



Cervical repair of congenital tracheoesophageal fistula: Complications lurking! ☆☆☆☆☆



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ABSTRACT

Aim: Esophageal atresia (EA) and tracheoesophageal fistula (TEF) consist of a spectrum of rare congenital abnormalities. Although EA surgical treatment is well established, the outcome of EA with proximal fistula (type B and D EA) or isolated H-type fistula (type E EA) is poorly explored. These forms of EA shared a common surgical step: the need of a cervical approach to close the fistula. Therefore, the aim of present study is to evaluate postoperative outcomes of patients treated for Gross type B–D and E EA, on regards of their cervical surgery.

Materials and methods: A retrospective case series analysis of all patients affected by type B–D and E EA, and admitted to our tertiary care center between January 2003 and December 2014 was performed. All patients underwent preoperative flexible laryngo-tracheobronchoscopy (LTBS) as part of our standardized preoperative diagnostic assessment to define the diagnosis, evaluate preoperative vocal cord motility and to cannulate the fistula when required. Fistula closure was always performed through a right cervical access. Analysis of all cases and comparison between type B–D and E EA were performed. Mann–Whitney test, Chi-squared test and unpaired t test were used as appropriate; $p < 0.05$ was considered significant.

Results: During the study period, 180 EA newborns were treated. Proximal or isolated TEF was found in 18 patients (10%): 7 type B, 11 type E EA. Patients affected by type B and E EA/TEF frequently present associated major malformations (27%), and major cardiac abnormalities (44%). Major postoperative complications were: vocal cord paralysis (5 patients), bilateral in 2 infants requiring tracheostomy, cerebral ischemia (1 patient), and cardiac failure (1 patient).

Conclusion: Patients affected by type B and E EA have a high rate of associated abnormalities, and risk of possible sequelae. Postoperative complications are common, with possible transient vocal cord dysmotility, but in some cases persistent paralysis may require tracheostomy. Therefore, both preoperative and postoperative LTBS is highly recommended to evaluate the presence of a proximal fistula, and vocal cord motility, even in asymptomatic patients, to rule out any possible intraoperative “surprise” and any vocal cord abnormality and to possibly define its pathogenesis (congenital vs. iatrogenic).

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Esophageal atresia (EA) with tracheoesophageal fistula (TEF) consists of a spectrum of rare congenital abnormalities that has an incredible attractiveness for pediatric surgical scientific community, with more than 1200 articles published in the last 10 years (PubMed

search 1st December 2015 “esophageal atresia”). Despite the broad interest on the condition, management and outcomes of patients treated for Gross type E EA type are poorly explored: a recent systematic review, found only 17 article reporting data on 90 patients treated for isolated tracheoesophageal fistula, between 1977 and 2012 [1]. Furthermore, most of those articles focused their attention on diagnosis and operative management rather than on postoperative outcomes.

No article compares postoperative outcomes of isolated TEF (type E EA) with TEF of proximal pouch (types B and D EA). This is particularly interesting since the vast majority of type E EA is repaired through cervical route as most of the proximal fistulas in case of types B and D EA.

Aim of the present study was to critically evaluate short-term postoperative outcomes of patients treated with a cervical approach to

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repair a TEF. The present study compares patients with EA types B and D, with those affected by type E EA.

1. Material and methods

All patients who had a hospital discharge with a IDC-9-CM code 7503 (tracheoesophageal fistula and esophageal atresia), between January 2003 and December 2014 entered the present study.

For the purpose of the present study, only those patients affected by EA type B, D and E, according to Gross classification were selected. The Institutional Review Board approved the study. Clinical files of all patients treated were evaluated: patient's characteristics and surgical data recorded. Main outcomes considered were: vocal cord (VC) motility at preoperative flexible laryngo-tracheobronchoscopy (LTBS), days of postoperative intubation, number of intubations, postoperative stridor, dysphonia, or respiratory symptoms, tracheostomy, postoperative VC paralysis, other postoperative major complications, length of hospital stay and mortality. Postoperative major complications included: esophageal, gastric, pulmonary, cardiac, renal, vascular problems before discharge. To analyze the outcome, patients were divided into two groups based on EA type: Group A – patients affected by type B, D EA; Group B – patients affected by type E EA.

1.1. Initial clinical workout

All patients admitted to our unit for the suspect of EA underwent preoperative blood analysis and a chest X-ray with a radiopaque 10 Ch nasogastric tube, to confirm/exclude proximal blind esophagus. Usually, patients with a suspected type E EA were referred to our hospital for choking during meal and/or recurrent pulmonary inhalations/infections.

All newborns were preoperatively screened for major associated abnormalities. In particular the screening included: chest and abdominal X-ray searching for vertebral anomalies, cerebral, renal and abdominal ultrasound for associated abnormalities, electrocardiographic and echocardiographic assessment to define out cardiac anatomy and function.

1.2. Laryngotracheoscopic evaluation and esophageal surgery

An educated/skilled team of neonatal otorhinolaryngologist/surgeons routinely performed a preoperative flexible LTBS, to define: the native vocal cord motility, the level and the number of esophageal fistulas, to rule out laryngotracheal malformations and to preoperatively assess esophageal gap in case of type D EA [2].

In case of suspected type E EA, preoperative LTBS was the gold standard to diagnose isolated TEF. Contrast study was never used in any type of suspected EA to show the blind upper pouch or to rule out the presence/absence of type E EA/TEF since the intrinsic risk of aspiration and the high rate false negative results.

Once type B or E EA was confirmed, a guide wire was inserted from the trachea into the fistula and withdrawn through the esophagus and the mouth: the guide wire allowed an easier intraoperative detection and identification of the fistula [3]. Preoperative fluoroscopy was performed to confirm the correct position of the guide wire into the fistula and show exactly the cervical/thoracic level of the “intubated” fistula.

Repetition of flexible laryngotracheoscopy was performed when stridor, dysphonia or respiratory symptoms were present postoperatively. Tracheostomy was also considered when ventilation weaning was not achievable after 4 weeks overall.

Esophageal surgery was performed in all cases as an elective procedure, according to our standardized approach [4].

1.3. Surgical technique

All patients underwent TEF closure by senior author (PB) or under his direct supervision.

A “J” shaped right cervical incision extending from jugulum to anterior aspect of sternocleidomastoid muscle was used in all cases. Carotid artery, internal jugular vein, and vagus nerve were gently retracted posteriorly. A minimal dissection, guided by the previously endoscopic positioned guide wire, was performed at the level of TEF, to isolate the esophageal aspect of the fistula. Once TEF was isolated and divided, both sides were separately closed with interrupted long-lasting absorbable monofilament stitches (5/0 PDS II, Ethicon, Inc., 2012). Contrast study was performed on postoperative day 6 to rule out possible esophageal leak.

All patients affected by type B EA, underwent a subsequent thoracotomy to correct the atresia. A delayed primary anastomosis was performed in all cases, and a nasogastric tube was left in place until a contrast swallow excluded esophageal leakage. The contrast study was usually performed on postoperative day 6.

1.4. Statistical analysis

Results were expressed as percentage, or medians and interquartile range. Mann–Whitney test, Chi-squared test and unpaired t-test were used as appropriate. *P* value less than 0.05 was considered significant. The statistical package used was Prism 5 for Mac OS X, GraphPad Software, Inc.

2. Results

One hundred-eighty EA-affected newborns were admitted during the study period. Proximal or isolated TEF was found in 18 patients (10%): 7 Gross type B EA (Group A), 11 Gross type E EA (Group B) [5]. No patient affected by type D EA/TEF was identified.

Demographic characteristics were summarized in Table 1. Birth weight was significantly lower and age at diagnosis significantly earlier in Group A. Preoperative LTBS ruled out congenital VC paralysis or cleft in all cases.

No differences were found between the two groups in the postoperative course (Table 2). In particular no differences concerning VC paralysis either monolateral [Group A 1 (14%), Group B 2 (18%): *p* 1.0] or bilateral [Group A 1 (14%), Group B 1 (9%): *p* 1.0], the side of monolateral VC (right sided in all cases) and the need for tracheostomy [Group A 1 (14%), Group B 1 (9%): *p* 1.0] were observed. Length of hospital stay was significantly longer for type B EA patients, since esophageal anastomosis is commonly delayed. No death was related to EA repair.

In all cases of postoperative VC paralysis, since the development of respiratory symptoms, primary treatment approach was “wait and see”, with elective temporary (2 to 4 days) oro-tracheal intubation to stent VC, associated with short-term endovenous steroids administration (dexamethasone 0.2 mg/kg/day). Repeated oro-tracheal intubations were used when deemed necessary, with no differences between the two groups, [Group A 34.5 days (IQR, 25–44) vs. 14 days (IQR, 2–30) in Group B: *p* 0.23].

Tracheostomy was selectively performed in the two patients with bilateral VC paralysis.

All symptomatic patients underwent elective LTBS 3–6 months later to re-assess VC motility. One patient with tracheostomy underwent CO₂ laser-posterior cordotomy with CO₂ laser at 2-year of age, and he was subsequently decannulated. The second patient with tracheostomy is still too young to undergo cordotomy. Spontaneous regression of VC dysmotility was observed in all cases of monolateral paralysis at 6 months LTBS.

In 3 patients (2 type B and 1 type E EA) temporary feeding and swallowing disorders were observed. Two of these patients had a bilateral VC impairment too. Feeding and swallowing problems were overcome by speech pathology rehabilitation in all cases.

A major complication was observed in one patient, affected also by trisomy 21, which experienced left hemisphere cerebral ischemia because of agenesis of left middle cerebral artery. West syndrome

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