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Short and long-term outcomes after pediatric redo fundoplication

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ABSTRACT

Background: Redo fundoplication (RF) is the most common surgical treatment for recurrent gastroesophageal reflux disease (GERD) in children, but outcomes after RF are rarely reported. The aim of this study was to assess short- and long-term outcomes after RF in childhood.

Methods: The study is a follow-up study of patients undergoing RF from 2002 to 2020 at a teriary care center. Patients/parents were sent questionnaires recording symptoms of recurrent GERD, troublesome side-effects and satisfaction. Retrospective chart review was also performed.

Results: 24/28 (86%) patients were included median 9 (1.6 months–17.7 years) years after RF. 16 (67%) had neurologic impairment. Indications for RF was recurrence of GERD ($n = 18$), discomfort or dysphagia from a herniated wrap ($n = 5$) and dysphagia from a slipped fundoplication ($n = 1$). Median operating time was 128 (95–250) min. Six (25%) patients experienced early major complications, of which two were gastrostomy related.

Five (21%) patients experienced recurrence after RF. Three of these were symptom free at follow-up with medical treatment or re-RF. The most common symptom at follow-up was stomach pain (37%) and excessive flatulence (38%). 18/22 (95%) patients/parents would choose RF again, and 21/22 would recommend RF to someone in a similar situation.

Conclusions: RF is successful in treating recurrent GERD after primary fundoplication, and patient/parental satisfaction is high.

1. Introduction

Fundoplication is one of the most commonly performed major gastrointestinal surgical procedures in pediatric patients. Indications for fundoplication are symptoms or complications of gastroesophageal reflux (GER) disease (GERD) not sufficiently relieved with conservative treatment [1]. Most patients report initial disappearance of GER symptoms after fundoplication, but a substantial number of patients experiences recurrent GERD after some time. A systematic review from 2011 including studies with at least six months follow-up after primary fundoplication, reported recurrence of symptoms in 4–30% [2]. When recurrence occurs, there is no agreed-on algorithm for treatment. Treatment options include conservative management, redo fundoplication (RF), jejunal feeding either through a transgastric jejunal (TGJ) tube or a jejunostomy, parenteral nutrition, or esophagogastric disconnection and Roux-en-Y esophagojejunostomy [1,3–5].

In most centers, RF is the most common surgical option to treat recurrent GERD. Despite being the primary choice of surgical treatment, the literature is sparse on outcome after RF in children. Most studies are small retrospective case series and often only re-RF rates are reported [3,6–11]. The re-RF rate varies from 6 to 26% [7–12]. However, the re-RF rate does not reflect the true recurrence rate since some patients are treated conservatively, and some undergo other interventions than re-RF. In line with this, when recurrence after RF was defined as vomiting and objectively proven GER, two studies including 81 and 35 children, reported higher recurrences rate after RF; 42% and 25%, respectively [3,6]. Only one previous study has assessed outcome after RF beyond retrospective chart reviews. In this study including 31 children, family members reported that 61% of the children were back on anti-secretory drugs, and 45% vomited regularly median 3.6 years after the RF [13]. Even more concerning was that only 17% of the parents were satisfied with the outcome of the RF [13].

Due to the paucity of studies on outcome after RF in children, and especially the lack of studies where patients' and parents' evaluation of outcome has been addressed, we undertook this study

Abbreviations: GER, gastroesophageal reflux; GERD, gastroesophageal reflux disease; NI, neurologic impairment; RF, redo fundoplication; TGJ, transgastric jejunal; UGI, upper gastrointestinal.

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where the main aim was to report frequency of recurrent GERD and troublesome symptoms after RF. Furthermore, we wanted to explore the short-term complication rate and patient and parental satisfaction with the RF.

2. Methods

2.1. Study design and patients

The current study is a cross sectional follow-up study of outcome in pediatric patients under 18 years undergoing RF between January 2002 and July 2020 at Oslo University Hospital Rikshospitalet. This is a tertiary pediatric surgery referral center, with an average of twelve primary fundoplications per year, and neurologically impaired (NI) children constitute around half of the patients [14]. The patients were identified from a surgical logbook. Patients who had undergone primary fundoplication at other hospitals were also included. Exclusion criteria were patients who were no longer alive or had emigrated. Data were collected by questionnaires and chart reviews. All fundoplications, both primary and redo, were performed with the Nissen technique by at least one attending surgeon.

Patients and/or parents got the questionnaires by mail in the period from November 2019 to August 2020. The questionnaires were filled out by parents of NI children and non-NI children below 12 years, by non-NI patients over 16 years, and by both non-NI patients between 12 and 16 years and their parents. Patients or parents were contacted by one of the authors if they had questions or symptoms that might be related to the fundoplication, and they were offered appropriate investigations if indicated.

Patients that already were included in a randomized controlled trial comparing laparoscopic and open fundoplication at our center, were not separately approached for this study since questions about postoperative outcome covered the same topics in the randomized trial as in this study [14-16]. In the randomized trial, patients were interviewed one, two, four and twelve years postoperatively, recording signs of recurrent GERD, troublesome side effects and parental/patient satisfaction.

2.2. Questionnaire

The questionnaire sent to patients and parents included questions about heartburn, vomiting, regurgitation, abdominal pain, dysphagia, discomfort during meals, retching, belching, and excessive flatulence. The questions were derived from the Pediatric Quality of Life Inventory Gastrointestinal Symptoms Scale [17]. Answers were given on a 5-point Likert scale (0: never, 1: Almost never, 2: Sometimes, 3: Often, 4: Almost always). When analyzing the results, the answers were transformed into dichotomous variables; 3 and 4 were reported as “yes” and 0, 1 and 2 as “no”. The questionnaire also addressed use of anti-secretory drugs (yes/no), if the patients had undergone any investigations for recurrent GERD or experienced any side effect of the fundoplication (yes/no and if yes, what had been done). The patient or parents were also asked if they had any questions related to the surgery or if they wanted investigations for symptoms that may be related to the fundoplication. Lastly, the questionnaire recorded satisfaction with the redo NF: “With the benefit of being able to look back, would you choose a RF again?” and “Would you recommend a RF to others in a similar situation?” (yes/probably yes/do not think so/no/unsure).

2.3. Retrospective chart review

Patient demographics, perioperative data regarding both the primary fundoplication and the RF, time from primary fundoplication to RF, and results from all investigations for GERD after the primary fundoplication and RF were recorded retrospectively. Recurrence of GERD was defined as symptoms of GERD and investigations confirming GER and/or herniated wrap. Indication for RF was registered as free text and categorized based on main complaint. NI was defined as a static or progressive, central or peripheral neurologic condition associated with chronic functional or intellectual impairment [18]. Complications during the first 30 postoperative days after the RF were graded according to the Clavien Dindo classification [19]. Grade 3 complications or higher were considered major complications.

2.4. Ethics

The study was approved by the hospital’s commission for personal security (19/22925). Consent was obtained from parents of patients under 12 years and NI patients, from both parents and patients between 12 and 16 years, and from patients only if older than 16 years.

2.5. Statistics

Data were presented with descriptive statistics; categorical data as frequencies and percentages and numerical data as median and range using IBM SPSS Statistics for Windows Version 25.0 (IBM, Amonk, NY).

3. Results

3.1. Patients

31 patients underwent a RF during the study period. Three were excluded; one had emigrated, and two were no longer alive. Six patients were already included in the above-mentioned randomized trial. Consequently, questionnaires were sent to 22 patients, and replies received from 18 (2 by patient only, 4 by both patient and parents and 12 by parents only). Thus, 24 out of 28 eligible patients (86%) were included in the current study, and the median follow up time was 9 (1.6 months-17.7 years) years.

The majority (67%) of patients had NI, and only 21% had no comorbidity (Table 1). The primary fundoplications were performed according to the Nissen technique, and hiatal closure was performed in all. A gastrostomy was established before or concomitantly with the RF in 17 (74%) patients. The median time from primary fundoplication to RF was 2.8 years (2 days-13 years). The RFs were performed by seven consultants. There were almost always two consultants performing the procedure, and senior author K.B was involved in 18/24 procedures.

3.2. Investigations before RF

All 24 patients had at least one investigation showing recurrent GER before the RF. Eight had a reflux index >4 on 24-h pH monitoring, eight patients had GER on upper gastrointestinal (UGI) contrast study, and radiographic or endoscopic wrap herniation was demonstrated in 18 patients.

3.3. Indications for RF and perioperative data

The most common reason for RF was recurrence of GER symptoms such as heartburn, regurgitation and/or vomiting not satisfactorily relieved by conservative treatment ($n = 18$, 75%). In five patients, the indication for RF was discomfort or dysphagia assumed to be caused by a herniated wrap. One of these patients presented with severe pain the first postoperative day and was reoperated the second day. Lastly, in one patient, the RF was performed because of dysphagia, and the UGI contrast study suggested that the

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Table 1
Demographics of 24 patients undergoing redo fundoplication at Oslo University Hospital in the period 2002–2020.

	$N = 24$	
Age at primary fundoplication, median (range)	3 years (2.6 months–14.3 years)	
Age at redo fundoplication, median (range)	9 years (1.0–16.0 years)	
Male, n (%)	14 (58%)	
Comorbidity		
	Neurologic impairment, n (%)	16 (67%)
	Repaired esophageal atresia, n (%)	3 (13%)
	No comorbidity, n (%)	5 (21%)
Primary fundoplication, open/laparoscopic	10/14	
Follow up time after redo fundoplication, median (range)	9 years (1.6 months–17.7 years)	

Table 2
Perioperative findings from redo fundoplication in children.

	$N = 24$
Intact and herniated wrap	9 (38%)
Disrupted and herniated wrap	9 (38%)
Disrupted wrap	5 (21%)
Fundoplication wrapped around upper part of stomach	1 (4%)

fundoplication was wrapped around the upper part of the stomach (slipped Nissen).

RF was performed laparoscopically in five patients, of whom all had the primary fundoplication done laparoscopically. One of these procedures was converted because of extensive adhesions. The operating time, available in 18 (85%) patients, was median 128 (95–250) min. Wrap herniation was confirmed in all 18 patients where it was demonstrated at preoperative investigations, and the wrap was intact in half of these (Table 2). In one patient the wrap had slipped and was situated around the upper part of the stomach as shown on the preoperative barium swallow. A hiatal closure was performed in 22 (92%) patients. A Gore-Tex mesh was used to reinforce the hiatal closure in two patients because of weak crural muscles.

Concomitant procedures were performed in seven (29%) patients; pyloroplasty ($n = 2$), pyloroplasty and gastrostomy ($n = 1$), umbilical hernia repair ($n = 1$), removal of a pyloric hamartoma ($n = 1$), endoscopic dilation of esophageal stricture ($n = 1$), and Roux-en-Y-jejunostomy ($n = 1$). The Roux-en-Y jejunostomy was performed to provide jejunal feeding in a patient with severe intestinal dysmotility. The RF in this patient was performed because a herniated wrap was believed to cause discomfort.

3.4. Early postoperative complications after RF

Twenty early postoperative complications occurred in 12 (50%) patients; four grade I complications (pneumothorax ($n = 1$), wound infection ($n = 1$), dislocated gastrostomy tube ($n = 1$), gastrostomy site infection ($n = 1$)), ten grade II complications (pneumonia ($n = 5$), infection of unknown origin ($n = 2$), bleeding needing transfusion ($n = 1$), wound infection ($n = 1$), central venous catheter related infection ($n = 1$)) and six grade IIIB complications (revision of a central venous catheter ($n = 2$), gastrointestinal bleeding necessitating endoscopy and blood transfusion ($n = 1$), gastrostomy revision with drainage of peristomal abscess ($n = 1$), dislocated gastrostomy tube reinserted under general anesthesia ($n = 1$), respiratory problems requiring reintubation ($n = 1$)).

3.5. Long term outcomes after RF

Five (21%) patients had recurrent GERD after redo NF (Table 3). Three had NI, one had previous esophageal atresia, and one had no comorbidity. Only one patient had more than one RF. This patient had no comorbidity, underwent a third RF due to recurrent symptoms and later a fourth RF due to a herniated wrap, and is now without symptoms. Based on the follow-up questionnaire, three of the five patients had, with appropriate treatment, either no symptoms or improved symptoms compared to before the RF (Table 4). The two patients with persisting symptoms had investigations confirming recurrent GERD and reported vomiting at follow-up. They did not receive satisfactory treatment for their symptoms. Furthermore, three patients used PPI for other indications than GERD; gastritis ($n = 2$), to reduce gastric secretion because of severe gastrointestinal dysmotility ($n = 1$).

The most reported gastrointestinal symptoms at follow-up were troublesome flatulence and abdominal pain (Table 4). One patient with new onset retching and one patient who got delayed gastric emptying after the RF had been successfully treated with insertion of a TGJ feeding tube. Four parents and one patient were contacted by telephone because they wanted further follow-up. In addition, one patient had been operated two months and two years after the RF because of adhesion ileus.

Questions regarding satisfaction with outcome after the RF were answered by 22/24 (92%). 18 (82%) would choose RF again. Three would not, two were parents of patients with stomach pain and a herniated wrap and one patient with uncharacteristic upper abdominal pain, where endoscopy was normal. One parent of a child without any symptoms was unsure if she would choose RF again. Lastly, 21 (96%) said they would recommend RF to someone in a similar situation.

4. Discussion

This study on short and long-term outcome after RF in childhood demonstrates that most patients were successfully treated for their recurrent GERD. A large majority of parents and patients was satisfied with the outcome and would recommend RF to others in a similar situation. Those who experienced recurrent GERD or troublesome side effects after the RF, could often be satisfactorily treated without a new RF.

The most commonly reported results after both primary fundoplication and RF are recurrence and re-RF rates. Previous studies have reported symptomatic recurrence after RF in 25–45% of patients [3,6,13]. Thus, our finding of recurrent GERD in 21% is comparable to what has been reported earlier. Only one patient in our study underwent a re-RF (4%), reflecting previous findings of re-RF in 6–26% of patients [7–12]. Many pediatricians and pediatric surgeons assume that the success rate is considerably lower after RF than after primary fundoplication [9,10,20]. Interestingly, the recurrence rate after RF was similar to the recurrence rate after primary fundoplication at our center, questioning the opinion of significantly poorer results after RF [15].

Most patients reported no GERD symptoms at follow-up. Some mentioned early satiety and excessive flatulence, which are common complaints also after primary fundoplication [21]. Another frequently reported symptom was stomach pain. This is a symptom

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Table 3
Patients with recurrent gastroesophageal reflux disease after redo fundoplication.

Patient	Presenting symptoms	Objective findings	Treatment	Status at follow up
#1	Vomiting	Reflux index >4. Macroscopic esophagitis on endoscopy.	PPI	No symptoms
#2	Vomiting and abdominal pain	GER and wrap herniation on UGI	PPI	Persistent symptoms, parents reluctant to further surgical treatment
#3	Vomiting and regurgitation	Reflux index >4.	Redo fundoplication x2	No symptoms
#4	Abdominal pain	Wrap herniation on UGI	PPI	Improved symptoms
#5	Abdominal pain, vomiting and respiratory symptoms	Wrap herniation on UGI	PPI	Persistent symptoms, referred for jejunal feeding

Table 4
Symptoms occurring "often" or "almost always" in patients median 9 years after redo Nissen fundoplication. Not all questions were applicable to all patients.

Symptoms	$N = 24$
Vomiting	8% (2/24)
Regurgitation	0% (0/22)
Heartburn	0% (0/8)
Stomach pain	37% (7/19)
Retching	17% (4/24)
Discomfort during meals	17% (3/18)
Excessive flatulence	39% (9/23)
Dysphagia	25% (2/8)
Belching	18% (3/17)

that is difficult to interpret, especially in NI patients. Retching in NI patients may be present before fundoplication or may occur after surgery [22], and has been linked to increased risk of wrap herniation or disruption [23]. One patient experience new onset retching after the RF, which was completely resolved after initiating jejunal feeding, supporting jejunal feeding as a treatment option for severe post-fundoplication retching [24].

Almost all patients and parents were satisfied with the outcome of the RF. In contrast, the only other study that has addressed satisfaction after RF in children, found that only 17% of parents were satisfied [13]. One explanation for the discrepancy may be the definition of satisfaction. Baerg and coworkers defined satisfaction as improvement of GERD symptoms and that parents would proceed with fundoplication again. We separated these two measures and found that most parents were satisfied even if their child had recurrence or troublesome side effects. Another explanation for this surprising finding could be that symptoms were less severe compared to before the RF and often could be managed successfully without surgery. Furthermore, all patients had tried conservative treatment without satisfactory effect, and this may have contributed to tolerance for mild GERD symptoms and troublesome side effects.

Deciding how to treat recurrent GERD after RF that is unresponsive to antireflux medication, is challenging. In this series, none but one patient had a third fundoplication. Reluctance to undertake a third fundoplication is in line with results from other series [6]. Jejunal feeding is an alternative to primary fundoplication in NI patients, and may be even more relevant if recurrence occur after RF [1,4,25]. Total esophagogastric dissociation is another treatment option [5,26].

RF is technically more challenging than primary fundoplication, particularly after open surgery. Consequently, operation time is longer and the complication rate higher. In this series the mean operation time of 128 min is comparable with other studies reporting mean operating time for RF from 82 to 177 min [10,27]. We previously reported operating times of 89 and 150 min for open and laparoscopic fundoplication, respectively, reflecting that RF is a more complicated procedure than a primary fundoplication [28].

Half of the patients experienced early postoperative complications, and a third of complications were major. There are few previous reports on complication rates after RF. In a study of 130 children undergoing RF, Dalla Vecchia and coauthors reported postoperative complications in 35% [9]. We have previously reported early complications in half (54%) of patients after primary fundoplication of whom one in ten were major, supporting the opinion of RF as a technically demanding procedure [28].

Five patients or parents wanted follow-up because of troublesome symptoms. Two adult NI patients were according to parents not properly handled by the local healthcare providers. Both patients had symptoms from a herniated wrap. These cases show that transition from pediatric to adult health care is important also for children having undergone fundoplication, since recurrence may occur many years after the RF, and symptoms, particularly in NI patients, may be difficult to interpret [29,30].

Major strengths of this study are that we obtained patient/parent reported outcome measures and the long follow-up. The main limitation is the low number of patients. It would have strengthened the study if all patients had undergone postoperative investigations and if validated questionnaires for GERD had been applied.

In conclusion, this study finds that most patients undergoing RF were successfully treated. Regardless of recurrence or not, the patient and parental satisfaction was high. Based on these results and what is known in the literature, we believe that RF is a suitable treatment alternative when primary fundoplications fails. Careful patient selection and involvement of patients and parents in shared decision-making is important.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Levels of evidence

Treatment study level IV

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PPI: Proton pump inhibitor
GER: Gastroesophageal reflux
UGI: Upper gastrointestinal contrast study

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