Case Report

Restrictive Feeding and Airway Obstruction from Sub-glossopalatal Synechiae with Cleft Palate: An African Case Report and Review of Literature

Abdur-Rahman LO[1], Nasir AA[1], Adeyemi MF[2]

[1]Department of Surgery, Division of Paediatric Surgery, University of Ilorin and University of Ilorin Teaching Hospital, [2]Department of Surgery, Maxillofacial Surgery Unit, University of Ilorin and University of Ilorin Teaching Hospital, Ilorin, Nigeria

ABSTRACT

Glossopalatal and congenital alveolar synechiae are rare and often occur in association with orofacial anomalies. The occurrence of sub-glossopalatal membrane causing restrictive mouth opening associated with complete cleft of hard palate has not been reported from Africa. A report of a case of the subglossopalaal membrane and its management in a term male child delivered per vaginum by a 24-year-old primiparous woman in a rural setting in Nigeria. All available English literature was search using Medline, PubMed, and Google scholar. A 3-day-old boy was presented because of a restrictive mouth opening due to sub-glossopalatal membrane associated with a palatal cleft that made him to choke when feeding and snores all the time. He was admitted and nursed prone to allow the tongue to fall forward and prevent airway obstruction. He was also fed through nasogastric tube. He had a successful excision of membrane under local anesthesia on the 10th day of life. He did well postexcision and was placed on cup and spoon feeding while awaiting a palatal cleft repair. He had an uneventful cleft palate repair at 10 months of age. This is the first African report of sub-glossopalatal membrane associated with a cleft palate. A careful management brings good outcome.

Key words: Airway obstruction, cleft palate, feeding difficulty, glossopalatal synechiae, sub-glossopalatal membrane

How to cite this article: Abdur-Rahman LO, Nasir AA, Adeyemi MF. Restrictive feeding and airway obstruction from sub-glossopalatal synechiae with cleft palate: An african case report and review of literature. Niger J Health Sci 2015;15:116-8.

INTRODUCTION

Glossopalatal and congenital alveolar synechiae are rare and often occur in association with orofacial anomalies.[1] Ten cases of isolated congenital alveolar synechiae and nine cases of those associated with other anomalies or syndromic synechiae were reported from Europe, Asia, and United States.[1-8] This is the first case of membranous subglossopalatal synechia with cleft palate being reported from Africa. We present the management challenges and review of the literature.

CASE REPORT

Patient B. T. was brought by parents at 3 days of live on account of difficulty in feeding, intermittent respiratory distress, snoring, and observation of fleshy tissue in the mouth. The patient chokes whenever he is fed and cries because of nonsatisfaction. He was a term and delivered in a private clinic per vaginum in a town about 80 km from our institution. He was irritable, weighed 3.6 kg, pink in room air, acyanosed, anicteric, afebrile, and mildly dehydrated. There was no craniofacial dysmorphism, and other body systems were essentially normal. Examination of oral cavity revealed membranous sheet arising from the floor of the mouth (below the tongue [Figure 1]) extending to the palate above and from the midline over the sublingual ducts to the incisive foramen anteriorly and to the molar margin posteriorly. There was a central opening about 4 cm × 2.5 cm revealing the tongue behind the membrane and complete secondary palatal cleft above. The jaw could be opened up to about 4 cm. No other abnormality in the

Access this article online

Quick Response Code:

Website: www.chs-journal.com

DOI: 10.4103/1596-4078.182326

Address for correspondence: Dr. Abdur-Rahman LO, Department of Surgery, Division of Paediatric Surgery, University of Ilorin and University of Ilorin Teaching Hospital, P. O. Box 5291, Ilorin 240001, Nigeria.
E-Mail: bolarjide@yahoo.com

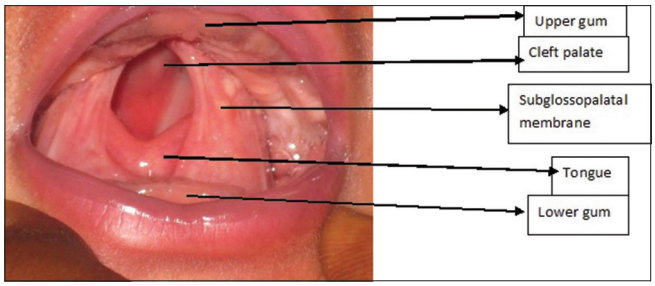

Figure 1: Subglossopalata membrane displacing the tongue backwards and also showing cleft palate above. See accumulated saliva in the vestibule on the right.

chest, abdomen, or extremities was noted. A diagnosis of subglossopalatal membrane with cleft palate was made.

He was fed through nasogastric tube, which made him calm. He also had intermittent suction of the oropharynx and was nursed in the prone position for the tongue to fall forward and saliva to drool outwards. The hemogram and blood chemistry were essentially normal. Informed consent was obtained from the parents for surgical intervention.

At 10 days of live, he had incision of the membrane under local infiltration with lignocaine and oxygen by nasal cannular because the tracheal could not be accessed and because we were afraid we might lose control of the airway when any form of general anesthesia is given [Figure 2]. Blood loss was minimal. Remnant of the membrane was left more toward the palate to give enough tissue during palatal repair in future. The tongue immediately returned anteriorly and there was free movement of the temporomandibular joint. Feeding was commenced a few minutes postoperative by cup and spoon because of the palatal cleft. He did remarkably well and had cleft of palate repaired at 10 months of age by Von Langebeck technique using mucoperiosteal palatal flaps.

DISCUSSION

Synechiae or ankyloses are adhesions between anatomic structures. In the maxillofacial region, synechiae most commonly arise between the upper and lower alveolar ridges (syngnathism) or between the tongue and margins of the palate or maxilla (glossopalatal ankylosis). Synechiae arising from the lower lip, the floor of the mouth, or at the oropharyngeal isthmus have been described.[5]

Congenital alveolar synechiae is a rare anomaly, which may be isolated or syndromic.[1-8] Isolated anomaly rarely occurs as they are almost always accompanied by one or more additional congenital defects, such as cleft palate, cleft lip, microglossia, micrognathia, temporomandibular joint disorders, or lip anomalies.[1,5,7-13] The common associated syndromes are Van der Woude and cleft palate-lateral alveolar synechiae syndrome.[1,5-7,11,12] The fusion may consist of membranes or bands of epithelium supported by various amounts of connective tissue ranging from fibrous or elastic tissue to even muscle or bone.[5] Cleft palates are the deformities most

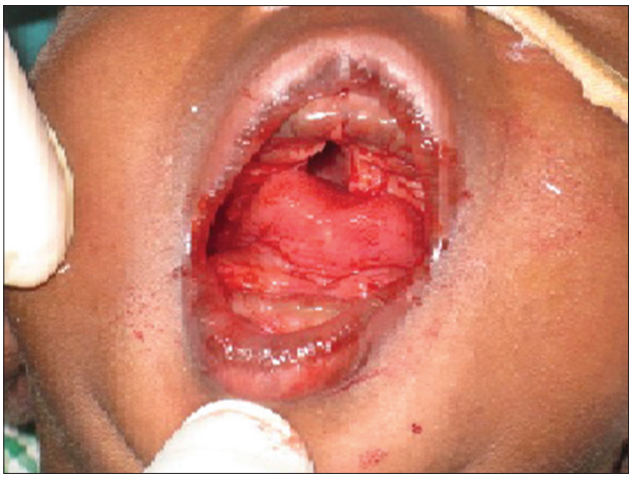

Figure 2: After the division of the membrane under local plain lignocaine infiltration. The tongue now moves more anteriorly and palatal cleft is shown.

frequently seen with alveolar synechiae.[5-8] In our patient, a membranous tissue arose from the floor of the mouth to the hard palate giving a jaw opening of about 4 cm in addition to a complete cleft of the secondary palate.

The cause of synechiae is unknown though theories have been postulated about its embryogenesis. During the 7th–8th week of embryological development, the alveolar ridges, tongue, and palatal shelves are in contact with each other. The ensuing palatal closure depends on downward contraction of the tongue, descent, and medial fusion of the palate. When the tongue protrudes from the mouth as a result of medial movements of the oral cavity walls, it prevents the alveolar ridges from fusing.[14] (ref Sadler embryology). Persistence of remnants of the buccopharyngeal membrane, contact between the epithelium of the gums or the palatine shelves and the floor of the mouth and presence of an ectopic membrane may result in abnormal fusion. These can be sub-classified as a sub-glossopalatal membrane, glossopalatal ankylosis, or syngnathia.[5] This membrane theory especially the ectopic membrane aspect may be a possible cause in our case because of the subglottic attachment of the membrane. However, the presence of the palatal cleft can be explained to be due to the inability of the tongue to contract downward and protrudes from the mouth because of the inhibition by the membrane.

Genetic, teratogenic, or mechanical insults during this critical stage may lead to periods of close, quiescent contact between oral structures, and this predisposes to abnormal fusion.[13] Familial, autosomal dorminant and recessive inheritance were reported previously based on the level of transmission and expression of the anomalies.[8,11,15]

The membranes in our patient were separated with incision after infiltration with a local anesthetic to ensure we do not inflict pain on this neonate because we are aware neonates also feel pain. This surgical division of the membrane allowed better coordination of swallow and gave room to normal mandibular function and growth. It also prevented upper airway obstruction

from posterior displacement of the tongue, and it was easy to nurse the baby in any position. This patient was changed to cup and spoon from nasogastric tube feeding, and he gained good weight while awaiting the repair of cleft palate.

CONCLUSION

Interalveolar synechiae though a rare condition has an excellent prognosis. Its presence necessitates careful examination for associated abnormalities and syndromes and diligent management of the patient.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Tomlinson JK, Liem NT, Savarirayan R, Meara JG. Isolated and syndromic syngnathism: Management, implications, and genetics. Ann Plast Surg 2006;57:231-5.
2. Sahin U, Ozdil K, Uscetin I, Saylkan S, Oktem F, Yuce S, *et al.* Cleft palate and congenital alveolar synechiae syndrome. Plast Reconstr Surg 2005;115:1212-3.
3. Tanrikulu R, Erol B, Görgün B, Ilhan O. Congenital alveolar synechiae - a case report. Br Dent J 2005;198:81-2.
4. Murphy SM, Rea S, McGovern E, Fleming P, Orr D. Cleft palate and congenital synechiae syndrome: A case report. Cleft Palate Craniofac J 2004;41:206-7.
5. Gartlan MG, Davies J, Smith RJ. Congenital oral synechiae. Ann Otol Rhinol Laryngol 1993;102(3 Pt 1):186-97.
6. Dinardo NM, Christian JM, Bennett JA, Shutack JG. Cleft palate lateral synechia syndrome. Review of the literature and case report. Oral Surg Oral Med Oral Pathol 1989;68:565-6.
7. Verdi GD, O'Neal B. Cleft palate and congenital alveolar synechia syndrome. Plast Reconstr Surg 1984;74:684-6.
8. Dalal M, Davison PM. Cleft palate congenital alveolar synechiae syndrome: Case reports and review. Br J Plast Surg 2002;55:256-7.
9. Denion E, Capon N, Martinot V, Pellerin P. Neonatal permanent jaw constriction because of oral synechiae and Pierre Robin sequence in a child with van der Woude syndrome. Cleft Palate Craniofac J 2002;39:115-9.
10. Knoll B, Karas D, Persing JA, Shin J. Complete congenital bony syngnathia in a case of oromandibular limb hypogenesis syndrome. J Craniofac Surg 2000;11:398-404.
11. Haramis HT, Apesos J. Cleft palate and congenital lateral alveolar synechia syndrome: Case presentation and literature review. Ann Plast Surg 1995;34:424-30.
12. Ergen C, Sayan NB. Van der Woude syndrome (report of a case). Ankara Univ Hekim Fak Derg 1985;12:163-9.
13. Sternberg N, Sagher U, Golan J, Eidelman AI, Ben-Hur N. Congenital fusion of the gums with bilateral fusion of the temporomandibular joints. Plast Reconstr Surg 1983;72:385-7.
14. 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.
15. Seres-Santamaria A, Arimany JL, Muñiz F. Two sibs with cleft palate, ankyloblepharon, alveolar synechiae, and ectodermal defects: A new recessive syndrome? J Med Genet 1993;30:793-5.