Laparoscopic Fundoplication after Esophageal Atresia Repair

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Abstract
To evaluate our experience with Gastro-Esophageal Reflux (GER) treatment after Esophageal Atresia (EA) repair. Esophageal Atresia (EA) is a rare congenital malformation. A high incidence of GER unresponsive to medical management is noted with EA. Literature suggests that complications from GER can persist in adulthood.

In pediatric age, laparoscopic treatment is a valid option even if recurrence rate is not negligible. We retrospectively analyzed 29 consecutive patients treated for EA at birth and studied for GER at our Institute in a period of 11 years. 24/29 (82.7%) cases had symptoms of reflux, 17/29 (58.6%) cases were treated with Laparoscopic Fundoplication (LF). Three infants were younger than 6 months and had Apparent Life Threatening Events (ALTE) condition as principal indication for surgery. No intra-operative complications occurred. 3/17 LF had opens surgical conversion due to technical problems. 2/17 cases required a second operation. At the last follow-up: - 6/17 (35.3%) of patients healed after the last operation!

Keywords: Esophageal atresia; Gastroesophageal reflux; Laparoscopic treatment

Abbreviations
EA: Esophageal Atresia; GER (D): Gastro-Esophageal Reflux (Disease); LES: Low Esophageal Sphincter; LF: Laparoscopic Fundoplication; ALTE: Apparent Life Threatening Events; G: Gastrostomy; J: Jejunostomy; CCAM: Congenital Cystic Adenomatoid Malformation; ARM: Ano-Rectal Malformation; TEF: Tracheo-Esophageal Fistula

Introduction
Esophageal Atresia (EA) is a rare congenital malformation (1: 3500 live births). Mortality rate after EA repair is low and depends on birth weight and other associated malformations, while long term morbidity is not to be neglected. Gastro-Esophageal Reflux (GER) is the most common long-term complication. The poor esophageal motility, the inadequate clearance of acid from the esophagus, the shortening of the esophagus and the abnormal Low Esophageal Sphincter (LES) pressure predispose patients to develop GER [1].

In literature, a clinical improvement after school age and adolescence is reported [2]. Due to many bias in the epidemiological studies, the actual prevalence of GER is not clear, it ranges from 25% to 75% of the cases [3,4] depending on anatomical changes of the gastroesophageal region after EA repair and on pathophysiological mechanisms like reduced esophageal clearance. Medical treatment with H2-antagonist, prokinetics and proton pump inhibitors can be efficient, but if no improvement occurs a surgical approach is suggested. Children with esophageal atresia have high incidence of symptomatic reflux that is not responsive to medical therapy [5]. According to Tovar more than 40% of these cases require surgical correction [6]. Among this group of patients, GER late recurrence rate after fundoplication procedures is high [7].

Materials and Methods
From January 2004 to September 2015, 29 consecutive patients treated for EA were studied for GER at our Institute and were available for follow-up. GER study included: contrast X-ray, endoscopy, 24 hours impedance-pH monitoring. The standard follow-up consisted on clinical evaluations 1 and 3 months after AE repair, contrast X-ray studies after 6 and 18 months and, endoscopic procedures after 12 and 24 months associated with esophageal 24 hour impedance-pH monitoring when possible, clinical evaluations and/or repetition of the examinations once a year in persistent GER cases. The medical records of all patients were reviewed and the demographic
and surgical data have been analyzed. Standard surgical treatment consisted in a complete or partial Laparoscopic Fundoplication (LF). Our aim was to evaluate the outcome of EA patients who received GER surgical treatment.

A visit or a telephone call represented the last follow-up. We considered:

- Resolution of GER
- Improvement of GER symptoms out of medical therapy
- Improvement of GER symptoms with medical therapy
- Recurrent GER requiring redo-operation.

We describe: EA type, associated conditions, age at surgery, presence of gastrostomy or jejunostomy, indications and type of surgical treatment.

**Results**

From January 2004 to September 2015, 29 EA patients were studied for GER. 28/29 newborns had type III (Gross type C) three of which were long gap form, 1/29 had type I (Gross type A) long gap atresia. Both X-ray contrast and endoscopic studies were performed in 25/29 cases; X-ray contrast studies, endoscopic procedures and esophageal 24 hr impedance–pH monitoring studies were performed in 14/29 cases. 18/29 children presented associated conditions like prematurity, other malformations, presence of ventricular-peritoneal derivation. 25/29 newborns belonged to group I according Spitz risk classification: patients’ birth weight was >1500 gr without major cardiac defect; 4/29 newborns belonged to group II having a birth weight <1500 gr. 12/29 had a gastrostomy (ten cases) or a jejunostomy (two cases). 24/29 patients (12 males, 12 females) presented symptomatic GER (82.7%) and 17/29 cases (5 males and 12 females), were surgically treated (58.6%). All long gap EA needed surgical therapy except for one (Gross C, III type). LF was the initial approach in all patients and skilled surgeons did it at a median age of 37.3 months (range 4-204 months). 3/29 infants were younger than 6 months (weight between 4 and 8 kg) with Apparent Life Threatening Events (ALTE) condition as principal indication for surgery. In all cases, an evaluation to rule out other causes of respiratory difficulties that could assume to be gastro-esophageal reflux was done. Surgical treatment of GER consisted in 6/17 complete fundoplication (Nissen), and 11/17 partial wrap (Thal or Toupet). 3/17 (17.6%) laparoscopic procedures had open surgical conversion due to technical problems: two of them were low weight infants, all three patients had with gastrostomy or jejunostomy. In one case an important hepatomegaly increased fundoplication failure rates in small children [13]. In our experience 3/17 cases (17.6%) were converted to open surgery: in all 3 cases a gastrostomy or a jejunostomy was present at the time of surgery. 2/17 cases (11.7%) had a redo-operation: both were patients with associated malformations; one of them was a small infant with jejunostomy at the time of operation Kubiak et al. Observed a high incidence of failure and redo fundoplication in infants with associated between the 3rd and 12th day after the operation: the longest hospital staying was not related to surgery.

2/17 (11.8%) patients with associated malformations required a second fundoplication: one of them was performed in a low weight infant (age <6 months) with ALTE, and presence of jejunostomy at time of surgery.

At the last follow-up (visit or call phone):

- 6/17 (35.3%) of patients healed after the last operation
- 4/17 (23.5%) have GER improvement out of medical therapy
- 4/17 (23.5%) have GER improvement still in medical therapy
- 2/17 (11.8%) have recurrent GER
- 1/17 (5.9%) died for causes not related to anti reflux surgery.

In summary 82.3% of patients had a good outcome after surgical treatment. The average follow-up was 80 months (range 1 to 140 months). In Figure 1 and Figure 2 we resume our results.

**Discussion**

Recent studies report a high percentage of GER complications in the adult age causing GER Disease (GERD) in patients operated of EA. In a Finnish adult study published in 2010, the occurrence of symptomatic GER is 34% and the occurrence of dysphagia is 85% compared with 8% and 2% respectively among general population [8]. According to Rintala [9], in adulthood approximately one fifth of EA patients develop epithelial metaplastic changes; one-third of these have intestinal metaplasia (Barrett’s esophagus). It generally occurs at a much younger age than general population [10]. In the long run, these patients have 50-fold higher risk of carcinoma than the control population [11].

According to Tovar [6] and to our experience more than 40% of EA patients are refractory to medication and require surgical correction. In a retrospective study published in 2010 by the Great Ormond Street Hospital group, the laparoscopic GER operation is considered an appropriate treatment for EA/GER. It is feasible and effective even in children under one year old if performed by skilled laparoscopic surgeons [12]. Anyway, other Authors report increased fundoplication failure rates in small children [13]. In our experience 3/17 cases (17.6%) were converted to open surgery: in all 3 cases a gastrostomy or a jejunostomy was present at the time of surgery. 2/17 cases (11.7%) had a redo-operation: both were patients with associated malformations; one of them was a small infant with jejunostomy at the time of operation Kubiak et al. Observed a high incidence of failure and redo fundoplication in infants with associated complications occurred. All patients were fed after 6 hrs from the surgical procedure and they were discharged from the hospital.
anomalies, particularly those with EA: almost half of these patients showed no improvement of their symptoms after fundoplication [14]. Tovar reports surgery failure rate in 30% of cases [11]. As far as the type of LF is concerned, Snyder reports that a complete wrap (Nissen) is inappropriate for GER associated with EA. Severe esophageal dysmotility may contribute to poor passage of food through the gastro-esophageal junction, especially against the increased resistance provided by a complete wrap, so partial fundoplication could be theoretically more attractive with a minor risk of prolonged dysphagia [1]. In literature, a clinical GER improvement after school age and adolescence is reported [2], anyway serious respiratory disorders related to GER are a strong indication for surgery even in very small babies [15]. Sload et al. in a systematic review in 2014 suggest that anti-reflux surgery is an effective and safe treatment for severe reflux related airway disease [16].

In our experience, one of the six Nissen operations required a conversion to open surgery and another one failed and required a redo-procedure: in both cases the patients were small infants with associated conditions, one of these cases had a very short esophagus due to a long gap Gross C, III type EA repair. Finally our GER improvement rate after surgery was acceptable.

**Conclusion**

Laparoscopic treatment for GER after EA repair is efficient and feasible. According to literature the risk of recurrence after antireflux surgery is higher for low weigh infants compared with general population. Our number of cases is small, but in this retrospective series, low weigh infants younger than 6 months old can be initially treated with LF even if both, the risk of conversion to laparotomy and the rate of failure increase, especially for long gap cases. Associated conditions like malformations, presence of gastrostomy/jejunostomy seems to increase the risk. Anyway, surgery is mandatory for low weight infants with serious respiratory disease related to reflux refractory to medical therapy. While mortality for low risk EA patients is negligible, morbidity is not so low. All individuals who have undergone surgery for EA should be assessed clinically in the adolescence and adult age [17,18]. A careful surveillance and further studies will be necessary to understand the outcome of EA/RGE patients.

**References**