

# Spontaneous Palatal Cleft Closure

Fabio G. Ritto, DDS, MD, MS, PhD<sup>1</sup> ,  
Geoffrey H. Sperber, BSc Hons, BDS, MS, PhD, FICD, Dr Med Dent  
(Honcausa), FRSSAF<sup>2</sup>, and Kevin S. Smith, DDS, FACS, FACD<sup>1</sup>

The Cleft Palate-Craniofacial Journal  
1-3

© 2020, American Cleft Palate-  
Craniofacial Association  
Article reuse guidelines:  
sagepub.com/journals-permissions  
DOI: 10.1177/1055665620960970  
journals.sagepub.com/home/cpc



## Abstract

This case report presents a palatal cleft that healed spontaneously, with complete formation of mucosa and bone. Even though the nasal structures could initially be observed through the cleft palate, a thin membrane sealed any communication between the oral and nasal cavities. The origin of this tenuous membrane cannot be fully understood with current discernment of palate formation, but it probably served as a basis for the formation of the other tissues. No previous record of nonintervened spontaneous closure of a cleft palate has been reported.

## Keywords

palatal development, scarring, craniofacial growth

## Introduction

Cleft palate can occur due to several phenomena that affect palatal closure during the 7th and 10th gestational weeks. More than 100 syndromes have been associated with cleft palates, as well as they may also occur as a primary and isolated condition. The interposition of any structure that will impair the palatal shelves to fuse on the midline will generate a palatal cleft. The Pierre Robin sequence perfectly exemplifies this mechanism, where the retropositioned tongue, due to mandibular underdevelopment, will prevent the palatal processes to fuse.

Human embryonic palatogenesis is a complex concatenation of both evolutionary and developmental phenomena what is exquisitely timed to divide the initial stomodeal oronasal cavity into separate upper nasal and lower oral cavities. This separation is peculiar to primates who evolved a need to create a suction apparatus during infant suckling, allowing concomitant breathing (upper compartment), and ingestion (lower compartment) prior to swallowing. The human palate during embryogenesis passes through stages representing divisions of the oronasal chamber found in primitive crossopterygian fishes, reptiles, and early mammals. Birds, snakes, and fish lack intact palates and swallow food untritured.

## Case Report

A 7-month-old girl was referred for cleft palate evaluation. The mother had 40 weeks of uneventful pregnancy, the child was born naturally, had no signs of disease, and the cleft palate was not diagnosed at birth. The mother reported normal

breastfeeding and bottle-feeding during the first months of life. There were no reports of traumatic events during pregnancy or after birth that could potentially result in malformation and oronasal communication. It was the older sister's observation that there was "a hole" on the patient's palate that initiated clinical examination at 7 months of age.

The patient was then examined and a round cleft measuring approximately 20 mm in diameter was observed. A thin membrane, which initially appeared to be just a saliva film, covered the cleft. It was possible to observe the nasal septum and the turbinates through the palate (Figure 1).

The protocol in our department is to perform hard palatal cleft closure between 12 and 18 months. As the patient presented normal growth, with adequate weight gain and within the 50th percentile for weight, height, and cephalic development, it was decided to wait for surgical closure of the cleft. Instructions were given on possible complications and foods to avoid, and a new appointment was scheduled for 5 months later.

<sup>1</sup> Cleft Palate and Craniofacial Surgery, University of Oklahoma Health Sciences Center, Oklahoma City, OK, USA

<sup>2</sup> University of Alberta, Edmonton, Alberta, Canada

## Corresponding Author:

Kevin S. Smith, Department of Oral and Maxillofacial Surgery, University of Oklahoma Health Sciences Center (HSC), 1201N. Stonewall Ave Suite 230, Oklahoma City, Norman, OK 73117, USA.

Email: kevin-smith@ouhsc.edu



**Figure 1.** Initial consult—7 months.



**Figure 2.** Second consult—12 months.



**Figure 3.** Final consult—2 years and 8 months.

When the 12-month-old patient returned for follow-up, the cleft was completely covered by mucosa (Figure 2). Palpation revealed absence of bone under the mucosa, but there was no sign of oronasal communication. The last follow-up was performed at 2 years and 8 months of age, and the palate appears absolutely normal on clinical examination, with solid bone under the palatal mucosa, no history of ear infection, and no speech difficulties (Figure 3).

## Discussion

Spontaneous bone regeneration is not a new topic for the mandible or maxilla. For the mandible, most reports occur after tumor resections in children (Kazanjian, 1948; Byars and Schatten, 1960), whereas spontaneous bone formation of the

maxilla is a common finding after maxillary sinus procedures, regardless of the patient's age (Jung et al., 2007; Lie et al., 2019).

Regeneration of bone after surgical palatal cleft closure has also been widely reported (Yin et al., 2005; Saijo et al., 2010). Some investigators have observed that the palatal bone defect will reduce, and sometimes, close after palatoplasty. The most accepted explanation for this phenomenon is the layer of blood that remains between nasal and oral mucosa after 2 plane closure of the palatal cleft. The blood layer stimulates osteoblasts to produce bone. However, the closure of a complete palate defect showing spontaneous regeneration of the oral mucosa, palatal bone, and nasal mucosa has never been previously described.

The conglomeration of the programmed concurrent and sequential gene expression patterns of CD44, PDGFC, IRF6, TGFβ3, TWIST, MSX1, PVRL1, TBX22, FGFR1, SATB2, and RUNX1 presents enormous challenges in dissecting the roles of individual genes in fusing the medial edge epithelia of embryonic elevated palatal shelves in the midline (Sperber and Sperber, 2018). Many studies have been devoted to exploring the unfolding phenomena of palatogenesis (Fitchett and Hay, 1989; Bush and Jiang, 2012; Neupane et al., 2018).

The role of variable penetrance of WNT9B expression levels in palatogenesis was explored by Fontura et al. (2015) and Green et al. (2019) who cite how methylation changes lead to morphological changes (Gonseth et al., 2019). A change in conditions postnatally may alter variance of penetrance of genes determining palate formation. Understanding how different genetic loci interact in different scenarios affected by environmental factors provide clues to the initially failed fusion of the palatal shelves at a critical time. Epithelial–mesenchymal interaction, whereby the complex biochemical signaling agents that allow palatal edge fusion to take place, was delayed in this case until 12 months postnatally.

This rare if not unique case report provides an opportunity to explicate the roles of different genes in the commixture of the embryonic palatal shelves in forming the roof of the mouth. How such a genetic analysis can be undertaken is a challenge for the future.

There seems to be a current tendency for numerous surgical procedures in patients with cleft lip and palate, with multiple revisions, in order to achieve the “perfect” result. This particular case presented here should be used to remind us of the extraordinary potentials that children have for creating new tissues and improving the quality of scars. Therefore, after performing the primary closure of the existing clefts on the adequate age, and if there are no functional problems such as breathing, eating, and speaking difficulties, we suggest providing time for the immature human apparatus to work on its own.

## Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

## Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

## ORCID iD

Fabio G. Ritto, DDS, MD, MS, PhD  <https://orcid.org/0000-0002-7293-1459>

## References

- Bush JO, Jiang R. Palatogenesis: morphogenetic and molecular mechanisms of secondary palate development. *Development*. 2012;139(2):231-243.
- Byars LT, Schatten WE. Subperiosteal segmental resection of the mandible. *Plast Reconstr Surg*. 1960;25:142-145.
- Fitchett JE, Hay ED. Medial edge epithelium transforms to mesenchyme after embryonic palatal shelves fuse. *Dev Biol*. 1989;131(2):455-474.
- Fontura C, Silva RM, Granjeiro JM, Letra A. Association of WNT9B gene polymorphisms with non syndromic cleft lip with or without cleft palate in Brazilian nuclear families. *Cleft Palate Craniofac J*. 2015;52(1):44-58.
- Gonseth S, Shaw GM, Roy R, Mark RS, Kripa A, Jasper R, Joseph W, Nicholas JM. Epigenomic profiling of newborns with isolated orofacial clefts reveals widespread DNA methylation changes and implicates metastable epiallele regions in disease risk. *Epigenetics*. 2019;14(2):198-213.
- Green RM, Leach CL, Diewert VM, Aponte JD, Schmidt EJ, Cheverud JM, Charles CR, Nathan MY, Ralph SM, Benedikt H. Nonlinear gene expression-phenotype relationships contribute to variation and clefting in the A/WySn mouse. *Dev Dyn*. 2019;248(12):1232-1242. doi:10.1002/dvdy.1110.
- Jung YS, Chung SW, Nam W, Cho IH, Cha IH, Park HS. Spontaneous bone formation on the maxillary sinus floor in association with an extraction socket. *Int J Oral Maxillofac Surg*. 2007;36(7):656-657.
- Kazanjian VH. Spontaneous regeneration of bone following excision of section of the mandible. *Am J Orthod Oral Surg*. 1948;32(4):242-248.
- Lie N, Merten HA, Yamauchi K, Wiltfang J, Kessler P. Pre-implantological bone formation in the floor of the maxillary sinus in a self-supporting space. *J Craniomaxillofac Surg*. 2019;47(3):454-460.
- Neupane S, Adhikari N, Jung JK, Chang-Hyeon A, Sanggyu L, Jong-Hwa J, Ji-Youn K, Youngkyun L, Wern-Joo S, Jae-Young K. Regulation of mesenchymal signaling in palatal mucosa differentiation. *Histochem Cell Biol*. 2018;149(2):143-152.
- Saijo H, Mori Y, Fujihara H, Kanno Y, Chikazu D, Ohkubo K, Hisako H, Mitsuyoshi I, Yoshiyuki Y, Tsuyoshi T. Evaluation and analysis of formation of bone at the palate in patients with cleft lip and palate after palatoplasty based on computed tomograms and three-dimensional data. *Scand J Plast Reconstr Surg Hand Surg*. 2010;44(1):21-25.
- Sperber GH, Sperber SM. *Craniofacial Embryogenetics & Development*. 3rd ed. People's Medical Publishing House; 2018.
- Yin N, Ma L, Zhang Z. Bone regeneration in the hard palate after cleft palate surgery. *Plast Reconstr Surg*. 2005;115(5):1239-1244.