Partial gastric pull-up in the treatment of patients with long-gap esophageal atresia

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Background: This study was to analyze outcomes of long-gap esophageal atresia (LGEA) treated with partial gastric pull-up (PGP) into the thorax.

Methods: The medical records of all children who had undergone PGP for LGEA from 1999 to 2012 were reviewed. Preoperative data, initial postoperative course, complications, time to full oral nutrition, follow-up diagnostics and nutritional status were assessed.

Results: Nine children who had undergone PGP were followed up for a mean period of 6.2 ± 3.1 years. Their median gestational age was 37+2 weeks, and mean birth weight 2462 ± 658 g. Eight children were primarily treated with a gastrostomy, their mean age at PGP was 11.4 ± 10.9 weeks and mean weight was 4484 ± 1966 g. Their mean operation time was 199 ± 51 minutes. Leakage was an early postoperative complication in three children, one of whom had a consecutive stricture resection. Late complications were stenosis (n=7) and gastro-esophageal reflux (n=5). The general status of the children was judged as "good" or "very good" on the last presentation. The median percentile of the body-mass-index was 25. Gastroscopy at 3.7 ± 3.2 years after the operation revealed a grade I esophagitis in two children. There was no death in this group of children.

Conclusions: Because of its high complication rate, partial gastric pull-up cannot be recommended as an alternative for the treatment of LGEA at present. A final judgment could be made on the basis of a comparative study.

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Introduction

Jejunal interposition (JI), colon interposition (CI) and gastric pull-up (GPU) are the most common surgical techniques for the treatment of long-gap esophageal atresia (LGEA).^[1] However, there is still no consensus on the optimal treatment for this entity. This disagreement concerning the best treatment option is eventually due to lack of evidence. There are just very few prospective studies on gastric pull-up with a total of 37 patients.^[2-4] Prospective comparative studies are lacking.^[1] The current evidence suggests that none of these most common techniques represents the optimal treatment option. The morbidity of the patients using these techniques is significant. In the currently available studies, the need of reoperation is probably not sufficiently outlined, as mentioned in a recent meta-analysis.^[1]

Surgical treatment for LGEA using the stomach without interposition of the intestine is a classical method which is used since the 1940s.^[5] Within the gastric transposition, the whole stomach is mobilized and brought into the thorax, where it is anastomosed with the cervical esophagus. This procedure can even be performed in the neonatal period without establishment of a gastrostomy.^[4] The method of GPU most does probably not reveal substantial disadvantages compared with JI and CI. Generally, the problems with all these methods on different levels do not imply any real drawback of GPU.^[1]

The possibility of a partial gastric pull-up (PGP) with incomplete positioning of the stomach in the thorax is generally not taken into consideration. Studies evaluating this method do not exist in the current literature. The reason for this is unclear. Possibly, first attempts with this technique have been unsuccessful and the method was not pursued. Another possibility might be that PGP could not build up an own "tradition" because of three already existing alternatives.

The method is technically simple and easy to learn compared with complete transposition of the stomach without the need for extensive resection. Becaue of the lack of adverse evidence, PGP has been performed at Karolinska University Children's Hospital in Stockholm as a treatment for patients with long-gap esophageal atresia in the past ten years.

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Methods

The medical records of all children who were operated on for long-gap esophageal atresia from 1999 to 2012 were reviewed. Long-gap esophageal atresia was defined as atresia with a long distance between the esophageal segments, which could not be brought close to each other within the definitive corrective procedure. Esophagostomies were not performed. The decision for definitive surgery was made when the patients were cardiorespiratory stable and had a weight of at least 2500 g.

Within the PGP procedure, the children were placed left side down. A right latero-dorsal thoracotomy was performed. Both ends of the esophagus dissected with preservation of the lower segment, and when judged incompatible with a direct anastomosis, up to half of the stomach was brought into the thorax via the transhiatal route, after which the two esophageal ends were anastomosed with resorbable sutures. If the access of the upper pouch was not possible due to a short segment, the cervical esophagus was mobilized through the neck. For the mobilization of the lower pouch, a laparotomy had to be performed. In contrast to the classical GPUprocedure,^[4] the vessel ligation was limited and the gastric perfusion via the celiac trunk was not affected. The stomach was just partly mobilized as far as necessary for the anastomosis and not additionally fixed. Tube gastroplasties or pyloromyotomies were not performed. A primary gastrostomy after birth was part of the concept. Nutrition via the gastrostomy was initiated shortly after the operation via gastrostomy. Jejunostomies were not performed. Fig. 1 shows an example of an upper contrast study 1.5 weeks after PGP.

Preoperative data records included gender, gestational age, birth weight, type of atresia, gastrostomy (yes/

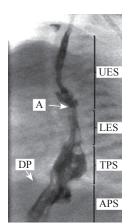


Fig. 1. Upper contrast study 1.5 weeks after partial gastric pull-up. DP: diaphragm; APS: abdominal portion of the stomach; TPS: thoracic portion of the stomach; LES: lower esophageal segment; UES: upper esophageal segment; A: anastomosis.

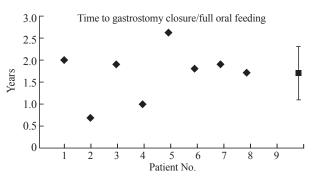


Fig. 2. Time to full oral nutrition (mean value) was generously defined as postoperathve time period till gastrostomy closure in order to refer to a definite time point.

no) and co-morbidities. The distance between the upper and lower segment under maximum tension was measured in vertebral bodies. Perioperative data records included age and weight at surgery, duration of operation and postoperative ventilation, early postoperative complications, early reoperation, time of discharge and weight at discharge. "Early complications" were defined as complications before discharge. Within the follow-up, late complications, reoperations and the general condition of the patients at last clinical control were analyzed. "Late complications" were defined as complications after discharge. The time to full oral nutrition was generously defined as a postoperative period till gastrostomy closure. This was due to an effort to have a definite time point of full oral nutrition. The body mass index at last presentation is referred to as percentile. An additional focus was the last gastroscopy with related histology and the last pH-measurement at least six months after PGP. Pathological reflux was defined as pH lower than 4 in more than 4% of measured time. The diagnosis of pneumonia was made by chest X-ray.

In the follow-up, the general status was judged by the treating physician as (very) good if the patients were thriving according to their age dependent growth curve (at least 10th percentile) with enteral nutrition and without respiratory or swallowing problems.

Data are shown as absolute values, mean values± standard deviation of the mean and median where appropriate.

Results

Partial gastric pull-up was performed in 10 children with long-gap esophageal atresia from 1999 to 2010. One of the children with severe malformations within a VACTERL association was treated especially for additional tracheal stenosis in other hospitals and could not be followed up properly. This child was excluded from the study.

| Patient No. | Gender | Gestational age (wk/d) | Birth weight (g) | Type of atresia | Gastrostomy | Co-morbidity | Gap length under max. Tension (vertebral bodies) |
|-------------------|--------|---------------------------|------------------|-----------------|-------------|--|---|
| 1 | Male | 37+2 | 3025 | Upper fistula | Yes | No | 2 |
| 2 | Male | 38+0 | 2600 | No fistula | Yes | Down's syndrom, duodenal atresia, ASD | 4 |
| 3 | Male | 39+0 | 2700 | No fistula | No | Down's syndrom, duodenal atresia, TAM | 1 ³ |
| 4 | Female | 30+5 | 1330 | Upper fistula | Yes | No | 6 |
| 5 | Male | 32+5 | 1550 | No fistula | Yes | Exomphalos | 3 |
| 6 | Male | 36+2 | 3015 | No fistula | Yes | Rectal atresia | 3 |
| 7 | Female | 38+0 | 3140 | No fistula | Yes | No | 2 |
| 8 | Female | 36+6 | 2100 | No fistula | Yes | Rectal atresia | 3 |
| 9 | Male | 38+4 | 2700 | No fistula | Yes | Down's syndrom | 1 |
| Mean value/median | - | 37+2 | 2462 ± 658 | - | - | - | 3 |

Table 1. Patients and preoperative data

ASD: atrial septal defect; TAM: transient abnormal myelopoeisis.

Table 2. Perioperative data and course

| Patient No. | Age at surgery (wk) | Weight at surgery (g) | Duration of ventilation (d) | Early postoperative complications before discharge | Early reoperation before discharge | Discharge (postop. wk) | Weight at discharge (g) |
|-------------|---------------------------|-----------------------------|-----------------------------------|--|------------------------------------|---------------------------|-------------------------------|
| 1 | 2.0 | 3100 | 6 | - | - | 4.6 | 3980 |
| 2 | 1.6 | 3800 | 7 | - | - | 3.1 | 4065 |
| 3 | 0.4 | 2705 | 2 | - | - | 2.1 | 2785 |
| 4 | 22.9 | 4780 | 2 | - | - | 3.0 | 5140 |
| 5 | 25.3 | 6520 | 1 | - | - | 1.3 | 6642 |
| 6 | 5.9 | 3750 | 11 | Leakage, stenosis | Stricture resection | 17.0 | 6455 |
| 7 | 27.4 | 8785 | 1 | - | - | 4.1 | 8420 |
| 8 | 11.6 | 3420 | 8 | Leakage | - | 5.3 | 3960 |
| 9 | 5.4 | 3500 | 5 | Leakage | - | 3.7 | 3660 |
| Mean value | 11.4±10.9 | 4484±1966 | 4.8±3.5 | - | - | 4.9±4.7 | 5012±1811 |

postop .: post-operative.

Characteristics and preoperative data of the children

At last, 9 children were included in the study: 6 were male and 3 female. Their median gestational age was 37+2 gestational weeks+days (range: 30+5 to 39+0) and their mean birth weight was 2462±658 g (range: 1330 to 3140). Two of the children had an esophageal atresia with upper fistula, and 7 had an atresia without fistula. The median distance between the upper and lower esophageal segment under tension was 3 vertebral bodies (range: 1-6). Three children had Down's syndrome, and 2 had additional duodenal atresia. Of the latter, one child had an atrial septal defect and the other a transient abnormal myelopoeisis. The third child with Down's syndrome was not affected by further comorbidities. Two of the chidren had rectal atresia and one had exomphalos, respectively. All children but one were primarily treated with a gastrostomy within the first two days of life. The detailed characteristics of patients are shown in Table 1.

Peri- and postoperative course

The mean age of the patients at surgery was 11.4 ± 10.9 weeks (range: 0.4 to 27.4) and the mean weight was 4484 ± 1966 g (range: 2705 to 8785). The mean operation time was 199 ± 51 minutes (range: 175-298 minutes). Intraoperative complications were not recorded. A pure

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thoracotomy was performed in 6 of the patients, a cervicothoracic access was done in one. A cervico-thoracicoabdominal access (separate incisions) and a thoracicoabdominal access were performed in each patient. The duration of postoperative ventilation was 4.8 ± 3.5 days (range: 1-11 days). Leakage was an early postoperative complication before discharge, which was observed in 3 patients with subsequent stricture resection.

The mean postoperative hospital stay of the patients was 4.9 ± 4.7 weeks (range: 1.3-17.0 days). The mean weight of the patients at discharge was 5012 ± 1811 g (range: 2785 to 8420) (Table 2).

Follow-up

The mean follow-up duration of the patients was 6.2 ± 3.1 years (range: 1.4 to 10.2). Late complications of the patients were observed: stenosis and subsequent leakage after stricture resection in one patient; isolated stenosi in 2 patients; isolated reflux that needed fundoplication in one patient; and stenosis with additional diagnosis of pathological gastro-esophageal reflux during the follow up in 4 patients. In all patients with stenosis, endoscopic dilatations were performed (Table 3).

Full oral nutrition was exceeded in 8 of the 9 patients. Only the patient who had been operated on the last did not attain full oral nutrition. The time to full oral

| Patient No. | Length of postop. clinical follow-up (y) | Late complications after discharge | Reoperation | General conditions at last clinical control | PPI |
|-------------|---|------------------------------------|--|--|-----|
| 1 | 1.4 | Stenosis, leakage | Stricture resection, operation for leakage, dilatation | Very good | Yes |
| 2 | 1.6 | Stenosis | Dilatation | Very good | Yes |
| 3 | 4.5 | Stenosis | Dilatation | Good | Yes |
| 4 | 9.4 | Stenosis, reflux | Dilatation | Good | Yes |
| 5 | 10.2 | - | - | Good | Yes |
| 6 | 6.8 | Reflux | Fundoplication | Very good | Yes |
| 7 | 7.3 | Stenosis, reflux | Dilatation | Very good | Yes |
| 8 | 6.8 | Stenosis, reflux | Dilatation | Good | Yes |
| 9 | 8.2 | Stenosis, reflux | Dilatation | Very good | Yes |
| Mean value | 6.2±3.1 | - | - | - | - |

Table 3. Course within a follow-up

PPI: proton pump inhibitors; postop.: post-operative.

feeding was generously defined as postoperative time till gastrostomy closure in order to refer to a definite time point. In one patient who did not undergo a gastrostomy, time to full oral feeding was documented in the medical charts. The mean time to full oral feeding of the patients was 1.7 ± 0.6 years (range: 0.7 to 2.6; Fig. 2).

In four patients (patients 3, 4, 6 and 8), at least one patient with pneumonia was documented. One patient (patient 6) had recurrent (>3) episodes of pneumonia. Two patients (patients 2 and 9) were diagnosed with infection-triggered asthma.

However, these respiratory problems were no longer an issue at last presentation. At this time, the median body mass index (BMI) was 25 percentile. The general status of the patients was judged as "good" (n=4) or "very good" (n=5). These patients received proton pump inhibitors immediately. There was no mortality in this series of patients.

Gastroscopy (n=8; mean time 3.7±3.2 years postoperatively, range: 0.9 to 9.1) and pH-measurement (n=7; 5.5±3.4 years, range: 1.7 to 10.5) were performed regularly during the follow-up. In one patient with excellent clinical outcome, neither gastroscopy nor pHmeasurement was performed because of absence of clinical indications over six months after repair. In another patient, pH measurement was not performed. Macroscopic signs of stenosis were not found. Grade I esophagitis was histologically confirmed in two patients. In all other patients, no inflammation was found. Pathological reflux was finally shown in three patients.

Discussion

The treatment of LGEA is often associated with complications and the best method has not yet been identified. The most widely used methods are JI,^[6,7] CI^[8,9] and GPU.^[10-12] These techniques do not substantially differ with respect to mortality.^[1]

However, their morbidity is significant. In a recent

review,^[1] there were 93 very early and 262 early gastrointestinal complications in 470 patients. In these patients 6% were reoperated because of graft loss or anastomotic leakage. According to one study, graft loss was found in 38% of patients who were operated upon with JI.^[7] Such devastating complications were not found in another study concerned with JI.^[6] In the present study, the percentages of anastomotic leakage (26%) and stricture (52%) were significant.

The technique of PGP was first described in the classic textbook by Robert E. Gross in 1953.^[13] PGP represents primarily a technical modification of the complete GPU into the thorax. Both methods share some features. A comparative study, however, does not exist. Within the traditional approach, the gastric vessels are extensively ligated.^[4,12-14] This might be a leading etiologic factor for anastomotic leakage, which was found in up to 36% of patients who were treated with GPU according to an retrospective analysis by Coran's group.^[15] The relative risk for anastomotic leakage in our considerably small series is comparable (33%, 3 out of 9 patients). The explanation for this observation has to remain hypothetical. But it can be assumed that anastomotic tension is considerably high in both methods. The proportion of patients with anastomotic stenosis is considerably higher in our patients compared with published data on GPU (78% vs. 49%).^[15] This might be at least partly due to our generous definition of stenosis, which is essentially based on radiological and endoscopical and not clinical findings. A technical explanation might be given by the use of the possibly narrow lower esophageal segment in case of PGP compared with a broader opening within classical GPU in combination with anastomotic tension.^[15] As in conventional gastric transposition, in PGP a loss of the "Angle of His" can be assumed, possibly favoring pathological reflux and, thus, microaspiration with respiratory problems.^[1] In all affected patients, stenosis could be managed with dilatations. Single patients had

temporarily problems with recurrent pneumonias and infection-induced asthma. However, at the last clinical presentations this was not a present concern.

More than half of our patients were in the lower percentiles for the body mass index. On the other hand, the 20th percentile is acceptable and problems have also been found in other methods including complete gastric transposition.^[12] However, a comprehensive evaluation of the patient situation could be optimally performed in a prospective study using standardized questionnaires. Medical assessment in the present study can be an orientation.

The incidence of stenosis and gastro-esophageal reflux after PGP is considerably high, even compared with alternative surgical techniques for the treatment of LGEA and with their high complication rates. Especially due to the limitations given by the low number of patients in our study, PGP cannot yet be generally recommended for the treatment of LGEA. However, systematic comparative studies evaluating the method are lacking.

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