, including sex, gestational age, age at procedure, serratus anterior muscle division, and chest tube placement.
Results: The study cohort consisted of 104 patients followed for a median of 10.8 years (range 3–21). A total of 56 MD developed in 41 patients (39%), including scapular winging (24; 23%), scoliosis (17; 16%), and chest wall anomalies (15; 14%). The majority of MD were subclinical, with only 8 patients [8% (6 thoracotomies, 2 thoracoscopies)] requiring intervention. Among patients who underwent thoracotomies (93, 89%), serratus anterior muscle division was the only significant predictor of the development of MD [OR 8.9; 95% CI 2.8–32.6].
Conclusion: Musculoskeletal deformities develop in a significant proportion of children following thoracic surgery, but most are subclinical. A muscle-sparing technique decreases the incidence of these deformities.
Type of Study: Prospective Cohort Study.
Level of Evidence: Level II.

Ample evidence exists associating pediatric thoracotomy with an increased risk of developing musculoskeletal deformities, including chest wall deformities such as pectus carinatum or pectus excavatum, scapular anomalies, and scoliosis, with a prevalence of up to 50% [1–5]. However, these studies typically reflect older thoracotomy techniques that were characterized by generous muscle division and rib fusion [1–10].

The standard thoracotomy approach in neonates and children is undertaken using a posterolateral incision extending from the anterior axillary line and curving behind the scapula. It also involves division of the latissimus dorsi and serratus anterior muscles. This may subsequently lead to injury of the long thoracic nerve and consequent atrophy of the serratus anterior muscle, resulting in many of the musculoskeletal sequelae associated with thoracotomies [1,4,6,7].

Modern pediatric thoracotomy techniques attempt to preserve the major chest wall muscles by either their retraction or disinsertion [11–13]. Ribs are never resected and approximation of the chest wall is carefully performed to avoid rib fusion. Moreover, the last two decades have seen an increased utilization of thoracoscopy by pediatric general and cardiac surgeons, with improvements in postoperative pain management, length of hospital stay, and cosmesis [14,15]. It has been assumed that these minimally invasive techniques will also significantly decrease or eliminate musculoskeletal defects. However, this assumption has not been studied. Rather, this inference stems from comparisons of thoracoscopic outcomes with thoracotomies performed in the 1980s and 90s [14–16].

Few studies have evaluated whether modern thoracotomy and thoracoscopy techniques are associated with the same risks of long-term musculoskeletal morbidities as compared with the traditional muscle-severing thoracotomy. We hypothesize that these modern techniques are associated with fewer musculoskeletal deformities compared to the standard thoracotomy. Therefore, the purpose of this study was to conduct a contemporary analysis and comparison of the incidence,

☆ *How this paper will improve care: This study demonstrates a high incidence of musculoskeletal deformities following thoracic surgery in children, but the majority are sub-clinical. A serratus anterior muscle-sparing approach significantly mitigates the incidence of these deformities, resulting in musculoskeletal outcomes similar to minimally invasive approaches.*

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severity, and predictors of musculoskeletal deformities in children who have undergone a thoracic procedure, specifically standard thoracotomy, muscle-sparing thoracotomy, and thoracoscopy.

1. Methods

1.1. Study population

The study population consisted of children who had undergone a thoracic procedure, either thoracotomy or thoracoscopy, prior to the age of 14 years during the 15-year period between January 1, 1997 and January 1, 2012, and who survived for a minimum of 3 years following the procedure. Patients with congenital anomalies known to predispose to musculoskeletal deformities (e.g. butterfly vertebrae, congenital diaphragmatic hernia, congenital chest wall deformity) were excluded. Patients who had undergone additional procedures (e.g. sternotomy, chest wall resection), and those with developmental conditions (e.g. cerebral palsy) that predispose to such anomalies were also excluded.

1.2. Patient follow-up

Eligible patients were sent a study invitation letter by mail that detailed the goals of the study and invited them to the research clinic in the Center for Innovative Medicine (CIM) at the McGill University Health Centre for a detailed musculoskeletal examination. Patients who did not respond were contacted again by telephone for participation. If patients did not answer, a message was left on their answering machine. Patients were classified as "could not be contacted" if the mailing address and phone number on file were not valid. Patients were reimbursed their parking costs for the day of their visit to the hospital.

Our clinical setting is unique due to the proximity and partnership between two university-affiliated children's hospitals, the Montreal Children's Hospital, and the Shriners Hospital for Children. The latter serves as a regional referral center for musculoskeletal deformities. The same teams of pediatric surgeons and pediatric orthopedic surgeons, as well as medical subspecialists, staff both hospitals. Patients diagnosed with scoliosis or chest wall anomalies following a thoracic procedure are referred for consultation at the Shriners Hospital with an orthopedic surgeon or in a multidisciplinary chest wall anomaly clinic, respectively.

For some patients, longitudinal follow-up data was obtained through two alternative sources. First, in our institution, patients with esophageal atresia are followed in a multidisciplinary clinic staffed by a number of health professionals, including pediatric surgeons. At each follow-up visit, a detailed musculoskeletal examination is performed. Therefore, eligible patients with esophageal atresia were examined in this clinic. Second, longitudinal data for patients who were had already been referred to the Shriners Hospital for musculoskeletal deformities were obtained from their detailed examinations during those clinic visits. This avoided the need for patients to return for a repeat musculoskeletal examination at the CIM.

1.3. Patient data

For each patient, their hospital data and outpatient charts from the Montreal Children's Hospital and the Shriner's Hospital for Children, if applicable, were retrospectively reviewed. Data collected included patient characteristics, operative details documented in the operative procedure note and operative report (including whether the chest wall muscles were divided, retracted, or disinserted during the operative approach), and post-operative outcomes. Only the division or preservation of the serratus anterior muscle was clearly reported in the majority of operative reports, while the division or preservation of the latissimus dorsi was rarely mentioned and was not used in the analysis. Longitudinal follow-up data was obtained from a follow-up clinic visit at the CIM, the multidisciplinary esophageal atresia clinic, or the orthopedic clinic or chest wall anomaly center at the Shriner's Hospital. All patients were examined standing up to assess for chest wall deformities (pectus carinatum and pectus excavatum). The wall push-up test, where the patient was asked to push against a wall, was used to identify a medial deviation of the scapula consistent with scapular winging. An Adam's forward bend test was performed to assess for scoliosis, and a scoliometer was used to measure the curvature of the spine. Secondary signs of scoliosis (e.g. shoulder height or scapular spine as asymmetry, deviation of the spine in the coronal plane) were also noted. If there was significant clinical suspicion of a musculoskeletal deformity, subsequent imaging (e.g. scoliosis series, chest x-ray) was obtained. Evaluation for scoliosis on radiologic examination was done by a fellowship-trained spine surgeon, and the Scoliosis Research Society definition of scoliosis was used as the clinical criterion for scoliosis (lateral curve of the spine greater than 10°) [17,18]. The need for intervention (e.g. physiotherapy, bracing, surgical procedure) was documented.

1.4. Statistical analyses

For descriptive purposes, statistical significance between means was determined using one-way ANOVA or Mann–Whitney rank tests for continuous variables, and the Fisher's exact text or χ^2 test for categorical variables, where applicable. Logistic regression was used to estimate the extent to which serratus anterior division, gestational age, sex, age at procedure, and placement of a post-operative chest tube predicted the presence of musculoskeletal deformities in general, and then more specifically scoliosis, chest wall anomalies, or scapular winging. Differences were considered significant at $p < 0.05$. Post-hoc power analysis for difference in proportions was performed to determine statistical power for the sample size used for analysis at the alpha $= 0.05$ level. Statistical analysis was performed using R version 3.5.1 [19].

1.5. Study approval

This study was approved by the Research Ethics Board of the Montreal Children's Hospital, McGill University Health Centre (14–530-PED), and at the Shriners Hospital by the McGill University Faculty of Medicine Institutional Review Board (A07-M41-15B).

2. Results

2.1. Patient cohort

A search through medical archives using diagnostic and procedure codes produced a list of 492 patients who underwent a thoracic procedure via a thoracotomy or thoracoscopy at the Montreal Children's Hospital during the study period. Of those, 58 patients could not be contacted by mail or telephone. A total of 434 patients received a study invitation letter. Of those, 104 patients either responded and were examined in the research clinic or had long-term data available on musculoskeletal outcomes, as shown in Fig. 1. A total of 7 patients who underwent thoracotomy had missing operative reports and were therefore excluded from the regression analyses. The average follow-up period was 11 (range 3–21) years. A total of 37 patients (36%) were followed after the age of 12 years.

2.2. Clinical characteristics

Table 1 shows the clinical characteristics of the patient cohort. The diagnoses requiring thoracic surgery consisted of esophageal atresia with or without tracheo-esophageal fistula (51/104; 49%), patent ductus arteriosus (29/104; 28%), aortic coarctation (9/104; 9%), empyema (7/104; 6%), lung mass (6/104; 6%), and mediastinal mass (2/104; 2%). Fifty-one (49%) patients were male, and 38 patients (36%) were born preterm. The median age at the time of the follow-up examination was 11.1 years (range 3–30). Four patients (4%) underwent two thoracotomies, two to address complications of the primary repair, one

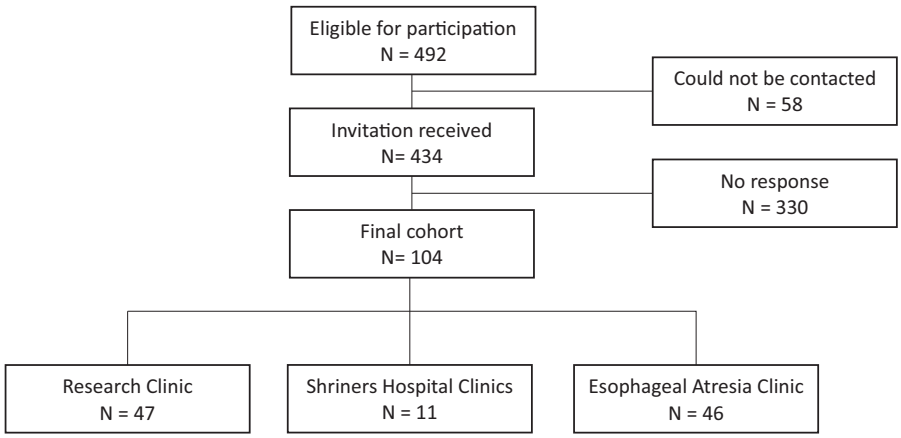

Fig. 1. Flow diagram of study cohort.

for a staged Foker procedure, and one for aortopexy to treat severe tracheomalacia. Patients' characteristics and outcomes, stratified by operative approach, are shown in Table 2. The difference in the overall incidence of musculoskeletal deformities between the operative approaches was significantly different ($p < 0.0001$).

Overall, 56 musculoskeletal deformities developed in 41 (39%) patients. The most frequent musculoskeletal deformity detected was scapular winging (24; 23%), followed by scoliosis (17; 16%) and chest wall anomalies (15; 14%). Of those patients with scoliosis, 11 (65%) had scoliosis <20°, 4 (23%) had scoliosis between 20–45°, and 2 (12%) had scoliosis >45°. When all thoracotomies were compared to thoracoscopy, no significant difference in the incidence of musculoskeletal deformities was found between the two groups (Table 3).

A total of 8 patients (8%), 6 who underwent thoracotomies and 2 who underwent thoracoscopies, required some type of active treatment in the context of a musculoskeletal deformity during the follow-up period. Figs. 2 and 3 depict four such patients. A brace was prescribed to two patients with scoliosis and two patients with pectus carinatum. Two patients with scoliosis and one patient with scapular winging and shoulder asymmetry underwent physiotherapy. One patient with severe scoliosis was followed for approximately 6 years and was then scheduled to undergo spinal fusion surgery after failure of bracing to prevent the progression of the spine deformity. Five patients with mild scoliosis, including one patient with a Cobb angle of 9°, continue to require follow-up with a pediatric orthopedic surgeon.

Among the 92 patients who underwent thoracotomy, after adjusting for the potential confounders, serratus anterior muscle division was found to be independently associated with considerably higher risk for developing a musculoskeletal deformity compared with a muscle-sparing thoracotomy (odds ratio 8.9, 95% confidence interval 2.8–32.6) (Table 4). Post-hoc power analysis for the observed difference in proportions estimated power > 0.99. All but one of the patients who developed scoliosis after a thoracotomy underwent a standard posterolateral thoracotomy with muscle division, and serratus division was independently associated with a higher risk for developing scoliosis (odds ratio 15.6, 95% confidence interval 2.4–308.2). Serratus division was also significantly associated with the development of scapular winging. Although incidence of chest wall anomalies was higher in the patients who had serratus division, serratus division was not independently associated with a higher risk of pectus deformities.

Table 1
Patient cohort.

Variable	% (N)
Sex	
Male	49% (51)
Female	51% (53)
Gestational age (weeks)	
<32	30% (31)
32–37	12% (13)
>37	58% (60)
Surgical approach	
Thoracotomy	89% (93)
Thoracoscopy	11% (11)
Serratus anterior [a]	
Divided	46% (48)
Spared	47% (49)
Unknown	7% (7)
Number of thoracotomies [a]	
1	96% (100)
2	4% (4)
Post-operative chest tube	
Yes	66% (69)
No	29% (30)
Unknown	5% (5)
Intraoperative complications	
Yes	7% (7)
No	88% (92)
Unknown	5% (5)

[a] Thoracotomy cohort (N = 93)

3. Discussion

The findings of the present study emphasize the importance of early and longitudinal screening for musculoskeletal deformities in children who undergo thoracic procedures. Nearly 39% of patients in our cohort were found to have some type of obvious musculoskeletal deformity. Serratus anterior muscle division was found to be the only strong independent risk factor associated with an increased risk of the development of musculoskeletal deformities, most notably scoliosis, on multivariate analysis. The results of this study demonstrate that a muscle sparing technique minimizes the incidence of musculoskeletal deformities following thoracotomy in children. In the last 30 years, the muscle-sparing thoracotomy technique has gained popularity over the traditional posterolateral thoracotomy as the standard approach for most thoracic surgical procedures in children for many reasons, including less postoperative pain, improved functional recovery, and earlier discharge [11,12]. With a proper understanding of the anatomy of the chest wall, stepwise mobilization and proper retraction of the latissimus dorsi and serratus anterior muscles can facilitate entry into the chest without difficulty and spare the chest wall muscles. Specifically, posterior retraction of the anterior border of the latissimus dorsi exposes the serratus anterior muscle, which must then be partially disinserted and retracted at its inferolateral border to expose the intercostal muscles and ribs. If necessary for adequate rib retraction, the anterior border of the latissimus dorsi can be divided for a distance of 2–3 cm. The

Table 2
Patient characteristics and outcomes by operative approach.

Variable	Standard thoracotomy (N = 48)	Muscle-sparing thoracotomy (N = 38)	Thoracoscopy (N = 11)	p-Value
Sex (Male)	50% (24)	49% (19)	27% (3)	0.369
Gestational age (weeks); median (IQR)	29.5 (15)	39 (2.8)	40 (0)	<0.0001 *
Age at surgery (days); median (IQR)	25 (39)	1 (1)	989 (564.5)	<0.0001 *
Post-operative chest tube; % (n)	54% (26)	87% (33)	91% (10)	0.0012 *
Follow-up time (years); median (IQR)	11.5 (5.5)	9.2 (4.2)	11.3 (6.9)	0.0048 *
Any musculoskeletal deformity; % (n) [a]	60% (29)	16% (6)	27% (3)	<0.0001 *

IQR, Interquartile Range
[a] Scapular winging, pectus carinatum, pectus excavatum, or scoliosis.

additional time needed for proper muscle mobilization and retraction to achieve a true muscle-sparing technique is compensated for at the time of closure, where the muscles are left to naturally fall back into place.

With the introduction of thoracoscopy two decades ago, many studies have examined and compared patient outcomes after thoracoscopic and thoracotomy techniques, largely with focus on patient safety, complication rates, and post-operative surgical outcomes [20]. Despite the widespread view that thoracoscopy decreases subsequent development of scoliosis and chest wall deformities, we could not identify any long-term studies that proved this benefit. The potential advantages of thoracoscopy were not demonstrated in our study. This may be a function of the small number of thoracoscopic procedures included, but it is a finding that certainly warrants further studies. The musculoskeletal advantages of thoracoscopy compared to a well-performed muscle-sparing thoracotomy remain to be proven.

In 2009, Lawal et al. compared the musculoskeletal sequelae in 62 children who underwent either video-assisted thoracic surgery (VATS) or thoracotomy via muscle-sparing technique for lobectomy, esophageal atresia repair, or other pleural surgery [21]. After a mean post-operative follow-up of 3.8 years, 53.8% of the thoracotomy group and 9.7% of the VATS group developed subclinical (curvature <20 degrees) scoliosis. In 2018, Bastard et al. performed a national multicenter retrospective study that examined the incidence of thoracic skeletal anomalies in 322 patients from 32 centers in France after VATS, conventional thoracotomy with muscle division, or muscle-sparing thoracotomy for esophageal atresia [22]. They reported a total of 187 patients (58.1%) with sequelae after surgery, including 2 patients who underwent thoracoscopy, 32 patients who underwent muscle-sparing thoracotomy, and 62 patients who had a conventional thoracotomy with muscle-division. They found a significantly higher number of musculoskeletal deformities in patients who underwent thoracotomy compared to thoracoscopy, and no advantage conferred by muscle-sparing technique. However, follow up was limited to an average of 2.75 years post-operatively, and information on type of thoracotomy was missing in approximately 50% of patients who went on to develop musculoskeletal sequelae.

Table 3
Incidence of musculoskeletal deformities (MDs).

	Thoracotomy (N = 93)	Thoracoscopy (N = 11)	p-Value
Length of follow-up [a]	10.9 (5.1)	11.3 (6.9)	0.657
Any MDs	41% (38)	27% (3)	0.521
Winging of scapula	25% (23)	9% (1)	0.450
Chest wall anomaly	14% (13)	18% (2)	0.658
Scoliosis	16% (15)	18% (2)	1.000

[a] Years: median (interquartile range).

In our patient cohort, scoliosis was present in 16% of patients, which far exceeds the range of reported prevalence of adolescent idiopathic scoliosis of 0.47–5.2% [23]. Unlike adolescent scoliosis that presents between 11 and 18 years, thoracogenic scoliosis has been reported to present at an average age of 6 years. [23,24]. Therefore, despite the relatively young age of our patient cohort at the time of follow-up examination, we believe that our length of follow-up time ensures an accurate depiction of the incidence of scoliosis in our cohort. In 2019, Soliman et al. reported on the development of scoliosis in 106 patients operated for esophageal atresia [25]. A muscle-sparing technique was used for all patients who had a right-sided thoracotomy (94/106 patients), while the remainder underwent right-sided thoracoscopy, right or left-sided cervicotomy, or left-sided thoracotomy. After a median follow-up of 6.5 years (range 5–14 years), 53 patients (49%) developed scoliosis, four of whom required operative management. While there exists a wide range in the reported incidence of scoliosis in the literature, it is certain that muscle division plays a significant contributory role through its association with denervation, muscle atrophy and subsequent fibrosis [1–4,6,9,10,26].

In 2017, we performed a retrospective study to evaluate the incidence of musculoskeletal deformities following neonatal thoracotomy for patients with esophageal atresia, and reported that 13 out of 52 patients (25%) developed a musculoskeletal deformity in the post-operative period [27]. In that study, serratus anterior muscle division was associated with significantly higher probability of developing these deformities. The incidence of scoliosis was 8%, and all cases were subclinical. The present study is an extension of our previous study, and includes the previous esophageal atresia cohort. This current study reports outcomes in a larger and more diverse group of patients with additional diagnoses warranting a thoracic procedure in order to more accurately describe the incidence and clinical significance of musculoskeletal deformities after thoracic procedures in children.

There is a paucity of recent studies that have examined whether multiple thoracotomies contribute to an increased occurrence of musculoskeletal deformities. In a review of long-term outcomes among patients with esophageal atresia who reached adulthood, Rintala reports an increased incidence of chest wall anomalies in patients who have undergone more than one thoracotomy [28]. However, much of this evidence comes from older cohorts where rib resection was commonly performed, which may have been a more significant predisposing factor than the number of thoracotomies [2,6,7]. Only four patients in our current cohort underwent more than one thoracotomy, and none of these patients went on to develop a musculoskeletal deformity. However, the limited number of cases makes it difficult to conclude whether a relationship exists between the number of thoracotomies and the development of subsequent musculoskeletal sequelae.

While a large proportion of children following a thoracic procedure may go on to develop musculoskeletal deformities, the clinical course for the majority of the anomalies is usually subclinical by surgical definitions. In our cohort, only 8% of patients required some type of treatment. This is in keeping with what is reported in other contemporary series [8,21,25–27,29]. Though most of these deformities do not meet the threshold for treatment, they still present as obvious abnormalities, and may have a negative impact on the child's body image and quality of life. Therefore, they should still be considered significant longitudinal sequelae post-operatively after thoracic interventions, and may be best studied in the future as a patient-reported outcome.

The present study has several limitations. First, we were able to capture long-term outcomes in only approximately 21% of the entire cohort. Patients and families who responded to the study invitation letter may have represented a group more inclined to participate in the study due to the presence of a visible deformity, leading to a volunteer bias. This may have resulted in an overestimation of the true incidence of musculoskeletal deformities in this patient population. Nevertheless, post-hoc power analysis demonstrated that the sample size was adequate to detect observed differences between standard thoracotomy and muscle-

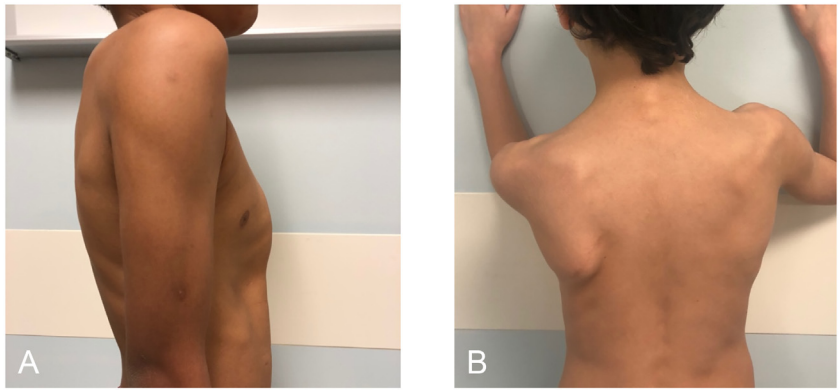

Fig. 2. Two patients with obvious musculoskeletal deformities after left posterolateral thoracotomy. **A,** Pectus carinatum deformity. **B,** Scapular winging and left posterolateral thoracotomy scar.

sparing thoracotomy groups. Second, the cross-sectional design of the study may have introduced confounders that may have influenced the patients' risk for developing a musculoskeletal deformity. We attempted to control for several confounders with multivariate logistic regression analysis, but the possibility of residual confounding remains. It is difficult to make assumptions about the patients who could not be contacted or who did not respond to our invitation letter, so it is unclear how the addition of those patients may have influenced the results presented in this study. However, our reported 39% incidence of musculoskeletal anomalies following thoracic procedures is in keeping with previously published literature, so it is likely that our patient cohort accurately represents the study population.

Despite these limitations our study provides accurate long-term outcome data following pediatric thoracic procedure. These data confirm a high incidence of musculoskeletal deformities. Most of these are not clinically relevant by surgical definition, but their impact on the patient is unknown. A muscle-sparing technique mitigates the incidence of these deformities. The additional benefits of thoracoscopy were not demonstrated in this study. However, the sample size of patients undergoing thoracoscopy was small, warranting further long-term studies to confirm the presumed musculoskeletal benefits of minimally invasive techniques.

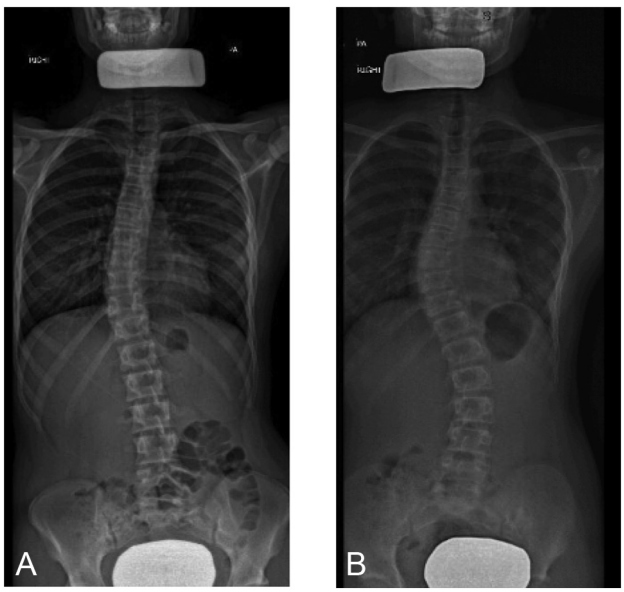

Fig. 3. Scoliosis in patients who underwent left thoracotomy for repair of aortic coarctation (A) and decortication of empyema (B).

Table 4
Risk of musculoskeletal deformities (MD) by serratus anterior muscle division using logistic regression[a].

MD	Unadjusted OR (95% CI)	Adjusted OR (95% CI)
Any MD	8.1 (3.0–25.1)	8.9 (2.8–32.6)
Winging of scapula	3.3 (1.1–11.1)	4.2 (1.2–16.5)
Scoliosis	15.2 (1.9–122.1)	15.6 (2.4–308.2)
Chest wall anomaly	4.7 (1.1–32.2)	3.9 (0.7–31.3)

OR: odds ratio; CI: confidence interval.
[a] Serratus muscle division compared to muscle-sparing (reference).

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