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Thoracic skeletal anomalies following surgical treatment of esophageal atresia. Lessons from a national cohort

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ABSTRACT

Introduction: Thoracotomy as surgical approach for esophageal atresia treatment entails the risk of deformation of the rib cage and consequently secondary thoracogenic scoliosis. The aim of our study was to assess these thoracic wall anomalies on a large national cohort and search for factors influencing this morbidity.
Materials and methods: Pediatric surgery departments from our national network were asked to send recent thoracic X-ray and operative reports for patients born between 2008 and 2010 with esophageal atresia. The X-rays were read in a double-blind manner to detect costal and vertebral anomalies.
Results: Among 322 inclusions from 32 centers, 110 (34.2%) X-rays were normal and 25 (7.7%) displayed thoracic malformations, including 14 hemivertebrae. We found 187 (58.1%) sequelae of surgery, including 85 costal hypoplasia, 47 other types of costal anomalies, 46 intercostal space anomalies, 21 costal fusions and 12 scoliosis, with some patients suffering from several lesions. The rate of patients with these sequelae was not influenced by age at intervention, weight at birth, type of atresia, number of thoracotomy or size of the center. The rate of sequelae was higher following a classical thoracotomy (59.1%), whatever the way that thoracotomy was performed, compared to nonconverted thoracoscopy (22.2%; $p = 0.04$).
Conclusion: About 60 % of the patients suffered from a thoracic wall morbidity caused by the thoracotomy performed as part of surgical treatment of esophageal atresia. Minimally invasive techniques reduced thoracic wall morbidity. Further studies should be carried out to assess the potential benefit of minimally invasive approaches to patient pulmonary functions and on the occurrence of thoracogenic scoliosis in adulthood.
Levels of evidence: Level III retrospective comparative treatment study.

Esophageal atresia is a well-known congenital anomaly that affects around 150 children per year in France (1.8/10,000 births). The current survival rate is higher than 95% [1]. Among associated malformations occurring in more than half of esophageal atresia cases, vertebral and musculoskeletal malformations are well described, with rib cage deformations occurring in 20% to 50% of adult patients in the historical series [2–5]. Vertebral malformations are frequent as well, but we also know that secondary scoliosis occurs in 14% of cases without any initial vertebral malformation seen on X-ray at birth [5].

Right thoracotomy performed during surgical treatment of esophageal atresia is the classical explanation for these secondary skeletal anomalies. New minimally invasive approaches by thoracoscopy have recently been developed [6,7], in order to reduce the parietal impact of classical thoracotomy for these infants and thereby decrease the long-term rate of thoracogenic scoliosis.

To date, there are no large-scale studies of thoracic skeletal anomalies among esophageal atresia patients that make a clear distinction between bone malformations and sequelae from surgical approaches, or that evaluate the impact of new minimally invasive techniques. We therefore performed a national multicenter retrospective study in order to assess thoracic skeletal anomalies in patients included in the French national register for esophageal atresia. We then looked for what surgical factors from the different operative approaches influenced rates of thoracic skeletal sequelae.

1. Materials and methods

Pediatric surgery departments from the French CRACMO network (CRACMO: « Centre de Référence des Affections Congénitales et Malformatives de l'œsophage »: Reference Center for esophageal congenital malformations, Lille, France) were asked to include all patients born between 01/01/2008 and 12/31/2010 and registered in CRACMO files for esophageal atresia. For each patient, the national register had prospectively recorded the birth date, pregnancy term and birth weight; the esophageal type according to the Ladd classification; associated malformations; date of surgery, type of surgical approach (thoracotomy, thoracoscopy), type of surgical treatment (suture, esophagoplasty). These data from the register for the years 2008, 2009 and 2010 were controlled, sealed and available for studies. The Ethics Committee of CRACMO approved the study protocol.

Each local CRACMO referred investigator was requested to send to the investigating center, electronically and anonymously, operative reports for thoracic procedures and the most recent chest X-ray results available to them, ensuring that the documents were labeled with the national register patient number. X-rays were read in a double-blind manner by a pediatric surgeon and a radiologist, and classified as normal (normal group), abnormal with malformations seen on vertebrae or ribs from a site other than the surgical approach (malformation group), and abnormal on the surgical site without malformations (sequelae group). The type of anomaly (vertebral, costal or from intercostal space), site and side were registered for each abnormality. In the event of discordance between the surgical and radiological interpretations of the chest X-ray, the two readers examined the image together once more in order to reach a consensus.

The information recorded from operative reports for each thoracic surgical procedure was, when available, each step of surgical approach including skin incision, muscle opening technique, intercostal space opening technique and intra- or extrapleural dissection, and the type of surgical sutures used for intercostal closure.

Statistical analysis was performed using Student's t-test for quantitative data and Fisher exact test for qualitative data. P values below 0.05 were considered statistically significant. Data were processed using the software Stata12.1 (StataCorp, Texas).

2. Results

Among the 37 centers belonging to the CRACMO Network, 32 centers participated, allowing for the enrollment of 339 patients for the study, from a total of 397 registered patients born between 2008 and 2010 (85.3%). Seventeen X-rays were excluded owing to poor quality or date prior to surgery. Among the 322 inclusions, 110 (34.2%) X-rays were normal and 25 (7.7%) displayed thoracic malformations, including 14 hemivertebrae (Fig. 1). We found 187 (58.1%) sequelae of surgery, including 85 costal hypoplasia (rib smaller an thinner than these in their neighborhood), 47 other types of costal anomalies, 46 intercostal space anomalies, 21 costal fusions and 12 scoliosis, with some patients suffering from several lesions (Fig. 1).

Sequelae after surgery were not affected by age at procedure (average age in group of patients respectively with and without sequelae: 8.36 ± 26 *versus* (*vs*) 4.93 ± 20 days, $p = 0.24$), weight at birth (2580 ± 685 g *vs* 2620 ± 706 g; $p = 0.2$), type of atresia (Rate of Type III–IV: 165/187 (88.2%) *vs* 126/135 (93.3%); $p = 0.2$).

The average age of patients at the time of the chest X-ray was 2.75 ± 1.92 years (Table 1). Age was older for the sequelae group (2.92 ± 1.88