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Management of laryngeal cleft in mechanically ventilated children with severe comorbidities☆

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ABSTRACT

Laryngeal clefts are rare congenital malformations of the posterior part of the larynx. The severities are correlated with the downward extension of the cleft and can involve numerous clinical symptoms including dysphagia and respiratory distress. As significant comorbidities may be present, individual treatments depend on the child's general condition and type of cleft involved. Herein, we describe two cases of children with laryngeal clefts and severe comorbidities requiring mechanical ventilation. One child with type III laryngeal cleft was successfully managed with the lateral pharyngotomy approach. The other child with type II laryngeal cleft has not been able to undergo cleft-closure surgery because of severe general conditions, therefore has continued training for feeding and swallowing.

1. Introduction

Laryngotracheoesophageal clefts (laryngeal clefts) are rare congenital anomalies and result from fusion failure of the tracheoesophageal septum or the posterior cricoid cartilage during embryological development. Some cases are associated with congenital anomalies, while most are sporadic [1].

Clefts range from deep interarytenoid notches to those extending below the vocal cords, through the cricoid and into the trachea [1–3]. The Benjamin and Inglis classification system [3] is most commonly used, and cleft grade (type I–IV) correlates with the severity of symptoms. Patients with laryngeal clefts commonly present with airway and/or swallowing impairment [1–4]. These symptoms are influenced both by the depth of the cleft and by the underlying medical status of the patient.

The timing of the diagnosis of laryngeal clefts is crucial to successful treatment, which requires diverse surgical approaches for different types of clefts [5,6]. However, in children with severe comorbidities, such as severe congenital anomalies or severe motor and intellectual disabilities, immediate surgical treatment is often not undertaken due to patient poor physical status. Herein, we report two cases of children with laryngeal clefts and severe comorbidities who were dependent on mechanical ventilation.

2. Case reports

2.1. Case 1

This boy was born at a gestational age of 34 weeks and weighed 2154 g at birth. He had multiple congenital conditions

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including cardiac, respiratory, and digestive system anomalies. Immediately after birth, a tracheal tube was inserted for artificial respiratory management due to severe respiratory distress. Tracheostomy was performed at the age of 4 months. As he had experienced recurrent aspiration pneumonia and hypersecretion from the airway since infancy, he was referred to the Department of Otolaryngology for evaluation of swallowing function at the age of 10 months. We conducted laryngoscopic evaluation under general anesthesia, and laryngeal cleft, vocal cord immobility on the left side, and saliva aspiration to the trachea were detected. We initially employed a conservative treatment for laryngeal cleft management because treatments for multiple congenital anomalies were deemed to be of higher priority. At the age of 3 years, he was discharged with medical care, but motor and intellectual disabilities remained. At 5 years of age, we decided to perform surgical treatment for laryngeal cleft, after consideration of his general condition. Preoperative videofluoroscopic examination of swallowing (VF) revealed aspiration of contrast agent from the laryngeal cleft into the trachea (Fig. 1A). We performed a detailed laryngoscopic assessment under general anesthesia to examine the depth of the cleft and the condition of the surrounding tissue. The examination revealed a type III laryngeal cleft extending below the cricoid cartilage and thickening mucosa herniating through the posterior cleft into the laryngeal lumen (Fig. 1B, C). As the herniated mucosal tissue extended beyond the cricoid cartilage, we surmised that cleft closure via a transoral approach would be challenging. Thus, we decided to utilize a transcervical approach.

2.1.1. Surgical procedure: lateral approach with lateral pharyngotomy

Surgery was performed under general anesthesia (Fig. 2). The skin was incised along the anterior border of the left sternocleidomastoid muscle (Fig. 2A). The strap and omohyoid muscles were divided along the posterior border of the thyroid cartilage. The inferior pharyngeal constrictor muscle was incised along the posterior border of the thyroid cartilage to the lower border of the thyroid cartilage (Fig. 2B). After identifying the pharyngeal wall, it was sectioned to open the pharyngeal cavity (Fig. 2C). The laryngeal structure on the left side was retracted to the anterior right side. The laryngeal cleft and the herniating mucosal tissue were visualized after retraction. Excessive mucosal tissue overlying the cleft was dissected along the border of the cleft (Fig. 2D). After exposing the whole cleft, it was closed in two layers under direct vision. First, tracheolaryngeal closure was performed in the inner side. Sutures were tied to obtain meticulous mucosal contact with the cut surfaces facing inward (Fig. 2E). A second layer of suturing was placed for the pharyngoesophageal repair (Fig. 2F). With this approach, mucosal suturing could be performed easily without anatomical distortion of the laryngeal framework.

VF 1 month after the operation revealed no penetration or aspiration upon thickened fluid swallowing (Fig. 3A) but silent aspiration upon liquid swallowing was present. In addition, due to a lack of oral feeding experience over a prolonged period, the patient's oral and oropharyngeal swallowing remained underdeveloped. Given these results, swallowing exercises using thickened liquids were initiated. He also practiced oral motor

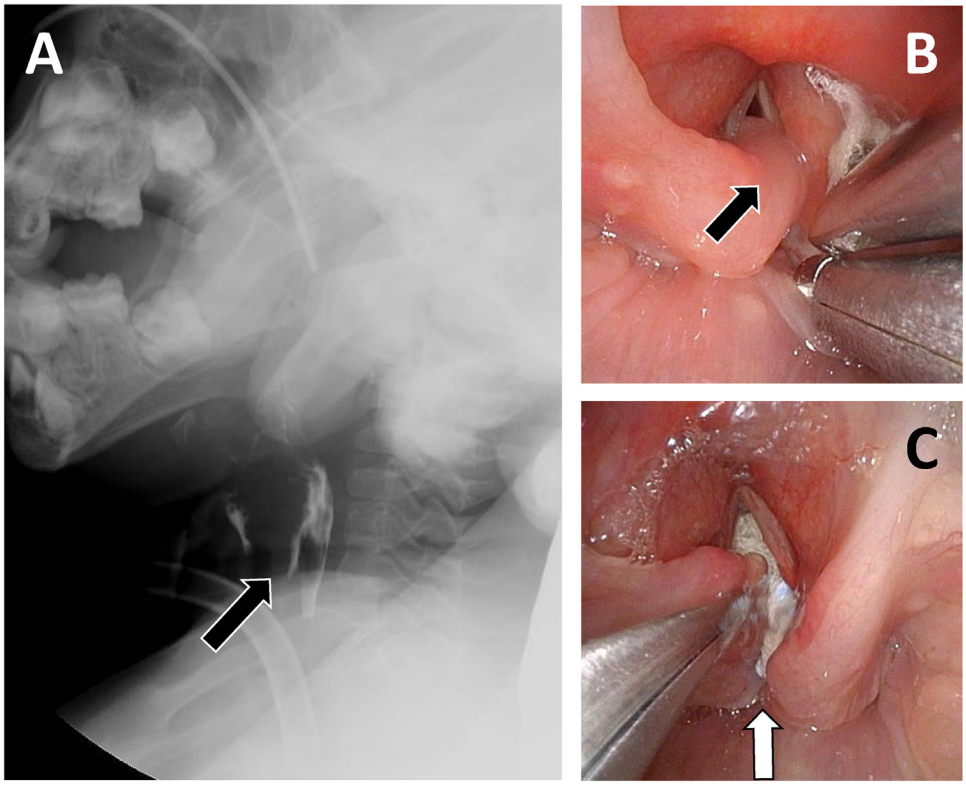

Fig. 1. (A) The first videofluoroscopic examination of swallowing in case 1 showed aspiration of contrast agent from the laryngeal cleft into the trachea (black arrow). (B, C) Laryngoscopic examination views of the type III laryngeal cleft in case 1. (B) Thickening mucosa herniating through the posterior cleft into the laryngeal lumen (black arrow). (C) The cleft extending below the cricoid cartilage (white arrow).

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