Clinical outcomes of tracheoesophageal diversion and laryngotracheal separation in neurologically impaired children

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ABSTRACT

Objective: Outcomes of tracheoesophageal diversion and laryngotracheal separation were evaluated in 15 neurologically impaired children treated for intractable aspiration.
Methods: A retrospective analysis of the hospital records was carried out in 15 consecutive pediatric patients who underwent either tracheoesophageal diversion or laryngotracheal separation with trumpet-shaped tracheotomy from 1999 to 2006 in Kagawa Children's Hospital.
Results: The number of hospital admissions for aspiration pneumonia after surgery was significantly decreased in 6 patients who were cared for at home. The parent-reported number of secretion suctioning was decreased after surgery especially in patients with pre-operative tracheotomy or intubation. Four patients fed through naso-gastric tube progressed with oral diet post-operatively, whereas 3 patients who had pre-operative tracheotomy developed temporary post-operative tracheocutaneous fistula that was managed by local wound care.
Conclusion: It is suggested that tracheoesophageal diversion and laryngotracheal separation decrease the morbidity of pediatric patients and improve their quality of life and that of their parents. It is suggested that the risk of developing post-operative fistula is higher in pediatric patients with than without pre-operative tracheotomy.

1. Introduction

Chronic pulmonary aspiration is frequently observed in neurologically impaired children. The intractable aspiration is a life-threatening medical problem and often results in increased suction requirement, repeated hospitalizations for aspiration pneumonia, expensive nursing care and decreased the quality of life in both patients and their parents.

Various techniques have been proposed to control intractable aspiration. These include the tracheoesophageal diversion reported by Lindeman in 1975 [1] and its variant, the laryngotracheal separation reported in 1976 [2]. These two procedures have become the standard procedures to intractable/chronic aspiration of oral/gastric secretions into the lower respiratory system by the separation of the digestive and respiratory tracts. Tracheoesophageal diversion connects the upper trachea to the cervical esophagus, while laryngotracheal separation includes

closure of the proximal trachea. Additionally, both procedures that preserve the structural integrity and motor and sensory innervations of the larynx are potentially reversible.

In the present study, outcomes of tracheoesophageal diversion and laryngotracheal separation were evaluated in 15 neurologically impaired children. The number of hospital admissions for aspiration pneumonia, the frequency of secretion suctioning by their parents, dietary route and post-operative complications were examined retrospectively.

2. Methods

2.1. Patients

A retrospective analysis of the hospital records was carried out in 15 consecutive neurologically impaired children who underwent either tracheoesophageal diversion or laryngotracheal separation for the treatment of intractable aspiration pneumonia from 1999 to 2006 in Kagawa Children's Hospital. Ten patients (9 males and 1 females; 3–20 years old; mean age: 12.3 years) were submitted to tracheoesophageal diversion and five patients (5 males; 4–14 years old; mean age: 8.6 years) to laryngotracheal

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separation. Their underlying medical conditions contributing to aspiration include anoxic injury at birth (3 cases), anoxic injury after near drowning (3 cases), anoxic injury during cardiac operation (1 case), cerebral palsy (4 cases), congenital hydrocephalus (2 cases) and Gaucher's disease (2 cases).

2.2. Operative procedures

Tracheoesophageal diversion was performed in 10 patients, including 4 patients who underwent tracheotomy prior to the surgery. Laryngotracheal separation was performed in 5 patients, including 3 patients with pre-operative tracheotomy.

The tracheoesophageal diversion was preferred initially, because the tracheoesphageal anastomosis allows the drainage of secretions from the larynx into the esophagus. The laryngotracheal separation was chosen in patients with high tracheotomy [3] or those with a thoracic deformity such as scoliosis or retroflexion to avoid the tension of tracheoesphageal anastomosis.

Under general anesthesia, inverse triangular skin flap was made between the caudal end of the thyroid cartilage and the rostral end of the sternum. After the thyroid isthmus was divided in the midline and its lobes separated, the recurrent laryngeal nerves were identified and carefully preserved. During the tracheoesophageal diversion, the trachea was divided at the level between the third and fourth tracheal rings obliquely upward on the second tracheal ring or at the level of the tracheostomy after exposing the cervical esophagus posterior to it (Figs. 1a and 2a). After an inverse U-shaped incision was made in the anterior cervical esophagus (Fig. 1b), the mucomuscular flap of the cervical esophagus was anastomosed to the proximal trachea reflected superiorly in an end-to-side fashion (Fig. 1c and d). The anastomosis was then reinforced with the thyroid lobes (Fig. 1e) and covered with the cervical inverse triangular skin flap (Fig. 1f). The inferior cervical skin was invertedly sutured to cover the cartilaginous portion of the distal trachea. A midline vertical incision (1 cm) was made in the membranous portion of the distal trachea (Fig. 2b) and the V-shaped superior skin flap was inserted into the incision and sutured to the membranous flaps (Y–V plasty) (Fig. 2c), creating a trumpet-shaped tracheostomy (Fig. 2d).

During the laryngotracheal separation, the trachea was divided horizontally between the first and second tracheal rings or at the level of the tracheostomy. The proximal tracheal lumen was sutured to close posterioanteriorly in several layers so as to invert the tracheal mucosa into the tracheal lumen. The closure was then reinforced by strap muscles and subcutaneous tissues over the tracheal stump. The inferior cervical skin was invertedly sutured to cover the cartilaginous portion of the distal trachea. A midline vertical incision (1 cm) was made in the membranous portion of the distal trachea and the V-shaped superior skin flap was inserted into the incision and sutured to the membranous flaps (Y–V plasty), creating a trumpet-shaped tracheostomy.
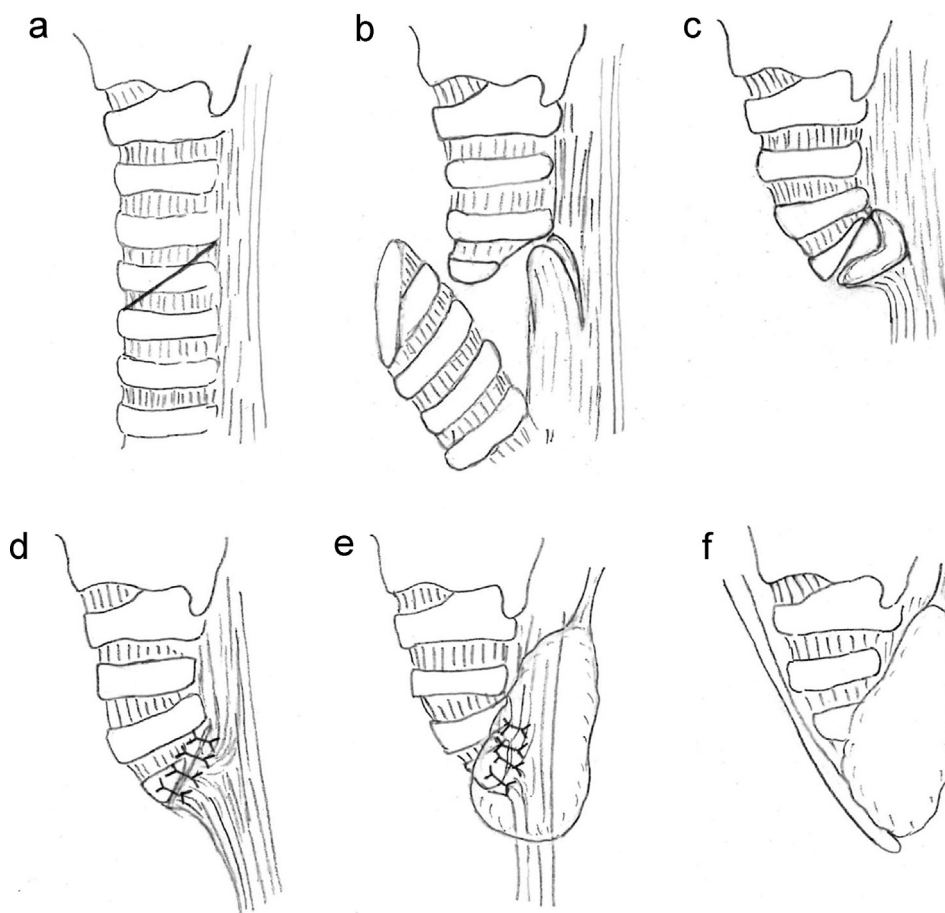

Fig. 1. Operative procedures tracheoesophageal diversion in the proximal trachea. (a) The trachea was divided obliquely after exposing the cervical esophagus posterior to it. (b) An inverse U-shaped incision was made in the anterior cervical esophagus. (c and d) The mucomuscular flap of the cervical esophagus was anastomosed to the proximal trachea reflected superiorly in an end-to-side fashion. (e and f) The anastomosis was then reinforced with the thyroid lobes and covered with the cervical inverse triangular skin flap.
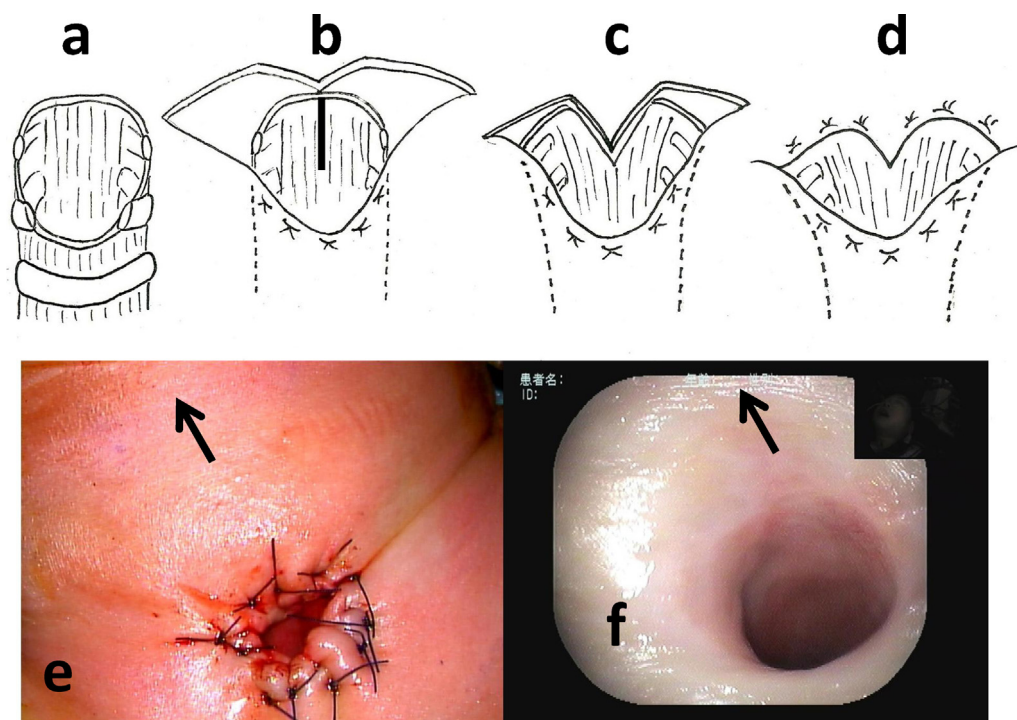
Fig. 2. Operative procedures of trumpet-shaped tracheotomy. (a) The trachea was divided obliquely after exposing the cervical esophagus posterior to it. (b) A midline vertical incision was made in the membranous portion of the distal trachea. (c) V-shaped superior skin flap was inserted into the incision and sutured to the membranous flaps (Y–V plasty). (d) A trumpet-shaped tracheal stoma was created. (e) The tracheal stoma immediately after trumpet-shaped tracheotomy. (b) The tracheal stoma 6 months after trumpet-shaped tracheotomy. The arrow indicates rostral direction.

The selection of a tracheotomy tube that properly fits the airway is important to prevent granulation formation with resultant tracheal stenosis. But, any tracheal tubes cannot conform to the airway anatomy in some neurologically impaired children. The trumpet-shaped tracheostomy we created in the present study can reduce the pressure or rubbing of the tracheal wall by the curve or tip of the tracheal tube in such children (Fig. 2e and f).

2.3. Factors reviewed

Chart reviews were used to evaluate the number of hospital admissions for aspiration pneumonia prior to and after the surgery in 6 patients (case 2, 3, 5, 11, 12, 15) who were cared for at home. Medical questionnaire in the charts were used to examine the parent-reported frequency of suctioning for removal of secretions prior to and after the surgery in 8 patients (case 2, 4–6, 8, 11, 12,

15). Dietary route and post-operative complications were also determined by chart reviews.

2.4. Statistical analysis

Student's *t*-test and chi-squared analysis were used for statistical analysis. $p < 0.05$ was considered significant.

3. Results

Among 15 neurologically impaired children, 10 underwent tracheoesophageal diversion and 5 laryngotracheal separation to prevent intractable chronic aspiration (Table 1). Seven patients (47%) had a tracheotomy prior to the surgery. Three patients (20%) developed temporary tracheocutaneous fistula post-operatively, which was managed by local wound care. Among them, a patient underwent tracheoesophageal diversion and two laryngotracheal

Table 1
Tracheoesophageal diversion and laryngotracheal separation in children with severe neurological disorders.

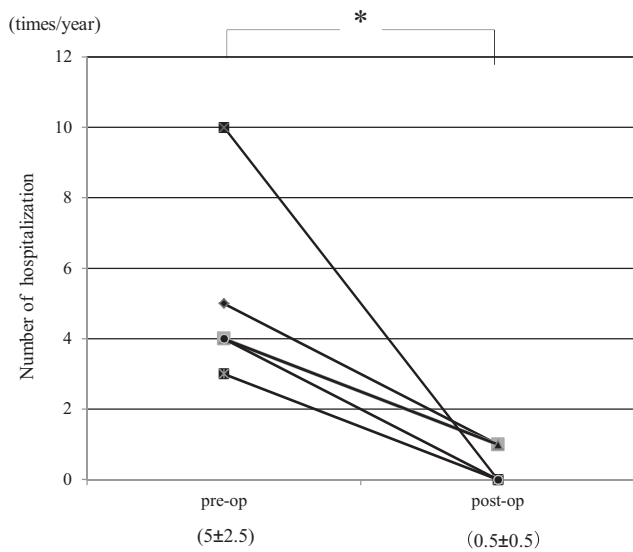| Case	Age	Sex	Operative method	Tracheotomy	Complication
1	3	M	Tracheoesophageal diversion	−	−
2	4	M	Tracheoesophageal diversion	−	−
3	8	M	Tracheoesophageal diversion	−	−
4	14	M	Tracheoesophageal diversion	−	−
5	16	F	Tracheoesophageal diversion	−	−
6	20	M	Tracheoesophageal diversion	−	−
7	12	M	Tracheoesophageal diversion	+	−
8	12	M	Tracheoesophageal diversion	+	−
9	14	M	Tracheoesophageal diversion	+	−
10	20	M	Tracheoesophageal diversion	+	+
11	6	M	Laryngotracheal separation	−	−
12	9	M	Laryngotracheal separation	−	−
13	4	M	Laryngotracheal separation	+	−
14	10	M	Laryngotracheal separation	+	+
15	14	M	Laryngotracheal separation	+	+

Fig. 3. Effects of tracheoesophageal diversion and laryngotracheal separation on the number of hospital admissions for aspiration pneumonia in 6 neurologically impaired children who were cared for at home postoperatively. *$p < 0.05$.

separation. On the other hand, all three patients had a tracheotomy prior to the surgery, indicating the tendency for pre-operative tracheotomy to be associated with this complication ($p = 0.0549$). Moreover, their tracheotomy was infected by methicillin-resistant *Staphylococcus aureus* (MRSA). No patients developed granulation formation around the stoma of trumpet-shaped tracheostomy and tracheal stenosis after the surgery. Four patients (Case 5, 6, 8 and 15) had no cannula through post-operative tracheotomy stoma.

The number of hospital admissions for aspiration pneumonia after surgery was significantly decreased in 6 patients who were cared for at home. Pre-operatively, they were hospitalized for aspiration pneumonia 5.0 ± 2.5 (mean $\pm$ S.D.) times/year, which was significantly decreased to 0.5 ± 0.5 times/year post-operatively (Fig. 3).

Medical questionnaires in the charts of 8 patients indicated that the parent-reported number of secretion suctioning (times/day) was decreased after the surgery (Fig. 4). The decrease in suctioning frequency was more remarkable in patients with tracheotomy (case 8, 15) or intubation (case 2, 6, 12) prior to surgery.

Pre-operatively, 4 patients were fed through gastrostomy and 11 through naso-gastric tube. Among them, 4 patients (26.7%) fed through naso-gastric tube progressed to oral diet post-operatively.

4. Discussion

Intractable aspiration is a life-threatening medical problem in neurologically impaired children. Several surgical procedures have been reported in an attempt to prevent it. These included total laryngectomy, glottis closure and tracheotomy [4–6]. In 1975, Lindeman reported tracheoesophageal diversion, a surgical procedure for directing aspirated saliva/gastric juice into the cervical esophagus via an end-to-side tracheoesophageal anastomosis [1]. In 1976, he modified the procedure by closing the proximal trachea as a blind pouch [2]. These procedures give a good control of the aspiration, but sacrifice verbal communication. The loss of speech is a major handicap in normal patients, but most neurologically impaired children were essentially unable to communicate verbally. So, tracheoesophageal diversion and laryngotracheal separation are commonly used nowadays by pediatric otolaryngologist for the treatment of intractable aspiration in neurologically impaired children [7]. The procedures have proved to be reliable in preventing aspiration with a low risk complications. Moreover, both procedures are more acceptable for patients' parents and family, because the larynx is preserved.

In the present study, tracheoesophageal diversion and laryngotracheal separation significantly decreased the number of hospital admissions for aspiration pneumonia in patients who were cared for at home. This finding indicates that these procedures are effective in preventing aspiration and thereby decrease morbidity in these patients. The number of secretion suctioning was also decreased after the surgery especially in patients with pre-operative tracheotomy or intubation. Four patients who were unable ingest food orally could tolerate oral diet post-operatively. These findings are in line with previous
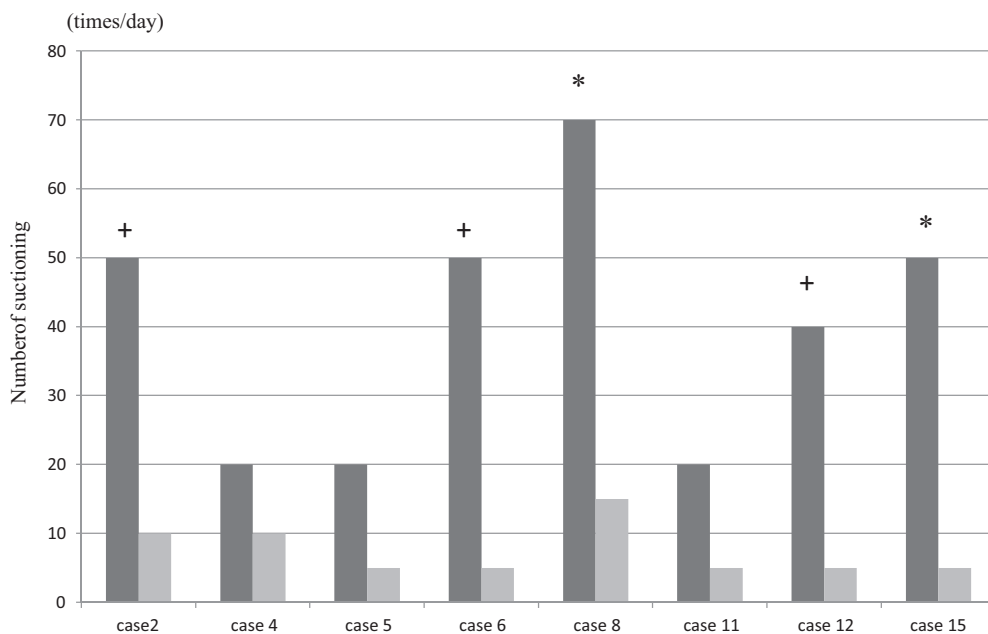

Fig. 4. Effects of tracheoesophageal diversion and laryngotracheal separation on the number of suctioning in 8 neurologically impaired children. *Tracheotomy prior to the surgery, +intubation prior to the surgery.

reports showing that some patients processed with oral diet instead of naso-gastric tube or gastorostomy after surgery [8,10]. They also suggest that tracheoesophageal diversion and laryngotracheal separation improved the quality of life not only in the patients and their parents, but also reduced the burden of work for nursing-care helpers. Indeed, it was reported that after laryngotracheal separation in neurologically impaired children, the satisfaction of their parents was increased because of reduced hospitalizations and home care requirements [8–11].

Ttracheocutaneous fistula was the most frequent complication after tracheal diversion or separation. Indeed, in the present study, three patients (20%) developed temporary tracheocutaneous fistula in the early post-operative period. This complication has been reported on other studies, being observed in 17.6% of patients in the study of Eisele et al. [12], 38% in that of Eibling et al. [13], 22% in that of Yamana et al. [14] and 23% in that of Zocratto et al. [15]. In children, the incident of fistula was 20% in the present study as compared to 11% of pediatric patients in the study in Cook [8]. Thus, there seems to be no difference in the incidence of this complication between adults and children. In the present study, three patients who developed temporary tracheocutaneous fistula post-operatively had undergone a tracheotomy prior to the surgery. Therefore, there was a tendency to develop fistula in patients with prior tracheotomy, but statistical analysis indicates a borderline p value ($p = 0.0549$), probably because of small samples. The risk of developing the post-operative fistula in pediatric patients who had prior tracheotomy may be higher than that in children without tracheotomy, as reported previously [8,13,14], probably due to the tracheotomy wound infection. In fact, in the present study, tracheotomy of all three patients with post-operative tracheocutaneous fistula was infected by MRSA. On the other hand, the presence of fistula was not related to diversion or separation.

In the present study, we initially recommended tracheoesophageal diversion to prevent intractable aspiration of their oral/gastric secretion in neurologically impaired children, because the diversion drains the secretion into esophagus through the anastomosis. However, we chose the laryngotracheal separation in patients with high tracheotomy [3] or the deformity of thorax due to scoliosis or retroflexion to avoid the tension of tracheoesophageal anastomosis, Baron and Dedo have demonstrated that any secretion that pooled in the pouch of laryngotracheal separation would drain when the patients was recumbent [16]. Moreover, we used V-shaped skin flap and made a Y–V plasty to create a trumpet-shaped tracheal stoma. This surgical technique is useful to prevent the development of tracheostomy stenosis by the development of granulation around it or a lethal artery fistula in neurologically impaired children, because the trumpet-shaped tracheotomy reduces the depression or rubbing of the tracheal wall by the curve or tip of the tracheal tube. In fact, no granulation formation around the stoma of trumpet-shaped tracheostomy and tracheal stenosis were observed in the present study, although it was reported that stenosis of tracheostomy stoma developed after laryngotracheal separation in 4 of 56 neurologically impaired children [8].

In conclusion, both tracheoesophageal diversion and laryngotracheal separation in the present study are effective surgical procedures in preventing aspiration pneumonia in neurologically impaired children as reported previously. They not only decrease the morbidity of pediatric patients, but also improve the quality of life in the patients and their parents. In the present study, we developed and created a trumpet-shaped tracheal stoma to prevent granulation formation around the stoma and tracheal stenosis. We also emphasized that the risk of developing the post-operative fistula in pediatric patients who underwent pre-operative tracheotomy may be higher compared to those who did not. Tracheoesophageal diversion and laryngotracheal separation should be considered in the management of neurologically impaired children with intractable aspiration before the performance of tracheotomy.

Conflict of interest

We declare that we have no conflicts of interest.

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