Original Article

Laparoscopic Fundoplication after Oesophageal Atresia Repair

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Abstract

Background: 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 paediatric age, laparoscopic treatment is a valid option even if recurrence rate is not negligible. **Aims and Objectives:** To evaluate our experience with gastro-esophageal reflux (GER) treatment after esophageal atresia (EA) repair. **Materials and Methods:** We retrospectively analysed 29 consecutive patients treated for EA at birth and studied for GER at our Institute in a period of 11 years. **Results:** 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 open surgical conversion due to technical problems. 2/17 cases required a second operation. At the last follow-up: (1) 6/17 (35,3%) of patients healed after the last operation, (2) 8/17 (47,1%) have GER improvement (four still in medical treatment), (3) 2/17 (11,8%) have persistent GER, (4) 1/17 (5,9%) died for causes not related to antireflux surgery. **Conclusions:** According to literature and to our retrospective analysis, LF for GER after EA repair is feasible, even if recurrence risk is not negligible. Infants less than 6 months old with associated conditions (malformations, gastrostomy/jejunostomy) seem to have a higher failure rate with a greater risk of conversion. Longer follow-up and multicenter experiences would guarantee an adequate surveillance for patients with EA.

Keywords: Oesophageal atresia, gastro-oesophageal reflux, laparoscopic fundoplication

Introduction

Oesophageal atresia (EA) is a rare congenital malformation with significant long-term morbidity. The most common complication is gastro-oesophageal reflux (GER) due to poor oesophageal motility, inadequate acid clearance, shortening of the oesophagus and abnormal lower oesophageal sphincter (LES) pressure.[1] A clinical improvement after school age has been reported.[2] The actual prevalence of GER ranges from 25% to 75% of the cases.[3,4] Children with EA have a high incidence of symptomatic GER unresponsive to medical therapy.[5] More than 40% of them require surgical correction[6] and the GER late recurrence rate after fundoplication procedures is high.[7] The objective of this study was to confirm the feasibility of laparoscopic fundoplication (LF) after EA repair and the medium-term outcomes.

Materials and Methods

This study was based on the medical records of all consecutive patients treated for EA and studied for GER from January 2004 to September 2015 in a single centre.

During the same period, 31 patients were operated for EA.

GER studies included: Contrast X-ray, endoscopy, 24-h impendance pH monitoring.

The standard follow-up consisted of clinical evaluations 1 and 3 months after EA repair, contrast X-ray studies at 6 and 18 months and endoscopic procedures associated with oesophageal 24-h impendance–pH monitoring, when possible,

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at 12 and 24 months, 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 were analysed.

The institutional policy during the study period indicated a surgical approach in case of failure of a well-conducted medical treatment based on posture, diet and appropriate antacid and/or prokinetic medication. Standard surgical treatment consisted of a complete or partial LF. A visit or a telephone call represented the last follow-up. The outcomes considered were as follows:

1. Resolution of GER
2. Improvement of GER symptoms without medical therapy
3. Improvement of GER symptoms with medical therapy
4. Current GER requiring redo-operation [Table 1].

Statistical analysis was performed using the Statistical Package SAS 9.4 (SAS Institute, Inc., Carey, NC, USA). For the categorical variables, Fisher's exact analysis was used. For the continuous variables, the Wilcoxon test was applied. Two-sided $P < 0.05$ was considered statistically significant.

Results

From January 2004 to September 2015, 29 patients were managed for EA and studied for GER. Twenty-eight new-borns had type III (Gross type C) atresia, three of which were long-gap form and one had type I (Gross type A) long-gap atresia.

Both X-ray contrast and endoscopic studies were performed in 25 cases; X-ray contrast studies, endoscopic procedures and oesophageal 24-h impedance–pH monitoring studies were performed in 14 cases.

Eighteen children presented with associated conditions such as prematurity, other malformations and ventriculoperitoneal shunt. Twenty-five belonged to Group I according to the Spitz risk classification (patients' birth weight was >1500 g without major cardiac defect) and 4 belonged to Group II (birth weight <1500 g). Twelve had a gastrostomy (ten cases) or a jejunostomy (two cases). Twenty-four patients (12 males and 12 females) presented with symptomatic GER (82.7%) and 17 cases (5 males and 12 females) were surgically treated (58.6%). All long gap EA needed surgical therapy except one case (Gross C, III type). However, the need for fundoplication did not depend on the type of AE ($P = 0.621$). LF was the initial approach in all patients and skilled surgeons performed the procedure at a median patient age of 37.3 months (range 4–204 months). Three infants were younger than 6 months (weight between 4 and 8 kg) with apparent life-threatening events (ALTE) as the principal indication for surgery. In all cases, an evaluation was performed to exclude other causes of respiratory difficulties than GER. The surgical treatment of six GER patients was a Nissen complete fundoplication and the other 11 were treated with Thal or Toupet partial fundoplication. The rate of gastrostomy performed at the time of EA repair (66.6% and 45.4%, respectively) did not influence the type of fundoplication ($P = 0.59$).

Table 1: Follow-up data related to the last visit or telephone call

Outcome	Patients, *n* (%)
Healing	6 (35.3)
GER symptoms improvement (without therapy)	4 (23.5)
GER symptoms improvement (with therapy)	4 (23.5)
Redo-surgery	2 (11.8)
Death	1 (5.9)
Total	17 (100)

GER: Gastro-oesophageal reflux

Three (17.6%) laparoscopic procedures were converted to open surgery due to technical problems: Two were low weight infants and all three patients had gastrostomy or jejunostomy. In one case, hepatomegaly was the reason for conversion; in the second, it was due to anaesthesiologic difficulties with poor compliance to pneumoperitoneum; in the third, the reason was the presence of gastrostomy and a small stomach. No intraoperative complications occurred.

All patients were fed after 6 h from the surgical procedure, and they were discharged from the hospital between the 3rd and 12th day after the operation: The longest hospital stay was not related to surgery.

Two (11.8%) patients with associated malformations required a second fundoplication: One was a low-weight infant (age <6 months) with ALTE, and the presence of jejunostomy at the time of surgery. A summary of our cases is reported in Table 2.

The median follow-up was 80 months (range 1–140 months). In summary, 82.3% of patients had a good outcome after surgical treatment (healing or improvement of the symptoms) [Figures 1 and 2].

Discussion

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

According to Tovar and Fragoso,[6] in 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

Table 2: Summary of our cases

EA type	Associated conditions	Age at first surgery in months (weight in kg reported in only small infants)	Indications for surgery	Stomy (gastrostomy/ jejunostomy)	Type of surgery
IIIC	Prematurity, PS	4 (4)	ALTE - no growth	Jejunostomy	Thal
IIIC	Hydrocephalus - VP shunt	5 (8)	ALTE - hiatus hernia	Gastrostomy	Nissen
IIIC LG	Prematurity, PS	5.5 (4.5)	ALTE - hiatus hernia - no growth	Jejunostomy	Nissen*
IIIC	PS-tracheostomy	1	Hiatus hernia - recurrent respiratory infections	Gastrostomy	Toupet
IIIC	PS	12	No growth - recurrent respiratory infections	Gastrostomy	Nissen
IIIC LG	No	17	No growths - no medical response	Gastrostomy	Thal
IIIC	No	17 (9)	Hiatus hernia - recurrent respiratory infections - no growth	No	Thal
IIIC	Prematurity	23	No medical response	No	Toupet
IIIC	Cleft palat, convulsions	24 (11)	Hiatus hernia - esophagitis	No	Toupet
IIIC	Anorectal malfromation	30	GERD - no medical response	No	Nissen
IIIC	/	36	GERD - no medical response	Gastrostomy	Thal
IA LG	Anorectal malformation, PS	36	GERD - no medical response	Gastrostomy	Thal
IIIC	No	60	Hiatus hernia - recurrent respiratory infections - esophagitis	No	Toupet
IIIC	Recurrent TEF	84	GERD - no medical response	No	Nissen
IIIC	PS	84	Persistent dysphagia and recurrent GERD	No	Thal**
IIIC	No	120	GERD - no medical response	No	Nissen
IIIC	Hydrocephalus – ventriculoperitoneal shunt	204	Barret	No	Toupet

*Patient underwent Redo-Nissen, **Patient underwent Redo-Thal. TEF: Tracheo-esophageal fistula, ALTE: Apparent life-threatening events, LG: Long-gap, GERD: Gastro-oesophageal reflux disease, VP: Ventriculoperitoneal, PS: Polymalformative syndromes

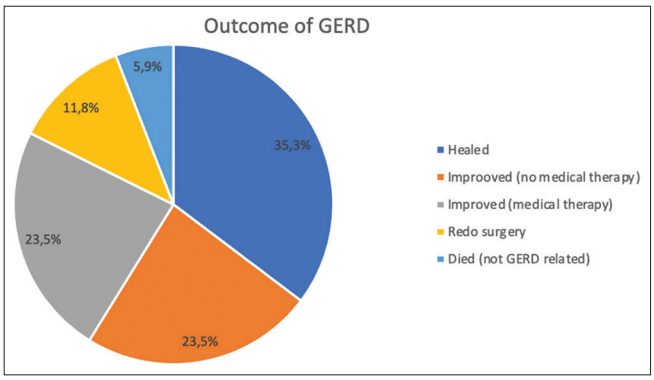

Figure 1: Outcomes after fundoplication

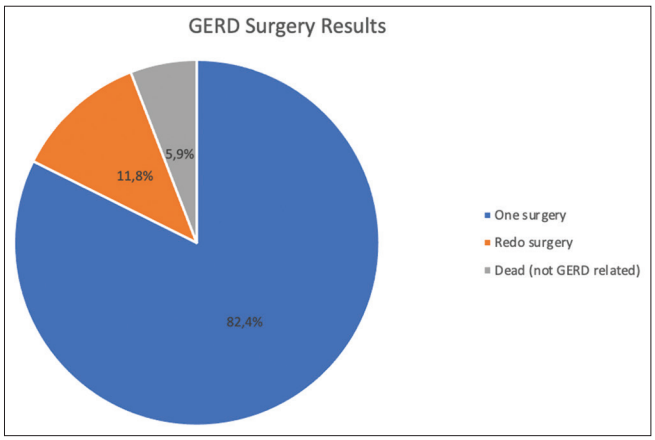

Figure 2: Fundoplication success after primary surgery

operation is considered an appropriate treatment for EA with GER. It is feasible and effective even in children under 1 year old if performed by skilled laparoscopic surgeons.[12] However, other authors have reported increased fundoplication failure rates in small children.[13] In our experience, three cases (17.6%) were converted to open surgery: In all three cases, a gastrostomy or a jejunostomy was present at the time of surgery.

Kubiak *et al*. observed a high incidence of failure and repeated fundoplication in infants with associated anomalies, particularly those with EA. Almost half of these patients showed no improvement of their symptoms after fundoplication.[14] Tovar and Fragoso[11] reported surgery failure rate in 30% of cases.

Regarding the type of LF, Snyder *et al*. reported that a complete wrap (Nissen) is inappropriate for GER associated with EA. Severe oesophageal dysmotility may contribute to poor passage of food through the gastro-oesophageal junction, especially against the increased resistance provided by a complete wrap, so partial fundoplication could theoretically be more attractive, with a minor risk of prolonged dysphagia.[1] Although it is well known that Thal method can be considered advantageous because of maintenance of the LES and low anatomical dissection, in our experience, there was no statistical difference in failure of fundoplication between the two groups. However, it is important to bear in mind that our case series had a small sample size.

A clinical GER improvement after school age and adolescence has been reported.[2] However, serious respiratory disorders related to GER are a strong indication for surgery even in

very small babies.[15] In a systematic review in 2014, Sload and Brigger[16] suggested that antireflux surgery is an effective and safe treatment for severe reflux-related airway disease.

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 had a very short oesophagus due to a long-gap Gross C, III type EA repair [Table 2]. Finally, our GER improvement rate after surgery was acceptable. However, complete control of symptoms was not achieved, especially in patients for whom the indication of fundoplication was ALTE or respiratory symptoms. This finding might be explained by the multifactorial aetiology of ALTE in AE, including GERD, tracheomalacia and greater reactivity of the respiratory tract.[17] The main limitation of this study is the small sample of patients. Its strength is the fact that these patients were consecutively included in a specific follow-up schedule. A multicentre longitudinal prospective follow-up might help to define the most appropriate treatment in such cases since AE patients can manifest lifelong symptoms.

Conclusion

Laparoscopic treatment for GER after EA repair is efficient and feasible. According to the literature, the risk of relapses after antireflux surgery is higher for low-weight infants compared with the general population. Both the risk of conversion to open surgery and the rate of failure are increased in long-gap cases and the presence of associated conditions (malformations and gastrostomy/jejunostomy). Morbidity for EA patients is relevant and requires clinical assessment in adolescence and adulthood.[18,19] A careful follow-up and further multicentre studies will be necessary to understand the outcomes of patients affected by GERD after EA repair.

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Conflicts of interest

There are no conflicts of interest.

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