Original Articles

Fundoplication in Children with Neurological Impairment: A Worthwhile Surgical Treatment?

NSY CHAO, MWY LEUNG, M POON, BPY WONG, KW CHUNG, WK KWOK, KKW LIU

Abstract

Purpose: To review the treatment and outcome of children with neurological impairment (NI) undergoing fundoplication in a paediatric surgery centre. Methods: Records of children with NI undergoing fundoplication from 1996 to 2004 were reviewed regarding impact on feeding, nutrition, respiratory symptoms and operative technique. Results: Forty-six NI children underwent fundoplication with gastrostomy. Thirty-nine records were retrievable. Recurrent vomiting and aspiration-pneumonia were the dominant indications for operation. Nissen's wrap replaced partial wraps since 2002. Laparoscopic Nissen's became the procedure of choice since 2003. Post-operative complication rate was 53%. Mean follow-up was 25 months with 8% gastroesophageal reflux disease recurrence and 13% mortality. Mean hospital-stay relating to pneumonia during the 6 months pre- and post-operation were 82 and 40 days respectively (p<0.001). Mean weight increased from 15th to 17th percentile. Conclusions: Reflux-related respiratory symptoms in the NI children were significantly improved by fundoplication. Co-morbidities contribute to post-operative complications, ongoing airway symptoms, failure-to-thrive and mortalities. Laparoscopic approach offers comparable outcome.

Key words: Fundoplication; Neurologically impaired children

Introduction

Gastroesophageal reflux disease (GERD) is common among neurologically impaired (NI) children. GERD causes significant morbidity in these NI children by way of feeding problems, respiratory tract complications, failure to thrive and prolonged hospitalisation. Over the last two decades, fundoplication has been increasingly recognised as the anti-reflux treatment of choice, replacing medical therapy, both in adults and children. Nissen's complete wrap has been the gold standard operation, although partial wraps and variants have also claimed good outcomes. In recent years, many centres have chosen laparoscopic approach to be the new standard, and laparoscopic fundoplication (Figure 1) has become one of the most commonly performed operations in some paediatric surgical centres. While many large series reported excellent outcomes for children after anti-reflux surgery, others questioned the high complication and GERD recurrence rates in the NI group. In the past ten years an increasing number of NI children had anti-reflux surgery performed in our centre. This study reviews the treatment indications, operative procedures and outcomes of this NI group of children in our locality undergoing anti-reflux surgery.
Materials and Methods

In our centre, consecutive cases of paediatric fundoplications from January 1996 to March 2004 were included. Their hospital records were reviewed, with regard to the cause of neurological impairment, presence of other associated physical conditions and co-morbidities. Feeding patterns, GERD symptoms during the six months pre-versus post-operatively including admission episodes for aspiration pneumonia, the number of days of in-hospital treatment, frequency of intensive care unit (ICU) admissions, bleeding oesophagitis and blood transfusions were recorded. GERD diagnostic work-up included 24-hour oesophageal pH-study, isotope milk-scan and contrast gastrogram, gastric motility and occasionally endoscopy. Drug therapies, nutritional status in terms of body weight percentile and serum markers at pre-operation versus last follow-up were reviewed. Methods of fundoplication, with or without gastrostomy, operating time, duration of recovery, number of days until full enteral feeding, presence of post-operative complications and gastrostomy complications, additional symptoms such as delayed gastric emptying and dumping, and hospital mortality were charted.

Data recorded were analysed via SPSS version 12.0. Student's t-tests were applied with statistical significance assigned at p-values being less than 0.05.

Results

Demography

A total of 46 infants and children with neurological impairment underwent fundoplication during this eight year period. Seven patients defaulted from follow-up or having incomplete records were also excluded. A total of 39 cases completed the review. There were 20 boys and 19 girls. The age at operation ranged from four months to 15 years with a mean age of 6.8 years. Post-operative follow-up ranged from seven months to six years, the mean follow-up being 25 months.

Neurological and Physical Conditions

In 13 patients (33%) the neurological impairment was acquired cerebral palsy from perinatal anoxia; another 13 had associated hydrocephalus or cerebral atrophy from various congenital causes; ten were syndromic. Other associated physical conditions directly contributing to GERD such as hiatus hernia and oesophageal atresia were present in three patients. Major congenital heart disease and upper airway anomalies like laryngomalacia were present in 19 patients (48%). Thirty-five patients (90%) were chair-bound or bed-bound. Thirty-two patients (81%) were completely or partially dependent on tube-feedings. Seven children who did not use a feeding-nasogastric tube were feeding poorly and failing to thrive. Thirty-four patients (87%) had significant vomiting and regurgitations during feeds.

Lower Respiratory Tract Symptoms before Operation

These children were admitted to acute hospital beds for pneumonia symptoms for a mean of 1.6 times during the sixth months before anti-reflux surgery. Related in-hospital stays averaged 83 days. The number of related admissions to intensive care unit averaged 0.45 times.

GERD Investigation Work-up

In 16 children, the clinical diagnoses of GERD were confirmed by at least one of three tests: 24-hour oesophageal pH-study, radionuclide milk-scan and contrast gastrogram. 24-hour pH-study showed positive reflux in six of 23 patients (pH <4 for 5% or more of study-period). Nine of 18 milk-scans and eight of 18 gastrograms were positive, with impaired gastric motility demonstrated in four patients. Upper endoscopy was performed in those with recurrent anaemia, haematemesis or passage of malaena; hiatus hernia with oesophagitis was demonstrated in two patients.
Nutritional Status before Surgery

The mean body-weight of this group of children was at 15th percentile pre-surgery. The mean serum albumin was 38 g/L.

Failure of Medical Treatment

Ranitidine was the most commonly prescribed medical therapy (33%), followed by common prokinetics such as metoclopramide or domperidone (31%). Cisapride, erythromycin and proton-pump-inhibitors were infrequently used. Despite combination or sequential therapies, all subjects in this series had persistent symptoms necessitating anti-reflux surgery.

Operations

There were 39 anti-reflux operations with concomitant gastrostomies. Twenty-nine were open procedures with four partial and 25 complete wraps. Ten were laparoscopic Nissen’s, with one conversion to open procedure due to intra-operative bleeding. Drainage procedures were only performed during open operations and none were in the laparoscopic group; there were 14 pyloromyotomies and seven pyloroplasties. The operation time ranged from 90 minutes to 450 minutes overall, averaging 139 minutes for open approach and 310 minutes for laparoscopic approach.

Post-operative Course and Complications

Post-operative hospital stays varied from 8 to 227 days, with a mean of 69 days. Time to full enteral feeding varied from one to 47 days with a mean of nine days. Overall post-operative complication rate was 53%. Pneumonia occurred in 9 patients and was the most frequent complication. Other medical complications include line sepsis (one), infective enteritis (two), and syndrome of inappropriate ADH (two). There were eight surgical complications that mandated surgical management, including pyloric obstruction, functional or by migrating gastrostomy-balloon, adhesive intestinal obstruction, wound infection, gastrostomy leakage with peritonitis, broken or slipped-out gastrostomy tube. Other minor gastrostomy complications such as superficial leakage and troublesome granulomata were encountered in 33% of cases. Symptoms relating to delayed gastric emptying or dumping were documented in 31% of cases.

Recurrent GERD and Mortalities

Significant vomiting and pneumonic episodes recurred in three patients (all open approaches). Wrap dehiscence was radiologically proven in two. There were five hospital mortalities (12.8%), none within the 30-day post-operative period. Two of these had major congenital heart disease and other syndromic co-morbidities. One death was related to tracheo-esophageal fistula recurrence. In the other two, there were no other significant associated co-morbidities.

Comparing Feeding Problems and Lower Respiratory Tract Symptoms before Versus after Anti-reflux Surgery

Four children (10%) still suffered from frequent vomiting and regurgitations after surgery. The other 90% children had occasional or rare feeding problems. Admission to acute hospital beds for pneumonic symptoms during the sixth months after surgery averaged 0.6 times amongst these children. Related in-hospital stays averaged 40 days. Compared with the pre-surgery admission frequencies and length of in-hospital treatment with paired t-tests, p-values were <0.001 and 0.02 respectively (Figures 2 and 3).
number of related admissions to intensive care unit averaged 0.13 times. For paired t-tests against the pre-surgery intensive-care admissions, p-value was 0.006.

Comparing Nutritional Status before and after Anti-reflux Surgery

The mean body weight percentile at the last follow-up was 17th. Serum albumin averaged 41 g/L. For respective paired t-tests against pre-surgery status, p-values were 0.64 and 0.093 respectively (Figures 4 and 5). In four children body weight percentiles or even actual weight fell after surgery. Nonetheless in eight children (20%) there was a greater than ten-percentile weight increase.

Discussions

Neurological impairment was severe in the majority in this study series. Over 90% of the subjects had significant ambulatory problems; all of them had feeding difficulties and over 80% require tube-feeding at least partially or completely. Cerebral palsy due to perinatal hypoxia was the commonest cause.

The diagnosis of GERD is mainly clinical. Pre-operative investigations confirmed the diagnosis in 16 (41%) of cases. Ambulatory pH-study remains the standard investigation tool. Radionuclide milk-scan and contrast-gastrogram may yield additional information regarding gastric motility in some cases. Endoscopy was only indicated for upper gastrointestinal bleeding which occurred in two children; hiatus hernia was the cause in both and bleeding oesophagitis was their first presenting symptom of GERD. Broncho-alveolar lavage for fat-laden macrophage is a diagnostic modality with high specificity but was not frequently performed in our centre.

Although pH-study is the standard investigation with the best positive predictive value it is not always well tolerated by young un-cooperative subjects. Other tests have higher false negative rates. Fifteen patients had completely negative tests. But as significant GERD symptoms prevailed, empirical anti-reflux surgery was still indicated in these subjects. At other times anti-reflux surgery may be prophylactic, when gastrostomy for feeding purpose was the primary indication for surgery. The risk of secondary GERD due to hiatus tenting is notorious in these NI children after gastrostomy. Two of our subjects who were indicated for feeding gastrostomy had prophylactic fundoplication.

Majority of the operations were performed after 1999 following an increased number of referrals and surgical consultations. All children in this series received concomitant feeding gastrostomy with anti-reflux surgery. There were 29 open fundoplications, of which four were partial wraps. Open complete anterior wrap replaced partial wraps after 2001. Laparoscopic Nissen’s became the procedure of choice since 2003 unless contraindicated. Operating time for laparoscopic approach has been significantly longer than open approaches so far during our early experience with the first 10 cases.

Drainage procedures were performed during majority of the open fundoplications, mostly in the light of pre-operative isotope or radiographic evidence of decreased gastric motility. One child had pyloromyotomy and subsequent pyloroplasty at a repeat operation because of persistent gastric stasis. However seven children experienced dumping after open procedure with pyloric drainage. The overall benefit of drainage procedures seemed outweighed, and indeed there have been no consistent evidence that drainage procedures were essential or
beneficial in preventing gastric outlet problems with anti-reflux surgery.\textsuperscript{4-6} Drainage procedures were not routinely performed since 2003.

One of the main goals of anti-reflux surgery and gastrostomy is to alleviate feeding problems, associated pulmonary aspirations and minimise hospitalisation. Surgery had improved the feeding patterns in 90\% of the children with major vomiting. Only four of the vomiting children continued to have feeding problems. Indeed many carers at follow-up had reported improved satisfaction in managing these children since surgery. Comparing the frequency and duration of hospitalisation in treating severe pneumonic episodes, the improvement post anti-reflux surgery is statistically significant. Many of these children however continued to suffer minor pneumonic episodes post-surgery and chronic respiratory symptoms. Chronic aspiration of oral pharyngeal secretions is a likely cause as these NI children have life-long swallowing problems.

Improving the nutritional status and alleviating failure-to-thrive were the other main goal of surgery. Reviewing weight gain and serum parameters post-operation there was a trend of improvement but it was not statistically significant. Serum albumin and lymphocyte counts were the chosen parameters prospectively, but these fluctuate with acute morbidities and may not have reflected long term nutritional status accurately. Growth-rate had been used by other investigators who demonstrated statistically significant nutritional improvement with anti-reflux surgery.\textsuperscript{7} The lack of weight-percentile gain in this series may be attributed to several causes. Firstly failure to thrive is often resistant in the syndromic group; other chronic co-morbidities may have also attributed. Secondly, from an unfortunate and sinister perspective, progressive weight gain and advancing feeding-needs for the severely NI child may not be the universal goal among carers, especially when parents or infirmaries are under-resourced. The burden of care unfortunately 'grows' with the growing mentally handicapped.

Without anti-reflux treatment, many of these NI children probably might not have thrived at all and their weight percentile would have fallen progressively. The fact that most children in this series had sustained thriving along their respective weight percentile suggested surgery had already improved nutritional potential. Those 20\% children whose weight caught up by more than 10 percentiles were clearly benefited.

High post-operative complication rates have been reported by many authors.\textsuperscript{8,9} In this series it was over 50\%. More than half of these were related to airway problems, pneumonia and non-surgical sepsis. Technical operative complications were not common. Frequent post-operative pneumonia was likely related to prolonged atelectasis associated with large upper laparotomy wounds in open surgery. Unfortunately the early laparoscopic cases with prolonged operating-time suffered the same problem. Anecdotally in the later series of the learning curve with shorter operating-time, the advantages of minimally access surgery conferred with less post-operative intubations and respiratory complications. In many large series of paediatric laparoscopic funduplications elsewhere, other authors had claimed significantly reduced post-operative morbidities, analgesia requirements and shortened recovery in these children.\textsuperscript{10-13}

Significant reflux symptoms recurred in three patients, two were proven radiologically with anatomical failure of fundal wrap, one migration and one dehiscence. In other series for anti-reflux surgery in the NI children with longer follow-up, recurrence rates of up to 25\% have been reported.\textsuperscript{7,9} Oesophageal dysmotility, raised intra-abdominal pressures and gastric outlet obstruction are among many theories proposed to explain the causes of wrap failures. Two of the three patients in this series had notable problems with gastric emptying post-operatively. All were treated conservatively with H2-blockers or prokinetics. There may prove to be greater number of GERD recurrence in this series during longer term follow-up. Some of these may require follow-up 'redo' operation, an operation much more amenable if the primary operation was done via minimal access.

Other series have reported peri-operative mortalities of 9\% or less.\textsuperscript{7,9} There was no operative mortality in our series, but a much higher rate hospital-mortality. It may be postulated that syndromic children and those with multiple co-morbidities may be associated with higher mortality. In the Columbus series, only 51\% of syndromic children with anti-reflux surgery survived, but there was no statistical relationship between aetiological factors for NI and survival or mortality.\textsuperscript{4} Likewise, of the five deaths in our series, two did not have syndromic or other congenital co-morbidities.

**Conclusions**

Children with profound neurological impairment may be severely handicapped by their gastrointestinal and respiratory complications, with otherwise poor chance of thriving. Fundoplication with feeding-gastrostomy alleviates their feeding problems, significantly reduces their respiratory morbidities and enable thriving with sustained
nutritional status in most cases. Surgery can improve their survival and some of their debilitating symptoms, and facilitate the care of these children. Although technically a straight-forward operation, fundoplication in this group of children has been associated with high post-operative complication rates and high mortality. It is hoped that with increased experience with the laparoscopic approach, post-operative morbidities can be minimised and overall surgical outcome can be improved. Continued longer-term follow-up and review will be necessary to validate this.

References