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Case Report

Airway management for rigid bronchoscopy via a freshly performed tracheostomy in a child with Goldenhar syndrome

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Abstract A case of tooth aspiration in a 6 year old boy with Goldenhar syndrome and known difficult intubation is presented. A fresh tracheostomy was performed after a failed fiberoptic intubation and dental aspiration. The patient was transferred to our tertiary-care children's hospital for emergency bronchoscopy through the fresh tracheostomy for removal of an aspirated tooth. Rigid bronchoscopy performed via a fresh tracheostomy presents several challenges. The major complications associated with bronchoscopy performed via a fresh tracheostomy, as well as management of airway emergencies are discussed.

1. Introduction

Aspiration of teeth during induction and intubation is a recognized anesthetic risk, especially in pediatric patients between the ages of 6 and 12 years. A case of aspiration of a tooth in a 6 year old boy with Goldenhar syndrome and known difficult airway is presented. Following a freshly performed tracheostomy at an outside hospital, the patient was transferred to our tertiary-care center for retrieval of an aspirated tooth. The issues regarding airway management during rigid bronchoscopy through a recently performed tracheostomy, as well as possible related complications, are presented.

2. Case report

A 6 year old Caucasian boy was scheduled for elective placement of tissue expanders for correction of left ear

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microtia. His past medical history included a diagnosis of Goldenhar syndrome with left hemifacial microsomia and left microtia. He had significant limitations of mouth opening combined with fusion between skull and C_1 vertebrae and C_2-C_3 vertebrae. Also he had a bicuspid aortic valve, mild mitral stenosis, and obstructive sleep apnea. Two loose teeth were seen during preoperative assessment. Past surgical history included multiple plastic surgical procedures for which his airway was managed by elective fiberoptic intubations during general anesthesia without incident.

At the outside hospital, patient had an easy induction with easy mask ventilation. During elective fiberoptic intubation to secure the airway, both loose upper incisors were dislodged. The anesthesiologist immediately retrieved the first tooth from the patient's oropharynx. However, the second tooth was not found. Subsequently, the ear, nose, and throat (ENT) surgeon performed an urgent elective tracheostomy. A chest radiograph performed in the operating room (OR; Fig. 1) showed the second tooth to be located in the right lower lobe bronchus.

The patient was transferred to Children's Hospital of Michigan for further management. He arrived 5 hours after the tracheostomy in stable condition, breathing spontaneously, and sedated with a 200 μg/kg/min propofol infusion. Routine ASA monitors were applied. After the tracheostomy stoma was connected to the breathing circuit, correct tracheostomy tube location was confirmed by end-tidal CO_2 ($ETCO_2$). Sevoflurane was introduced in addition to the propofol infusion; inspired oxygen concentration was maintained at 100%. In a discussion with the ENT and the pediatric surgical services, the decision was made to perform a rigid bronchoscopy initially via the fresh tracheostomy site to retrieve the tooth. The tracheostomy tube was removed by the ENT service and a size 3.5 mm rigid bronchoscope was introduced via the fresh stoma, but the tooth was not found. The bronchoscope was then removed and the tracheal tissues were manually spread apart to reinsert the tracheostomy tube. There were no stay sutures in the tracheostomy site. As the tracheostomy tube was being repositioned, anatomical structures were unidentifiable for a brief period. A tracheostomy set was available but within a few seconds the lumen of the trachea was visualized again and the tracheostomy tube was placed correctly with reappearance of the $ETCO_2$ on capnogram and confirmation of breath sounds by auscultation. Stay sutures were then placed in the tracheostomy at that time.

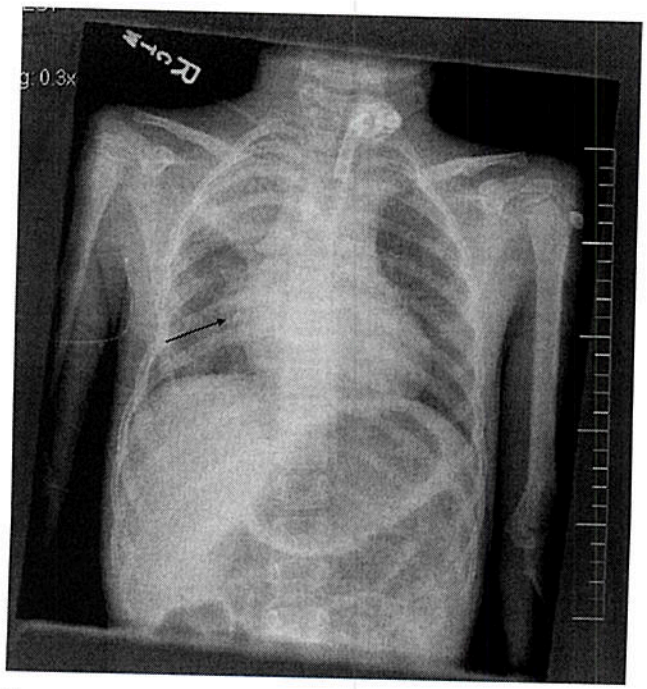

Fig. 1 Chest radiograph of the patient showing the tooth (arrow) dislodged in the right lower distal lobar bronchus.

A broad sweep of the upper airway with the direct laryngoscope, per the ENT service, was negative for any foreign body. Another chest radiograph obtained in the OR showed the foreign body still located in the right lower distal bronchus (similar location to what was seen on chest radiograph from the outside hospital). A smaller size 3.0 mm rigid ventilating bronchoscope was inserted through the tracheostomy stoma. At this time, the missing tooth was noted to be deeply impacted distally in the right lower lobe bronchus. There was minimal mucosal swelling proximal to the tooth but no bleeding was noted. A 3.5 mm rigid bronchoscope was then reinserted through the fresh tracheotomy stoma after location of the aspirated tooth was verified by the size 3.0 bronchoscope. Several attempts were made to retrieve the tooth, including optical forceps, with peanut grasper, small cup grasper, alligator graspers, three Fogarty catheters, and a urethral basket. An attempt was made to dislodge the tooth by placing the patient in the right lateral position and performing chest percussions. Finally, with small alligator optical forceps the surgeon grasped the tooth past the segmental juncture where it was lodged. The foreign body and bronchoscope were then removed from the trachea in one motion. The bronchoscope was reinserted to check for any residual foreign bodies. The tracheostomy tube was reinserted and secured. The patient tolerated the procedure well, spontaneously breathing throughout the procedure with an $ETCO_2$ between 53-56 mmHg and respiratory rate of 25 to 28 breaths/min. There were no episodes of desaturation during the case and vital signs were normal at all times. Muscle relaxation was considered initially, but the small size of the bronchoscope and the fact that the patient was tolerating the procedure prevented us from using muscle relaxants.

At the end of the case, the patient was taken to the pediatric intensive care unit in satisfactory condition. The duration of the procedure was approximately three hours. Postoperative chest radiography showed no foreign body.

The postoperative course was uneventful. The patient was transferred out of the pediatric intensive care unit on postoperative day (POD) 2. The patient's tracheostomy

tube was decreased in size to 3.0 mm and he was eventually decannulated on POD 5. The patient was discharged home on POD 6 in stable condition.

3. Discussion

Goldenhar syndrome is a spectrum of oculoauriculovertebral anomalies. It is a rare syndrome with an incidence of 1/5,000, with male predominance [1]. It is associated with difficult intubation in 40% of cases [2]. Difficult intubation has been attributed to a combination of hemifacial microsomia, micrognathia, mandibular hypoplasia, vertebral anomalies, and craniofacial anomalies [3,4]. Difficult mask ventilation has been reported in Goldenhar syndrome secondary to soft-tissue cleft extending from the lateral aspect of the mouth to the mid-cheek on the affected side. Several case reports have described techniques for intubation in children with Goldenhar syndrome, including the Laryngeal Mask Airway, fiberoptic bronchoscopy (FOB), and tracheostomy with retrograde intubation [5-7].

Tracheostomy is required in approximately 22% of children with Goldenhar syndrome for upper airway obstruction [8]. In our case, tracheostomy was done to secure the airway after failed intubation with FOB secondary to loss of two teeth. In a review of bronchoscopy-related complications in children, two cases of bronchoscopy were performed through a fresh tracheostomy, during which both patients developed pneumothorax followed by death. Unfortunately, the authors did not describe the anesthesia management for the two patients [9].

Rigid bronchoscopy performed via an established stoma versus a fresh stoma requires different considerations. A tracheostomy takes approximately 10 days to mature and a stoma to be established. A fresh tracheostomy does not have healed margins or well-demarcated structures for safe manipulation. Insertion of instruments into a fresh friable tracheostomy site may contribute to life-threatening complications, including pneumothorax, subcutaneous emphysema, hemorrhage, creation of false passages, and

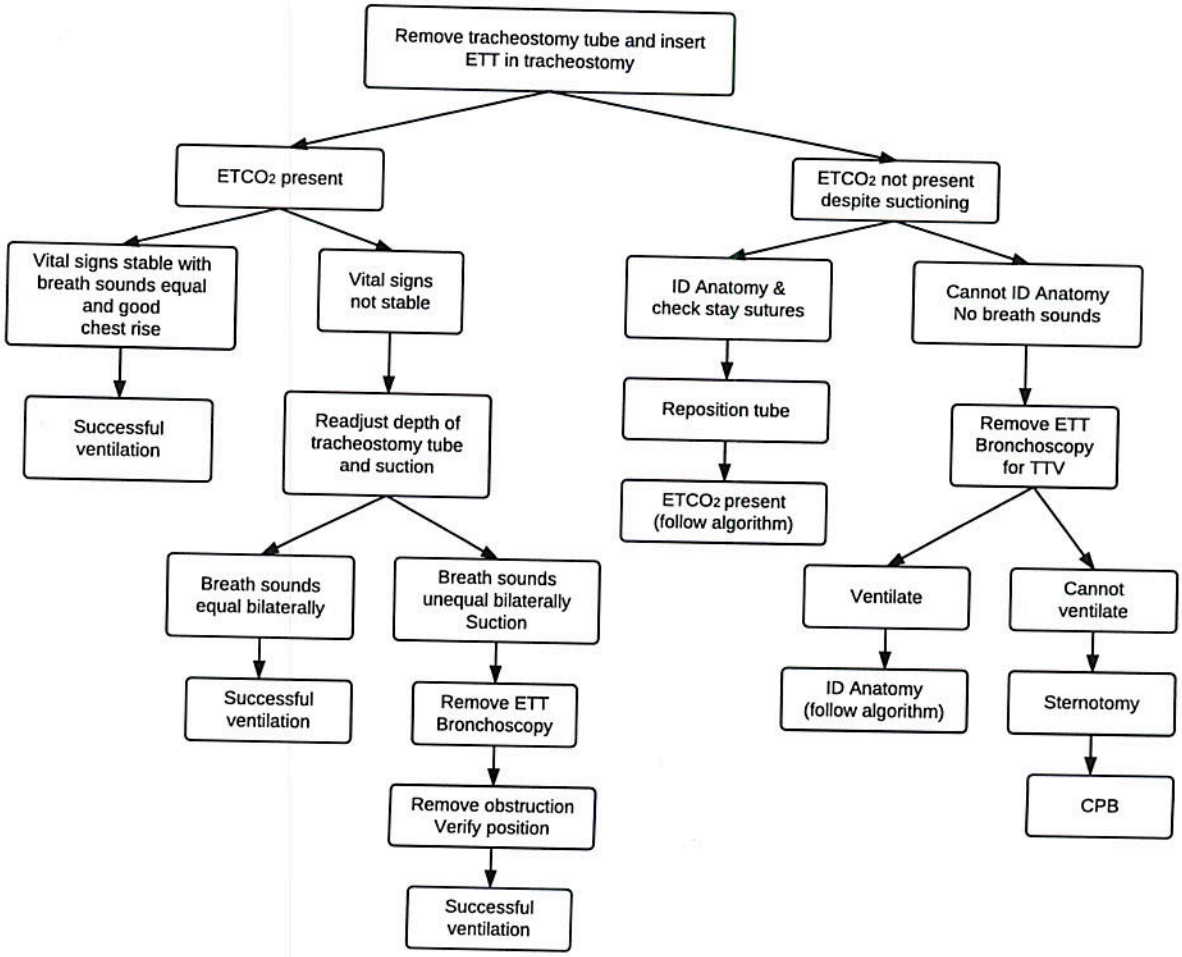

Fig. 2 Suggested algorithm for loss of airway in fresh tracheostomy. ETT=endotracheal tube, $ETCO_2$=end-tidal carbon dioxide, ID=identify, TTV=transtracheal ventilation, CPB=cardiopulmonary bypass.

pneumomediastinum (Fig. 2). An organized plan should be outlined by the surgeon and the anesthesiologist regarding anticipation of the complications its management. Special attention should be paid to early detection and treatment of pneumothorax. Intraoperatively, close monitoring of $ETCO_2$, oxygen saturation, blood pressure, and heart rate are essential. Continuous auscultation of breath sounds by precordial stethoscope, inspection of the lungs for symmetrical excursion, and continuous assessment of airway compliance are of utmost importance. In case of suspected pneumothorax, a portable chest radiograph and chest tube should be available in the OR for prompt diagnosis and treatment. The anesthesiologist may also palpate the neck and shoulders for possible subcutaneous emphysema. Postoperatively, the patient should be monitored closely and chest radiography should be followed.

Several measures are described in the literature regarding management of loss of airway or false passage of the tracheostomy. These include insertion of a Cook exchange catheter through the tracheostomy and keeping it as a rescue airway in case of false passage and airway loss [10], insertion of a jet ventilation-airway exchange (JVAE) catheter through an endotracheal tube (ETT) prior to incision and keeping the JVAE distal to the tracheostomy. The latter may serve as a stylet for reintubation and may also be connected to a jet ventilation device for oxygenation of the lungs [11]. Since our patient was known to be difficult to intubate with a fresh tracheostomy, our plan was careful manipulation of the fresh stoma by direct visualization. If all of these measures fail to provide adequate ventilation and oxygenation, there are several invasive options. These include insertion of a large-bore intravenous catheter into the tracheal lumen in the sternal notch distal to the tracheal opening and connecting it to a jet ventilation device, and finally sternotomy, followed by carinal transtracheal jet ventilation. If oxygen is not delivered into the lungs, emergency cardiopulmonary bypass may be indicated as the last resort [11]. Finally, we suggest placing stay sutures in the tracheal cartilage laterally to prevent loss of the stoma and provide better visualization of the tracheal lumen at the stoma site during the bronchoscopy. Another alternative is to perform a starplasty tracheotomy to create a permanent and secure tracheotomy stoma, but this technique is more time consuming.

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