

Pediatr Surg Int (2014) 30:997–1001
DOI 10.1007/s00383-014-3584-9

ORIGINAL ARTICLE

Esophagogastric dissociation reduces the re-operation rate for persistent gastroesophageal reflux in severely neurologically impaired children

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Accepted: 5 August 2014 / Published online: 20 August 2014
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Abstract

Purpose In this study we want to demonstrate the effectiveness of the esophagogastric dissociation as a first level operation in treatment of the gastroesophageal reflux in severe neurologically impaired children, in term of a reduction of reoperation rate.

Methods We divided patients operated from 1998 to 2005 in a group A, composed by children treated with fundoplication, and in a group AR, composed by the patients of group A who had a recurrence of reflux and that was treated with esophagogastric dissociation. Patients operated from 2005 to 2013 were selected on the basis of the severity of the neurological impairment and were divided in a group B, treated with fundoplication, and in a group C of more severe impaired children, treated with esophagogastric dissociation. Data regarding the complications of the A and C groups were analyzed with Fisher's test.

Results We evaluated 63 patients: 34 (54 %) in group A, 11 in group AR, 15 (23.6 %) in group B, 14 (22.4 %) in group C. The Fisher's test showed a non significant difference with a *p* value of 0.2.

Conclusion Despite of statistic result we believe that TOGD is a useful procedure as the first choice of surgical management in severe neurological impaired children affected by gastroesophageal reflux.

Keywords Gastroesophageal reflux · Neurologically impaired children · Fundoplication · Esophagogastric dissociation

Introduction

Gastroesophageal reflux disease (GERD) is a common problem in childhood, especially in those cases with severe neurological impairment. In these children the prevalence of GERD is reported to be between 33 and 75 % [1, 2].

Fundoplication and gastrostomy may be effective in resolving symptoms, in facilitating nutrition, reducing the risk of aspiration pneumonia, and thus improving quality of life of these patients. However, symptom recurrence often heralds failure of fundoplication. The failure rate in neurologically impaired children has been reported to be between 12 and 45 % [3–5].

The failing outcome of the fundoplication in this kind of patients is consequence of a variety of factors (seizure activity, constipation, aerophagia, gut dysmotility) which, through the increase the intraabdominal pressure, can stress the fundoplication wrap. A re-do Nissen fundoplication has been reported to have a failure rate of 15–28 % [3, 6, 7].

In the last decades, Bianchi proposed esophagogastric dissociation not only as a second level surgery after a failed fundoplication, but also as a first practice to totally eliminate the gastroesophageal reflux in the severe neurologically impaired children [8].

In this retrospective study, we report an experience of 15 years of antireflux surgery, to demonstrate how the management of GERD change in relation to the neurological impairment considered. Thus, on the basis of this change we started to practice esophagogastric dissociation as a good practice in treatment of gastroesophageal reflux

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Table 1 Neurological impairment of the patients

Neurological impairment	Group A	Group AR	Group C	Group D
Neuromotor delay	11 (32.3 %)	6 (54.5 %)	4 (26.6 %)	
Epileptic encephalopathy	7 (20.5 %)		3 (20 %)	
Spastic quadriplegia	5 (14.8 %)	2 (18.2 %)	4 (26.6 %)	
Congenital malformation syndrome	5 (14.8 %)	2 (18.2 %)	3 (20 %)	
Rett syndrome	2 (5.9 %)			3 (21.5 %)
Cerebral palsy				4 (28.6 %)
Autism				2 (14.3 %)
Idrocephalus	2 (5.9 %)			
SMA	1 (2.9 %)	1 (9.1 %)		1 (7.1 %)
Muscular dystrophy				2 (14.3 %)
Anencephaly	1 (2.9 %)			1 (7.1 %)
CHARGE syndrome			1 (6.8 %)	
PESS				1 (7.1 %)

Table 2 Preoperative symptoms in patients of group C

Symptoms	Patients (Group C)
Failure to thrive	4 (28.6 %)
Recurrent chest infection	5 (35.9 %)
Dysphagia	2 (14.2 %)
Vomiting	1 (7.1 %)
Retching	2 (14.2 %)

in severe neurologically impaired children, especially in term of a reduction of reoperation rate and of morbidity and mortality.

Materials and methods

At the department of Clinical Pediatric Surgery in Siena, from January 1998 to March 2013, 63 neurologically impaired children were surgically treated for GERD.

From January 1998 to January 2005 we treated patients with laparoscopic fundoplication as a procedure of first level, and with TOGD (total oesophago-gastric dissociation) only as a rescue surgery to treat GERD's recurrences. Then, until March 2011, we considered TOGD also as a first procedure in selected cases. On the basis of this idea, we previously identified the patients affected by serious neurological disease, on the basis of underlying neurological pathology, clinical features and the degree of disability. For this scope we took as a parameter the GMFCS-E&R (Gross Motor Function Classification System Expanded and Revised), which considers ambulation, functional limitations, the need for devices for mobility or mobility in a wheelchair and to a lesser extent on the quality of the movement. It consists of five levels, of which the fifth (to be transported in a wheelchair) is what

we used to distinguish patients with more severe neurological deficit. In addition to this assessment scale we took into account the degree of feeding difficulties and breathing problems, carefully assessing their clinical condition.

The neurological problems in our population are shown in Table 1. As we can see until 2005 the patients are not divided on the basis of their impairment, from this date, instead, there is a distinction basing on the severity of the handicap considered. The main symptoms we valued crucial to consider more severe a patient, were: failure to thrive, recurrent chest infection, vomiting, dysphagia and retching (Table 2).

We divided all patients in four groups: group A with patients treated with fundoplication from 1998 to 2005, group AR with patients treated with TOGD as a redo reoperation from 1998 to 2005, group B with patients treated with fundoplication from 2005 to 2013, group C with patients treated with TOGD as a first level surgery from 2005 to 2013.

For each patient of each group, information was gathered about the neurological disease associated with the preoperative symptoms, diagnostic tests performed and postoperative complications.

We evaluated the diagnostic procedure followed by patients (upper gastrointestinal X-ray and esophagogastroduodenoscopy before surgery and 24 h esophageal pH monitoring in a part of patients of group C) and postoperative complications by analyzing their performance over time and comparing them to the type of surgery performed. We considered positive for GERD patients with a DeMeester score >18, with esophageal acid exposure values measured by the integral H^+ >110 mmol/l/min, with integral over threshold to pH <4 resulted >34 pH/min.

The data regarding the complications of the A and B groups were analyzed with Fisher's test.

Table 3 Complications during follow-up

Complications	Group A	Group AR	Group B	Group C
Recurrence of gastroesophageal reflux	11 (32.3 %)		2 (13.4 %)	
Dehiscence		1 (9 %)		
Bleeding		1 (9 %)		1 (7.1 %)
Stricture of anastomosis				1 (7.1 %)
Dumping syndrome				1 (7.1 %)

Results

In our study we evaluated 63 patients who underwent antireflux surgery. Of the 63 patients, 40 (63.4 %) were males and 23 (36.6 %) females. The mean age was 10, 5 years (8–13 years).

Group A was composed by 34 (54 %) patients, group AR by 11 patients, group B by 15 (23.6 %) patients, group C by 14 (22.4 %) patients. All patients followed a diagnostic which included the execution of an upper gastrointestinal X-ray and esofagogastrosocopy; 10 (15.8 %) patients of group B also performed 24-h esophageal pH-metry which resulted positive in all patients.

Patients in group A was treated with 18 Nissen fundoplication, 10 Toupet and 6 Thal. In group B, nine patients were treated with Nissen fundoplication, four with Toupet and two with Thal. Patients in group AR and C was treated with a TOGD as described by Bianchi.

After surgery the mean follow-up was 2 years (range 6 months–5 years). In group A, 11 patients had a recurrence of GERD and they were treated with TOGD as a redo operation; these patients composed group AR (Table 3). In group AR, two had complications: a bleeding from anastomosis and a dehiscence (Table 3). In group B, two patients had a recurrence of GERD which was treated with TOGD (Table 3). In group C, all patients were free of GERD during follow-up. In this group, three patients had complications: one stricture of anastomosis, one dumping syndrome and one bleeding from anastomosis. No patients died.

The Fisher's, about the complications of group A and group B, showed a statistically significant difference with a *p* value of 0.0128.

Discussion

Gastroesophageal reflux disease in neurologically impaired children is a challenging surgical problem.

The standard approach to GERD has always been fundoplication, but, during the last years many studies have shown that the results in neurologically impaired children are generally unsatisfactory.

Martinez et al. [5] has reported a postoperative return of symptoms in 71 % of patients and an operative failure rate of 25 % as documented by upper gastrointestinal series or pH probe test. Moreover, the postoperative morbidity can be two times greater and the mortality four times greater in the neurologically impaired [9].

In case of failure of an antireflux plastic surgery, what is performed is usually a redo fundoplication. The literature shows a high failure rate of this procedure (around 25 %), due to repeated vomiting and convulsions, and is reported increased complications due to adhesions and bleeding [10, 11].

In 1997 Bianchi presented his data regarding the intervention of disconnection esophagogastric not only as an alternative approach in place of a redo, in cases in which problems feeding persist due to a failure of the sleeve or symptoms such as retching and vomiting, but also as the first level of intervention in patients with severe neurological deficits [8].

In this study, the author explained how patients undergoing this type of surgery have shown several benefits, such as increased growth in the period of 30 months after the operation, the absence of recurrent reflux, aspiration of contents of the stomach, vomiting. All subjects recovered the ability to feed orally, limiting their underlying disease, and have recovered body weight [12].

This surgical option, therefore, has opened a new path in the management of neurological patients with gastroesophageal reflux disease, providing a procedure that is a valid treatment in severe neurological patients.

This was subsequently confirmed by other studies [3, 13] that have found that, when the ability to swallow is clearly compromised, primary definitive TOGD and gastrotomy offer a superior solution in eliminating reflux and feed aspiration with no possible risk of gastroesophageal reflux recurrence.

In this report, we presented our experience and analyzed the results of treatment of gastroesophageal reflux in patients with neurological deficits, highlighting what was the best treatment according to the severity of the disease they were suffering from.

We set out our analysis on some scales that assessed the impact of neurological disease on motor skills and severity

of gastroesophageal reflux of the patients [14]. We also evaluate the clinical behavior and condition of subjects, basing our choice on the presence of failure to thrive and chronic respiratory disorders as recurrent aspiration pneumonitis.

At the beginning of our experience we considered fundoplication a good choice, as operation of first level, for all neurological patients but during the follow-up we found out that among these subjects there was a significant rate of complications, overall recurrence of gastroesophageal reflux symptoms that required reoperation.

Therefore, according to the guidelines previously mentioned, we observed that some kind of neurological impairment for its severity exposed the patient to a higher failure rate of the fundoplication.

In particular, diseases like Rett syndrome, SMA and cerebral palsy determine a high reduction of the motor abilities of the patient, also as regards the gastrointestinal motility, and lead to serious deficiency of nutrition already a few years of age [15, 16] as it was explained in the NASPGHAN/ESPGHAN GERD guidelines.

According to guidelines and to our observations, from 2005 we started to distinguish the patients in relation to the severity of their neurological impairment and to treat these affected by a higher disability with the TOGD, as first level operation.

In the group of patients operated with fundoplication before 2005 there was a rate of reoperation of 32, 3 % in opposite to the rate of 13, 4 % in the group of the patients treated with a fundoplication after we started to select patients on the base of the severity of neurological impairment.

The difference in terms of complications and reoperation rates were demonstrated not statistically significant by the analysis with the Fisher's test (p value = 0.2), despite of our observations of better results of TOGD in the severe neurological impairment patients.

This result may be explained by the small number of patients in our group, especially with regard to the sample of subjects undergoing TOGD. Although statistical results, we observed in our experience an effective reduction in terms of reduction of GERD recurrence and we believe that our hypothesis could be result statistically significant in a more extensive study.

However, on the basis of this data, TOGD has revealed an acceptable complication rate, that was lower than some operation rates after fundoplication. In conclusion, we believe that TOGD is a useful procedure as the first choice of surgical management in severe neurologically impaired children affected by gastroesophageal reflux.

Also the review of the available literature [17, 18] and our own series showed that TOGD should not only be considered as a procedure of rescue, but as the first

surgical choice in selected patients, because the risk of recurrence is appreciable and secondary surgery is far more difficult.

TOGD, as a primary procedure in children with severe neurological impairment with pharyngeal neuromuscular incoordination and severe gastroesophageal reflux, offers the benefits of eliminating the reflux and avoiding recurrence with consequent antireflux surgeries.

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