Difficult airway management in an infant with bilateral Tessier number 4 cleft

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Abstract: This case report describes the airway management of a three month old child presenting with a severe facial congenital malformation, a bilateral Tessier number 4 cleft, and scheduled to undergo benign eye surgery. The use of a conventional laryngeal mask allowed successful management, despite the severity of the facial abnormality. This case is then discussed at the light of the literature.

Key words:

Introduction

The incidence of rare craniofacial clefts, which include craniofacial dysraphia and orbitomaxillary and lateral facial clefts, is reported to range between 1.43 and 4.85 per 100,000 births (1). Tessier number 4 clefts are one of the most disfiguring and rare craniofacial clefts. Approximately 50 cases of this defect have been reported in the literature. This pathology is characterized by gross dysmorphism of the face, and a cleft that is either unilateral or bilateral. The cleft involves the orbit, the maxilla and the palate, running through the face. Consequently to these anatomical anomalies, airway manipulation and securing during anesthesia induction may be considerably difficult in these patients. The encountered problems include mask holding for pre-oxygenation and induction of anesthesia, laryngoscopy, and intubation. We present the case of a child presenting to us for an eye surgery under general anesthesia. Consent for the reporting of the case and its description, including publication of pictures, have been obtained from the parents of the child.

Case History

A three month old, male child, weighing 5.5 Kg, and with a diagnosis of bilateral Tessier number 4 cleft was scheduled for left eye lower lid coloboma repair under general anesthesia. The mother’s antenatal history was unremarkable, and the child was born at term through a caesarean section. The past medical history was not of birth asphyxia, and the birth weight was 2.5 kilograms. The child was being fed using artificial top feeds by spoon, and his body weight increase was within normal limits. On examination, the child had gross facial dysmorphism, with bilateral facial clefts involving the orbit, the maxilla, and the palate. He also had a complete cleft palate and a protruding premaxilla (Fig. 1). His left eye (the only functional eye) had started developing exposure keratitis due to the coloboma, and needed urgent repair. Blood test profile was within normal limits. Chest x-ray and echocardiographic evaluation revealed no congenital cardiac abnormality.

On the day of surgery, oral feeding was suspended four hours prior to shifting to the operating theatre. In view of anticipated difficult airway, sedative premedication was avoided. The child was placed on the operating table, and a number 4 transparent anesthesia mask (Intersurgical®, Berkshire, UK) was gently applied on the face, taking care of not injuring the eye (Fig. 2). The mask covered the child’s face completely. Sevoflurane in 100% oxygen, with a fresh gas flow rate of 5 L/min was used in incremental doses for induction of anesthesia. During induction, necessary monitors, including electrocardiography (ECG), peripheral saturation in oxygen (SpO2), and non-invasive blood pressure (NIBP) were used.
Intravenous access was achieved using a 24 G intravenous cannula. It was inserted after the child had lost consciousness. The child’s airway was maintained patent throughout the induction period. A 1.5 size Classic™ Laryngeal Mask Airway (LMA) device was then gently introduced using a lateral approach. Adequate ventilation could be achieved without any significant leak, and the LMA was secured. Anesthesia was maintained with oxygen, nitrous oxide, and sevoflurane. Targeted sevoflurane a minimal alveolar concentration ranged between 1 and 1.2. Analgesia was achieved using 10 mcg of intravenous fentanyl. This dose had to be supplemented intraoperatively with another 5 mcg. Ventilation was controlled. The surgery lasted 45 minutes and was uneventful. The child was allowed to breathe 100% oxygen after completion of surgery. Once spontaneous ventilation had adequately resumed, and once the child was fully awake, the LMA was removed. The postoperative period was uneventful and the child was discharged on the second postoperative day.

**DISCUSSION**

In 1976, Paul Tessier defined facial cleft nomenclature, numbering them according to anatomical findings and surgical dissection. He used number zero as the median facial dysrrhaphia, and determined fifteen locations for clefts (number 0 to 14), using the orbit as the point of reference (2). According to this classification, number 4 cleft is a central oribo-maxillary cleft (Fig. 3). It comes almost vertically through the lacrimal portion of the lower lid, lateral to the punctum, through the infraorbital rim and orbital floor, medial to the infraorbital nerve, through the maxillary sinus, and
cheek and continues through the lip midway between the philateral crest and the labial commissure. Alveolar cleft is usually present, and is located at the usual position for a complete cleft palate. In bilateral cases, the nose is smaller than normal, and the premaxilla is protuberant. The defect involves the orbit, cheeks, lip and the palate, whose reconstruction requires multiple surgeries over a prolonged period of time. However, protection of the eye is an early concern, and may require surgery as soon as possible to avoid exposure keratitis and its sequela. In our patient, due to the coloboma of the lower lid, the left eye was at risk of keratomalacia and corneal erosion. The anesthetic management of our patient, and of children with such facial and palatal deformities in general, is a challenge. In order to optimize the anesthetic management of these children, a thorough physical examination and radiological investigations to rule out cardiac anomalies are necessary.

The child had a wide uncorrected craniofacial cleft. Mask ventilation is a major concern in these patients, because achieving leak-proof mask ventilation is very difficult with such facial dysmorphism. The protruding left eye was also prone to injury and abrasions by the placement of the face mask. Carenze et al, in their description of anesthetizing a child with Tessier number 3 and 4, had also used a size 4 adult sized facemask for preoxygenation (3). However, to control the airway, they had inserted the LMA after sedation with transmucosal midazolam and topical anesthesia of the pharynx using a lidocaine spray. Our patient was very active, with an obviously difficult airway. An awake method of controlling the airway would obviously not have been possible. Since the mask facial interface was leak proof enough, anesthesia could be induced with sevoflurane in oxygen, using the same number 4 face mask. Spontaneous ventilation was maintained throughout the induction period. With such an induction method, we could avoid the risk of inducing any laryngospasm by spraying the airway of an awake child, or trying to insert the LMA at a lighter depth of anesthesia (4). We did not perform direct laryngoscopy, insofar as our airway management plan included maintaining spontaneous ventilation. Laryngoscopy itself could have produced laryngospasm and trauma in case of a too light anesthetic depth. In case oxygenation and ventilation through the LMA would have been found less than perfect, our back up plan was to maintain spontaneous ventilation, and perform fiberoptic-guided intubation through the already in place LMA.

Insertion of a LMA in a child with complete cleft palate may be complicated by the inadvertent entrapment of the LMA within the palatal cleft. Hence, to avoid this complication, we used a lateral method of insertion. This method has been reported as safe and effective in various studies in both children (5) and adults (6). Insertion of a LMA in a child with a cleft palate using a laryngoscope has previously been described (7). The LMA has now found a strong foothold in the field of difficult airway management in the pediatric age group (8, 9). Many authors have described airway management in children with anticipated difficult airways using an LMA, either as a sole airway device, or as a conduit for endotracheal intubation (10).

Endotracheal intubation in this child may have been a problem, insofar as the procedure would have required muscle relaxation or deepening of anesthesia using intravenous anesthetic agents such as propofol or remifentanil. This could have caused the tongue to fall posteriorly through the palatal cleft, leading to complete obstruction of the oropharynx. Although this can be overcome by inserting an oropharyngeal airway in a child with normal midface, it would not have been possible in this case because of the presence of a protruding premaxilla. The premaxilla can hamper the insertion of a laryngoscope blade, as well as obstruct the anesthesiologist line of sight during laryngoscopy and intubation attempts (11). The blade of the laryngoscope can also get entrapped in the palatal cleft. Therefore, to avoid all these problems, we decided to go further in the procedure using the LMA itself. In case of problem, we could have used the same LMA as a conduit for fiberoptic-guided endotracheal intubation. This was our plan in case the LMA was unable to provide satisfactory ventilation. A cannot ventilate cannot intubate scenario in this child, however, would have occurred in the event of a laryngospasm. In such a scenario, we were prepared to use succinylcholine to relieve the laryngospasm. Would an immediate airway control have been necessary, the use of muscle relaxation would have provided an optimized laryngoscopic view, with glottis relaxation (12).

To conclude, the successful airway management of a child with craniofacial cleft requires careful planning and use of innovative techniques, devices and resources to avoid major morbidity. Maintaining spontaneous ventilation during the inhalational induction of anesthesia, using large size masks, and inserting a LMA using a lateral method provided an appropriate anesthetic management of this patient.
References

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