Simple fundoplication versus additional vagotomy and pyloroplasty in neurologically impaired children – a single centre experience

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A B S T R A C T

Background and aims: Gastrooesophageal reflux disease (GERD) is a significant problem in children with neurological impairment (NI) with high failure rates for fundoplication. Fundoplication with vagotomy and pyloroplasty (FVP) can improve the outcome by altering the sensory or motor dysfunction associated with the reflux. We report our comparative outcomes for simple fundoplication (SF) and FVP in NI children.

Methods: Case records of all patients having fundoplication under a single consultant at a tertiary UK paediatric surgical centre between January 1997 and December 2012 were retrospectively assessed for recurrent symptoms and redo surgery. The data were collected using a Microsoft Excel database and analysed on Graphpad prism software program. Data are median (range). P value < 0.05 was considered significant.

Results: Data were available for 244 out of 275 patients who underwent fundoplication during this period (157 SF and 87 FVP). Neurological disease or known syndromes were recorded in 158 patients. Thirty-five children had congenital anatomical abnormalities. Laparoscopic fundoplication was done in 37 cases. Revisional surgery for recurrent symptoms was performed in 22 patients. In the neurologically normal children, all of whom had SF, the revision rate was 6.5%. In the NI children the revision rates were 18.5% for SF and 3.9% for FVP, respectively (Fisher’s exact, P < 0.05). The median time to redo surgery was 10 (1–63) months, and the median time to follow up was 19.5 (2–177) months.

Conclusions: There appears to be a significantly lower need for redo surgery following FVP than SF in children with NI.

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Gastrooesophageal reflux disease is symptomatic reflux of gastric contents into the oesophagus [1–3]. Indications for surgical management include inadequate response to medical therapy, development of complications secondary to acid reflux or occurrence of repeated significant respiratory complications [4]. A variety of operative techniques exist for the surgical management of GORD [5–11].

The high incidence of GORD in neurologically impaired children is well recognised, and this cohort makes up the largest group undergoing anti-reflux surgery [12,13]. Factors predisposing to GORD in the NI population include spasticity resulting in increased intra-abdominal pressure, anatomical variation of the gastro-oesophageal junction secondary to kypho-scoliosis, delayed gastric emptying and constipation secondary to gut motility disorders and long term supine positioning [1,14]. Although few studies have directly compared outcomes of these groups [15–17], higher failure rates have been reported in children with NI following anti-reflux surgery, with higher morbidity, mortality and requirement for redo operation [1,17,18]. Gastrointestinal dysfunction in the NI children manifests primarily as foregut dysmotility because the foregut has a great density of extrinsic innervation [19]. Vagotomy and pyloroplasty could therefore help alleviate these symptoms while a fundoplication would form an anatomical solution to the reflux itself. We report comparative outcomes of simple fundoplication (SF) and fundoplication with vagotomy and pyloroplasty (FVP) in children with NI.

1. Methods

1.1. Patient identification

All children undergoing fundoplication under the care of a single consultant paediatric surgeon at our regional tertiary referral paediatric unit over a fifteen year period between January 1997 and December 2012 were identified using the OPCS coding. Following Institutional Audit Committee review (#3560), a retrospective case-note review was performed to determine pre-operative comorbidities, peri-operative complications, and long term outcomes. Specifically the effect of surgery on pre-operative symptoms and the requirement for revisional surgery were determined. Patients undergoing open or laparoscopic simple fundoplication (SF), and fundoplication with vagotomy and pyloroplasty (FVP) were included, irrespective of the indication for anti-reflux surgery. The decision to proceed to surgical management was that of the senior author in all cases. For every patient the clinical decision was based upon symptom severity, oesophageal pathology or...
a history of respiratory compromise secondary to severe reflux disease. The only radiological investigation invariably used was an upper gastrointestinal contrast study, which was performed to exclude malrotation or other anatomical disorders.

1.2. Surgical technique

Fundoplication was done using a standard technique which included a crural repair when the hiatus was noted to be lax and a Nissen wrap secured with a double layer of non-absorbable sutures. In FVP the vagal trunks and any additional fibres were divided along the abdominal oesophagus and a Heineke–Mikulicz pyloroplasty was done with absorbable sutures and (occasionally) an overlaid omental patch to protect against any leaks.

1.3. Statistical analysis

Information collected retrospectively was stored in a Microsoft Excel (2007) database. Statistical analysis was done using Graphpad Prism contingency tables (v6 for Windows). Contingency tables for Fisher’s exact test were used to compare categorical data and Mann Whitney test was used to compare continuous data. Data are quoted as median (range). A P value of <0.05 was considered significant.

2. Results

Two hundred and seventy five children had been coded as having undergone fundoplication during the period of this study (Fig. 1). Thirty one patients were excluded from the final analysis for the following reasons. Ten procedures were wrongly coded and four patients had fundoplication with other procedures (Heller’s myotomy for achalasia cardia or colonic transposition for oesophageal atresia). Another 17 were excluded due to missing post-operative clinic letters, although none of these 17 patients underwent revisionary surgery. Of these, there were seven patients who did not have any clinical documentation to confirm their neurological status. An additional eight patients had neurological impairment and one had a syndrome and two patients were documented to have anatomical reasons for surgery according to the nursing records.

Primary surgical procedures performed were simple fundoplication alone (n = 157) and fundoplication, vagotomy and pyloroplasty (n = 87).

Of the 244 children, 132 children (54%) had known neurological impairment (NI), 26 (11%) had specific syndromes (SS) and 86 (35%) were neurologically normal (NN). All neurologically normal children underwent SF, 23 and 63 patients underwent the laparoscopic and open Nissen fundoplications respectively. In the NN group, 35 children had congenital anatomical abnormalities predisposing to GORD, including oesophageal atresia (OA = 22), congenital diaphragmatic hernia (CDH = 3) and hiatus hernias (HH = 10).

The NI group included 132 children with cerebral palsy and developmental delay with spasticity or seizures. The diagnoses of the children in SS group are listed in Table 1. In the NI group, 54 children underwent SF and 78 had a primary FVP. In the SS group 18 children underwent SF and 9 children had an FVP. Mean age at the time of the initial antireflux procedure was 3.4 years in the NN group and 4.7 years in the NI group (p = 0.01, Mann Whitney test).

Revisional surgery was required in a total of 23 patients in whom recurrent symptoms were evident at follow-up (Fig. 1). Of these patients, 13 had re-do SF, 1 patient had a re-do fundoplication with additional vagotomy and pyloroplasty, 4 patients underwent formation of surgical feeding jejunostomy and 5 patients underwent oesophagogastric disconnection with Roux-en-Y jejunostomy as described by Bianchi [8].

Six NN children (6.98%) had a redo fundoplication following initial SF. In the NI group, revisional surgery was less commonly required following FVP than with SF [10 (18.5%) SF vs 3 (3.8%) FVP, P = 0.01]. In the SS group, redo surgery was required in 2 (11%) children in SF and 1 (11%) child in FVP.

[Fig. 1. Flowchart of included patients and their outcomes. Abbreviations: SF = simple fundoplication; FVP = Fundoplication, vagotomy and pyloroplasty; OGD = oesophago-gastric disconnection; jej = feeding jejunostomy.]
Following initial fundoplication, 19 NN patients required oesophageal balloon dilatations; the primary pathology in 12 of these was OA and in a further 2, severe reflux stricture. Two patients underwent gastrostomy placement for venting for symptoms of gas bloat and another 2 NI patients had symptoms of dumping following FVP.

The median duration of follow-up was 41 (2–117) months at the time of data collection. At the most recent follow up, only 83 patients were on full oral feeds, the rest needed varying amounts of gastrostomy feeds, and 2 children were on parenteral nutrition due to severe intestinal dysmotility and malabsorption. The symptoms reported by patients at the most recent follow up are summarised in Table 2.

Thirty-six patients (24 NI and 10 NN and 2 SS) died during follow-up; median time from primary surgery was 15 months (3 days to 13 years). Six deaths in the NN group had severe congenital heart disease. Medium-term mortality in NI children was 18.8%. Of the NI group deaths, 12 patients had undergone SF and 12 had undergone FVP. There was no statistically significant difference in death rates between NI children who underwent SF and FVP (22.2% vs 15.4%, P = 0.36).

### 3. Discussion

The moral, ethical and timing questions surrounding the decision to perform antireflux surgery in a NI child are not new. A recent retrospective cohort study from the US of nearly 12,000 antireflux procedures criticised the decision to perform fundoplications on neurologically normal children below the age of 6 months — a time during which gastrointestinal reflux, without progression to GORD, is physiologically normal [20]. The higher median age of the patients in our series lends support to this argument and the need to have explored medical therapies before a decision for antireflux surgery is made. The main indications for surgery in our series were based on symptomatic assessment and a failure to respond to medical therapy. The only infants in our series who underwent surgery did so due to life threatening airway compromise secondary to severe GORD. The only routine pre-operative investigation performed was an upper GI contrast study in order to exclude malrotation or other anatomical abnormalities. A recent survey reported that most paediatric surgeons do not rely on pre-operative investigations other than a thorough history and physical examination to decide about the need for antireflux surgery in a child [21].

The principal aim of this work was to assess for potential differences in symptom recurrence and need for revisional surgery in NI children who undergo SF or FVP. Although the potential merits and shortcomings of vagotomy and pyloroplasty, both individually and combined in the same procedure, have long been described in published literature [22,23], to the authors’ knowledge no previously published work has specifically studied the use of this technique in the paediatric population. Ackerman et al., found no significant decrease in recurrent reflux symptoms following FVP compared to SF in neurologically normal adults [22].

Our overall results indicate a success rate of 91% for fundoplication in our paediatric population which compares favourably with previously published series. A recently published systematic review of antireflux procedures in children with GORD by Mauritz et al. [16] reported a median success rate of 86% (57%–100%).

Failure of fundoplication has long been recognised and investigated with attempts at defining the cohort of patients most likely to develop failure. Those routinely identified are patients with diffuse motility disorders, hypertonic cerebral palsy, severe learning difficulties, oesophageal atresia and tracheomalacia [5,9]. A recent systematic review of prospective studies of anti-reflux procedures showed a median success rate of 70% in NI patients with a range of 57%–79% and a statistically significant recurrence rate in NI patients of 18% compared to 2% in NN patients (p < 0.01) [16]. In contrast to Engelman et al. in 2010 who reported a better outcome in the NI patients [10], our results support the previous findings of increased failure in those with a neurological diagnosis who undergo a simple fundoplication.

In our series, success was defined as “improvement of symptoms following surgery”. This was a subjective assessment made without the use of a validated “quality of life” questionnaire, and as such could be seen as a significant limitation of our work. The assessment was however made by a single surgeon in all cases and this consistency lends weight to our results. The main indication for revisional surgery was a sustained failure of symptomatic and functional improvement during follow-up; further diagnostic investigations to assess the function of the wrap were performed when deemed appropriate. The decision to undertake a revisional procedure was made by the same Consultant Clinician in all cases which would tend to reduce any inter-operator bias. The findings that we present show a statistically significant difference in favour of fundoplication with vagotomy and pyloroplasty in patients with non-syndromic neuro-disability.

The success rate for FVP of 95.4% observed in our study is much higher than achieved by other antireflux procedures in previous reports reviewing NI children [6,7,15,16,24–27]. The extrinsic innervation to the foregut arises either from the spinal cord via the prevertebral ganglia or directly from the medulla via the vagus nerve. In NN patients, vagal nerve dysfunction can cause a relaxation of the proximal stomach and retroperistalsis secondary to inhibition of the gastric pacemaker [19]. The persistent symptoms of retching, pain and discomfort following simple fundoplication in NI children may be partly explained by this antinuclal ventral dysmotility, with delayed gastric emptying and constipation contributing to the wider symptomatology [18]. The global autonomic dysfunction seen in CP may also result in the inappropriate secretion of gastric acid which could further exacerbate the symptoms of reflux in these patients. Previous studies have argued for and against the addition of a pyloroplasty, however consistent improvements in gastric emptying have not been reliably demonstrated [28–31]. Our results suggest, however, that the addition of vagotomy and pyloroplasty to SF may be a significant step in reducing the need for revisional surgery in these patients.

### Table 1

<table>
<thead>
<tr>
<th>Underlying Condition</th>
<th>Number of Patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cerebral Palsy +/- developmental delay</td>
<td>132</td>
</tr>
<tr>
<td>Syndromes</td>
<td></td>
</tr>
<tr>
<td>Trisomy 21</td>
<td>12</td>
</tr>
<tr>
<td>Cornelia De Lange syndrome</td>
<td>5</td>
</tr>
<tr>
<td>Aicardi Goutieres syndrome</td>
<td>2</td>
</tr>
<tr>
<td>Cri du chat syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Williams syndrome</td>
<td>1</td>
</tr>
<tr>
<td>X-linked myotubular myopathy</td>
<td>1</td>
</tr>
<tr>
<td>Smith–Magenis syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Moebius syndrome</td>
<td>1</td>
</tr>
<tr>
<td>DiGeorge syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Wolf–Hirschhorn syndrome</td>
<td>1</td>
</tr>
</tbody>
</table>

### Table 2

<table>
<thead>
<tr>
<th>Symptoms</th>
<th>NN with SF (n = 54)</th>
<th>NI with SF (n = 78)</th>
<th>SS with SF (n = 9)</th>
<th>SS with FVP (n = 8)</th>
</tr>
</thead>
<tbody>
<tr>
<td>dead</td>
<td>10</td>
<td>12</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>none</td>
<td>64</td>
<td>33</td>
<td>41</td>
<td>7</td>
</tr>
<tr>
<td>vomiting</td>
<td>1</td>
<td>1</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>retching</td>
<td>2</td>
<td>1</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>occasional retch/vomit</td>
<td>5</td>
<td>3</td>
<td>9</td>
<td>0</td>
</tr>
<tr>
<td>dumping symptoms</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>with oral feeds</td>
<td>3</td>
<td>3</td>
<td>5</td>
<td>0</td>
</tr>
<tr>
<td>need meds</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>dysphagia</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Tpn</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

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The failure rates of 11.1% in FVP and SF patients with specific syndromic disabilities support the previous suggestions that GER in these patients is part of a generalised pan-intestinal dysmotility [14,18]. Many patients in this group have features of hypotonia rather than the hypertonia which is more typical in patients with CP or other UMN pathologies.

Previous studies into the cause for fundoplication failure have reported the predominant cause as a herniation of the wrap into the posterior mediastinum which could be prevented with better apposition of the crura at the index procedure [32]. This finding was most commonly observed in patients with hypertonic cerebral palsy, and included patients that had fundoplication and pyloroplasty [32]. The absence of this finding in our secondary operations lends weight to the suggestion that without vagotomy these patients still experience discomfort and retching due to antroduodenal dysmotility and inappropriate acid secretion which then leads to wrap failure as opposed to a true technical failure being the causative mechanism for the persisting symptoms.

Dumpling syndrome is another known complication of fundoplication characterised by early postprandial hypoglycaemia as a result of an exaggerated insulin response, triggered by early hyperglycaemia. The frequency of dumping syndrome following fundoplication has been variously reported between 2% and 30% [33,34]. We observed this in 2 patients following FVP. However neither of these patients had hypoglycaemia on further investigation.

Overall medium-term mortality in NI children regardless of the surgical procedure was in keeping with previously reported studies in this group [35,36].

There appears to be a significantly lower need for redo surgery following FVP than SF in children with NI. We suggest that it should be considered as a treatment option in these patients.

Appendix A. BAPS Discussion

Presented by: William Calvert

Discussant: Aruna Abyankhar (Cardiff) This is excellent data, and a single surgeon series which is even more important. Looking at the recurrences, could you tell us if they occur more in the early part of the career and if the FVPs were done more in the later part of his career.

Response: William Calvert There was a number who had a simple fundoplication within the neurologically impaired group and this was more apparent early on. These had a higher failure rate and then as the trend towards appreciable improvement in symptoms appeared in the FVP group with a decrease in the failure rate then that became the method of correction used towards the later part of this 15 year study.

Discussant: Aruna Abyankhar (Cardiff) So could there be some effect of the surgeons own skills and modifications on the recurrence rates?

Response: William Calvert Matthew Jones, the senior surgeon in this series had been doing the FVP prior to 1997.

Discussant: Richard Lindley (Sheffield) First of all I think the recognition and treatment of dumping syndrome after fundoplication is often not picked up on; so what was the incidence of dumping in the two groups? And related to that, is how were the FVP children being fed, because if they are being fed continuously as opposed to bolus fed that’s quite a significant lifestyle change for the parents.

Response: William Calvert There were two cases of dumping syndrome postoperatively, although I can’t recall the feeding methods post-operatively.

Discussant: Nitin Patwardan (Leicester) What was the incidence of post-operative retching in your children that required a revision? Because if you are doing a pyloroplasty which is increasing the gastric drainage that begs the question about the incidence of retching and the reason for the revision?

Response: William Calvert The decision to do the FVP was based on the improvements therein were seen in neurologically impaired children that have undergone the oesophago-gastric disconnection operation for reflux. Our question was what part of the oesophago-gastric disconnection was most likely to help reduce these troublesome symptoms and the conclusion we came to was that it was the vagotomy. So could we do a vagotomy with a more physiologically normal procedure which led us to use the FVP. The pyloroplasty was done in this context because the vagotomy was performed. The decision to revise the surgery went through the same multidisciplinary process as they had gone through for the index procedure. The decision to re-operate on those with symptoms of retching was when the medical management of had failed and it was a consensus decision from there on.

Discusant: David Crabbe (Leeds) Dealing with the children who had recurrent reflux, neurologically impaired children who had recurrent reflux following a simple fundoplication, I may have missed it but were you revising those children by redoing the fundoplication or were you revising them by doing a FVP? If it was a combination of both can you discuss the outcomes for the two different revisional operations?

Response: William Calvert For the neurologically impaired children there were six who had initial simple fundoplications who were revised with a further simple fundoplication. There was only one child who initially had a simple fundoplication who had it revised to a FVP and that was one of the syndromic children. If the neurologically impaired child had an initial FVP they went on to have either a feeding jejunostomy or an oesophago-gastric disconnection as their second line management.

References


