JK SCIENCE pISSN: 0972-1177

CASE REPORT

Airway Management of Palatoglossal Bands and Cleft Palate in An Infant in A Resource Limited Set Up – A Challenge to An Anaesthesiologist: A Case Report

Meenakshi Devi, Ashwani Kumar, Mushtaq Ahmed Wani, Heena Saini, Sanjay Dhar*

Abstract

Palatoglossal bands are one of the very rare congenital anomaly with very few documented cases worldwide. They can present with feeding difficulties or respiratory distress which requires immediate surgical intervention. The management of such patient is a challenge to any anaesthesiologist because of inability to perform conventional laryngoscopy, associated cardiac or digital anomalies and non-availability of pediatric flexible fibreoptic bronchoscope. We will discuss here the management of an infant who presented at one year of age with cough, recurrent upper respiratory tract infections, poor weight gain and feeding difficulties.

Key words

Difficult Airway in Neonates, Palatoglossal Bands, Conventional laryngoscopy

Introduction

Palatoglossal bands are rare congenital anomalies with very few reported cases.[1] They present with either respiratory distress at birth or feeding difficulties. They are usually associated with a variety of congenital defects such as cleft lip and palate, microglossia, micrognathia and temporomandibular disorders.[2] The cause of these bands is still unknown and presumed to be genetic, teratogenic, or mechanical insult in early gestation[3]. These defects are commonly associated with many congenital syndromes such as Moebius' syndrome, hypoglossia–hypodactylia syndrome, Hanhart's syndrome, cleft palate medial synechiae syndrome etc.[4,5] Our patient was a case of nonsyndromic palatoglossal bands and cleft palate as she did not have any other associated congenital anomaly. The definitive surgery includes excision of palatoglossal bands under anaesthesia.[6]

From:the Department of Anaesthesia . Intensive Care and Pain Medicine,and General Surgery*, GMC Jammu J&K India.

Correspondence to: Dr Meemakshi Devi , Senior Resident Department of Anaesthesia . Intensive Cre and Pain Medicine, GMC Jammu J&K India.

Manuscript Received: 31.01.2024; **Revision Accepted**: 01.04.2024;

Published Online First: 10 July, 2024

Open Access at: https://journal.jkscience.org

Case Presentation

A one year old child presented in the pediatric outpatient department with chief complaints of cough, recurrent upper respiratory tract infections, poor weight gain and feeding difficulties. She had recurrent episodes of vomiting after feeds. She was born full-term via normal vaginal delivery at home. Upon oral examination, the dorsum of the infant's tongue was adhered to posterior edge of hard palate and full thickness of the anterior soft palate [*Fig 1*]. A diagnosis of palatoglossal bands was made. History and examination revealed low weight as per age, delayed motor milestones, active cry and no cyanosis. Associated features present were hypoplasia of left horizontal process of maxilla and left micropthalmos with absent left eye ball. The child underwent an MRI of the head and neck region, a 2D echocardiogram, and an

Cite this article as: Devi M, Kumar A, Ahmed Wani MA, Saini H, Dhar S. Airway Management of Palatoglossal Bands and Cleft Palate in An Infant in A Resource Limited Set up – A Challenge to An Anaesthesiologist: A Case Report. JK Science 2024; 26(3):193-95

ultrasound of the abdomen for evaluation of associated conditions. The MRI was suggestive of a cleft palate. The ultrasound of the abdomen and 2D echocardiogram were normal. No cytogenetic analysis was done as the child did not show features of any named syndrome warranting this. Preoperative instructions were given to the parents to keep the infant fasting according to 2-4-6 rule, arrange blood and informed written high risk consent was obtained from the parents.

On the day of surgery Injection glycopyrrolate 5 mcg/kg was given in the preoperative room. A difficult airway cart was kept ready, and an ENT specialist was on standby as we were not having pediatric flexible fibreoptic bronchoscope in our setup. Monitors were applied. The patient was induced with sevoflurane 5% in a 50% mixture of N20 and oxygen. The IV cannula was secured, and fluid was started according to the Holliday-Segar formula. Anaesthesia was maintained on N2O, oxygen, isoflurane with spontaneous respiration till the confirmation of endotracheal tube.

We were able to ventilate the patient and we avoided muscle relaxant till we confirmed the position of ETT. Through external laryngeal manipulation and neck extension, the patient was intubated under direct laryngoscopy using a Miller blade approached from the right side of the fusion with a 3.5 mm uncuffed PVC endotracheal tube. When intubation was confirmed by chest auscultation and capnography, we administered muscle relaxant, injection atracurium 0.5mg/kg as a loading dose. Maintenance of anesthesia was done with oxygen, nitrous oxide, isoflurane, and injection atracurium 0.1 mg/kg. Adequate analgesia was provided with fentanyl 1mcg/kg and paracetamol 15mg/kg. The patient was then handed over to the plastic surgeon, who released the fusion of the tongue with the palate. After release, the full thickness defect was corrected by VWK Palatoplasty and intra velar veloplasty. The patient was successfully extubated after the surgery and shifted to the PACU for observation. During the follow-up period, the baby thrived well with adequate intake of milk. This was a novel approach such that patients' safety was not compromised as the patient was on spontaneous ventilation with adequate depth of anaesthesia. Second, we could guide our endotracheal tube under vision into the larynx, thus avoiding any trauma and bleeding. The ideal airway management in the presence of palatoglossal bands is through nasotracheal fibreoptic intubation.[7] However, we did not have a pediatric fibreoptic bronchoscope available with us in our setup.

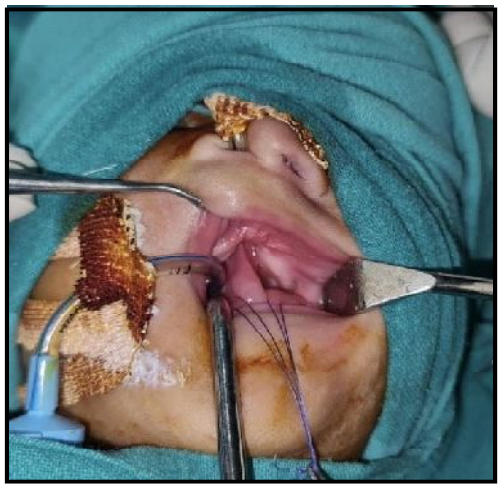

Fig 1: Palatoglossal Bands Extending From the Tongue to the Palate

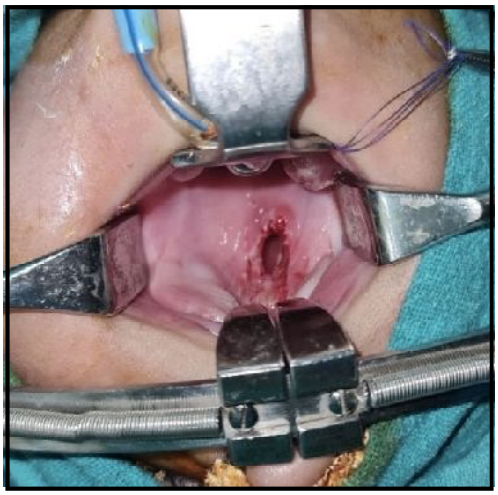

Fig. 2 Immediate Postoprative Image After Release of Tounge from Palate

Discussion

The airway management is always crucial for an anesthesiologist, particularly in case of intraoral pathologies where airway is anticipated to be difficult. Supra glottic airway devices cannot be used in such cases, which may lead to fatal outcomes when intubation fails. In our case, an ENT specialist was kept on standby for emergency tracheostomy in case intubation failed.

Palatoglossal synechia is a rare anomaly and its cause is still unknown.

Pandey *et al* did flexible fibreoptic nasopharyngoscope guided intubation under spontaneous ventilation and it was uneventful and smooth[8]. However, we were not having flexible fibreoptic nasopharyngoscope in our set up. So,

we proceeded with conventional laryngoscopy under sedation with the ENT team kept stand by all the time. Review of literature suggests a role for adhesion release in emergency, under local anaesthesia. Release under local anaesthesia was not tried in this case as it was thought to be risk with unprotected airway. We suggest complete evaluation of the child before major surgical intervention; because depending on the severity, these patients may present formidable anaesthetic challenges.

Conclusion

To conclude, in a resource limited setting, where the pediatric flexible fibreoptic bronchoscope may not be available, the conventional laryngoscopy through one side of the band under adequate sedation without muscle relaxation can be a feasible alternative for securing a definitive airway.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient's parents have given their consent for her images and other clinical information to be reported in the journal.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Naidoo S, Bütow KW. Nonsyndromic palate synechia with floor of mouth. Ann Maxillofac Surg 2015;5:100 3.
2. Solanki S, Babu MN, Gowrishankar, Ramesh S. Palatoglossal fusion with cleft palate and hypoplasia of cerebellar vermis. Indian J Plast Surg 2016;49:106 8.
3. Haydar SG, Tercan A, Uckan S, Gurakan B. Congenital gum synechiae as an isolated anomaly: A case report. J Clin Pediatr Dent 2003;28:81 3.
4. Hall BD. Aglossia adactylia. Birth defects Orig Artic Ser 1971;7:233 6.
5. Kaplan P, Cummings C, Fraser FC. A 'community' of face limb malformation syndromes. J Pediatr 1976;89:241 7.
6. Rai A, Bhola N, Yadav A, Sheorain A, Datarkar A, Jaju S, *et al*. Intra oral epithelial bands in a new born: Report of two cases and review of literature. J Maxillofac Oral Surg 2012;11:351 3.
7. Rani A, Sunder R, Kumar MM. Airway management of a neonate with a rare congenital palatoglossal synechiae. Pediatric Anesth 2008;18:980-2.
8. Pandey V, Waindeskar V, Katiyar R, Agrawal S. Airway management of palatoglossal bands – A challenge to an anaesthesiologist. Saudi J Anaesth 2019;13 (4):338-9.