Failed Intubation Secondary to Congenital Subglottic Stenozis with Vacterl Association

Eurasian Clinical and Analytical Medicine Case Report

Failed Intubation in a Patient with Congenital Subglottic Stenosis

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Abstract
Congenital subglottic stenosis (CSS) is the third most common laryngeal anomaly. Difficult tracheal intubation in anesthesized patients is estimated to be 1-3%. Failed intubation rate is approximately 0.05-0.2%. We report a case of failed intubation in 2 day-old newborn with CSS and multiple congenital anomalies. Two day old male newborn was referred to our center due to anal atresia and associated anomalies. Facomelia, finger anomalia, subglottic stenosis, hydronephrosis, cardiac, anal atresia and sacral defect were present in our patient and these anomalies were matched with VACTERL association. We report a case of a failed intubation secondary to CSS and the successful use of LMA during failed intubation and tracheostomy for CSS patients. We thought that VACTERL association with the presence of congenital subglottic stenosis is a condition which is rare and LMA to be effective on failed intubation.

Keywords
Congenital Subglottic Stenosis; Difficult Intubation; LMA

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Introduction

Congenital subglottic stenosis (CSS) is the third most common laryngeal anomaly. The typical patient with CSS is a newborn without a history of endotracheal intubation or trauma who has a subglottic airway lumen of less than 3.5 mm diamete r[1]. Depending on its severity it may present with simple stridor or with near-death episodes requiring resuscitation at home, or worse, inability to ventilate[2]. Difficult tracheal intubation in anesthesized patients is estimated to be 1-3%. Failed intubation rate is approximately 0.05-0.2% [3]. We report a case of failed intubation in 2 day-old newborn with CSS and multiple congenital anomalies.

Case Report

Two day old male newborn was referred to our center due to anal atresia and associated anomalies. He was born at 40 weeks 5 days gestational age (height 50 cm, weight 3.21 kg and APGAR score 7/8). Preoperative vital signs included BP 80/45 mmHg, heart rate of 154 b/min) and regular respiratory rate of 54 and temperature of 36.1°C. The physical examination showed left facomelia, right finger anomaly, bifid scrotum, high level anal atresia with flat bottom deformity. Babygram showed sacral defect and costal defect and ultrasonography showed mild right hydronephrosis. Transthoracic echocardiography revealed a patent foramen ovale (PFO) and anomalies of pulmonary venous return. A systolic ejection murmur of 2-3/6 was audible at the upper sternal border with no associated thrill or gallop. He was breathing spontaneously with equal breath sounds and retractions and stridor were noted. Pulse oximetry revealed an oxygen saturation of 91% in room air. A 24-gauge intravenous access line had been placed in her left foot. A sigmoid colostomy was planned for high level anal atresia. In the operating room standard monitors were placed including an electrocardiogram monitor, noninvasive blood pressure monitor and pulse oximetry. Anesthesia was induced using mask sevoflurane until an adequate depth of anesthesia. Direct laryngoscopy was performed using a Miller blade 0 and the airway was judged as a Cormack grade I. A size 3 uncuffed tracheal tube with no stylet could not be passed beyond the vocal cords. The tracheal tube was changed to size 2.5 and later to size 2.0 but intubation could not be accomplished. A laryngeal mask airway (LMA) size 1 was inserted and the infant was easily ventilated. We decided to postpone the operation for advanced to examine of failed intubation. An otolaryngologist was consulted and a computerized tomography (CT) scan of the trachea and larynx performed. Stenosis was seen just below the vocal cords with the narrowest lumen of 1,9 mm (Figure1). The decision was made to tracheostomy. One day later anesthesia could be induced using mask sevoflurane and LMA 1 was inserted. Tracheostomy was performed between 2-3 intercartilagenous tracheal rings, three cuffed tracheostomy canulla inserted and LMA was removed. Following completion of the cholostomy, rigid direct laryngoscopy was performed via a 2.5 mm endoscope. Stenosis were observed in the subglottic portion of the trachea. The 2.5 mm endoscope probe could not be passed trachea because of the severe subglottic narrowing (Figure 2). He was awake and breating spontaneously. Retractions and stridor were improve. He was transported to neonatal intensive care with spontaneously breathing. Postoperative mechanical ventilation carried on and cardiopulmonary arrest evolved on postoperative 4th day.

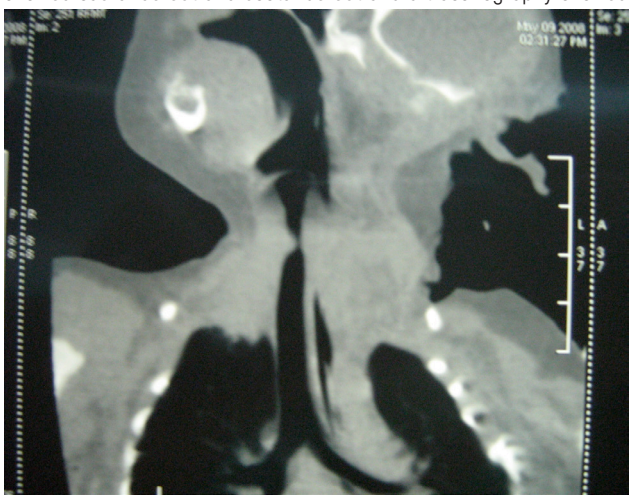

Figure 1. Computed tomography view of subglottic stenosis

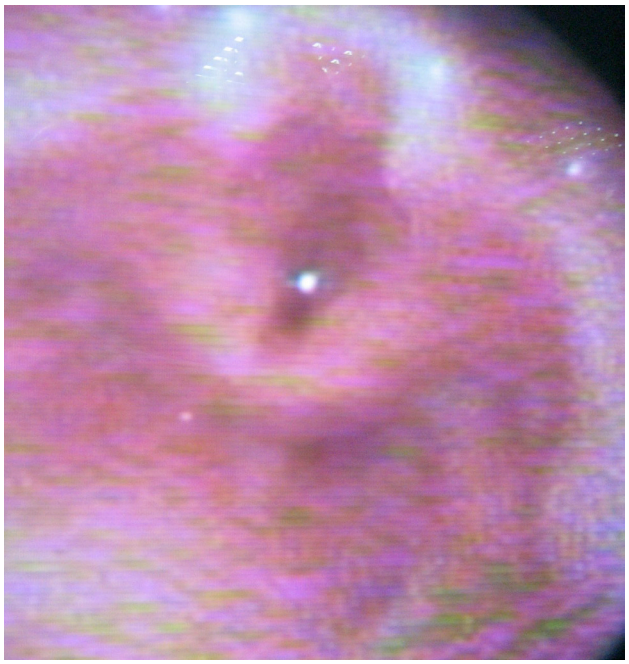

Figure 2. Endoscopic viev of subglottic stenosis

Discussion

The VACTERL association is a group of congenital malformations including vertebral, anal, cardiac, tracheoesophageal, renal and limb abnormalities which originate at this point from unknown mechanisms about the sixth week of gestastion [4]. Facomelia, finger anomalia, subglottic stenosis, hydronephrosis, cardiac, anal atresia and sacral defect were present in our patient and these anomalies were matched with VACTERL association.

CSS are believed to be the results of failure or incomplete recanalization of the laryngeal lumen by the 10th week of gestation [1]. CSS is considered to exist when the lumen of the cricoid region of the airway measure less than 4 mm in a full-term infant or 3 mm in a premature infant with no previous history of intubation [1]. The Cotton-Myer grading system is most widely used criteria for evalution of CSS (TABLE 1), [5], however endotracheal tube sizing is among most widely used means of grading and assesing the degree of stenosis [1]. This newborn was classified as grade III CSS. The diagnosis of CSS suggested CT with contrast may delineate associated intrathoracic anomalies such

Table 1. Cotton- Myer subglottic stenosis grading system [5].

Grade I	0% to 50 % obstruction
Grade II	51% to 75 % obstruction
Grade III	71 % to 99% obstruction
Grade IV	No detectable lumen

as pulmonary artery sling which can occur in up to one-third of the patients. Bronchoscopy is the best method to determine the extent of the tracheal stenosis [3]. However we used rigit endoscope to determine subglottic stenosis.

We report a case of a failed intubation secondary to CSS and the successful use of LMA during failed intubation and tracheostomy for CSS patients. We thought that VACTERL association with the presence of congenital subglottic stenosis is a condition which is rare and LMA to be effective on failed intubation.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

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Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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