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# Outcome of primary repair in extremely and very low-birth-weight infants with esophageal atresia/distal tracheoesophageal fistula

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ABSTRACT

*Purpose:* The optimal surgical management of extremely (ELBW) and very low-birth-weight (VLBW) neonates with esophageal atresia and distal tracheoesophageal fistula (EA/TEF) (Gross type C) is still debated. The aim of this study was to evaluate the surgical outcome of primary repair in these patients and compare it to ≥1500 g neonates.
*Methods:* Medical records of neonates with repaired EA from 2002 to 2016 were reviewed.
*Results:* 4 ELBW, 7 VLBW, and 24 ≥1500 g infants had type C EA/TEF and underwent primary repair. Anastomotic leakage occurred in 0% ELBW, 0% VLBW and 8.3% ≥1500 g patients and anastomotic stricture in 25% ELBW, 28.5% VLBW and 37.5% ≥1500 g patients. 50% ELBW, 14.2% VLBW and 20.8% ≥1500 g patients underwent secondary fundoplication. One patient of the VLBW group and one patient of the ≥1500 g group died postoperatively of causes not related to EA/TEF.
*Conclusions:* In extremely and very low-birth-weight neonates with type C EA/TEF surgical outcome after primary repair is comparable to the outcome in ≥1500 g neonates. Primary repair can be performed in most of these patients and staged repair can be restricted to unstable patients.
*Level of evidence:* Treatment study level III.



Birth weight as risk factor for survival of neonates with EA has been addressed in numerous studies. Low birth weight was identified as risk factor for survival [1–4]. Spitz [2] established a birth weight of 1500 g as cut-off value to separate risk groups. Further studies have shown that associated cardiac and chromosomal anomalies [5] and preoperative ventilator dependence and severe associated anomalies [6] could be more important for survival than body weight. For <1500 g neonates major cardiac anomalies were found to be more decisive for survival than weight [7]. In general, because of progress in pediatric surgery and neonatal and anesthetic care survival rates of neonates with EA including those of very low birth weight have increased [7–11].

Fewer data are available on the impact of birth weight on the surgical complications associated with different surgical procedures. The surgical management of ELBW and VLBW neonates with EA/TEF can be performed by primary or staged repair. Data on the outcome with these methods are not conclusive [12–14]. We analyzed the surgical outcome in ELBW and VLBW neonates with type C EA/TEF who underwent primary repair. For comparison, a group of ≥1500 g neonates were included.

## 1. Patients and methods

### 1.1. Patients and data preparation

The ethics committee of the University of Tuebingen approved the use of patient data for this study (Ref. 659/2016BO2). A retrospective review of medical records of all neonates with EA operated on at our institution between 2002 and 2016 was performed. During this period two patients referred to our hospital died before surgery could be performed and therefore they are not included in the analyses. Data of patients with distal tracheoesophageal fistula (Gross type C) who underwent open repair were analyzed for this study. Parameters considered were surgical procedure, birth weight, gestational age, associated anomalies, intra- and postoperative complications, and death. Of the parameters analyzed anastomotic leakage and stricture are the parameters most closely linked to surgery and therefore they were considered to be the

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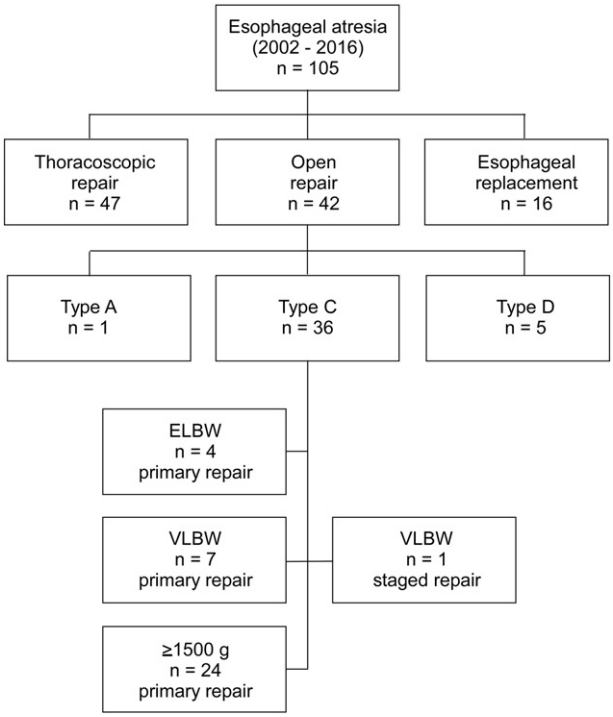


**Fig. 1.** Type of esophageal atresia and surgical techniques.

primary outcome parameters. *Leakage* was defined as the detection of a contrast by esophagography performed routinely 10 days after surgery. *Stricture* was defined if at least one balloon dilatation was necessary. Non-cyanotic and cyanotic type anomalies requiring medical or surgical treatment for heart failure were classified as major cardiac anomalies [2]. Fundoplication was performed if esophagography indicated gastro-esophageal reflux disease and reflux symptoms could not be managed by medical treatment.

Patients were divided into three groups depending on birth weight: <1000 g (ELBW), 1000 g to <1500 g (VLBW) and ≥1500 g. Data are presented as median (range). Given the low number of cases, no statistical analyses were performed.

### *1.2. Patient allocation to surgical procedures*

Thoracoscopy for EA repair is well established in our department and preferred to thoracotomy if type of EA, weight (>2 kg), patient conditions such as pulmonary and cardiac stability allow this approach and if there are no interfering associated malformations. We prefer primary repair to staged repair if all parameters indicate that the patient will tolerate the longer duration of anesthesia linked to primary repair compared to fistula ligation, the first step of staged repair. In case of long gap EA we perform esophageal replacement by gastric pull-up, if possible laparoscopically assisted.

### *1.3. Primary repair of Gross type C esophageal atresia*

Primary repair was performed electively as soon as possible, usually within the first 24 to 48 h of life. For closure of the fistula and primary esophageal anastomosis a right lateral thoracotomy in the 4th intercostal space was performed. Whenever possible, an extrapleural approach was used. After ligation of the azygos vein and closure of the fistula the proximal and distal pouches were mobilized and end-to-end single-layer anastomosis was performed. For splinting, feeding and for the esophagography a small feeding tube was placed nasally through the esophagus into the stomach prior to completing the anastomosis. A mediastinal drain was inserted in most cases, which was removed if no leakage was evident on the esophagogram at day 10 postoperatively. Over the time period covered surgery was performed by different pediatric surgery consultants under supervision of the senior author.

## 2. Results

### *2.1. Patient characteristics*

From 2002 to 2016 105 neonates with EA were operated on at our institution. Thoracoscopic repair was performed in 47 patients, open repair in 42 patients and esophageal replacement, open or laparoscopic-assisted, in 16 patients. 36 patients of the open-repair group were diagnosed with type C EA/TEF. 4 of these patients were allocated to the ELBW, 8 to the VLBW and 24 to the ≥1500 g group. Primary repair was performed on all but one neonate of the VLBW group (Fig. 1). Because of the severity of the major cardiac malformation (tricuspid atresia, pulmonary atresia with ductal-dependent pulmonary blood flow) of this patient a staged approach was intended. Therefore, TEF ligation was performed first. Before the date of the final repair the patient died of his cardiac anomalies.

For neonates with type C EA/TEF and primary repair the median weight (range) was 840 (780–980) g, 1300 (1000–1495) g and 2525 (1770–3440) g and the range of the gestational age was 27.7 to 31.7 weeks, 29.9 to 32.3 weeks and 33.1 to 40.4 weeks, respectively (Table 1). 20 out of 35 patients (57%) had associated malformations, 2 out of 4 (50%) of the ELBW, 5 out of 7 (71%) of the VLBW and 13 out of 24 (54%) of the ≥1500 g patients (Table 1). Follow-up for the ELBW group was 57.5 (1–112) months, for the VLBW group 25 (10–79) months and for the ≥1500 g group 27 (3–156) months (Table 1).

### *2.2. Intra- and postoperative complications*

In one patient of the ≥1500 g group an intraoperative complication occurred (injury of the trachea). The incidence of anastomotic stricture was 25% (1/4) in the ELBW, 28.5% (2/7) in the VLBW and 37.5% (9/24) in

**Table 1**
Characteristics of patients with primary repair.

| | ELBW ($n = 4$) | VLBW ($n = 7$) | ≥1500 g ($n = 24$) |
|---|---|---|---|
| Gestational age [wk] | 28.5 (27.7–31.7) | 31.6 (29.9–32.3) | 37.3 (33.1–40.4) |
| Birth weight [g] | 840 (780–980) | 1300 (1000–1495) | 2525 (1770–3440) |
| Associated malformations | $n = 2$ | $n = 5$ | $n = 13$ |
| | • Hypospadia $n = 1$<br>• PFO $n = 1$ | • Laryngotracheal cleft $n = 1$<br>• Major cardiac anomaly $n = 2$<br>• Imperforate anus $n = 2$<br>• VACTERL $n = 1$ | • Major cardiac anomaly $n = 9$<br>• Imperforate anus $n = 4$<br>• Duodenal atresia $n = 1$<br>• Tethered cord $n = 3$<br>• VACTERL $n = 4$ |
| Follow-up [mo] | 57.5 (1–112) | 25 (10–79) | 27 (3–156) |

PFO = patent foramen ovale; VACTERL = vertebral, anorectal, cardiac, tracheoesophageal, renal and limb anomalies.