

Esophagogastric Dissociation Versus Fundoplication: Which Is Best for Severely Neurologically Impaired Children?

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Purpose: Neurologically impaired children (NIC) often have swallowing difficulties, severe gastroesophageal reflux, recurrent respiratory infections, and malnutrition. Bianchi proposed esophagogastric dissociation (EGD) as an alternative to fundoplication and gastrostomy. The authors compared these 2 approaches.

Methods: Twenty-nine consecutive symptomatic NIC refractory to medical therapy were enrolled in a prospective study and divided into 2 groups: A (n = 12), NIC who underwent fundoplication and gastrostomy; B (n = 14), NIC who underwent EGD. Three were excluded because of previous fundoplication. Anthropometric (percentage of the 50th percentile/age of healthy children) and biochemical parameters, respiratory infections per year, hospitalization (days per year), feeding time (minutes), and "quality of life" (parental psychological questionnaire, range 0 to 60), were analyzed (*t* test and Mann-Whitney test) preoperatively and 1 year postoperatively. Complications were recorded.

Results: Compared with group A, group B presented a statistically significant increase of all anthropometric and nearly all biochemical parameters with a statistical difference in terms of respiratory infections, hospital stay, feeding time, and psychological questionnaire. In group A, 2 bowel obstructions, 1 tight fundoplication, 1 dumping syndrome, and 3 failures of fundoplication occurred. Group B presented 1 anastomotic stricture, 1 paraesophageal hernia, and 1 bowel obstruction.

Conclusions: Compared with fundoplication and gastrostomy, EGD offered better nutritional rehabilitation, reduction in respiratory infections, and improved quality of life. EGD can be rightfully chosen as a primary procedure.
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INDEX WORDS: Gastroesophageal reflux, neurologically impaired children, fundoplication, quality of life.

SEVERE NEUROLOGICALLY impaired children (NIC) often have pharyngeal neuromuscular incoordination and difficulty with swallowing, severe gastroesophageal reflux (GER), recurrent respiratory infections, and, consequently, severe malnutrition. In these patients, a unique guideline regarding the surgical treatment of GER disease does not exist, even if fundoplication, usually combined with gastrostomy¹ (tube/PEG or button), is the most widely adopted procedure.

In the literature, several reports have been published reporting the association of fundoplication with a 7% to 35% recurrence of reflux that required additional surgery^{2,3} in NIC. Because NIC have a limited quality of life and are poor anesthetic risks, a "once and for all, effective therapy" is demanded by the surgeon.

In 1997 Bianchi⁴ proposed esophagogastric dissociation (EGD) as a valid alternative to fundoplication and gastrostomy to totally eliminate the risk of recurrence of

GER. Only 2 reports have been published regarding this technique.^{5,6} Nowadays, nothing has been written specifically comparing the results of EGD versus fundoplication and gastrostomy in NIC. The current study was therefore undertaken to compare these 2 procedures especially examining their effectiveness (definitive resolution of GER, nutritional rehabilitation, and quality of life improvement for the child and the caregiver).

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Table 1. Parental Questionnaire (Scale 1-10) Based on the O'Neill Questionnaire

1. Easiness of caring for your child
2. Quality of time spent with your child
3. Level of frustration in taking care of your child
4. Overall enjoyment of your child
5. Time spent with your other members of your family
6. Overall quality of life

Data from O'Neill et al.⁹

MATERIALS AND METHODS

From 1997 to 1999, among all patients referred to our unit for surgical treatment of GER, 29 homogeneous patients presented the following baseline characteristics and indications for operation: severe neurological impairment as graded by the American Psychiatric Association,⁷ swallowing difficulties, severe GER with persisting esophagitis refractory to medical therapy, recurrent respiratory infections, and failure to thrive. Twenty-nine children were enrolled in 2 groups in an open prospective study: group A (n = 12), consecutive NIC who underwent fundoplication (Nissen) and gastrostomy (mean \pm SD age, 7.8 \pm 2.7 years; range, 1.5 to 17 years); group B (n = 14), consecutive NIC who underwent EGD (mean \pm SD age, 7.1 \pm 2.4 years; range, 1.7 to 16 years). Three children were excluded because of previous fundoplication.

All patients underwent a preoperative workup with a barium swallow, an esophagogastroduodenoscopy, a technetium tc99m gastric scintigraphy or 13C acetate breath test,⁸ a nutritional assessment, and a parental psychological interview.

Anthropometric (weight and triceps skinfold thickness [TSF] expressed as percent of 50th percentile for age of healthy children) and biochemical (hematocrit, lymphocyte counts, total protein, transferrin) parameters, number of respiratory infections per year, days of hospitalization per year and feeding time (in minutes) were recorded pre- and 1 year postoperatively. "Quality of life," based on parent's comments regarding children's care, was determined by a preoperative psychological questionnaire based on the questionnaire by O'Neill et al⁹ regarding NIC candidates for antireflux surgery. The questionnaire included 6 questions. A score variable from 1 to 10 points was assigned for each answer (Table 1). The questionnaire was repeated 1 year postoperatively.

Postoperative complications and deaths were recorded. *T* test for independent samples was used to compare anthropometric and biochemical parameters, feeding time (minutes), and questionnaire after the 2 surgical procedures. Mann Whitney test was used to evaluate the percent of variation in number of respiratory infections per year and days of hospitalization per year after the 2 surgical procedures. *P* value of less than .05 was considered significant. All parents gave their written informed consent.

RESULTS

In group A, before the operation, all patients presented relative weights less than 100% of normal, with a mean relative weight of 66.53% \pm 15.05%; after fundoplication the relative mean weight was 69.2% \pm 16.34%. Before the operation mean relative TSF was 69.93% \pm 11.09%, after 1 year follow-up mean relative TSF was 72.86% \pm 11.38%.

Preoperative mean hematocrit and lymphocyte count were 31% \pm 4.81%, 1,230.06 \pm 244.53/mm³, respectively; after operation these parameters were 31.6% \pm 5.13%, 1,342.2 \pm 232.17/mm³. Total protein and trans-

ferrin increased from 4.81 \pm 1.04 g/dL and 172.86 \pm 16.83 mg% to 5.15 \pm 0.72 and 277 \pm 79.40 after fundoplication and gastrostomy.

Before the operation all selected children presented 4.93 \pm 1.57 respiratory infections per year with a mean hospitalization time of 45.06 \pm 24.98 days per year; after 1 year, mean respiratory infections were 2.26 \pm 1.66 with a slightly decreased hospitalization time (21.33 \pm 12.48 d/yr).

Before fundoplication and gastrostomy, mean feeding time was 39.13 \pm 18.14 minutes; after 1-year follow-up it was 27.26 \pm 18.37 minutes. Mean questionnaire score increased from 27.2 \pm 7 points in the preoperative to 37.66 \pm 18.22 points in the postoperative period.

In group B, mean relative weight varied from 65% \pm 17.53% before EGD to 96.47% \pm 8.48% after operation. Before EGD, mean relative TSF was 64.40% \pm 13.81%; after 1-year follow-up, mean relative TSF was 93.13% \pm 7.93%.

Preoperative mean hematocrit and lymphocyte count were 29.6% \pm 3.4% and 1,504.87 \pm 254.96/mm³, respectively; after operation all parameters increased significantly (35.8% \pm 3.3% and 1,812 \pm 190.49/mm³). Total protein and transferrin increased from 5.25 \pm 0.71 g/dL and 184.07 \pm 19.21 mg% to 6.51 \pm 0.73 and 362.13 \pm 31.03, respectively after EGD.

Before the operation, respiratory infections per 1 year were 5.2 \pm 1.69 with a mean hospitalization time of 69.66 \pm 46.64 days per year; after 1 year mean respiratory infections greatly decreased (0.26 \pm 0.45) with a minimal hospitalization time (3.73 \pm 2.15 days per year). Mean feeding time varied from 48.47 \pm 23.73 minutes before EGD to 10.33 \pm 2.99 minutes after 1-year follow-up. Mean questionnaire score increased from 25.33 \pm 6.75 to 53.75 \pm 3.13 points after the operation, reflecting a considerable improvement in quality of life.

Table 2 summarizes the comparison between group A and B regarding the mean of differences between preop-

Table 2. Comparison Between Nissen and Gastrostomy Versus Esophago-gastric Dissociation Regarding Anthropometric and Biochemical Parameters, Feeding Time, and Parental Psychological Questionnaire

Parameters	Mean Difference Group A	Mean Difference Group B	t Value	P Value
Weight (%)	.027	.315	7.014	.0000
TSF (%)	.029	.287	7.806	.0000
Hematocrit level	.600	6.200	4.701	.0001
Lymphocyte counts/mm ³	112.133	307.133	4.265	.0002
Total protein (g/dL)	.717	1.260	1.080	.2894
Transferrin (mg%)	104.133	178.067	3.190	.0035
Feeding time (min)	-11.867	-38.133	-3.720	.0009
Questionnaire points	10.467	27.467	4.133	.0003

NOTE. *P* value is less than .005.

erative and postoperative values in anthropometric and biochemical parameters, feeding time, and questionnaire score. Group B increases were all statistically significant compared with those of group A, except for total protein.

Comparing the 2 surgical procedures, a statistically significant percentage decrease in number of respiratory infections per year ($U = 28.50$; $P = .0004$) and in days of hospitalization per year ($U = .00$; $P = .0000$) was noticed in group B toward group A.

Two bowel obstructions, 1 dumping syndrome, and 1 tight fundoplication occurred as postoperative complications in Group A. Failure of fundoplication requiring EGD occurred in 3 patients (22%). In group B 1 paraesophageal hernia, 2 bowel obstructions, and 1 anastomotic stricture, treated successfully with repeated dilations and stent positioning, were recorded. In group A one patient died of *ab ingestis* pneumonia 1 year after fundoplication; in group B 1 child died of progressive neurologic deterioration unrelated to EGD operation.

DISCUSSION

The reported incidence of GER in NIC ranges from 15% to 75%.¹⁰ Despite the wide variety of procedures advocated, surgical construction of a Nissen fundoplication with a concomitant feeding gastrostomy has evolved rapidly as the definitive therapy for persistently symptomatic GER disease in NIC refractory to medical treatment.

However, in these patients, this is at a cost of high morbidity and recurrence. Pearl et al¹¹ reported that in NIC the postoperative morbidity rate was 2 times greater and the mortality rate 4 times greater than in neurologically normal patients. In these children, significant complication rates are reported in up to 59% of patients, and a significant risk of failure is seen in up to 25%.^{1,2,12} The most frequent complication is recurrence of symptoms owing to herniation or failure of fundoplication wrap. Other complications that can accompany fundoplication are the gas-bloat syndrome,¹³ the dumping syndrome,^{1,14} and retching.¹⁵

These problems occur, even if particular attention is directed to correct or improve factors that favor breakdown of the fundoplication, including poor nutrition, gastric distention, delayed gastric emptying, mechanical small bowel obstruction, chronic lung disease, and seizures with retching. To definitively prevent reflux, Bianchi,⁴ Danielson and Emmens,⁵ and Dall'Oglio et al⁶ utilized esophagogastric dissociation as an alternative procedure to conventional antireflux surgery in selected NIC and achieved good results in terms of postoperative complications and nutritional rehabilitation.

However, it was clear that further studies were necessary to show the true efficacy of this technique as primary and not only as a "rescue" procedure for recur-

rent GER. The current study addresses the real effectiveness of Bianchi esophagogastric dissociation with respect to Nissen and gastrostomy, performing a prospective comparative analysis in selected NIC. It is important to define our concept of effectiveness based on nutritional rehabilitation, definitive reflux control with reduction of respiratory symptoms, and improvement in quality of life for the child as well as the caregiver.

Particular attention has been placed in patient selection. Twenty-six patients is a relatively small number, so it was important to enroll homogeneous patients to exclude bias. All our patients presented severe neurologic impairment as graded by the American Psychiatric Association,⁷ swallowing difficulties, severe GER with persisting esophagitis refractory to medical therapy, recurrent respiratory infections, and severe malnutrition defined as weight and triceps skinfold thickness below the fifth percentile for age and sex. After the parental informed consent had been obtained, children were enrolled in a nonrandomized, prospective study. Compared with NIC who underwent conventional surgery, patients who underwent esophagogastric dissociation presented a better and faster nutritional rehabilitation, testified by a more considerable increase in all anthropometric and biochemical parameters, except total protein.

Respiratory infections remained practically unvaried after conventional surgery, whereas a considerable reduction was recorded after EGD. Because the days of hospitalization in the preoperative period differ considerably between the 2 groups, a nonparametric test has been used, focusing on percent of decrease. After Nissen and gastrostomy, hospitalization became less frequent, but an extraordinary decrease with a significant improvement in quality of life has been recorded only after EGD.

Because a large part of a caregiver's life is involved in attending to NIC, quality-of-life analysis must be considered a crucial point of our study. Our questionnaire, based on the experience of O'Neill et al,⁹ tries to define parental satisfaction with the 2 procedures, even if this evaluation is subjective. Easiness of childcare, quality of time spent with the child, and expectations regarding surgical procedure are focused on in the questionnaire. After esophagogastric dissociation, the caregivers have shown to be less frustrated in caring for the child and spend less time for the child's "special" needs (especially for aspiration of oral secretions). Regarding postoperative complications, 2 bowel obstructions, 1 dumping syndrome, and 1 tight fundoplication occurred in group A.

Moreover, 3 patients, after Nissen and gastrostomy operation, presented wrap failure within a few months. These patients underwent reoperation and had esophagogastric dissociation with good results.

Esophagogastric dissociation is a more complex procedure than fundoplication with a potential for more

complications. In our group we recorded only 1 complication (1 paraesophageal hernia) strictly related to operation. Anastomotic stricture was related to a preexistent peptic esophageal stricture, and the small bowel obstructions presented in each group are totally unspecific complications.

The reported 1-year follow-up is relatively a short period to evaluate these types of patients; however, we

have formed the view that esophagogastric dissociation can have a definitive role as a primary form of management for severe NIC with pharyngeal neuromuscular incoordination and GER.

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