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Collis-Nissen Procedure in Patients with Esophageal Atresia: Long-term Evaluation

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Abstract. A subset of patients with esophageal atresia (EA) have an associated short esophagus and require an antireflux procedure. The Collis-Nissen procedure, which consists of a combination of Collis gastroplasty and Nissen fundoplication, is considered an option in such conditions. The long-term results of EA patients undergoing this procedure have rarely been reported. The results of the Collis-Nissen procedure were examined in four EA patients with follow-up for an average of 9 years. The Collis-Nissen procedure was conducted concurrently with segmental esophageal resection for an associated congenital esophageal stenosis close to the esophagogastric junction in two postoperative Gross type C patients (7 months, 2 years), as an antireflux operation in a postoperative Gross type A patient with an unreducible sliding hiatal hernia (10 years), and with primary repair of EA in a Gross type A patient with a long gap (4 months). There were no significant complications except minor anastomotic leakage and an anastomotic stenosis that required postoperative dilation. Significant reflux was negative, as shown by an upper gastrointestinal study and scintigraphy. Limited esophagitis just above the esophagogastric junction was observed in a Gross type A patient with a long mediastinal gastric segment. Swallow-related pressure reduction at the wrapping cuff was detected by manometric examination. The Collis-Nissen procedure is a useful option for treating EA patients who require esophageal lengthening and control of gastroesophageal reflux.

A subset of patients with esophageal atresia (EA) require esophageal lengthening in various situations because of a short esophagus. It is often difficult to ensure the safety of the intraabdominal esophagus by mobilizing the distal esophagus during esophageal surgery following primary repair of EA because of the upward tension due to tight adhesion of the esophagus to the mediastinum [1].

It has been a therapeutic challenge for pediatric surgeons to repair Gross type A EA with a long gap between the proximal and distal ends of the esophagus. Various procedures have been performed for surgical treatment of EA with a long gap, but the long-term results of those procedures remain controversial [2].

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Collis gastroplasty, which creates a gastric tubular segment from the lesser curve of the stomach, is an option for lengthening the esophagus [3]. This procedure has been reported to be useful to control gastroesophageal reflux (GER) in patients with a short esophagus when conducted concurrently with Nissen fundoplication [4, 5]. The results of EA patients undergoing this procedure have rarely been reported [6]. We report here the long-term results of the Collis-Nissen procedure applied in EA patients.

Materials and Methods

Subjects

During 1989–1999 the Collis-Nissen procedure was performed in four EA patients by one pediatric surgeon (H.K.) in Osaka University Hospital and Osaka Medical Center for Maternal and Child Health. The clinical details of each patient were as follows.

Case 1. A female baby was born at 39 weeks' gestation weighing 2821 g. She was diagnosed with Gross type C EA and underwent primary repair at 4 days of age (Fig. 1). The association of congenital esophageal stenosis (CES) was suspected during the operation because of difficulty inserting an 8F catheter into the stomach, which was confirmed by postoperative esophagram at 17 days of age. Postprandial emesis and stridor became significant at 2 months of age. A 2 cm segmental resection of the esophageal narrowing and primary end-to-end anastomosis were performed at 7 months of age, when she weighed 7 kg. A Collis-Nissen procedure was conducted concurrently with the repair of the CES. The tracheobronchial remnants, including cartilage, were documented histologically in the resected specimens.

Case 2. A male baby was born at 37 weeks' gestation weighing 3046 g. He was diagnosed with Gross type C EA and underwent primary repair at 2 days of age (Fig. 2). CES was suspected during the operation because an 8F catheter could not pass through the distal esophagus into the stomach; it was confirmed when an esophagram showed a tapered narrowing close to the esophagogastric junction at 14 days of age. The frequency of food impaction

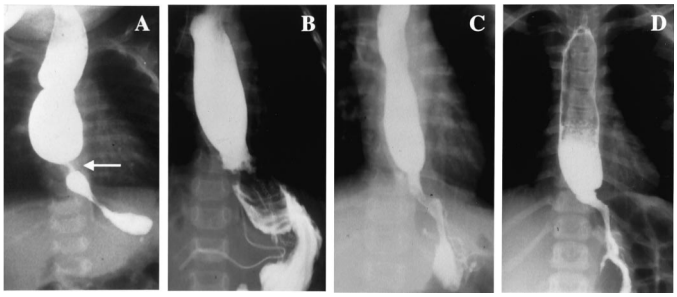

Fig. 1. Case 1. **A.** Free gastroesophageal reflux (GER) and stasis of contrast medium in the esophageal body proximal to the narrowing (white arrow) were observed at 2 months of age. **B.** A 2 cm segmental resection of the narrowing and primary end-to-end anastomosis with Collis-Nissen procedure was performed at 7 months of age. **C.** After repeated balloon dilatation for 1 year, swallowing difficulties and postprandial stridor were decreased at 19 months of age. **D.** Esophagram at 9 years of age shows an intact wrapping cuff and the appropriate passage of contrast medium from the esophagus into the intraabdominal stomach.

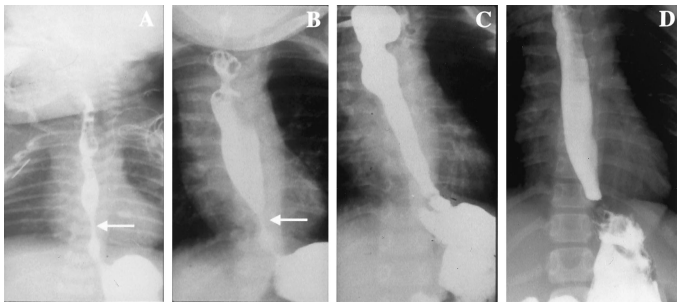

Fig. 2. Case 2. **A.** Initial esophagram at 14 days of age showed a tapered narrowing (white arrow) close to the esophagogastric junction. **B.** Persistent narrowing (white arrow) and free GER of contrast medium were observed at 20 months of age, when he had frequent respiratory infections and emesis. **C.** Same procedure (3 cm segmental resection) as in case 1 was performed at 2 years of age, and his symptoms were relieved by the operation. **D.** Esophagram at 10 years of age shows an intact wrapping cuff and no esophageal dilatation.

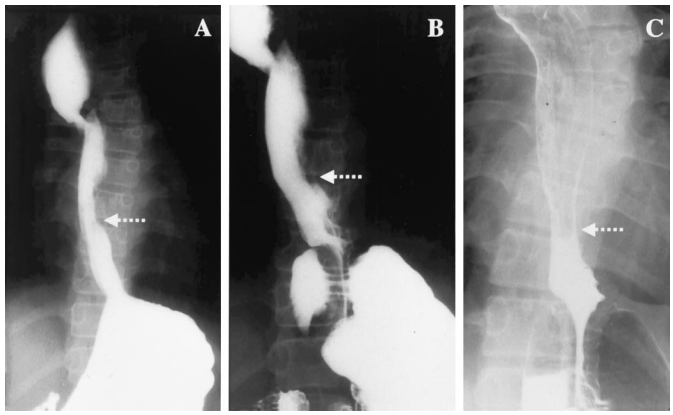

Fig. 3. Case 3. **A.** Esophagram at 9 years of age demonstrated GER in the upper esophagus and an unreducible sliding hernia. **B.** Collis-Nissen procedure performed at 10 years of age constructed a 7 cm gastric neoesophagus. As a result, an approximately 8 cm tubular stomach was located in the mediastinum. **C.** Appropriate esophagogastric passage and no reflux from the intraabdominal stomach into the neoesophagus were observed at 17 years of age. Dotted white arrows show the location of the esophagogastric junction.

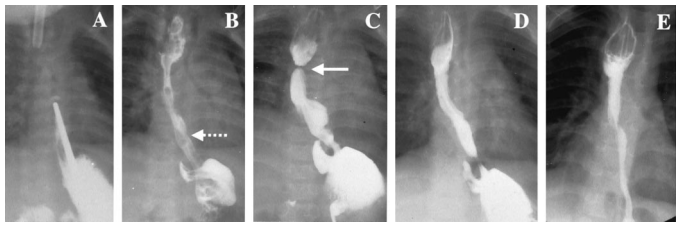

Fig. 4. Case 4. **A.** Four-vertebral-body length gap between the proximal and distal ends of the esophagus remained after 3-month bougienage to lengthen the esophagus. **B.** Primary repair with Collis-Nissen procedure was performed at 4 months of age. Mild anastomotic leakage was noted on an esophagram 2 weeks after the operation. **C.** Swallowing difficulties due to anastomotic stricture developed at 9 months of age. **D.** Esophageal balloon dilatation performed intermittently over 18 months eliminated the stricture. **E.** Esophagram at 3 years of age showed appropriate esophagogastric passage of contrast medium given orally. Dotted arrow shows the esophagogastric junction; white arrow shows an anastomotic stricture.

at the site of the narrowing increased after the age of 16 months. The same procedure (3 cm segmental resection) as in case 1 was performed at 2 years of age, which relieved his preoperative symptoms. The tracheobronchial remnants, including cartilage, were documented histologically in the resected specimens.

Case 3. A female patient with a Gross type A EA and a five-vertebral body length gap underwent primary repair at 5 months of age by right thoracotomy and reoperation for anastomotic stricture at 2 years of age by left thoracotomy (Fig. 3). She had complained of frequent emesis, especially during sleeping, and had been unable to sleep in the flat, supine position. An esophagram at 9 years of age demonstrated GER into the upper esophagus and an unreducible sliding hernia, indicating that the esophagus was too short to conduct Nissen fundoplication without esophageal lengthening. The acid exposure value was 12% in the distal esophagus and 55% in the stomach herniated into the mediastinum by 24-hour pH monitoring. Endoscopic examination showed advanced esophagitis in the distal esophagus. The Collis-Nissen procedure was conducted at 10 years of age. A 7 cm gastric neoesophagus was constructed; and as a result, an approximately 8 cm tubular stomach was located in the mediastinum.

Case 4. A male infant had a four-vertebral body length gap between the proximal and distal ends of the esophagus after 3-month esophageal elongation with bougienage (Fig. 4). He underwent primary repair combined with the Collis-Nissen procedure at 4 months of age, when he weighed 6 kg. Livaditis myotomy in the proximal pouch of the esophagus and pyloromyotomy were performed concurrently. Esophageal lengthening with Collis gastroplasty and Livaditis myotomy made it possible to perform primary esophageal end-to-end anastomosis.

Comments. The characteristics of each subject are summarized in Table 1. The age when the procedure was conducted ranged from 4 months to 10 years. Cases 1, 2, and 3 were diagnosed preoperatively with GER, as the acid exposure value in the distal esophagus was higher than 5% during 24-hour esophageal pH monitor-

Table 1. Characteristics of subjects.

Case	Gross type	Age at operation[a]	Follow-up (years)	Indications for Collis-Nissen procedure	Concurrent operative procedures[b]	Current condition
1	C	7 months	11	Associated CES at distal esophagus	Segmental resection of CES	Asymptomatic
2	C	2 years	12	Associated CES at distal esophagus	Segmental resection of CES	Asymptomatic
3	A	10 years	9	GER with unreducible sliding hernia	—	Chest pain (infrequent)
4	A	4 months	3	Long gap at primary repair for EA	Esophageal anastomosis with Livaditis myotomy	Emesis (infrequent)

CES: congenital esophageal stenosis; GER: gastroesophageal reflux; EA: esophageal atresia.
[a]Age when subjects underwent Collis-Nissen procedure.
[b]Operative procedures performed concurrently with Collis-Nissen procedure.

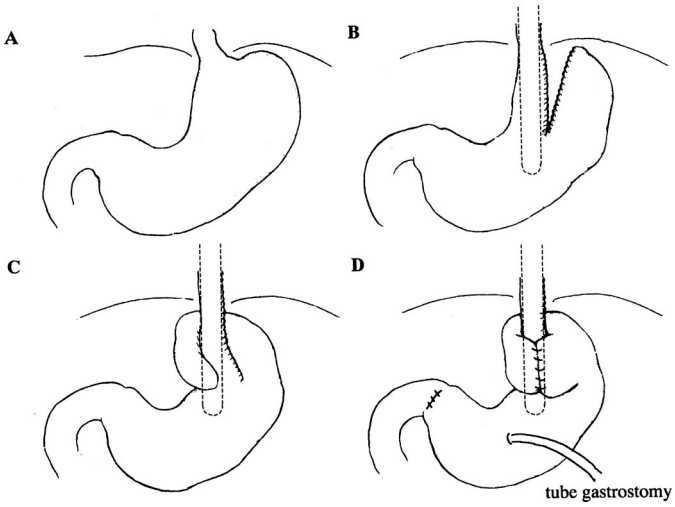

Fig. 5. Standard Collis-Nissen procedure. **A.** Operation is performed through a left subcostal incision. **B.** Vertical gastroplasty is created with staplers parallel to the lesser curve with the esophageal bougie in place in the esophagus. **C.** Fundoplication is wrapped loosely 360 degrees around the gastric neoesophagus with five nonabsorbable sutures. **D.** Heineke-Mikulicz's pyloroplasty is added when damage of the vagal nerve along the esophagus is considered. Tube gastrostomy is carried out routinely.

ing. The Collis-Nissen procedure was done to: (1) reconstruct the distal esophagus and fundus after segmental resection of the CES lesion located close to the esophagogastric junction (cases 1 and 2); (2) construct an antireflux barrier below the diaphragmatic hiatus to control advanced GER associated with a short esophagus (case 3); and (3) create an end-to-end anastomosis between the ends of the esophagus with a long gap (case 4). During the study period, Nissen fundoplication alone was used in other EA patients who required antireflux surgery, and primary esophageal end-to-end anastomosis with Livaditis myotomy was performed following esophageal elongation with bougienage in other Gross type A EA patients. The interposition of the intestine or the colon was not chosen as a definitive operation for Gross type A EA.

Collis-Nissen Procedure

The Collis-Nissen operation is performed through a left subcostal incision (Fig. 5). The esophagus is mobilized at the hiatus after division of all the short gastric vessels. Vertical gastroplasty is created with staplers (Autosuture TA 55; US Surgical, Norwalk, CT, USA) parallel to the lesser curve with the esophageal bougie in place in the esophagus. The fundoplication is wrapped loosely 360 degrees around the gastric neoesophagus with five nonabsorbable sutures. The total length of the wrap depends on the age of the patient, but it is usually 3 to 5 cm. Fixation of the wrap to the diaphragm and approximation of the crus are routinely performed. Heineke-Mikulicz's pyloroplasty is added when damage to the vagal nerve along the esophagus is considered. The pyloroplasty was conducted because of the possible vagal damage during segmental esophageal resection in cases 1 and 2 and extensive dissection of the distal esophagus in case 4. Tube gastrostomy is carried out routinely to decompress the stomach during the early postoperative stage and for supplemental feeding after enteral nutrition is commenced.

Follow-up Study

The subjects were followed up for 3 to 12 years. Clinical symptoms and radiographic findings were analyzed in all subjects. Endoscopy, esophageal pH monitoring, esophageal manometry, and scintigraphy were performed on clinical demand. Esophageal manometry was performed with the infusion method using a multichannel assembly associated with a sleeve sensor, the details of which were reported previously [7]. Manometric recording was performed for 30 minutes in each subject after a 4-hour fast.

Results

There were no deaths in this series. Early postoperative complications were minor leakage in the anastomosis in one case (case 4) and anastomotic stenosis in three cases (cases 1, 2, 4). The anastomotic leakage was treated conservatively with total parenteral nutrition for 2 weeks in case 4.

Esophageal balloon dilatation was performed repeatedly for anastomotic stenosis located just above the wrapping cuff over 1 year and over 3 months in cases 1 and 2, respectively (Figs. 1, 2). Swallowing difficulty and postprandial stridor continued until 19 months of age in case 1. Cases 1 and 2 were asymptomatic at the end of the follow-up period.

Case 3 had an uneventful course during the early postoperative period, and the regurgitation of intragastric contents experienced preoperatively was completely eliminated (Fig. 3). However, she developed a postoperative complaint of occasional nocturnal chest pain a couple of months after the operation. Twenty-four hour pH monitoring revealed that the pH at the esophageal anastomotic site remained higher than 4.0, whereas the acid exposure value was 81% in the gastric neoesophagus and 75% just above the esophagogastric junction. Scintigraphic examination with ^{99m}Tc-labeled Sn-colloid in the intraabdominal stomach

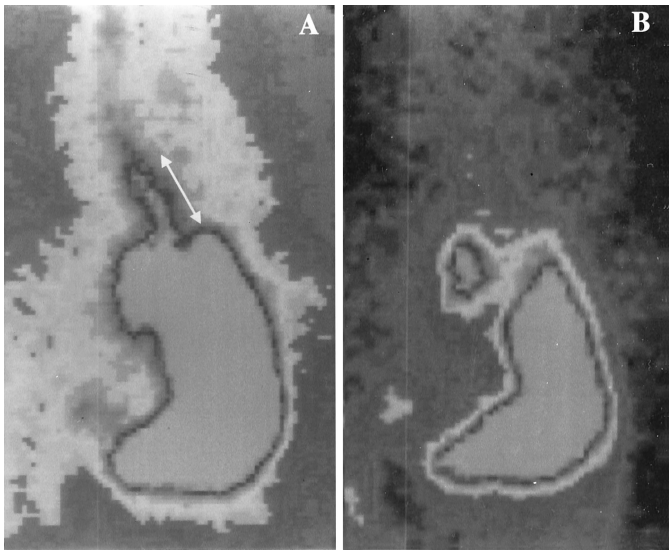

Fig. 6. Scintigraphic examination of case 3. **A.** Intravenous injection of ^{99m}Tc-pertechnetate showed the location of the gastric mucosa in the neoesophagus; it was located close to the level of the tracheal carina (white arrow) and in the intraabdominal stomach. **B.** Administration of ^{99m}Tc-labeled Sn-colloid into the intraabdominal stomach showed no reflux into the neoesophagus during the 60-minute measurement.

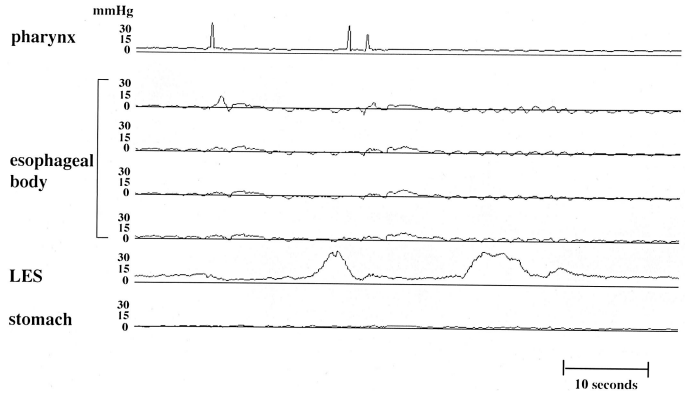

Fig. 7. Representative manometric tracing after Collis-Nissen procedure in case 1. The neo-lower esophageal sphincter (LES) at a wrapping cuff had 10 mmHg or less basal tone, as shown in the LES channel. Transient neo-LES pressure reductions occurred concurrently with swallow signals shown in the pharyngeal channel (pharynx). The amplitude of contractions was usually less than 30 mmHg, as shown in the esophageal and neoesophageal channels.

showed no reflux from the intraabdominal stomach into the gastric neoesophagus during a 60-minute measurement (Fig. 6). Intravenous injection of ^{99m}Tc pertechnetate showed the location of gastric mucosa in the neoesophagus, which reached the level of the tracheal carina and the intraabdominal stomach (Fig. 6). Ulcerative esophagitis of less than 1 cm was continuously observed just above the esophagogastric junction by repeated endoscopic examinations during the follow-up period, whereas an endoscopic retrograde view from the intraabdominal stomach confirmed intact cuff wrapping over the gastric neoesophagus. Microscopic findings in esophageal specimens obtained 5 years after the operation showed a low-degree dysplastic change in the area of the esophageal ulcer despite continuous administration of histamine-2 antagonist agents.

Esophageal balloon dilatation was performed intermittently for stricture in the anastomosis of the proximal and distal ends of esophageal atresia over 18 months in case 4 (Fig. 4). Case 4 required additional feeding via gastrostomy until the age of 3 years, when he could ingest the proper volume of food orally. He rarely exhibits emesis, except when he develops respiratory tract infections. His physical development remained approximately 1 SD below the average value for age compared with the Japanese standard.

Manometric examinations were performed in cases 1, 2, and 4. Basal pressure of the neo-lower esophageal sphincter (LES) at the wrapping cuff was usually 10 mmHg or less. Swallow-associated transient pressure reductions in the neo-LES were observed (Fig. 7). The amplitude of contractions was usually less than 30 mmHg in the esophagus and neoesophagus.

Discussion

We previously reported that an association of CES is a more important factor that influences the patients' subsequent clinical course after the repair of EA than has generally been appreciated [1]. In patients with a narrow segment in the distal esophagus, possible ways to reach the lesion are a transthoracic or transabdominal approach. A transabdominal approach was chosen for treating the CES in cases 1 and 2, as a right thoracotomy is usually performed to repair the EA before treating the CES, and postoperative EA patients might develop a chest deformity or respiratory malfunction by repeated (ipsilateral or bilateral) thoracotomy. Considering the poor acid-clearing capacity frequently associated with EA [8] and the impaired anti-reflux LES function caused by abdominal dissection of the distal esophagus, fundoplication is a useful adjunct for preventing postoperative GER when a distal esophageal lesion is treated by the transabdominal approach [9, 10]. In addition to cases 1 and 2, we have treated a postoperative Gross type A EA patient with CES, who underwent Nissen fundoplication following resection of a narrowing segment and primary end-to-end anastomosis [1]. This patient subsequently exhibited wrap herniation and swallowing difficulty. The upward tension on the esophagogastric junction caused by esophageal shortening after segmental resection of the distal esophagus may induce wrap failure. Esophageal lengthening with a Collis gastroplasty should be considered with Nissen fundoplication during surgical treatment of CES located in the distal esophagus in postoperative EA patients.

It is well known that GER is a major cause of postoperative complications seen in EA patients. The usefulness of Nissen fundoplication in the treatment of EA patients with GER has gained wide acceptance [11–13]. Evans reported a Gross type A EA patient with a 6 cm gap, who underwent primary repair after lengthening the distal esophagus with Collis gastroplasty at 3 months of age and who subsequently required Nissen fundoplication at 6 months of age [14]. Orringer et al. reported a 16-year-old EA patient with recurrent anastomotic stricture whose symptoms were eliminated by intraoperative esophageal dilatation and a combined Collis-Besley hiatal hernia repair [15]. Cameron et al. reported application of the Collis-Nissen procedure in EA patients but not in those with a congenital short esophagus, such as case 3 [6]. From a technical point of view, the standard Nissen

procedure was not applicable in case 3 because of the short esophagus and the tight adhesion of the esophagus with the mediastinal tissue, which was caused by the primary repair and the second operation for anastomotic stricture. The possible antireflux procedures for patients with such a short esophagus may be the Collis-Nissen procedure or the intrathoracic fundoplication procedure. Collis-Nissen procedure was considered preferable for case 3, as fundoplication procedures that leave the stomach in the chest exposes the patient to the well documented serious complications associated with paraesophageal hiatal hernia [16].

The cause of the low-degree dysplasia seen just above the esophagogastric junction in case 3 remains unclear. Advanced endoscopic esophagitis was observed before the Collis-Nissen procedure, but histologic examination was not performed at that time. These mucosal changes are possibly attributable to the esophageal acid exposure induced by GER prior to the Collis-Nissen procedure, which had continued for 10 years until the procedure was performed. However, the influence of the gastric juice secreted from the tubular gastric segment on the esophageal mucosa cannot be ignored. The scintigraphic and pH data in this case indicated a continuous acid environment in the tubular gastric segment consisting of the gastric neoesophagus and the original tubular stomach located in the mediastinum. The esophageal mucosa close to the esophagogastric junction may be frequently exposed to gastric acid, even when reflux from the intraabdominal stomach across the wrapping cuff to the distal esophagus was prevented. Collis stated that the area of the cardia and lesser curve has the definite ability to produce acid, but that it is deficient in its ability to produce pepsin [3, 17]. However, Lindahl et al. reported that gastric metaplasia in the cervical esophagus is frequent in gastric tube patients and is probably caused by the acid secreted by the parietal cells of the tubular stomach [18]. It may be speculated that pepsin and gastric acid secreted in the gastric tubular segment proximal to the wrapping cuff may damage the mucosa of the esophagus close to the gastric tubular segment when this segment is too long, such as in case 3. Parietal cell vagotomy combined with the Collis-Nissen procedure was reported to reduce acid secretion in the mediastinal tubular gastric segment [19]. However, acid secretion is not completely abolished even after parietal cell vagotomy because gastrin stimulates acid secretion in the vagotomized gastric neoesophagus [18]. Long-term endoscopic follow-up was necessary to detect the malignant changes that developed in the distal esophagus in case 3.

There are many options for the surgical management of EA with a long gap: circular or spiral esophageal myotomies, interposition with colon or jejunum, gastric transposition, gastric tube esophagoplasty, extrathoracic esophageal elongation [2, 20–24]. Despite various surgical procedures, no consensus has been reached with respect to the treatment of choice for EA patients with a long gap. We propose a primary esophageal anastomosis concurrent with Collis-Nissen procedure as a reasonable option. Collis gastroplasty provides sufficient length of the neoesophagus and prevents excessive GER when conducted with Nissen fundoplication. This procedure is not difficult in EA patients weighing more than 6 kg when the stomach has been used for enteral nutrition preoperatively. In case 4, there have been no significant postoperative symptoms except infrequent emesis. Further experience with the Collis-Nissen procedure concurrent with primary esophageal repair of EA may be necessary to assess the usefulness of this procedure as the definitive operation for Gross type A EA with a long gap.

Manometric examinations to evaluate esophageal motor function have rarely been performed after the Collis-Nissen procedure. Cooper et al. demonstrated a transient pressure reduction in the neo-LES coincident with peristaltic esophageal contractions using a three-transducer built-in catheter [25]. We have shown pressure changes in the esophagus, including the neoesophagus and neo-LES, using a manometric assembly with a sleeve sensor, which has been widely accepted as the most appropriate method for measuring sphincter pressure changes [26]. The postoperative basal pressure in the neo-LES reported by Cooper et al. was approximately 20 mmHg [25], whereas it was mostly 10 mmHg or less in our patients. Considering esophageal dysmotility in EA patients, a loose fundoplication may be better for avoiding postoperative swallowing difficulties.

Conclusions

The Collis-Nissen procedure is a safe, useful option for surgical treatment of EA patients who require esophageal lengthening with reflux control, so long as the length of the mediastinal gastric segment is limited.

Résumé. Parmi les patients présentant une atrésie de l'œsophage (AO), il existe un sous groupe avec œsophage court et qui nécessite un procédé anti-reflux. Le procédé de Collis-Nissen, qui consiste en la combinaison d'une gastroplastie de type Collis combinée à une fundoplicature de type Nissen est considérée comme une option valable dans ces conditions. Les résultats à long terme chez les patients porteurs d'AO n'ont été que rarement rapportés. On a analysé les résultats du procédé de Collis-Nissen chez quatre patients ayant une AO ayant un suivi d'un moyen de 9 ans. Le procédé de Collis-Nissen a été réalisé en même temps qu'une résection segmentaire de l'œsophage pour sténose oesophagienne congénitale associée située près de la jonction oesogastrique chez deux patients de type C de Gross postopératoires (7 mois, 2 ans), comme procédé anti-reflux chez un patient de type A de Gross postopératoire ayant une hernie hiatale par glissement irréductible (10 ans), et comme méthode de cure primitive d'AO chez un patient de type A de Gross avec un intervalle long (4 mois). Il n'y avait aucune complication importante, sauf un cas de fistule anastomotique et une sténose anastomotique nécessitant une dilatation postopératoire. On n'a pas mis en évidence de reflux sur le transit eosphagogastrique ou la scintigraphie. Une oesophagite limitée juste au-dessus de la jonction a été observée chez un patient Gross-A porteur d'un long segment gastrique dans le médiastin. Une réduction des pressions au niveau de la valve a été détectée par la manométrie en rapport avec la déglutition. Le procédé de Collis-Nissen est une option utile dans le traitement d'AO nécessitant un rallongement de l'œsophage et le contrôle d'un reflux gastro-oesophagien.

Resumen. Un subgrupo de pacientes con atresia esofágica (AE) presenta esófago corto y requiere elongación esofágica más un procedimiento anti-reflujo. El procedimiento de Collis-Nissen, que consiste en la combinación de la gastroplastia de Collis (que crea un tubo gástrico a partir de la curvatura menor del estómago para elongamiento esofágico) con la fundoplicación de Nissen, es considerado como una opción para ser aplicada en tal condición. Se dispone de escasos informes sobre resultados a largo plazo en pacientes con AE sometidos a este procedimiento. Los resultados a largo plazo con el procedimiento de Collis-Nissen fueron analizados en 4 pacientes con AE con un seguimiento promedio de 9 años. El procedimiento de Collis-Nissen fue practicado concomitantemente con una resección segmentaria del esófago por estenosis congénita asociada del esófago en la región vecina a la unión gastroesofágica en 2 pacientes postoperatoirs con AE del tipo Gross C (7 meses, 2 años), como una operación anti-reflujo en un paciente postoperatorio con AE del tipo Gross A con una hernia por deslizamiento irreductible (10 años) y con una reparación primaria de la AE en un

paciente del tipo Gross A con un defecto largo (4 meses). No se presentaron complicaciones importantes excepto escape anastomótico menor y estenosis anastomótica que requirió dilatación postoperatoria. No se detectó reflujo significante ni en el estudio radiográfico del tracto GI superior ni en la escintigrafía. Se observó esofagitis limitada en el paciente del tipo Gross A con un largo segmento gástrico mediastinal. Se registró reducción de la presión de deglución a nivel del manguito antireflujo mediante examen manométrico. El procedimiento de Collis-Nissen es una opción útil en el tratamiento de pacientes con AE que requieren alargamiento esofágico y el control de reflujo gastroesofágico.

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