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ORIGINAL ARTICLE

Laryngotracheal separation in pediatric patients: 13-year experience in a reference service

Separação laringotraqueal em pacientes pediátricos: 13 anos de experiência em um serviço de referência

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ABSTRACT

Objective: To evaluate clinical stability of neurologically impaired children and adolescents with recurrent pneumonia submitted to laryngotracheal separation. **Methods:** Between October 2002 and June 2015, 92 neurologically impaired children from a reference service, with median age of 68.5 months were submitted to laryngotracheal separation. Data were evaluated and statistical analysis was made by Student's *t* test and Pearson's χ^2 test (significance level adopted of 95%). **Results:** Fifty-three children were male (57.6%). Forty-six children required admission to intensive care, and 42.4% needed mechanical ventilation. We observed that 90.2% of patients were exclusively fed by gastrostomy and 72.4% of the gastrostomies were performed before the tracheal surgery. Thirteen (14.1%) children had postoperative complications as follows: fistulae (5.4%), bleeding (4.3%), granuloma (2.2%) and stenosis (3.2%). A total of 24 patients had pneumonia in the postoperative period (26.1%), but there was a significant drop in occurrence of this condition after surgery (100% *versus* 26.1%; $p<0.001$). Twenty-three patients (25%) died. Postoperative complications were similar when comparing patients who died and those that presented good outcome (16.7% *versus* 13.2%; $p=0.73$). **Conclusion:** When well-indicated, the laryngotracheal separation reduces the incidence of postoperative pulmonary infections, thus improving quality of life and reducing admissions to hospital. Laryngotracheal separation should be indicated as a primary procedure in patients with cerebral palsy and recurrent aspiration pneumonia.

Keywords: Larynx/surgery; Trachea/surgery; Pneumonia, aspiration; Nervous system diseases; Child

RESUMO

Objetivo: Avaliar a estabilidade clínica da criança e do adolescente neuropata com episódios de pneumonia de repetição submetidos a procedimento cirúrgico de separação laringotraqueal. **Métodos:** Entre outubro 2002 a junho 2015, 92 crianças neuropatas de um único serviço com idade mediana de 68,5 meses foram submetidas à separação laringotraqueal. Os dados foram avaliados e foi realizada análise estatística pelo teste *t* de Student e pelo teste do χ^2 de Pearson, com nível de significância adotado de 95%. **Resultados:** Dentre as 92 crianças, 53 eram do sexo masculino (57,6%). Quarenta e seis crianças necessitaram de internação em unidade de terapia intensiva, e 42,4% fizeram uso de ventilação mecânica. Dessas crianças, 90,2% alimentavam-se exclusivamente via gastrostomia, e 72,4% foram realizadas antes da separação laringotraqueal. As complicações pós-operatórias ocorreram em 13 crianças (14,1%), na seguinte ordem: fístula (5,4%), sangramento (4,3%), granuloma (2,2%) e estenose (3,2%). Observaram-se 24 episódios de pneumonia no período pós-operatório (26,1%). Houve diminuição significativa de ocorrência

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de pneumonias após a cirurgia (100% *versus* 26,1%, p<0,001). Óbito foi registrado em 23 pacientes (25%). A frequência de complicações pós-operatórias foi semelhante entre os pacientes que evoluíram ou não para óbito (16,7% *versus* 13,2%; p=0,73). **Conclusão:** A cirurgia bem indicada reduz o número de infecção pulmonar após o procedimento, melhorando a qualidade de vida desses pacientes e, consequentemente, reduzindo o número de internações. A separação laringotraqueal deve ser indicada como procedimento primário nos pacientes com paralisia cerebral e episódios repetidos de pneumonia aspirativa.

Descritores: Laringe/cirurgia; Traqueia/cirurgia; Pneumonia aspirativa; Doenças do sistema nervoso; Criança

INTRODUCTION

Chronic pulmonary aspiration is frequently observed in children with chronic non-progressive encephalopathy. It has high morbidity and mortality rates, and results in increased need for tracheal aspiration, repeated admissions, high cost with medical care, and decreased quality of life of patients and their parents.[1,2]

The ideal surgery to treat chronic aspiration, especially in neuropathic children, should completely avoid aspiration, be a single procedure, safely performed in small structures, cause no damage or scars that would avoid larynx growth and integrity, allow phonation and be reversible. Considering all these requirements, the technique preferred by many physicians and used in children, is the laryngotracheal separation (LTS), first described by Lindeman, in 1975, and modified in 1976.[3-5]

The technique consists of producing a blind pouch closing the proximal trachea and maturation of the trachea distal to the skin, creating an ample tracheostomy, as shown in figure 1.[5,6]

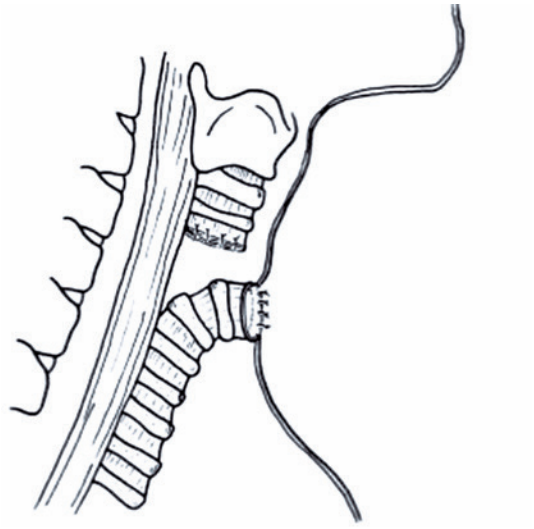


Figure 1. Lindeman surgery, modified in 1976

The most significant morbidity of this procedure is the loss of phonation, but it is more indicated in children with neurologic abnormalities, in whom phonation is no longer an option. Some patients communicate with their caregiver by making shrill sounds, and this may hinder authorization by the caregiver, since communication can be interrupted. Thus, it is important that the surgery details be fully explained to the family.[3]

OBJECTIVE

To evaluate the clinical stability of the neuropathic child and adolescent with episodes of repeated pneumonia submitted to the surgical procedure of laryngotracheal separation.

METHODS

A retrospective study conducted by analysis of medical records of 92 children with median age of 68.5 months, varying from 2 months to 17.8 years, submitted to LTS between October 2002 and June 2015. All children presented with delayed development, and the most common causes were perinatal asphyxia (59.8%), West syndrome (9.8%), laryngotracheomalacia (3.3%), microcephalus (3.3%), mitochondriopathy (2.2%), Dandy-Walker syndrome (2.2%), and posterior fossa tumor in 2.2%. No patient presented with verbal communication. All patients had had prior hospitalization due to aspiration pneumonia (> 5 episodes), and these data was one of the inclusion criteria in this study.

Modified Lindeman (1976) was the surgical technique used. An anterior cross-sectional incision was made on the skin, 2cm above the sternal furculum. The adipose tissue around the incision was removed, and the trachea was exposed. The trachea was then divided between the second and third tracheal cartilage. The proximal trachea was sutured with Vicryl 2.0 and covered with a muscular flap, while the distal extremity of the trachea was sutured to the skin, creating an ample tracheostomy, which is maintained with a tracheal cannula (Figure 2).

The data collected were feeding route, postoperative complications, pneumonia episodes in the postoperative period, and death. The data were evaluated, statistically analyzed by Student's *t* test and Pearson's χ^2 test, with significance level of 95%. This project was approved that the Ethics Committee of *Hospital Pequeno Príncipe*, opinion number 1.549.548, CAAE: 47646815.9.0000.0097.

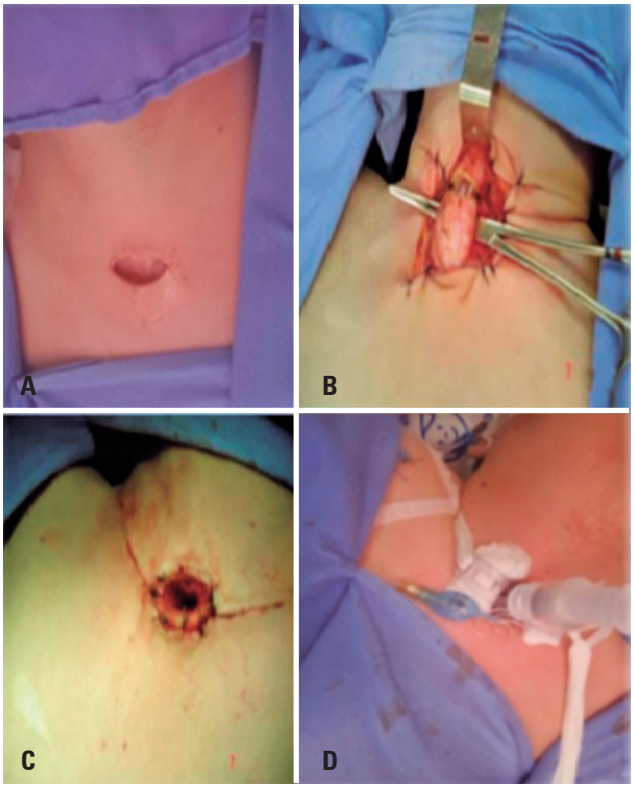


Figure 2. Intraoperative view of laryngotracheal separation. (A) Incision 2 cm above the sternal furculum with removal of adipose tissue. (B) Trachea is isolated and sectioned at the second tracheal ring, with suture of the proximal portion. (C) Trachea matured distal to the skin. (D) Final aspect with the tracheostomy cannula

RESULTS

Among the 92 children, 53 were male (57.6%). The clinical characteristics prior to LTS are displayed on table 1.

Postoperative complications occurred in 13 children (14.1%), three of them presented with more than one complication, as per table 2.

Four patients presented with bleeding through the tracheal cannula, and two died because of a voluminous bleed, probably associated with the presence of a fistula between the trachea and the innominate artery. Among the patients who presented with a tracheocutaneous fistula, only one had tracheostomy prior to LTS.

Twenty-four episodes of pneumonia were noted during the postoperative period (26.1%), and some children presented with repeated pneumonia: 1 episode (29.2%), 2 episodes (33.3%), and 3 episodes (25.0%), 4 episodes in 1 patient, 7 episodes in um patient, and 8 episodes in 1 patient. Children with more than 4 pneumonia episodes had a confirmed diagnosis of bronchodysplasia. There was a significant decrease in the occurrence of pneumonias after surgery (100% *versus* 26.1%; $p<0.001$), as shown on figure 3.

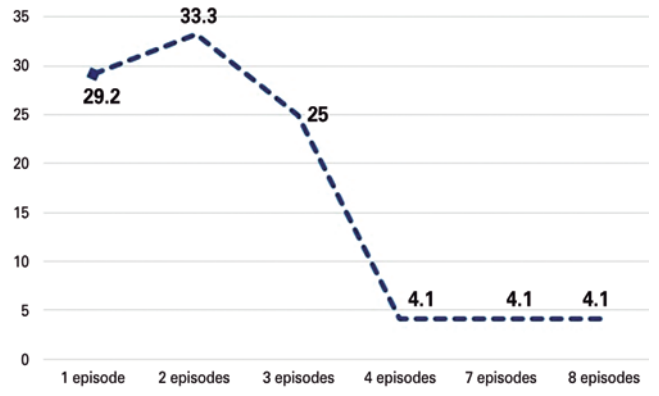
Table 1. Characteristics of children submitted to separation laryngotracheal

Characteristics	n (%)
Admission to ICU due to pneumonia	46 (50.0)
Need of mechanical ventilation	39 (42.4)
Previous tracheostomy	16 (17.4)
Feeding route	
Oral and Gastrostomy	4 (4.3)
Gastrostomy	83 (90.2)
Oral	2 (2.2)
Nasogastric tube	3 (3.3)
Gastrostomy	
Before LTS	63 (72.4)
Simultaneous with LTS	24 (27.6)

ICU: intensive care unit; LTS: separation laryngotracheal.

Table 2. Postoperative complications

Complications	n (%)
Fistula	5 (5.4)
Bleeding	4 (4.3)
Stenosis	3 (3.2)
Granuloma	2 (2.2)
Others	2 (2.2)



p<0.001.

Figure 3. Reduced episodes of pneumonia after separation laryngotracheal

Death was recorded in 23 patients (25%), 11 of them dependent on mechanical ventilation, who experienced respiratory failure (47.8%); 4 cases due to sepsis (17.4%), 5 patients with complications of the underlying disease (21.7%), 1 child due to subdural empyema (4.3%), and 2 patients because of pulmonary hemorrhage (8.7%) due to a probable tracheoinnominate fistula. Death occurred, as a median, at 2.5 years, varying from 6 months to 18 years (95% confidence interval – 95%IC: 8-128).

The occurrence of postoperative complications was similar among the patients who evolved, or not,

to death (16.7% *versus* 13.2%; p=0.73); however, the most severe complications, such as bleeding, occurred in patients who evolved to death.

DISCUSSION

Aspiration pneumonia, mostly due to aspiration of the individual´s own saliva, is considered the most frequent cause of recurring pneumonias in the pediatric population, and is responsible for 8% of children hospitalized with pneumonia. Chronic aspiration is a long-term problem in many children with neurologic deficiency. These patients generally experience recurring episodes of aspiration, which can be extremely debilitating.[2]

Several surgical procedures have been used to prevent aspiration, including total laryngectomy, closing of the glottis, tracheostomy, and Teflon injection into the vocal folds. Although each procedure can reach a good result, these operations may be incomplete to avoid aspiration and/or because there is no possibility of reversal.[6]

Described by Lindeman[4] and Yarington et al.,[5] LTS is an ideal procedure for recurring chronic aspiration. The main disadvantage is the loss of phonation, but there are reports of successful placement of prostheses. With the addition of the valve, the patients can experience phonation and adequately communicate. In addition, the prosthesis also can be used to drain the accumulation of secretions in the proximal tracheal pouch. Laryngotracheal separation completely preserves the integrity of the larynx, and is, therefore, more easily reversible.[2]

In the study by Gelfand et al., as well as in our study, all patients present neurological damage, with no hope of recovery, and therefore, are no longer candidates to reversal. Gelfand et al., reported two deaths during postoperative follow-up. As in other studies, death resulted from progression of their underlying disease, and not as a direct complication of surgery. Various patients required maintenance of the tracheostomy cannula secondary to their dependence on ventilation. The rate of death in this study was 25%, but only in 2 cases they were related to the surgical procedure of LTS, *i.e.*, the tracheoinnominate fistula, which is one of the most feared complications, leading to abundant bleeding within a short interval of time, and in most cases, and irreparable complication.[2]

The major risk factors that contribute towards the formation of the tracheoinnominate fistula are low tracheostomy, cannula cuff overly insufflated, and chest deformities, such as scoliosis. The last factor, scoliosis, is a characteristic finding in children with severe physical and mental deficiencies, resulting in a trachea that is closer to the sternum, compressing the innominate artery.[7]

If a tracheoinnominate fistula occurs, the first approach should be to insufflate the endotracheal cannula balloon to compress the fistula and control bleeding. If bleeding is controlled, the separation from the brachiocephalic trunk and/or endovascular embolization of the innominate artery should be done as quickly as possible.[7] The 2 patients seen at our service died during pre-hospitalization care, and there was no possibility of surgical intervention.

Chida et al.,[1] showed that surgery significantly decreases the number of hospital admissions for aspiration pneumonia in patients treated at home. This result indicates that these procedures are effective in preventing aspiration, decreasing morbidity in these patients. It was reported that, after LTS in neuropathic children, the satisfaction of their parents was better, and there was improved quality of life for the patients, especially considering reduced hospitalization and the need for care at home. Takamizawa et al.,[6] reported 11 patients submitted to LTS, aged 9 months to 16 years, and only 1 patient presented with postoperative pneumonia. All parents evaluated LTS as excellent or good in terms of improved quality of life. Gelfand et al.,[2] performed 12 LTS in neuropathic patients aged 3 to 12 years, with an important reduction in admissions to hospital due to aspiration pneumonia (5.4 *versus* 1.1; p<0.001). There were no major complications, except for 2 cases of wound dehiscence, one of tracheitis, and 2 of tracheal stenosis. All parents/caregivers agreed that LTS led to general flexibilization of care for these patients, primarily because there were smaller pulmonary complications. Hara et al.,[3] assessed 21 patients submitted to LTS who presented with a significant reduction in episodes of pneumonia (3.38 *versus* 0.52; p<0.01) and in need of aspiration (4.0 *versus* 0.84; p<0.01), in which 17 patients had no episodes of pneumonia up to 6 months after surgery.[1,2,6]

Although in literature regarding adults, the rate of formation of tracheocutaneous fistula varies from 16.6% to 38.2%, the procedure seems to be more secure in the pediatric population. In a study of 23 procedures published by Manrique et al., the rate of tracheocutaneous fistula was 17.4%; Hara et al., presented 3 cases of fistula of the proximal stump (14.3%); Chida et al., 20%; Eisele et al., 17.6%; Eibling et al., 17.6%; Yamana et al., 38%; Zocratto et al., 22%; Cook, 23%. Takamizawa et al., and Gelfand et al., presented no cases of fistula in their work. Our study reported a low rate of fistulas (5.4%), and all were treated by conservative approach.[1-3,6,8-13]

CONCLUSION

When well indicated, surgery reduces the number of pulmonary infections and of hospital admisisons, leading to better quality of life of these patients. Chronic broncoaspiration increases mortality of these patients, since they are already debilitated by their underlying disease. Thus, laryngotracheal separation should be indicated as a primary procedure in patients with chronic non-progressive encephalopathy and repeated episodes of aspiration pneumonia.

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