CASE REPORT

Difficult Airway Management in a Neonate with Giant Sublingual Cyst: A Case Report

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ABSTRACT **Introduction:** Congenital ranula cysts are rare epithelial retention cysts originating from major sublingual or submandibular salivary glands, with an incidence of 0.74%. Early presentation often involves severe symptoms, including airway compromise and feeding difficulties. Management options include needle aspiration, surgical excision, marsupialization, sclerotherapy, laser excision, and cryotherapy. Significant airway compromises due to these congenital ranulas often occur in neonates; however, limited studies address their airway management.

Case Presentation: We present the management of a 2-week-old neonate with a huge congenital ranula with sublingual swelling, poor feeding, sneezing, inability to cry post-delivery, and had surgery done following initial resuscitation and nasogastric tube insertion. Intubation using a video laryngoscope with lidocaine and adrenaline instillation was successfully carried out to maintain the airway. The cyst was then successfully removed and marsupialization performed under general anaesthesia. Subsequently, the neonate was extubated 24 hours postoperatively without complications and discharged home after an uneventful recovery.

Conclusion: Giant congenital ranulas pose significant challenges for airway management. First-intention use of a video laryngoscope with lidocaine and adrenaline instillation can be an effective technique to manage an anticipated difficult neonatal airway when performed meticulously.

Keywords: Lidocaine instillation, Difficult airway, Video laryngoscopy, Congenital ranula cyst, Neonatal airway management.

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INTRODUCTION

Congenital ranula cysts, also known as alimentary tract cyst or mucoceles, are not common, although an incidence of 0.74% has been reported, with prenatal diagnosis rarely reported.[1,2] In Northwestern Nigeria, of the 29 cases of ranula seen over a period of 19 years, only 8 cases were congenital ranulas, with the lowest age of presentation being 3 days.[3] Most cases that present early are associated with severe symptoms.

Ranulas are retention cyst, which is derived from the major sublingual or submandibular salivary glands in the sublingual space between the mylohyoid muscle and the tongue.[1] They are thought to occur following obstruction of the main sublingual duct, which causes extravasation of mucus into the submucosal tissues, as a result, may rupture and extend into the surrounding soft tissues as a pseudocyst.[1,4] They may also occur due to sublingual gland ductal atresia or failure of embryological canalisation.[1]

Mostly self-limiting and without symptoms, however some may present with airway compromise, feeding difficulties and facial distortion where the mass is large. Needle aspiration, surgical excision of the cyst, sublingual gland excision along with cyst, marsupialization with or without 'stitch and stab' technique, sclerotherapy, laser excision or cryotherapy

are some of the existing options for managing intraoral ranulas.[5,6]

Although surgery is an important management pathway for ranulas,.[3,6,7] there are limited studies addressing often challenging airway management in congenital ranulas, particularly in cases with significant airway compromise. We here-in report the first intention use of video laryngoscope with adrenalized lidocaine instillation for managing a difficult airway in a neonate with a giant congenital ranula.

CASE PRESENTATION

A 2-week-old term neonate weighing 3kg, who was admitted to the emergency paediatric unit one day after birth. Symptoms exhibited include a sublingual swelling, poor feeding, sneezing, poor sleep quality and an inability to cry immediately after delivery, he was admitted and Immediate resuscitation with oxygen and fluids was initiated, and a nasogastric tube for feeding with expressed breast milk was inserted. Surgery was scheduled at 2 weeks of age. The preanesthetic review noted a history of poor feeding and sneezing, with the mother reporting poor sleep quality for the neonate. The prenatal history was uneventful. Upon examination, the neonate appeared calm, afebrile, not dehydrated, not cyanotic, and not pale, with a significant sublingual swelling measuring approximately 5cm by 5cm (Figure 1).

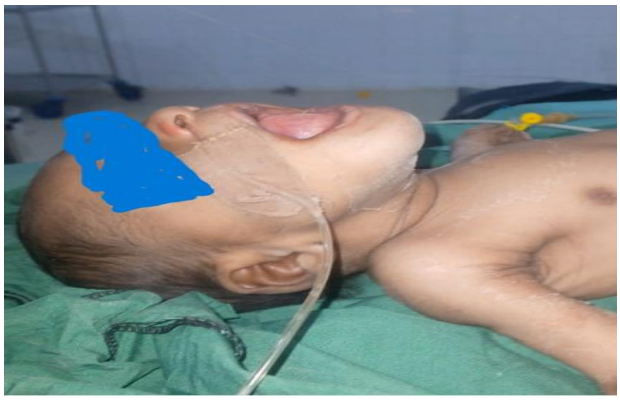

Figure 1: Giant Congenital Ranula Cyst.

Haematocrit, electrolytes, urea and creatinine estimations were within normal limits; however, no radiological investigation was done. The total bilirubin estimation was elevated, whereas the unconjugated bilirubin was normal. The plan included intubation using a video laryngoscope of appropriate size (BESDATA VL Mac2), with lidocaine plus adrenaline instillation, after obtaining written informed consent from the mother. A difficult airway cart including tracheostomy set was ordered for the surgery, and the ENT surgeon placed on standby in case of emergency surgical airway.

In the operating theatre, following a cockpit drill ensuring safety, the patient was given supplemental oxygen via nasal prongs after being gently positioned on the operating table. An intravenous access was secured with a size 24G cannula and Premedication was administered with intravenous atropine at a dose of 0.1 mg/kg before halothane in oxygen was delivered through nasal prongs connected to the anaesthetic machine. A bolus 5mg/kg of hydrocortisone was also administered to reduce airway oedema and inflammation in anticipation of tissue manipulation by the surgeon. The concentration of halothane was gradually titrated between 0.5% to 2% to adequately sedate the patient (COMFORTneo score of 11 was targeted) and facilitate intubation. A mixture of lidocaine and adrenaline (7 mg/kg) was prepared in a 2 ml syringe. Approximately 20 mg of the mixture was diluted to 2 ml with normal saline, and half of this solution was instilled directly into the mouth using a 2 ml syringe with a curved needle (Figure 2).

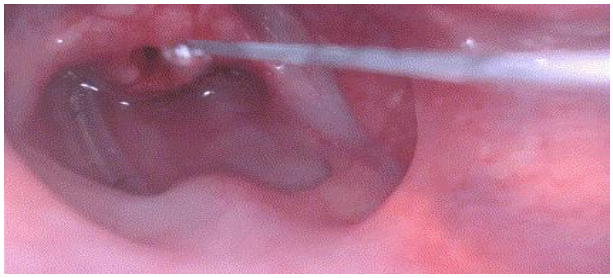

Figure 2: Lidocaine Instillation during the Airway Management.

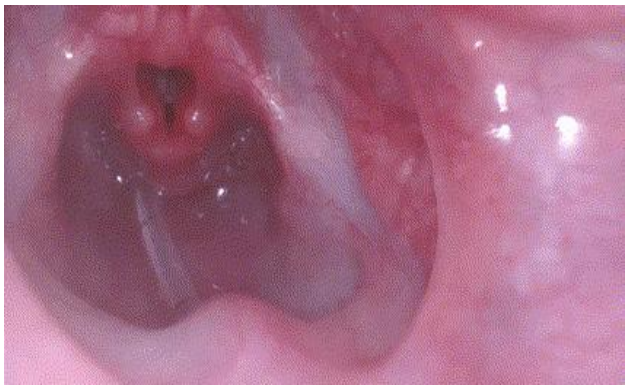

Figure 3: Insertion of the ETT during the airway management.

A gentle laryngoscopy was performed by a physician anaesthesiologist with the patient spontaneously breathing using a Macintosh-style video laryngoscope with a size 2 blade. Additional solution was instilled gradually while navigating around the mass to visualize the back of the tongue (Figure 2). Upon successfully viewing the glottis with the video laryngoscope, a 22G hypodermic needle attached to the 2 ml syringe with the lidocaine and adrenaline solution was slightly curved to avoid injuring the patient while manoeuvring through the pharynx. The solution was then carefully instilled over the pharynx and glottis before intubating the trachea with an uncuffed 3 mm endotracheal tube (ETT) (Figures 2 & 3). After successful intubation, gauze was packed into the throat under the guidance of the video laryngoscope, and the ETT was secured with adhesive plaster on the cheek (Figure 4). The cyst was successfully removed and marsupialization was performed under general anaesthesia maintained with 1% halothane in oxygen,

spontaneous breathing was assisted with Ayre's T-piece while intravenous paracetamol at a dose of 20mg/kg was administered for analgesia. The vital signs (pulse rate and respiratory rate) were stable throughout the surgery.

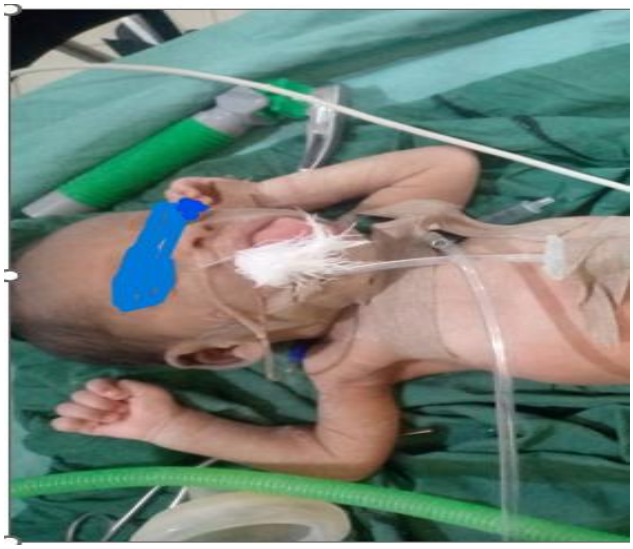

Figure 4: The Neonate after a successful ETT placement.

Following an uneventful surgery, halothane was discontinued and the patient was transferred to the post-anaesthesia care unit (PACU) with the ETT in situ after showing signs of recovery on the operating table. Oxygen supplementation was entrained through the ETT with a size 5 nasogastric tube at 2L/m at the PACU until the patient recovered fully and then transferred to the special care baby unit (SCBU) of the hospital for postoperative care. The patient was extubated 24 hours after surgery at the SCBU. The patient recovered fully without any incidence. Failed extubation due to airway oedema from tissue manipulation by the surgeon and inability to maintain adequate oxygen saturation in room air were anticipated perioperative concerns.

DISCUSSION

Huge congenital ranula may be associated with airway compromise and poor sleep quality as a result of the tongue falling backwards during sleep. These may present a significant challenge to anaesthesiologists, as securing the airway using conventional laryngoscopy is difficult and carries the risk of rupturing the cyst, leading to aspiration and bleeding. Besides the danger of cyst aspiration, the retromolar approach for laryngoscopy and intubation may be unfeasible due to the size and location of the intraoral mass. [8] Additionally, the swelling in the oral cavity complicates the use of a facemask and the insertion of a laryngeal mask airway. Consequently, in this patient, nasal prong connected to the anaesthetic machine was used to administer anaesthetic gases until tracheal intubation was achieved. Sevoflurane use is considered the gold standard for paediatric anaesthesia.[9], however, due to non-availability, we used halothane to manage our patient.

While a paediatric fibreoptic bronchoscope could be beneficial, its use is often limited by its availability and the required expertise in our setting, a paediatric fibreoptic was not available. Employing a Macintosh-style videolaryngoscope as the primary device for tracheal intubation in the operating room has been associated with a notable increase in the proportion of easy intubations compared to using a standard Macintosh laryngoscope. [10] However, the use of the video laryngoscope as a first option is not common in our setting due to its unavailability and lack of accessibility. When available, it is typically used only after the standard Macintosh laryngoscope has failed. Our use of a video laryngoscope as the initial option likely contributed to the successful tracheal intubation in this patient.

In a similar case reported by Singh et al, [11] an initial attempt to use a standard laryngoscope with a size 0 Millers blade was unsuccessful. Their second attempt succeeded only after aspirating the cyst to reduce the size of the swelling. Although in the case discussed here, aspiration of the cyst was not done due to the fear of airway compromise, our successful first attempt at intubation likely reduced morbidity and mortality in this patient. A size 2 blade has been reported to significantly reduce time to a successful intubation compared to a size 1 blade in neonates. [9] Although the study was conducted with a GlideScope®, we similarly used a size 2 blade for this patient.

In this case, premedication with an antisialagogue (intravenous atropine) was administered to enhance visualization during laryngoscopy. In addition, halothane was selected for sedation because of the risk of airway compromise and its advantage of maintaining spontaneous breathing in the neonate while achieving adequate anaesthetic depth for airway procedures.

Instillation with a syringe and needle is a simple but infrequently used method of applying the local anaesthetic topically for airway management, where a spraying device is not available. Local anaesthetic instilled onto the back of the tongue is swallowed initially but ultimately pooled in the pharynx and is aspirated by the patient, thus facilitating instrumentation of the airway. In this case, the instillation of lidocaine and adrenaline solution was done with a curved needle and syringe, which was sufficient for intubating the patient.

Another important factor is the positioning. Intubation was successful with the patient in supine position, Kumar et al[8] also reported success with the use of lateral position for the intubation of a huge congenital sublingual cyst underscoring the importance of positioning in managing upper airway compromise. In their report, the baby was placed in lateral position to reduce the risk of aspiration and the tongue falling back. However, we were able to achieve optimal intubating conditions with the patient positioned supine.

Residual swelling in the lingual space after the removal of a huge congenital ranula may result in postoperative airway compromise [8] often necessitating applying a tongue stitch and using naso- or oropharyngeal airways. We extubated this patient 24 hours postoperatively. The Extubation process was uneventful, likely because the swelling subsided significantly after 24 hours. The patient was then discharged from the intensive care unit and transferred to the Special Care Baby Unit (SCBU) of the paediatric department for observation, and subsequently discharged home after a 3-week stay.

CONCLUSION

Despite the challenges posed by a huge congenital ranula cyst, first-intention use of a video laryngoscope with lidocaine plus adrenaline instillation, when done meticulously, presented a good technique for managing a difficult airway in a neonate.

Conflicts of Interest: No conflict of interest is declared.

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