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Musculoskeletal Outcomes Following Thoracoscopic Versus Conventional Open Repair of Esophageal Atresia: A Systematic Review and Meta-Analysis from Pediatric Surgery Meta-Analysis (PESMA) Study Group

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ABSTRACT

Background and objective: Thoracic musculoskeletal deformities are a recognized long-term complication after esophageal atresia (EA) repair. Although evidence remains limited, thoracoscopic repair (TR) is thought to reduce morbidity compared to conventional open repair (COR), with potential benefits including earlier recovery, reduced pain, improved cosmesis, and fewer musculoskeletal sequelae. This systematic review and meta-analysis aimed to compare the incidence of musculoskeletal deformities following EA repair via TR versus COR, emphasizing their long-term clinical implications.
Method: The protocol for this review was registered in PROSPERO(CRD42024576044). A comprehensive literature search was conducted across Ovid Medline, Cochrane, PubMed, Web of Science, EMBASE, SCOPUS, and Google scholar from inception to January 2025. The primary outcomes assessed were scoliosis, rib anomalies (including deformity, fusion, and adhesions), chest wall anomalies, and scapula alata. Statistical analysis was performed using Review Manager (RevMan) version 5.4.
Results: Four retrospective studies comprising 283 patients (TR: 96; COR: 187) were included. TR was associated with significantly lower rates of scoliosis (3.1 % vs. 16 %; RR 0.35, 95 % CI 0.14–0.84, $p = 0.02$) and rib anomalies (0 % vs. 41.5 %; RR 0.05, 95 % CI 0.01–0.25, $p = 0.0002$). No significant differences were observed for chest wall anomalies (RR 0.65, $p = 0.41$) or scapula alata (RR 0.37, $p = 0.30$). However, the small number of studies and variability in diagnostic methods limit the strength and generalizability of these findings.
Conclusion: TR of EA may reduce long-term musculoskeletal morbidity—particularly scoliosis and rib anomalies—when compared to COR. While promising, these results should be interpreted with caution given the limited sample size and methodological heterogeneity. Further multicenter studies with standardized outcome definitions are warranted to confirm these findings and explore the role of muscle-sparing techniques.

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1. Introduction

Esophageal atresia (EA) is a rare congenital anomaly characterized by the discontinuity of the esophagus, occurring in approximately 1 in every 3500 to 4500 live births [1–3]. Surgical intervention is essential for the management of EA, with various approaches described including right posterolateral thoracotomy or conventional open repair (COR), muscle-sparing thoracotomy, and thoracoscopic repair (TR) [3,4]. COR typically involves a posterolateral thoracotomy, which may entail division of the latissimus dorsi and serratus anterior muscles, potentially disrupting musculoskeletal integrity and thoracic growth. In contrast, TR is a minimally invasive alternative that avoids muscle division and is thought to minimize surgical trauma to the developing chest wall. Despite this theoretical advantage, evidence comparing the long-term musculoskeletal outcomes of TR versus COR remains scarce and heterogeneous. Advances in surgical techniques have significantly improved the survival rate of these children over the past decades, creating new challenges in managing long-term postoperative complications, such as musculoskeletal deformities [5,6].

The primary concern with COR in the neonatal period is the higher risk of long-term musculoskeletal deformities, which can significantly affect a child's quality of life [5–7]. Such deformities may include rib fusion, vertebral anomalies, and scoliosis, which have been reported to occur more frequently in patients undergoing open thoracic procedures compared to those treated with minimally invasive surgery (MIS) [2,6,8,9]. Moreover, damage to the serratus anterior and latissimus dorsi muscles, as well as denervation of the long thoracic nerve, may result in functional limitations and compromised upper body movement postoperatively [10]. Additionally, the incidence of scoliosis following esophageal atresia repair ranges from 6 % to 50 %, resulting from factors such as rib fusion, congenital spinal anomalies, or pleural scarring [9,10]. Previous studies suggest that COR may increase the risk of thoracic deformities, whereas muscle-sparing techniques may reduce this risk. However, most reports do not clearly distinguish between muscle-sparing and traditional thoracotomies, nor do they use standardized criteria for outcome assessment. Moreover, the timing of surgical approaches (e.g., historical controls for open repair) and variability in surgeon expertise may further confound interpretation.

Since the first published successful thoracoscopic repair in 1999 [11], TR has gained popularity and has become standard practice in numerous medical centers, due to its potential benefits as a result of its minimally invasive nature and the potential for reduced morbidity [12–15]. However, it is a complex procedure, technically demanding, requiring advanced skills and a steep learning curve [2].

Although the postoperative changes after open EA repair and the potential benefits of the thoracoscopic approach have been well described, a quantitative comparative analysis has so far not been performed. Given the lack of standardized, high-quality data in this domain, the present systematic review and meta-analysis aims to compare long-term musculoskeletal outcomes—specifically scoliosis, rib anomalies, scapula alata, and chest wall deformities—between patients undergoing TR versus COR for EA repair. By synthesizing the available evidence, we hope to clarify whether TR provides a long-term musculoskeletal benefit and to identify gaps for future research.

2. Materials and methods

2.1. Search strategy

The protocol of this review was prospectively registered in PROSPERO (CRD42024576044) (http://www.crd.york.ac.uk/PROSPERO/). We conducted a comprehensive literature search using several databases, including Ovid Medline, Cochrane, PubMed, Web of Science, EMBASE, SCOPUS, and Google scholar. Our search targeted keywords including “esophageal atresia”,”musculoskeletal abnormality”, “scoliosis”, “rib fusion”, “rib deformity” The extended search strategy was given in Appendix 1.

2.2. Study selection

Studies reporting musculoskeletal abnormalities following thoracoscopic or open (via thoracotomy) repair of esophageal atresia were included.

Inclusion criteria: Clinical studies comparing thoracoscopy or thoracotomy for esophageal atresia repair, and report any of the following outcomes: scoliosis, chest wall anomaly (asymmetry), rib deformities, scapula alata, or rib adhesions.

Exclusion criteria: The absence of comparative case series serving as control groups, and studies that either did not provide useable raw data or were instances of duplicate publications.

2.3. PICOS strategy

Participant/Population(s): Neonates diagnosed with EA.

Intervention(s): Patients who underwent thoracoscopic repair for EA repair.

Comparator(s)/Control: Patients who underwent thoracotomy for EA repair.

Outcome(s): Scoliosis, rib deformity, rib adhesions, chest wall anomaly, scapula alata.

Studies: Randomised controlled trials (RCT), prospective, and retrospective studies.

2.4. Data extraction

Three researchers (MA, EK, and TOK) independently evaluated the studies that surfaced from our search and selected them according to the above criteria to be included in this meta-analysis. Information on the number of participants, study design, country, and publication year was extracted from qualifying publications. Additionally, data on population characteristics, such as birth weight, age at the time of surgery, and surgical approach was gathered.

2.5. Risk of bias assessment

The Newcastle–Ottawa Scale (NOS), specifically designed for assessing observational studies, was used to evaluate the quality of the selected research. Three authors (MA, EK, and TOK) independently assessed the included studies. They used a predefined meta-analysis form to extract relevant data from each study. Any differences in opinion were resolved through discussion among the reviewers.

2.6. Statistical analysis

For statistical analysis, we employed Review Manager (RevMan) software, version 5.4. Continuous variables were analyzed using mean differences, while dichotomous variables were assessed using risk ratios. The I^2 statistic was used to quantify statistical heterogeneity, and the Chi-square test determined its presence. A significance level of $p < 0.05$ was established for the analysis. A fixed effects model was used to provide a comprehensive analysis of the data.

2.7. Reporting

The findings of this systematic review were reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [16].

3. Results

3.1. How were the outcomes measured?

In the study by Borselle et al. [10], musculoskeletal anomalies such as scoliosis and rib deformities were assessed through a combination of clinical examination, X-ray imaging, and, when available, CT scans. Scoliosis was defined as a lateral curvature of the spine $\geq 10°$ of Cobb angle with vertebral rotation. Rib fusion was identified based on CT scans or appropriate X-ray projections. Physical examinations were performed by experienced pediatric surgeons and radiologists during follow-up visits. Median age at evaluation was 5.5–6 years, with maximum follow-up of 15 years in the TR group and 13 years in the COR group.

Safa et al. [13] assesed musculoskeletal deformities, including scoliosis, chest wall anomalies, and scapular winging through a combination of clinical examinations and radiographic assessments during long-term follow-up. Mean follow-up was 10.9 years for the COR group and 11.3 years for the TR group. Patients underwent detailed musculoskeletal examinations in specialized clinics, where specific tests, such as the Adam's forward bend test and the wall push-up test, were performed to identify scoliosis and scapular winging. If clinical suspicion of a deformity was present, further imaging, such as a scoliosis series or chest X-ray, was conducted to confirm the diagnosis. A scoliometer was used to measure the curvature of the spine. The evaluation for scoliosis on radiological examination was conducted by a fellowship-trained spine surgeon. Scoliosis was defined as a lateral spinal curvature $>10°$, according to the criteria of the Scoliosis Research Society.

Hattori et al. [14] used follow-up chest radiographs to assess scoliosis, chest wall asymmetry, and rib deformities. Evaluations were conducted by pediatric surgeons. The mean follow-up was 8.7 years in the TR group and 11.5 years in the COR group.

Aubert et al. [15] used real-time MRI (3 T system) to evaluate thoracic structures during unsedated free breathing. This allowed identification of chest wall anomalies, vertebral malformations, and rib abnormalities. Thoracic cross-sectional area and lung volumes were also measured.

3.2. Details of selected studies

Figure 1 shows the PRISMA flowchart for the study screening process. The initial search yielded 240 titles, and after removing irrelevant entries and duplicates, 131 records remained. Following a review of titles and abstracts, 50 full-text articles were selected for in-depth evaluation of the full text. Of these, 46 were excluded for not meeting the inclusion criteria. As a result, four studies [10,13–15] comparing TR ($n = 96$) to COR ($n = 187$) were included in the meta-analysis. Table 1 provides an overview of the basic demographic information for each of the four studies, including the country where the study was conducted, the year of publication, study design, number of patients, and the Newcastle–Ottawa Scale (NOS) bias score. In contrast, Table 2 presents the outcomes reported in each study.

Musculoskeletal deformities were mainly assessed by clinical examination and X-ray [14,15]. In some cases, a CT scan of the thorax using a low dose scanning technique was performed [10]. Aubert et al. performed MRI scans on all their patients to evaluate potential musculoskeletal abnormalities [15].

4. Outcomes

4.1. Scoliosis

Reported in all four studies, including 96 patients in the TR group and 187 in the COR group. Our analysis revealed that the TR group had a significantly lower scoliosis rate compared to COR ($I^2 = 36$ %) (RR: 0.35, 95 % CI: 0.14 to 0.84; $p = 0.02$). Overall scoliosis rates were 3.1 % (TR) and 16 % (COR) (Fig. 2).

4.2. Rib deformity

Reported in three studies, including 85 patients in the TR group and 94 in the COR group. Our analysis revealed that the TR group had a significantly lower rib deformity rates compared to COR ($I^2 = 0$ %) (RR: 0.05, 95 % CI: 0.01 to 0.25; $p = 0.0002$), with overall rates of 0 % (TR) and 41.5 % (COR) (Fig. 3).

4.3. Chest wall anomaly

Reported in two studies, including 20 patients in the TR group and 101 in the COR group. No difference was found between groups in terms of chest wall anomaly rate ($I^2 = 54$ %) (RR: 0.65, 95 % CI: 0.23 to 1.81; $p = 0.41$). The overall chest wall anomaly rate were 15 % (TR) and 17 % (COR) (Fig. 4).

4.4. Scapula alata

Reported only in one study, including 11 patients in the TR group and 93 in the COR group. No difference was found between groups in terms of scapula alata rate (I^2=Not applicable) (RR: 0.37, 95 % CI: 0.05 to 2.46; $p = 0.30$). The overall scapula alata rates were 9 % (TR) and 25 % (COR) (Fig. 5).

5. Discussion

This systematic review and meta-analysis provide a nuanced understanding of the musculoskeletal outcomes following TR versus COR of EA in pediatric patients. While TR, with its minimally invasive nature, offers advantages like reduced postoperative pain, quicker recovery, and minimized scarring [2,17,18], its implications on the musculoskeletal system have been under debate. Our analysis shows that TR is associated with significantly lower rates of scoliosis and rib deformities compared to COR, suggesting a protective effect on the developing musculoskeletal system [2,10,15,19]. Specifically, our meta-analysis revealed a scoliosis rate of 3.1 % in the TR group versus 16 % in the COR group, indicating a substantial relative risk reduction that is both statistically and clinically significant [10,14,15].

The potential mechanisms behind the reduced scoliosis rates in TR are likely multifactorial. One hypothesis is that the smaller incisions and reduced chest wall disruption in thoracoscopic surgery result in less musculoskeletal damage, thereby decreasing the risk of scoliosis development. However, the occurrence of scoliosis even in the TR group suggests that other factors, such as primary scoliosis unrelated to surgical technique, may play a role [20]. None of the included studies systematically excluded patients with preexisting musculoskeletal anomalies or syndromes, potentially confounding the results. Therefore, further research is needed to distinguish between scoliosis directly related to surgical intervention and cases with different etiologies.

The discrepancies in rib fusion rates across studies highlight the impact of diagnostic modality on outcome reporting and the need for standardized assessment methods. While our analysis showed lower rates of rib deformities in the TR group (0 %) compared to

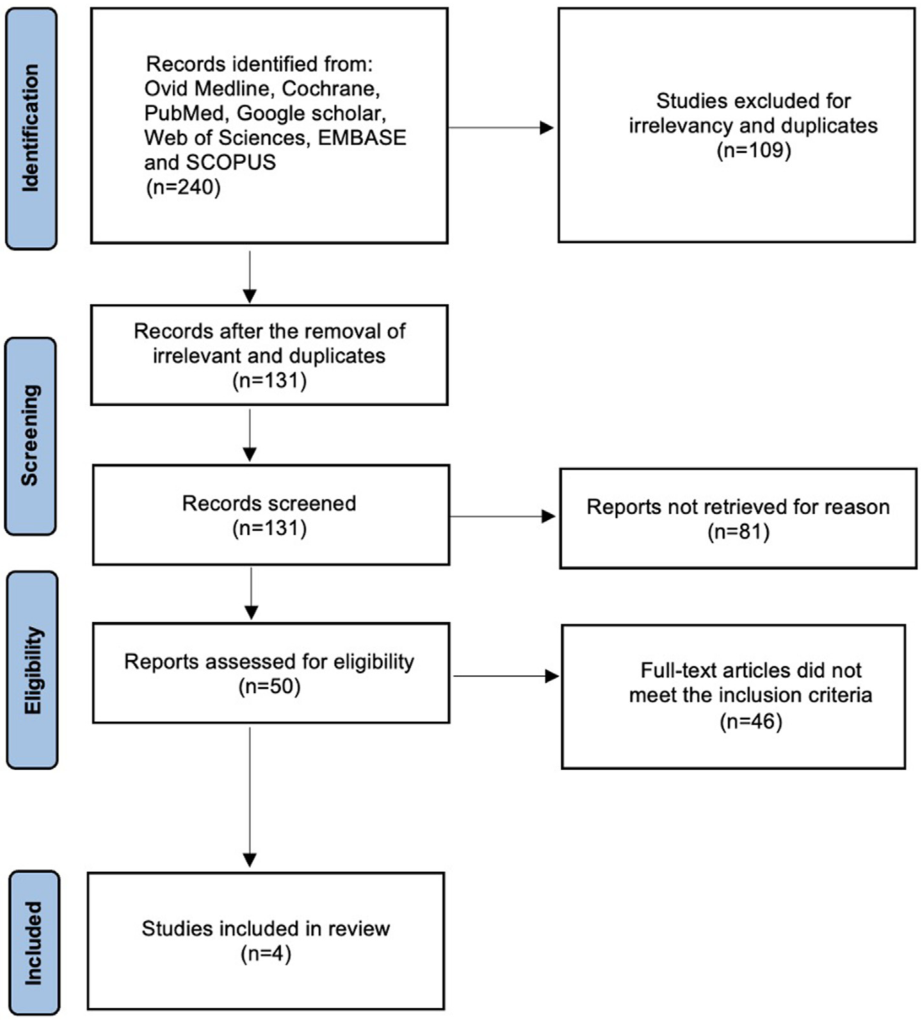

Fig. 1. PRISMA flow diagram of study selection.

Table 1
The characteristics of the included studies.

Authors	Study years	Type of study	Country	TR (n)	COR (n)	NOS
Aubert et al., 2024	2021–2023	Prospective	Germany	8	42	7
Borselle et al., 2024	2005–2021	Retrospective	Poland	68	44	8
Hattori et al., 2024	2003–2016	Retrospective	Japan	9	8	8
Safa et al., 2021	1997–2012	Retrospective	Canada	11	93	7

NOS: Newcastle Ottawa Scale; TR: Thoracoscopy; COR: Conventional open repair.

Table 2
The primary outcomes of included studies.

Authors	Scoliosis		Rib deformity		Chest wall anomaly		Scapula alata	
	TR	COR	TR	COR	TR	COR	TR	COR
Aubert et al., 2024	0	6	0	19	NR	NR	NR	NR
Borselle et al., 2024	1	6	0	13	NR	NR	NR	NR
Hattori et al., 2024	0	3	0	7	1	4	NR	NR
Safa et al., 2021	2	15	NR	NR	2	13	1	23

TR: Thoracoscopy; COR: Conventional open repair; NR: Not reported.

41.5 % in the COR group, Aubert et al. reported a much higher rib fusion rate of 78 % using rtMRI [15]. This suggests that advanced imaging may identify subtle abnormalities missed by conventional radiography, and that methodological differences among studies affect detection rates. This underreporting issue extends to scapular winging, which is almost impossible to occur after a minimally invasive repair but might be missed without a thorough and standardized clinical examination. Proper examination, including assessing winging with the child leaning forward and elevating both arms, is crucial for accurate diagnosis. Consequently, the prevalence of scapula alata reported in our meta-analysis likely underestimates its true incidence.

The musculoskeletal advantages of TR must be weighed against its technical challenges. Thoracoscopic repair demands a high level of skill and experience, with a steep learning curve that may lead to variability in outcomes among surgeons [2,12,14]. The clinical implementation of TR requires not only mastery of the technique but also access to specialized training and resources, particularly for

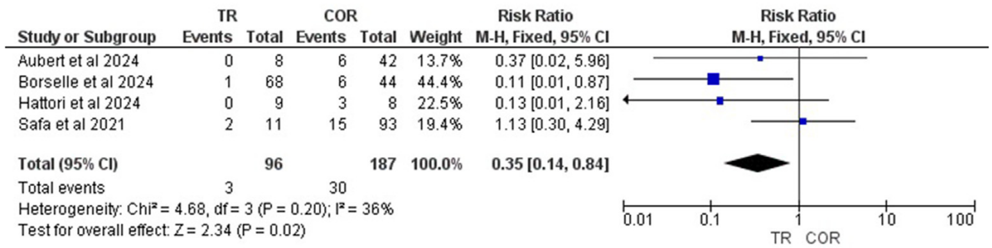

Fig. 2. Forest plot of scoliosis rate comparing TR and COR.

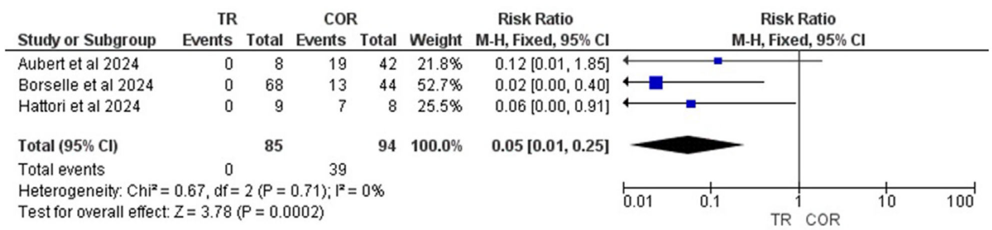

Fig. 3. Forest plot of rib deformity rate comparing TR and COR.

Fig. 4. Forest plot of chest wall anomaly rate comparing TR and COR.

Fig. 5. Forest plot of scapula alata rates comparing TRand COR.

centers with less experience in TR. As highlighted by prior literature, the precision required for thoracoscopic surgery necessitates structured training programs to overcome the learning curve and reduce the risk of complications [18,21]. Additionally, the retrospective nature of all included studies and potential surgeon-related variability in technique or experience were not consistently reported or controlled for, limiting our ability to interpret outcomes with certainty.

The consideration of muscle-sparing thoracotomy as an alternative to COR also warrants discussion. While not directly analyzed in our study, this approach could offer a middle ground, potentially reducing musculoskeletal sequelae without the technical demands of thoracoscopy [22]. Future research comparing muscle-sparing thoracotomy with both TR and COR is essential to provide a clearer understanding of its potential benefits and limitations [5,10,13].

The clinical implications of our findings are significant, as reducing the incidence of scoliosis and rib deformities can lead to improved long-term musculoskeletal health and quality of life for patients with EA [10,14]. These conditions, if left untreated, can result in chronic pain, reduced pulmonary function, and the need for additional surgical interventions [5,9]. Therefore, the adoption of TR in centers with the requisite expertise could play a crucial role in minimizing these long-term complications [2,5,10,13,15,19]. However, given the variability in outcomes and the need for further research, the choice between TR and COR should be individualized, taking into account patient anatomy, surgeon expertise, and the availability of resources [2,11].

This study has several limitations. The number of included studies was small ($n = 4$), all were retrospective in design, and the overall sample size was limited, reducing statistical power and generalizability. There was considerable heterogeneity in follow-up duration, imaging modalities (e.g., X-ray, CT, MRI), and outcome definitions, which limits the reliability of pooled estimates. In some cases, it was unclear whether patients had scoliosis or musculoskeletal anomalies preoperatively, making it difficult to distinguish new-onset complications from pre-existing conditions. The involvement of different clinical specialties (e.g., pediatric surgeons, spine surgeons, radiologists) in diagnostic screening may have introduced additional variability. Finally, the lack of information regarding the severity of musculoskeletal anomalies or whether they required further intervention represents an additional limitation. Moreover, none of the included studies reported on the use of muscle-sparing thoracotomy, a potentially relevant confounding factor. Future high-quality, multicenter studies with long-term follow-up and standardized outcome assessment are needed to clarify the true impact of surgical approach on musculoskeletal health in EA survivors.

6. Conclusions

This systematic review and meta-analysis suggests that thoracoscopic repair of esophageal atresia is associated with a lower incidence of scoliosis and rib deformities compared to conventional open thoracotomy. These findings support the hypothesis that minimally invasive techniques may reduce the risk of long-term musculoskeletal complications in pediatric patients. However, given the small number of available studies, methodological variability, and lack of standardized outcome definitions, these results should be interpreted with caution. Further high-quality, prospective multicenter studies with standardized assessment protocols are needed to validate these findings and to better define the long-term musculoskeletal impact of different surgical approaches for esophageal atresia repair.

Data availability

The datasets generated and analyzed during the current study are available from the corresponding author upon reasonable request.

Ethical approval

Ethical approval and written informed consent from patients were not required as the study is based on a systematic review of previously published data.

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None.

Conflict of interest

None declared.

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Appendix 1. Search Strategy

("musculoskletal"[Title/Abstract] OR "musculo"[Title/Abstract] OR "skeletal"[Title/Abstract] OR "musculo*"[Title/Abstract] OR "vertebrae"[Title/Abstract] OR "rib"[Title/Abstract] OR "costa"[Title/Abstract] OR "costae"[Title/Abstract] OR "costal"[Title/Abstract] OR "fracture"[Title/Abstract] OR "bone"[Title/Abstract] OR "scoliosis"[Title/Abstract]) AND ("deformity"[Title/Abstract] OR "deformities"[Title/Abstract] OR "abnormal"[Title/Abstract] OR "abnormality"[Title/Abstract] OR "anomaly"[Title/Abstract] OR "anomalies"[Title/Abstract] OR "problem"[Title/Abstract] OR "deform"[Title/Abstract] OR "deform*"[Title/Abstract] OR "issue"[Title/Abstract] OR "fusion"[Title/Abstract] OR "fusi*"[Title/Abstract]) AND ("esophageus"[Title/Abstract] OR "esophageal"[Title/Abstract] OR "esophagus"[Title/Abstract] OR "oesophageal"[Title/Abstract] OR "oesophagus"[Title/Abstract] OR "foregut"[Title/Abstract] OR "esophag*"[Title/Abstract]) AND ("atresia"[Title/Abstract] OR "atretic"[Title/Abstract] OR "atresy"[-Title/Abstract] OR "disconnection"[Title/Abstract] OR "disconnect*"[Title/Abstract] OR "abnormality"[Title/Abstract] OR "abnormalities"[Title/Abstract] OR "anomaly"[Title/Abstract] OR "anomalies"[Title/Abstract] OR "absent"[Title/Abstract] OR "absence"[Title/Abstract] OR "disconnected"[Title/Abstract] OR "disconn*"[Title/Abstract] OR "interrupted"[Title/Abstract] OR "interrupt*"[Title/Abstract]) AND ((("thoracoscopy"[MeSH Terms] OR "thoracoscopy"[All Fields] OR "thoracoscopies"[All Fields] OR ("thoracoscopes"[MeSH Terms] OR "thoracoscopes"[All Fields] OR "thoracoscope"[All Fields] OR "thoracoscopic"[All Fields] OR "thoracoscopical"[All Fields] OR "thoracoscopically"[All Fields]) OR ("minimal"[All Fields] OR "minimisation"[All Fields] OR "minimisations"[All Fields] OR "minimise"[All Fields] OR "minimised"[All Fields] OR "minimises"[All Fields] OR "minimising"[All Fields] OR "minimization"[All Fields] OR "minimizations"[All Fields] OR "minimize"[All Fields] OR "minimized"[All Fields] OR "minimizer"[All Fields] OR "minimizers"[All Fields] OR "minimizes"[All Fields] OR "minimizing"[All Fields]) OR ("invasibility"[All Fields] OR "invasible"[All Fields] OR "invasion"[All Fields] OR "invasions"[All Fields] OR "invasive"[All Fields] OR "invasively"[All Fields] OR "invasiveness"[All Fields] OR "invasives"[All Fields] OR "invasivity"[All Fields]) OR "thoracos*"[All Fields] OR ("robot"[All Fields] OR "robot s"[All Fields] OR "robotically"[All Fields] OR "robotics"[MeSH Terms] OR "robotics"[All Fields] OR "robotic"[All Fields] OR "robotization"[All Fields] OR "robotized"[All Fields] OR "robots"[All Fields]) OR "thoracosc*"[All Fields]) AND ("open"[All Fields] OR ("thoracotomy"[MeSH Terms] OR "thoracotomy"[All Fields] OR "thoracotomies"[All Fields]) OR ("classic"[All Fields] OR "classical"[All Fields] OR "classically"[All Fields] OR "classicals"[All Fields] OR "classics"[All Fields]) OR ("repairability"[All Fields] OR "repairable"[All Fields] OR "repaire"[All Fields] OR "repaired"[All Fields] OR "repairment"[All Fields] OR "wound healing"[MeSH Terms] OR ("wound"[All Fields] AND "healing"[All Fields]) OR "wound healing"[All Fields] OR "repair"[All Fields] OR "repairing"[All Fields] OR "repairs"[All Fields]))) OR ("thoracic surgery, video assisted"[MeSH Terms] OR ("thoracic"[All Fields] AND "surgery"[All Fields] AND "video-assisted"[All Fields]) OR "video-assisted thoracic surgery"[All Fields] OR "vats"[All Fields]) OR "video-assisted"[All Fields])

Translations

thoracoscopy: "thoracoscopy"[MeSH Terms] OR "thoracoscopy"[All Fields] OR "thoracoscopies"[All Fields]

thoracoscopic: "thoracoscopes"[MeSH Terms] OR "thoracoscopes"[All Fields] OR "thoracoscope"[All Fields] OR "thoracoscopic"[All Fields] OR "thoracoscopical"[All Fields] OR "thoracoscopically"[All Fields]

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VATS: "thoracic surgery, video-assisted"[MeSH Terms] OR ("thoracic"[All Fields] AND "surgery"[All Fields] AND "video-assisted"[All Fields]) OR "video-assisted thoracic surgery"[All Fields] OR "vats"[All Fields]

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