Case Report

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Sevoflurane as the Single Anesthetic Agent for Management of Anticipated Pediatric Difficult Airway

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Mucopolysaccharidoses (MPSs) are a group of hereditary diseases characterized by accumulation of Glycosaminoglycans (GAGs) due to deficiency or malfunctioning of lysosomal enzymes. Difficult intubation is anticipated in these patients because of a variety of anatomical and functional abnormalities. We report the successful use of sevoflurane and laryngeal mask for airway management of a 15 year-old boy requiring corneal transplantation surgery.

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INTRODUCTION

Mucopolysaccharidoses (MPSs) are a group of hereditary diseases caused by impaired breakdown of glycosaminoglycans (GAGs). This may result from either a deficiency or malfunctioning of lysosomal enzymes leading to accumulation of GAGs in the connective tissue, particularly in bone, brain, liver, blood vessels, skin, cartilage, airways, heart valves, and corneas (1).

Currently, there are seven types of MPSs categorized by clinical and laboratory findings (2). The different types of the disease share many clinical features but differ in severity. Usually, the patients appear normal at birth but symptoms begin to manifest as GAGs accumulate in the tissues.

The airways are not spared in the disease process and multiple anatomic changes are present. Short neck, rigid mandible and obstruction of the airways by the soft tissue of the pharynx and trachea are some changes that herald difficult intubation.

In this report, we present our experience with a teenage boy with MPS type VI (Maroteaux-Lamy Syndrome) whose surgery was deferred several times due to difficult intubation and difficult airway management (DAM).

CASE SUMMARIES

A 15-year-old boy was scheduled for keratoplasty due to severe corneal cloudiness. His procedure was postponed for three times in the past due to inability to maintain a secure airway. The ophthalmic surgeon insisted on general anesthesia and therefore local options were ruled out since the patient was too young to be cooperative.

He was 130cm in height and weighed 22 kilograms. Physical examination revealed short neck, large tongue, small mandible, inability to open mouth completely, a Mallampati score of IV, finger contracture and kyphoscoliosis. He had a history of repeated otitis media for which he underwent placement of ventilation tube (VT), and also sleep apnea, gastroesophageal reflux,

hydrocephalus, seizures and spinal abnormalities (corrected by surgery in early childhood) (Figures 1-4).

Laboratory examinations were normal. An echocardiography showed mild mitral valve prolapse (MVP) and regurgitation (MR). Pulmonary function test revealed a forced vital capacity (FVC) of 75% of the predicted value.

The physical examination predicted a difficult intubation and since prior attempts were unsuccessful, even with fiberoptic laryngoscopy, we decided to maintain spontaneous ventilation during anesthesia by a laryngeal mask airway (LMA) and sevoflurane. In the operating room, a 22-gauge intravenous line was established and infusion of 200 milliliters of Ringer solution was started. The patient was monitored with precordial stethoscope, ECG and pulse oximetry. The patient was pretreated with 0.2mg intravenous atropine and sedated with 25μg fentanyl and 0.3mg midazolam. Since the patient was a case of DAM we decided not to use an intravenous anesthetic that might compromise the airways. Therefore, anesthesia was induced by a Mapleson F system with inhalation of 50% N_2O/O_2 and very low concentration of sevoflurane. Sevoflurane was started at 0.5% concentration and increased by every 3-4 respirations. When anesthesia was deep enough, a size 2.5 LMA was introduced without use of muscle relaxants. The position of LMA was assessed clinically and its function was checked by assessing the quality of ventilation by pulse oximetry and measurement of airway pressure and bag compliance. Anesthesia was maintained by inhalation of sevoflurane and during the surgery, the patient received assisted ventilation with a low tidal volume at a rate of 20-25 breaths per minute. The operation proceeded smoothly and lasted 1.5 hours. Alfentanil (100μg) was administered 20 minutes before conclusion of the operation. Near the end of the operation, sevoflurane concentration was decreased gradually so that during the last 5 minutes the patient was breathing 100% O_2. Three to four minutes before the conclusion of the surgery, the LMA was removed on spontaneous respiration while the patient was completely awake. He was transferred to recovery room 7-8 minutes later and received 100% O_2 through a face mask. Two hours later the patient was transferred to the ward without any adverse events. He was visited a few hours later and since no complication was observed, we discharged him the next day.

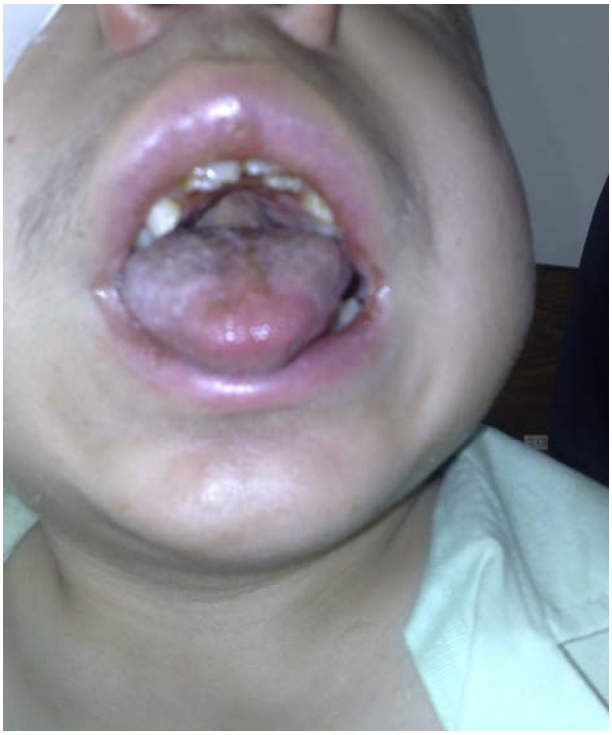

Figure 1. Physical examination revealed short neck and large tongue with small mandible.

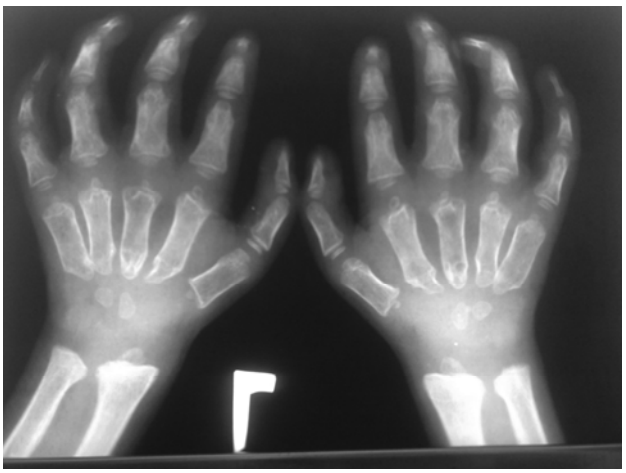

Figure 2. Radiography of patient's wrists.

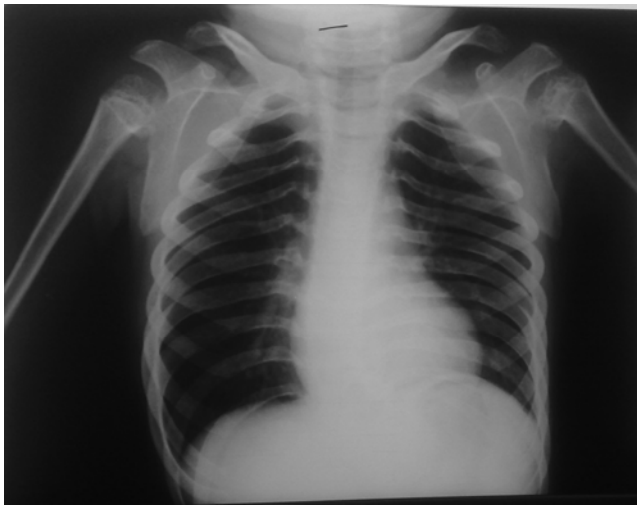

Figure 3. Chest radiography of patient.

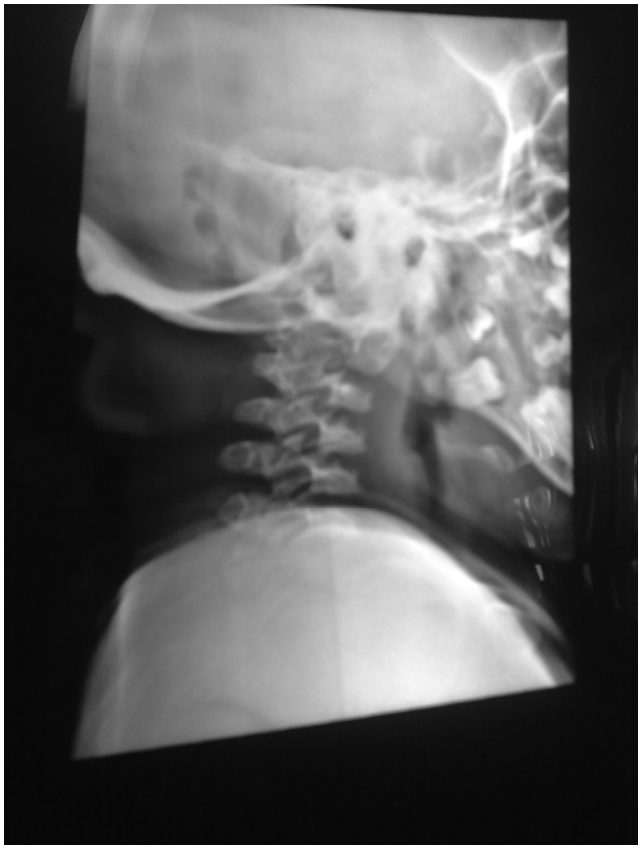

Figure 4. Lateral radiograph of neck.

DISCUSSION

The most important responsibility of an anesthesiologist is to maintain a reliable airway and adequate ventilation. This role becomes even more pronounced in pediatric difficult airway where rapid and correct actions and expert clinical judgment can reduce mortality and morbidity (3). A variety of anatomical and functional abnormalities in MPS turns the intubation of these patients into a challenge even for the most experienced anesthesiologists. In a review by Walker and colleagues on anesthetics in MPS patients, the incidence of difficult intubation and failed intubation was 25% and 8%, respectively. The incidence of difficult intubation was as high as 54% in some subtypes of the disease like Hurler's syndrome (4).

Our patient manifested several features that predicted difficult intubation including large tongue, short neck and stiff temporomandibular joint. Considering these factors and history of failed attempts of endotracheal intubation, we designed a special approach for management of the patient's airway based on the current literature. Most researchers recommend premedication with antimuscarinic agents to dry secretions (5). We used atropine for this purpose.

In MPS patients, the mainstay of the anesthetic technique is to maintain spontaneous ventilation by avoiding muscle relaxants that may compromise ventilation and lead to the dangerous "can't ventilate can't intubate" situation. Inhalation technique, particularly with sevoflurane, is the preferred technique in children since it induces a "smooth and rapid" anesthesia with maximal cardiopulmonary stability (6,7). Additionally, adequate muscle relaxation and fast recovery are possible with sevoflurane (8). Sevoflurane is the drug of choice in patients with cardiac abnormalities and DA because it does not cause bradycardia (like halothane) and does not reduce myocardial contractility. The change from halothane to sevoflurane decreased medication-related preoperative cardiac arrest (POCA) from 37% to 18%(9).

We decided to manage the airway with a laryngeal mask considering failed previous attempts of intubation and prediction of a difficult airway. It is recommended that anesthesiologists dealing with DAM use the method they

are more comfortable with since success is unlikely when using a less practiced method in a critical situation (10). In 2009, Taguchi and colleagues reported their experience in securing an airway using airway scope and an LCD monitor for a patient with Hunter syndrome; however, they reported this method to be difficult (11). The use of LMA has gained popularity for the management of difficult airway in children (12). Its use has been recommended when DA is suspected or a Cormack and Lehane grades 3 and 4 are present (13,14). Walker et al. used LMA in 34 children with craniofacial abnormalities and mucopolysaccharidosis and did not encounter any case of poor airway (15).

Due to the sensitive nature of our patient's operation, it was necessary to provide a smooth emergence. Therefore, we administered alfentanil before termination of the procedure that helped our patient to be transferred to the recovery room on complete consciousness.

CONCLUSION

In summary, we reported the successful anesthetic management of a patient with MPS by sevoflurane and LMA on spontaneous ventilation without unnecessary alteration of the airways.

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