Oral Teratoma in a Neonate - Anesthetic Challenge

Trishi Sharma, Neelam Dogra

Abstract: Congenital teratoma of oral cavity in a neonate is a rare condition associated with compromised airway and challenges anesthesiologist in airway management. We report a case of neonate with multiple oral teratoma, cleft palate and bifid tongue who presented with respiratory distress. A one day old 2.8 kg female baby with multiple oral mass protruding from mouth, cleft palate and bifid tongue was scheduled for excision of mass. Injection Atropine 0.01mg/kg was given through already secure i.v. line to limit secretions. Oxymetazoline nasal drop were put in both nostrils and then patency of right nare was checked by nasogastric feeding tube. A bigger size mask was selected to ensure proper bag and mask ventilation. Anesthesia was induced by sevoflurane and oxygen in gradual incremental concentration. A check laryngoscopy was done to visualize glottis. As glottis was visible injection succinylcholine 1.5 mg/kg was given and nasal intubation was done with the help of magill’s forcep from right nostril. Injection Fentanyl 2 microgram/kg was given and anesthesia was maintained with oxygen, nitrous oxide and sevoflurane. Recovery was uneventful after the surgical procedure and extubation was done.

Keywords: Oral mass, Congenital teratoma, Neonate difficult airway

1. Introduction

Newborn with congenital mass in oral cavity can present as challenges for anesthetist in airway management. The causes of difficult airway in newborn are different from that of adults. It mostly includes congenital anomalies such as laryngomalacia, hemangiomas, epulis, vascular ring, hypoplastic mandible. Oral teratoma is a rare congenital anomaly occurs in 1:35,000 to 1,200,000(9). The clinical presentation depends upon the size and location of mass. It may present as mass protruding from mouth with high chances of respiratory tract obstruction.

2. Case Presentation

A 2.8 kg one day old female baby was brought to the hospital with respiratory distress and multiple growth protruding from oral cavity. The child had macrostomia, cleft palate, bifid tongue and two oral growths one measuring approximately 25X20X15 mm attached to hard palate and other in the sublingual region measuring approximately 12X10X15 mm. C.T. finding showed large well defined cystic lesion of size 23X17X12 mm in left parasagittal palatal region with internal fat fluid density and calcified densities (Hypertrophied palatal bones). Similar small cystic lesion of size approximate 11X8X13 mm seen within oral cavity at sublingual region. On examination the child was vigorous, tachypnic, there was flaring of ala nasae. Mallampati grading could not be assessed.

Excision of the tumor was scheduled on the day 3rd of life of the baby.

On arrival in operating room, routine monitoring (ECG, Pulse Oxymeter, NIBP, Temperature) were attached. Oxymetazoline nasal drop was put in both nostril and patency of nare was checked by nasogastric feeding tube to rule out any extension of mass. Surgical team were kept ready to perform emergency tracheostomy if required. Injection atropine 0.01mg/kg was given through already secured i.v. access to limit secretions. For Preoxygenation, a bigger size transparent circular silicon face mask was selected along with surgical pads to form a seal around the nose, lower jaw and the mass to ensure proper bag and mask ventilation without causing any trauma to mass. Preoxygenation was done for 5 minutes. Once it was established that mask ventilation was possible anesthesia was induced by sevoflurane and oxygen in gradually increasing concentration. When the patient was adequately anesthetized the mass was covered with surgical pads to avoid any injury to it and a gentle check laryngoscopy was done to visualize the glottis with Miller’s blade 1.5, as glottis was visible, injection succinylcholine 1.5 mg/kg was given. After adequately lubricating the ET tube with 2 % lignocaine jelly, Right Nasal intubation was done with 3mm internal diameter uncuffed Endotracheal tube by using Magill’s forcep. Tube was fixed on the anterior surface of the nose. Injection fentanyl 2 microgram/kg and injection atracurium 0.5 miligram/kg was given. Anesthesia was maintained with oxygen, nitrous oxide, sevoflurane.

Pharyngeal packing was done by the surgeon before performing procedure. Sublingual mass was excised completely while mass at hard palate could be excised in piecemeal. There was not enough blood loss and haemostasis was done. The duration of surgery was 40 minutes. At the end of surgery neuromuscular blockade was reversed by injection Neostigmine 0.08 mg/kg and injection glycopyrrolate 0.02 mg/kg and extubation could be done smoothly. Patient was shifted to post-operative care unit. Nasogastric feeding was started on 2nd day and on fourth post-operative day oral feeding was started and patient was discharged on fifth post-operative day.
tumours may require emergent airway management after birth.

The term teratoma is derived from the Greek word teratōn, which means monster, and initially was used by Virchow in his first edition of his books on tumors, published in 1863 (1). Teratomas are neoplasms composed of tissue elements foreign to the anatomic site of origin (2). Epignathus are intraoral malformations present at birth and the point of origin, specifically the alveolar bone or jaw.

The etiology of epignathus is unknown and may arise from pluripotent cells in the region of Rathkes pouch that grow in a disorganized manner. Teratomas contain elements derived from all three embryonic germ layers: ectoderm, mesoderm, and endoderm. The reported incidence is 1:4000 births (2,3,4). They occur most commonly in the sacrococcygeal region, followed by the ovaries (5,6). Congenital oral teratoma (epignathus) occurs in 1:35,000 to 1:200,000 live births. This accounts for 2% to 9% of all teratoma. Teratomas are associated with concomitant malformations, with cleft palate being the most commonly associated anomaly. This is thought to be because of mechanical obstruction caused by the neoplasm, preventing closure of the palatal shelves. Other malformations associated with epignathus teratomas are bifid tongues and noses.

This is caused by a delayed swallowing reflex of the fetus and may be associated with a large oral mass. Polyhydramnios is often associated with an epignathus teratoma. Antenatal diagnosis by ultrasound imaging is essential for perioperative care and management. This would allow a team approach in planning the cesarean section and further perioperative care and management, which may include an Ex Utero Intrapartum Treatment (EXIT).

Neonatal oral teratomas are benign lesion having potential to cause respiratory obstruction so they should be diagnosed and excised earliest for good prognosis. This rare site accounts for 5% to 6% of teratomas, which generally present in the neonatal period with large tumors. Most are mature or immature teratomas, but up to 20% are malignant. A review of 20 neonates noted that 35% presented with airway obstruction.

The airway management in such type of cases is a challenge for anesthesiologist. An awake flexible fiberoptic intubation is often primary approach for management of difficult airway. Fiberoptic intubation requires considerable skill to perform it. In this presented case we planned to isolate the airway by nasotracheal intubation. Prior to induction of anesthesia all arrangement for emergency surgical tracheostomy and needle cricothyroidotomy are kept ready and necessary consent taken from the parents.

For visualizing the glottis, the approach that we used was different from the conventional one. In this, our blade entered through left side of the mouth, carefully and gently so as to avoid any injury to the mass. This approach helped us visualize the glottis with Miller's blade, and we didn’t need to use the fibroscope.

3. Discussion

Congenital oropharyngeal tumours derived from embryonic germ cells are very rare tumours. Large teratomas can cause respiratory tract obstruction at birth. Child presents with respiratory distress, difficulty in swallowing. Oropharyngeal tumours may require emergent airway management after birth.

The term teratoma is derived from the Greek word teratōn, which means monster, and initially was used by Virchow in his first edition of his books on tumors, published in 1863 (1). Teratomas are neoplasms composed of tissue elements foreign to the anatomic site of origin (2). Epignathus are intraoral malformations present at birth and the point of origin, specifically the alveolar bone or jaw.

The etiology of epignathus is unknown and may arise from pluripotent cells in the region of Rathkes pouch that grow in a disorganized manner. Teratomas contain elements derived from all three embryonic germ layers: ectoderm, mesoderm, and endoderm. The reported incidence is 1:4000 births (2,3,4). They occur most commonly in the sacrococcygeal region, followed by the ovaries (5,6). Congenital oral teratoma (epignathus) occurs in 1:35,000 to 1:200,000 live births. This accounts for 2% to 9% of all teratoma. Teratomas are associated with concomitant malformations, with cleft palate being the most commonly associated anomaly. This is thought to be because of mechanical obstruction caused by the neoplasm, preventing closure of the palatal shelves. Other malformations associated with epignathus teratomas are bifid tongues and noses.

This is caused by a delayed swallowing reflex of the fetus and may be associated with a large oral mass. Polyhydramnios is often associated with an epignathus teratoma. Antenatal diagnosis by ultrasound imaging is essential for perioperative care and management. This would allow a team approach in planning the cesarean section and further perioperative care and management, which may include an Ex Utero Intrapartum Treatment (EXIT).

Neonatal oral teratomas are benign lesion having potential to cause respiratory obstruction so they should be diagnosed and excised earliest for good prognosis. This rare site accounts for 5% to 6% of teratomas, which generally present in the neonatal period with large tumors. Most are mature or immature teratomas, but up to 20% are malignant. A review of 20 neonates noted that 35% presented with airway obstruction.

The airway management in such type of cases is a challenge for anesthesiologist. An awake flexible fiberoptic intubation is often primary approach for management of difficult airway. Fiberoptic intubation requires considerable skill to perform it. In this presented case we planned to isolate the airway by nasotracheal intubation. Prior to induction of anesthesia all arrangement for emergency surgical tracheostomy and needle cricothyroidotomy are kept ready and necessary consent taken from the parents.

For visualizing the glottis, the approach that we used was different from the conventional one. In this, our blade entered through left side of the mouth, carefully and gently so as to avoid any injury to the mass. This approach helped us visualize the glottis with Miller's blade, and we didn’t need to use the fibroscope.
The major anaesthetic concerns were as follows:
- Securing the airway by tracheal intubation and maintaining it perioperatively
- Doing intubation without causing any injury to the mass
- Maintaining homeostasis so as to prevent dribbling into airway.
- Prevention of post-operative respiratory difficulties

Our first major challenge was knowing the fact that the size of the mass was too big to make bag and mask ventilation achievable. To add to this concern was the vascularity of the mass, which needed us to carefully and gently handle the mask, while doing bag and mask ventilation.

To overcome our first hurdle, we chose a bigger size circular transparent mask to achieve a proper seal outside the mass and subsequently, bag and mask ventilation was possible.

Since we could do proper bag and mask ventilation, we ruled out the possibility of an awake technique approach and we carefully induced the patient.

But, since orotracheal intubation seemed remote in this patient, nasotracheal intubation was our first plan of management. And, since the baby was in respiratory difficulty prior to surgery also, we wanted to carefully handle nostrils, as practically, they were the only possible venue of ventilation for the baby. Keeping this in mind, necessary preparation for the use of fiberscope as well as for tracheostomy was done.

Though possibility of orotracheal intubation seemed remote, we performed a check laryngoscopy by entering from the left side of the mouth and visualization of the glottis was possible.

Adequate premedication with atropine and oxymetazoline drops effectively controlled secretions as well as bleeding while nasotracheal intubation.

Once we accomplished successful nasotracheal intubation using Magill's forceps, maintenance of anaesthesia was achieved using oxygen, nitrous, sevofluorane and atracurium.

Prior pharyngeal packing minimized trickling of blood intraoperatively into the airway. Intraoperatively, another major concern was the problem of maintaining homeostasis during the procedure. A possibility of potential severe haemorrhage was kept in mind and adequate blood was arranged. Fortunately, manipulation and excision of the mass by the surgical team was careful and homeostasis was well maintained throughout and blood loss was minimal.

Postoperatively, smooth extubation was carried out and the baby was comfortable in the post operative recovery room and manifested no signs and symptoms of respiratory difficulty. The baby was discharged on the fifth post-operative day, after achieving adequate oral feeding. The baby recovered excellently and will be planned up for palate repair at the age of 12 months.

4. Conclusion

Though airway compromise and its management are not infrequent, but, by appropriate prior planning, effective communication between anaesthetist as well as surgical team, it becomes far easier to manage such challenging cases and, therefore, successful outcome and optimal results become easily achievable.

References