# INTERDISCIPLINARY CLEFT CARE

## *GLOBAL PERSPECTIVES*

**Usama S. Hamdan**, MD, FICS
**Carolyn R. Rogers-Vizena**, MD
**Raj M. Vyas**, MD
**Brian C. Sommerlad**, MBBS, FRCS
**David W. Low**, MD

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e-mail: instructormaterials@pluralpublishing.com
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# Preface

The quest for optimal patient outcomes is a driving force for health care professionals. This is especially true for those caring for children and adults born with cleft lip and palate. Significant advances in interdisciplinary cleft care, coupled with exponentially expanding information dissemination, facilitate knowledge sharing and quality improvement on a global level. A remaining aspiration of those involved in cleft care is to "level the playing field" and achieve ideal outcomes at all income and resource levels. That goal inspired the current work.

Through print, illustration, and videography, this book presents the knowledge, skill, and evidence-based practice of an interdisciplinary group of cleft experts from around the world. Lessons from individuals practicing in a variety of cultures and resource environments have been thoughtfully assembled. The resulting work provides the reader with practical and personally vetted solutions for all aspects of cleft care. Highly accurate illustrations by surgeon-artist Dr. David Low and technical videos accompanying this book also enhance the reader's ability to understand and apply techniques used by the authors to achieve successful outcomes. A multidisciplinary team approach is essential for rendering the best possible cleft care, and we hope this book serves as a catalyst for development of comprehensive cleft programs worldwide.

# Acknowledgments

The editors are grateful to all the children and adults born with cleft lip and palate who were the inspiration for this project and for the lessons they have taught us in perseverance and humility. We are also indebted to the Global Smile Foundation postdoctoral fellows whose efforts made this book possible: Dr. Elsa Chahine, Dr. Antonio Melhem, Dr. Omar Al Abyad, Dr. Mario Haddad, and Dr. Robert Younan. Their initiative, dedication, and perseverance created the momentum needed to transform the idea for this book into reality. We are also incredibly appreciative of our coeditor, Dr. David Low, whose illustrations are found throughout this book. The anatomic detail and surgical accuracy rendered by a practicing cleft surgeon is truly a unique highlight of this book. Finally, we are thankful to our colleague authors, volunteers, and supporters whose tireless energy and focus on providing safe and comprehensive cleft care throughout the world have been instrumental in this undertaking.

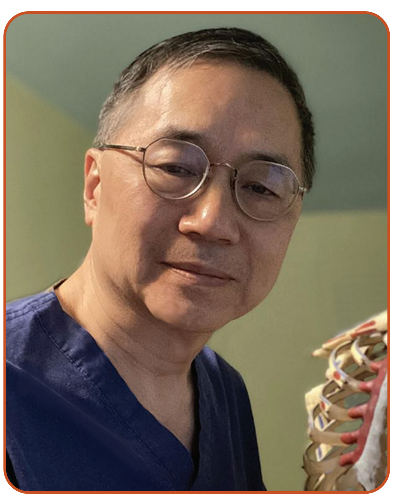
# About the Editors

**Usama S. Hamdan, MD, FICS,** is president and cofounder of Global Smile Foundation, a 501(c)(3) Boston-based nonprofit foundation that provides comprehensive and integrated pro bono cleft care for underserved patients throughout the world. He has been involved with outreach cleft programs for over three decades. Dr. Hamdan is an otolaryngologist/facial plastic surgeon with former university appointments at Harvard Medical School, Tufts University School of Medicine, and Boston University School of Medicine. He is also the founder of the *International Comprehensive Cleft Care Workshop* based on his special interests in simulation-based cleft training, empowerment, and sustainability initiatives for providing comprehensive cleft care as well as quality assurance strategies. For his philanthropic service to the people of Ecuador, he was awarded the knighthood, "Al Merito Atahualpa" En El Grado De Caballero, by the president of Ecuador in March 2005. He received honorary professorship at Universidad de Especialidades Espíritu Santo, School of Medicine, in Ecuador on March 5, 2015, for his contributions in the field of cleft lip and palate.

**Carolyn R. Rogers-Vizena, MD,** is a pediatric plastic and craniofacial surgeon at Boston Children's Hospital, assistant professor of surgery at Harvard Medical School, and associate clinical director for special effects and materials science in the Boston Children's Simulator program. Her clinical areas of expertise include cleft lip and palate, velopharyngeal dysfunction, facial trauma, adolescent breast surgery, and spina bifida. Dr. Rogers-Vizena leads Boston Children's cleft outcomes program with interinstitutional collaborations nationally and internationally through the International Consortium for Health Outcomes Measurement aimed at optimizing the physical and psychosocial impact of cleft lip and palate. In addition, she has partnered with the Global Smile Foundation to provide cleft care to underserved populations in the Middle East.

**Raj M. Vyas, MD,** earned his BS in biology with honors and distinction from Stanford University and completed his MD at UCLA, where he was awarded the Stafford L. Warren Medal for most outstanding medical student and the Longmire Medal for most outstanding student in surgery. He then completed integrated plastic surgery residency at Harvard and Craniofacial Fellowship at New York University. Dr. Vyas is a professor of plastic surgery at UC Irvine School of Medicine, where he serves as vice-chair for the Department of Plastic Surgery and chief of pediatric plastic surgery at CHOC Children's Hospital. His clinical and research interests include characterizing neonatal sleep and breathing disturbances, enhancing recovery after cleft/craniofacial surgery, understanding patient-reported psychosocial outcomes, and using technology to facilitate knowledge and skill transfer. Aligned with his passion for enhancing global capacity for interdisciplinary cleft care, Dr. Vyas is codirector of Global Smile Foundation's International Research Fellowship and codirector of Research for Plastic Surgery Foundation's SHARE program (Surgeons in Humanitarian Alliance for Reconstruction Research and Education). Dr. Vyas actively serves on dozens of regional, national, and international cleft committees, hospital consortia, and journal editorial boards.

**Brian C. Sommerlad, MBBS, FRCS,** qualified in medicine in Sydney, Australia. He went to the United Kingdom in 1968 to further his training and stayed. He is an honorary consultant plastic surgeon at Great Ormond Street Hospital for Children, London. He has been caring for children with clefts for 45 years—in the United Kingdom and by regularly working with colleagues in many less privileged countries over the past 22 years. His ongoing research interests have centered on palate anatomy and function and attempting to improve the speech outcomes of palate repair. He was cofounder in 2007 and is chairman of the U.K. charity CLEFT. which funds research into the causes and treatment of clefts and supports cleft centers in several low-resource countries.



**David W. Low, MD,** is a professor of surgery at the Perelman School of Medicine at the University of Pennsylvania and attending surgeon in the Division of Plastic Surgery at the Children's Hospital of Philadelphia. A graduate of Harvard College and Harvard Medical School, he completed all of his surgical training at Penn and was privileged to learn cleft care under the mentorship of Don LaRossa and Peter Randall. He is the medical director of his CHOP-based international cleft team Smiles for Guatemala. In addition to his teaching and patient care responsibilities, he is a board-certified medical illustrator whose work appears in many medical journals and textbooks.

# Contributors


**Shelly Abramowicz, DMD, MPH, FACS**
Associate Professor in Surgery and Pediatrics
Department of Surgery, Division of Oral and Maxillofacial Surgery
Emory University School of Medicine
Chief of Oral and Maxillofacial Surgery
Children's Healthcare of Atlanta
Atlanta, Georgia
*Chapter 35*

**Mohammad M. Ahmed, DDS, MS**
Diplomate
American Board of Pediatric Dentistry
Fellowship
Craniofacial Pediatric Dentistry
Consultant
Dubai Health Authority
Dubai, United Arab Emirates
*Chapters 7 and 9*

**Omar S. Al Abyad, MD**
Postdoctoral Research Fellow
Global Smile Foundation
Norwood, Massachusetts
*Chapters 17, 28, 36, 50, and 53*

**Alexander C. Allori, MD, MPH**
Assistant Professor of Surgery
Division of Plastic, Maxillofacial, and Oral Surgery
Duke University School of Medicine and Duke Children's Hospital
Durham, North Carolina
*Chapter 18*

**Nivaldo Alonso, MD, PhD**
Associate Professor
Faculda De Medicina Universidade de São Paulo
Responsible for Craniofacial Service–HRAC–BAURU
Universidade de São Paulo
São Paulo, Brazil
*Chapter 55*

**Mahmoud Issam Awad, MD**
Fellow in Facial Plastic and Reconstructive Surgery
Department of Otolaryngology-Head and Neck Surgery
Medical University of South Carolina
Charleston, South Carolina
*Chapter 27*

**Ruben E. Ayala, MD, MSc**
Chief Medical Officer
Operation Smile, Inc.
Virginia Beach, Virginia
President of the Permanent Council
The Global Alliance for Surgical, Obstetric, Trauma, and Anesthesia Care
Chicago, Illinois
*Chapter 49*

**America Ayuso Arce, DDS, MSc**
Private Practice
*Chapter 23*

**Babak Azizzadeh, MD, FACS**
Director
Center for Advanced Facial Plastic Surgery
Beverly Hills, California
*Chapter 38*

**Adriane L. Baylis, PhD, CCC-SLP**
Associate Professor-Clinical
Department of Plastic and Reconstructive Surgery
Nationwide Children's Hospital and The Ohio State University College of Medicine
Columbus, Ohio
*Chapter 3*

**Beatriz Berenguer, MD, PhD**
Assistant Professor
Division of Pediatric Plastic Surgery
Hospital General Universitario Gregorio Marañón
Madrid, Spain
*Chapter 37*

**Mikaela M. Bow, BSp Path, CPSP**
Speech Pathologist
Global Smile Foundation
University of Sydney
Sydney, Australia
*Chapters 39 and 40*

**Corstiaan Breugem, MD, PhD**
Professor in Plastic Surgery
Emma Children's Hospital, Amsterdam University Medical Center
University of Amsterdam
Amsterdam, Netherlands
*Chapter 53*

**Hugh T. Brewster, MEd**
Executive Director
Transforming Faces
Toronto, Ontario
*Chapter 49*

**Christopher J. M. Brooks, MD, FACS**
Chief of Plastic Surgery
Vice Chief of Surgery
Joe DiMaggio Children's Hospital
Hollywood, Florida
*Chapter 36*

**Anne M. Burrows, PhD**
Professor
Department of Physical Therapy
Duquesne University
Pittsburgh, Pennsylvania
*Chapter 1*

**Anna Rose Carlson, MD**
Fellow in Craniofacial Surgery
The Children's Hospital of Philadelphia
Division of Plastic and Reconstructive Surgery
Philadelphia, Pennsylvania
*Chapter 37*

**Karen Z. Carver, DDS, MD, MPH**
Cleft and Craniofacial Fellow
Department of Plastic and Oral Surgery
Boston Children's Hospital
Boston, Massachusetts
*Chapter 30*

**Elsa M. Chahine, MD**
Global Smile Foundation
Norwood, Massachusetts
*Chapters 17, 50, 51, and 52*

**Yu-Ray Chen, MD**
Professor
Department of Plastic and Reconstructive Surgery
Chang Gung University and Chang Gung Memorial Hospital
Taoyuan, Taiwan
*Chapter 33*

**Philip Kuo-Ting Chen, MD**
Professor in Surgery, Taipei Medical University
Director, Craniofacial Center
Taipei Medical University Hospital
Taipei, Taiwan
*Chapter 45*

**David K. Chong, MBBS, FACS**
Plastic and Maxillofacial Department
Royal Children's Hospital
Melbourne, Australia
*Chapter 15*

**Jennifer Co-Vu, MD, FACC, FAAP**
Associate Professor of Pediatrics, Radiology,
Obstetrics, and Gynecology
Director, Fetal Cardiac Program and Single Ventricle Program
University of Florida
Gainesville, Florida
*Chapter 11*

**Ryan J. Cody, DDS**
Dentist and Orthodontic Resident
Department of Orthodontics
University of Texas Health Science Center at Houston
Houston, Texas
*Chapter 29*

**Caitlin E. Cummings, MA, CCC-SLP**
Speech-Language Pathologist
Department of Speech Pathology
Nationwide Children's Hospital
Columbus, Ohio
*Chapter 3*

**Dima G. Daaboul, MD**
Associate in Cardiac Anesthesia
Department of Anesthesiology, Critical Care, and Pain Medicine
Instructor of Anesthesia, Harvard Medical School
Boston Children's Hospital
Boston, Massachusetts
*Chapter 10*

**Gareth Davies, BA(Hons)**
Executive Director
European Cleft Organisation
Rijswijk, Netherlands
*Chapter 49*

**James A. Davis Jr., DMD**
Maxillofacial Prosthodontist
Oro-facial Center
Adjunct Professor of Emory University
Department of Oral and Maxillofacial Surgery
Emory, Georgia
*Chapter 35*

**Tiago Turri de Castro Ribeiro, PhD, MSc, DDS**
Orthodontist
Department of Orthodontics
Hospital for Rehabilitation of Craniofacial Anomalies
University of São Paulo (HRAC-USP)
Bauru, São Paulo, Brazil
*Chapter 29*

**Gaurav Deshpande, MDS**
In-Charge, "Maaya" Cleft and Craniofacial Unit
MGM Hospital
Navi Mumbai, India
Associate Professor, Department of Oral and Maxillofacial Surgery
MGM Dental College and Hospital
Navi Mumbai, India
Clinical Assistant Professor
Division of Plastic and Reconstructive Surgery
Penn State Hershey School of Medicine
Hershey, Pennsylvania
*Chapter 54*

**J. Peter W. Don Griot**
Plastic Surgeon, Director of the Cleft Program Amsterdam
Department of Plastic, Reconstructive and Hand Surgery
Amsterdam UMC, location AMC
Amsterdam, The Netherlands
*Chapter 53*

**Briana Kelly Dornan, AuD, CCC-A**
Audiologist
Boston Children's Hospital
Boston, Massachusetts
*Chapter 4*

**Marcia R. Pérez Dosal, MD, MSc, PhD**
Assistant Professor
Department of Plastic Surgery
Pediatric National Institute
Mexico City, Mexico
*Chapter 23*

**Stephanie J. Drew, DMD, FACS**
Associate Professor Department of Surgery
Division of Oral and Maxillofacial Surgery
Emory School of Medicine
Atlanta, Georgia
*Chapter 35*

**Elcin Esenlik, DDS, PhD**
Professor Doctor and Chair of the Orthodontic Department
Faculty of Dentistry
Akdeniz University
Antalya, Turkey
*Chapter 32*

**David Malcolm Fisher, MB, BCh, FRCSC, FACS, BFA**
Plastic Surgeon, Hospital for Sick Children
Professor, Department of Surgery
University of Toronto
Toronto, Ontario
*Chapter 15*

**Roberto L. Flores, MD**
Joseph G. McCarthy Associate Professor of Reconstructive Plastic Surgery
Director, Cleft Lip and Palate
Hansjorg Wyss Department of Plastic Surgery
New York University Langone Health
New York, New York
*Chapter 14*

**Barry H. Grayson, DDS**
Clinical Associate Professor of Plastic Surgery (Craniofacial Orthodontics)
Department of Plastic Surgery
New York University Langone Medical Center
New York, New York
*Chapter 7*

**Usama S. Hamdan, MD, FICS**
President and Cofounder, Global Smile Foundation
Norwood, Massachusetts
*Chapters 12, 17, 28, 36, 38, 49, 50, 51, 52, 53, and 56*

**Oksana A. Jackson, MD**
Associate Professor of Surgery, Division of Plastic Surgery
Perelman School of Medicine at the University of Pennsylvania
The Children's Hospital of Philadelphia
Philadelphia, Pennsylvania
*Chapters 42 and 43*

**Daniel Jaffurs, MD, PhD**
Clinical Professor and Division Chief of Pediatric Plastic Surgery
Department of Plastic Surgery
University of California, Irvine
Orange, California
Children's Hospital of Orange County
Orange, California
*Chapter 50*

**Adam B. Johnson, MD, PhD**
Assistant Professor of Pediatric Otolaryngology
Department of Otolaryngology and Head and Neck Surgery
University of Arkansas for Medical Sciences
Arkansas Children's Hospital
Little Rock, Arkansas
*Chapter 17*

**Deborah S. F. Kacmarynski, MD, MS**
Paul N. Johnson Associate Professor of Craniofacial Abnormalities
Department of Otolaryngology-Head and Neck Surgery
University of Iowa
Iowa City, Iowa
*Chapter 46*

**Melissa D. Kanack, MD**
Resident Physician Department of Plastic Surgery
University of California, Irvine
Orange, California
*Chapter 5*

**Rami S. Kantar, MD, MPH**
Global Smile Foundation
Norwood, Massachusetts
Department of Plastic and Reconstructive Surgery
Amsterdam University Medical Center
Amsterdam, Netherlands
Surgery Resident, Department of Surgery
The University of Maryland Medical System/Shock Trauma Center
Baltimore, Maryland
*Chapters 12, 17, 28, 50, 53, and 56*

**Serena N. Kassam, DMD**
Assistant Clinical Professor
Department of Pediatric Dentistry
New York University College of Dentistry
New York, New York
Clinical Assistant Professor
Division of Pediatric Dentistry
Faculty of Dentistry
University of British Columbia
Vancouver, British Columbia
Clinical Attending
Department of Pediatric Dentistry
British Columbia Children's Hospital
Vancouver, British Columbia
Director Pre-Surgical and Dental Division
Global Smile Foundation
Norwood, Massachusetts
*Chapters 7, 9, and 49*

**Jennifer M. Kohler, MS, RD, LD**
Pediatric Clinical Dietitian
Cincinnati Children's Hospital Medical Center
Cincinnati, Ohio
*Chapter 2*

**Pete G. Kovatsis, MD, FAAP**
Senior Associate
Department of Anesthesiology
Critical Care and Pain Medicine
Boston Children's Hospital
Assistant Professor of Anesthesia
Harvard Medical School
Boston, Massachusetts
*Chapter 10*

**Ann W. Kummer, MAT, PhD, CCC-SLP, FASHA**
Professor of Clinical Pediatrics and Otolaryngology
University of Cincinnati College of Medicine
Cincinnati, Ohio
*Chapters 39 and 40*

**Suying Lam, MD**
Congenital and Pediatric Cardiology
Golisano Children's Hospital of Southwest Florida
Fort Myers, Florida
*Chapter 11*

**Jakob B. Lattanzi, BS**
Department of Biology
Tufts University

Medford, Massachusetts
*Chapter 34*

**Daniel Levy-Bercowski, DDS, MS, MSD**
Professor
Department of Orthodontics
The Dental College of Georgia, Augusta University
Augusta, Georgia
*Chapter 29*

**Yu-Fang Liao, DDS, PhD**
Professor
Department of Craniofacial Orthodontics
Chang Gung Memorial Hospital
Graduate Institute of Dental and Craniofacial Science
Chang Gung University
Taoyuan, Taiwan
*Chapter 33*

**Jan Lilja, MD, DDS, PhD**
Associate Professor
Department of Plastic Surgery
Sahlgrenska University Hospital
University of Gothenburg
Gothenburg, Sweden
*Chapter 26*

**Joseph Lopez, MD, MBA**
Craniomaxillofacial Fellow
Clinical Instructor
Yale-New Haven Children's Hospital
Yale School of Medicine
New Haven, Connecticut
*Chapter 34*

**Joseph E. Losee, MD, FACS, FAAP**
Dr. Ross H. Musgrave Endowed Chair in Pediatric Plastic Surgery
Associate Dean for Faculty Affairs
University of Pittsburgh School of Medicine
Professor and Executive Vice Chair
Department of Plastic Surgery
Division Chief
Pediatric Plastic Surgery
UPMC Children's Hospital of Pittsburgh
Pittsburgh, Pennsylvania
*Chapter 22*

**David W. Low, MD**
Professor of Surgery
Division of Plastic Surgery
Perelman School of Medicine at the University of Pennsylvania
The Children's Hospital of Philadelphia
Philadelphia, Pennsylvania
*Chapters 42 and 43*

**Tingchen Lu, MD**
Assistant Professor
Craniofacial Center
Department of Plastic and Reconstructive Surgery
Chang Gung Memorial Hospital
Linkou, Taiwan
*Chapter 33*

**Christopher Ma**
DMD Candidate
Harvard School of Dental Medicine
Boston, Massachusetts
*Chapter 8*

**James K. MacLaine, BDS, FDS Orth RCSEd**
Craniofacial Orthodontist
Boston Children's Hospital
Instructor
Harvard School of Dental Medicine
Boston, Massachusetts
*Chapter 8*

**Laura Mancini, DMD, MSD, FRCD(C)**
Craniofacial Orthodontist
CHU Sainte-Justine Hospital
Private Practice Ortho NDG
Montreal, Quebec, Canada
*Chapter 32*

**Robert Joseph Mann, MD**
Assistant Clinical Professor
Surgery and Pediatrics
Michigan State University
College of Human Medicine
Emeritus Director
Oral Cleft Program
Helen DeVos Children's Hospital
Grand Rapids, Michigan
*Chapter 25*

**Hans Mark, MD, PhD**
Associate Professor
Department of Plastic Surgery
Sahlgrenska University Hospital
Institute of Clinical Sciences at Sahlgrenska Academy
University of Gothenburg
Gothenburg, Sweden
*Chapter 26*

**Alexander P. Marston, MD**
Assistant Professor
Department of Otolaryngology-Head and Neck Surgery
Tufts University School of Medicine
Tufts Medical Center
Boston, Massachusetts
*Chapter 51*

**John G. Meara, MD, DMD, MBA**
Plastic Surgeon-in-Chief
Boston Children's Hospital
Kletjian Professor of Global Surgery
Harvard Medical School
Boston, Massachusetts
*Chapter 24*

**Antonio Melhem, MD**
Postdoctoral Research Fellow
Global Smile Foundation
Norwood, Massachusetts
*Chapters 12, 50, 52, 53, and 56*

**Claire Kane Miller, PhD, MHA**
Program Director
Aerodigestive and Esophageal Center
Cincinnati Children's Hospital Medical Center
Cincinnati, Ohio
*Chapter 2*

**Kerry A. Morrison, MD**
Plastic Surgery Resident Physician
Hansjörg Wyss Department of Plastic Surgery
New York University Langone Health
New York, New York
*Chapter 14*

**Amr M. Moursi, DDS, PhD**
Professor and Chair
Department of Pediatric Dentistry
College of Dentistry, New York University
New York, New York
*Chapter 9*

**Jennifer Munoz, MD**
Associate Professor
Pediatric Critical Care
University of Florida
Gainesville, Florida
*Chapters 11 and 53*

**Marie Nader, MD**
Pediatric Critical Care Fellow
Yale-New Haven Hospital
New Haven, Connecticut
*Chapters 11 and 53*

**Douglas P. Olson, DMD, MS**
Director
Children's Hospital of Orange County
Children's Division of Craniofacial Orthodontics
Orange, California
*Chapter 31*

**Bonnie L. Padwa, DMD, MD**
Oral Surgeon-in-Chief
Department of Plastic and Oral Surgery
Boston Children's Hospital
Professor of Oral and Maxillofacial Surgery
Harvard School of Dental Medicine and Harvard Medical School
Boston, Massachusetts
*Chapter 30*

**Vikram S. Pandit, BDS, MDS**
Consultant Oral and Maxillofacial Surgeon
Pandit Clinic
Pune, Maharashtra, India
*Chapter 45*

**Krishna G. Patel, MD, PhD**
Professor
Director of Facial Plastic and Reconstructive Surgery
Medical University of South Carolina
Charleston, South Carolina
*Chapter 27*

**Connor J. Peck, BS**
Department of Surgery, Division of Plastic Surgery
Yale School of Medicine
New Haven, Connecticut
*Chapter 34*

**Grace Lee Peng, MD, FACS**
Facial Plastic and Reconstructive Surgery
Beverly Hills, California
*Chapter 38*

**Valerie J. Pereira, PhD**
Senior Lecturer
Department of Otorhinolaryngology-Head and Neck Surgery
Faculty of Medicine
The Chinese University of Hong Kong

Hong Kong SAR, China
*Chapter 41*

**Jamie L. Perry, PhD, CCC-SLP**
Department Chair and Professor
Department of Communication Sciences and Disorders
East Carolina University
Greenville, North Carolina
*Chapter 49*

**Martin J. Persson, PhD, MPH, MSc**
Professor of Health Sciences
Faculty of Health Sciences
Kristianstad University
Kristianstad, Sweden
*Chapters 47 and 48*

**Miles J. Pfaff, MD, MHS**
Cleft and Craniofacial Surgery Fellow
Clinical Instructor
UPMC Children's Hospital of Pittsburgh
Pittsburgh, Pennsylvania
*Chapter 22*

**Elie P. Ramly, MD**
Surgery Resident
Department of Surgery
Oregon Health and Science University
Portland, Oregon
*Chapter 51*

**Cassio Eduardo Raposo-Amaral, MD, PhD**
Medical Director Hospital SOBRAPAR
President of Brazilian Association of Craniofacial Surgery
Professor of Neurosurgery Department University of Campinas–UNICAMP
Vice President of Latin American Craniofacial Association (LATICFA)
Campinas, São Paulo, Brazil
*Chapter 19*

**Cesar Augusto Raposo-Amaral, MD**
Plastic Surgeon
SOBRAPAR's Clinical Director
Campinas, São Paulo, Brazil
*Chapter 19*

**Tamara Rhodes, MS, RD, CSP, LD**
Pediatric Clinical Dietitian
Cincinnati Children's Hospital Medical Center
Cincinnati, Ohio
*Chapter 2*

**Matthew Ridley, PhD**
Chartered Research Psychologist
Norton, Stockton-on-Tees, England
*Chapters 47 and 48*

**Carolyn R. Rogers-Vizena, MD**
Attending in Plastic and Oral Surgery
Boston Children's Hospital
Assistant Professor
Harvard Medical School
Boston, Massachusetts
*Chapters 1, 8, 18, and 24*

**Steven M. Roser, DMD, MD, FACS, FRCS(Ed)**
DeLos Hill Chair and Professor of Surgery
Division of Oral and Maxillofacial Surgery
Department of Surgery
Emory University School of Medicine
Atlanta, Georgia
*Chapter 35*

**Elizabeth J. Ross, DDS, FRCS(C)**
Department of Dentistry
Boston Children's Hospital
Boston, Massachusetts
*Chapter 8*

**Monica K. Rossi Meyer, MD**
Resident Physician
Department of Otolaryngology-Head and Neck Surgery
University of Iowa
Iowa City, Iowa
*Chapter 46*

**Pedro E. Santiago, DMD, MBA**
Director of Oral Health
Director of Craniofacial Orthodontics
Division of Plastic, Maxillofacial and Oral Surgery
Duke University
Durham, North Carolina
*Chapters 7 and 29*

**Debbie Sell, OBE, PhD**
Senior Research Fellow
Independent Consultant Speech and Language Therapist
Codirector, Speech @ Home
London, United Kingdom
*Chapter 41*

**Navil F. Sethna, MD, MA, FAAP**
Professor of Anesthesia and Clinical Director
Mayo Family Pediatric Pain Rehabilitation Center
Department of Anesthesiology, Pain Medicine, and Critical Care
Harvard Medical School
Boston, Massachusetts
*Chapter 10*

**Nikhil Shah, BS**
Northwestern University Feinberg School of Medicine
Chicago, Illinois
*Chapter 50*

**Pradip R. Shetye, DDS, BDS, MDS**
Assistant Professor and Program Director (Craniofacial Orthodontics)
Hansjorg Wyss Department of Plastic Surgery
New York University Langone Health
New York, New York
*Chapter 31*

**Taylor Snodgrass, MS, CCC-SLP**
Speech-Language Pathologist and Research Assistant
Department of Communication Sciences and Disorders
East Carolina University
Greenville, North Carolina
*Chapter 49*

**Brian C. Sommerlad, MB, BS, FRCS Eng, FRCS Ed(Hon), DSc(Med)UCL(Hon), FRCPCH, FRCSLT(Hon)**
Honorary Consultant Plastic Surgeon
Great Ormond Street Hospital for Children NHS Foundation Trust
London, United Kingdom
*Chapters 16, 20, 21, and 44*

**Derek Steinbacher, DMD, MD, FACS**
Professor of Plastic Surgery
Chief of Oral and Maxillofacial Surgery
Chief of Dentistry, Director of Craniofacial Surgery
Department of Plastic and Reconstructive Surgery
Yale School of Medicine
New Haven, Connecticut
*Chapter 34*

**Erin Stieber, JD**
Senior Vice President, Programs
Smile Train
New York, New York
*Chapter 49*

**Jordan W. Swanson, MD, MSc**
Assistant Professor
University of Pennsylvania
Division of Plastic Surgery
Children's Hospital of Philadelphia
Philadelphia, Pennsylvania
*Chapter 54*

**Daniela Y. S. Tanikawa, PhD, MD**
Assistant Professor
Division of Plastic Surgery
University of São Paulo School of Medicine
Attending Physician
Cleft Lip and Palate Program
Hospital Municipal Infanti Menino Jesus
Instituto de Responsabilidade Social Sírio-Libanês
Research Physician
Instituto Sírio-Libanês de Ensino e Presquisa
Member, Brazilian Medical Advisory Council of Smile Train
São Paulo, Brazil
*Chapter 6*

**Jesse A. Taylor, MD**
Peter Randall Endowed Professor and Chief
Division of Plastic Surgery
The Children's Hospital of Philadelphia
Philadelphia, Pennsylvania
*Chapter 37*

**Brenda K. Thompson, MA, CCC-SLP, BCS-S, CNT**
Clinical Manager
Division of Speech-Language Pathology
Cincinnati Children's Hospital Medical Center
Cincinnati, Ohio
*Chapter 2*

**Cristiano Tonello, PhD, MD**
Doctor Professor and Hospital Director
Hospital for Rehabilitation of Craniofacial Anomalies
University of São Paulo
Bauru, São Paulo, Brazil
*Chapter 55*

**Raymond W. Tse, MD, FACS**
Division of Craniofacial and Pediatric Plastic Surgery
Seattle Children's Hospital

Associate Professor
University of Washington
Seattle, Washington
*Chapter 15*

**Charanya Vijayakumar, BDS, MDS**
Senior Lecturer in Cleft and Craniofacial Surgery
Cleft and Craniofacial Center
Department of Plastic and Reconstructive Surgery
Sri Ramachandra Institute of Higher Education and Research (SRIHER)
Porur, Chennai, India
*Chapter 50*

**Rafael Villaseñor Caloca, MD, MHA**
Assistant Professor
Department of Plastic and Reconstructive Surgery
Instituto Nacional de Pediatría
Mexico City, Mexico
*Chapter 23*

**Raj M. Vyas, MD, FACS**
Vice-Chairman and Professor of Plastic Surgery
Department of Plastic Surgery
University of California, Irvine
Irvine, California
Chief
Children's Division of Pediatric Plastic Surgery
Children's Hospital of Orange County
Orange, California
*Chapters 5, 13, 50, 51, 52, 53, and 56*

**Chuan-Fong Yao, MD**
Craniofacial Center
Department of Plastic and Reconstructive Surgery
Chang Gung Memorial Hospital
Linkou, Taiwan
*Chapter 33*

**Touran M. Zadeh, MD**
Division of Medical Genetics
Children's Hospital of Orange County Genetics Center
Orange, California
*Chapter 5*

**Neda Zadeh, MD**
Division of Medical Genetics
Children's Hospital of Orange County Genetics Center
Orange, California
*Chapter 5*

# 1

# Embryology, Anatomy, and Classification of Cleft Lip and Palate

Carolyn R. Rogers-Vizena and Anne M. Burrows

## Introduction

Shape and form of the human face is rooted in early embryonic development. Similarly, cleft lip and/or palate (CL/P) begins with malformation early in embryonic development that persists as tissues differentiate and development progresses. This chapter will provide the embryologic and anatomic foundation necessary to understand CL/P pathology. In addition, common classification systems used to describe and document the extent of anatomic findings in CL/P will be presented to foster consistent language for communicating CL/P phenotype.

## Embryology

Knowledge of normal facial embryology is important for understanding the underlying basis of craniofacial anomalies such as CL/P. Basic morphology of the face is established between the 4th and 8th embryonic weeks, developing from five primordia surrounding a central stomodeum (primitive mouth). These primordia include a single midline frontonasal prominence (FNP), paired maxillary prominences, and paired mandibular prominences. These prominences undergo mostly symmetrical growth followed by subsequent fusion to form the face.

### Development of the Face

During the 4th week, the stomodeum invaginates, and nasal pits, or placodes, form in the FNP. In the 5th week, medial and lateral nasal processes form around the nasal pits (Figure 1–1). The FNP gives rise to the nose, philtrum, primary palate, perpendicular plate of the ethmoid bone and vomer (nasal septum), cribriform plates, and forehead. The medial nasal processes go on to form the nasal tip, columella, philtrum, and premaxilla/primary palate (central portion of the alveolus and anterior hard palate, including the central and lateral incisors). The lateral nasal processes form the nasal alae.

Concomitant with FNP development, the first pharyngeal arch forms paired maxillary prominences that enlarge and migrate ventrally. The maxillary prominences give rise to the upper cheek and most of the upper lip, maxilla, zygoma, and secondary palate. The medial nasal processes and maxillary prominences fuse to form a continuous upper lip and primary palate by the end of the 6th week (Sperber et al., 2001). Disrupted fusion of the medial nasal process and maxillary prominence results in cleft lip and alveolus (Larrabee et al., 2004).

### Development of the Palate

The palate develops between the 6th and 12th weeks from a single midline nasal septum originating on the

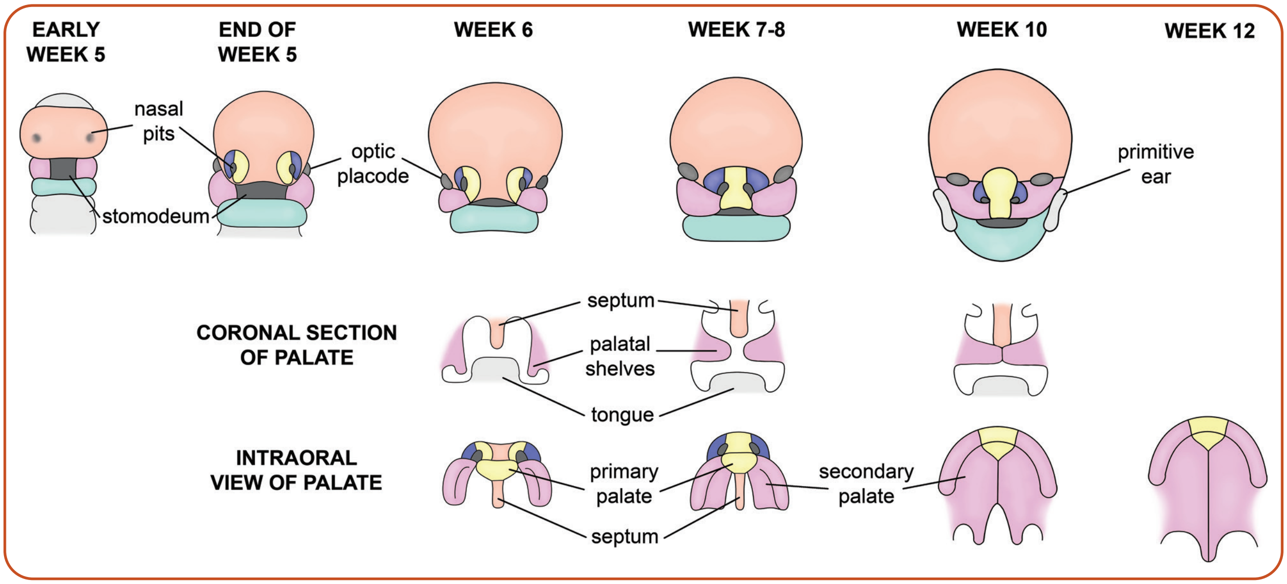



**FIGURE 1–1.** Embryologic and fetal facial development. The face forms from the frontonasal prominence (FNP, orange), paired maxillary prominences (pink), and paired mandibular prominences (green). At the beginning of week 5, the mandibular prominences have merged, a stomodeum begins to invaginate, and nasal pits start forming in the FNP. By the end of week 5, medial (yellow) and lateral (blue) nasal processes form around each invaginating nasal pit and the optic placodes form. In week 6, the medial nasal process and maxillary prominence fuse and the stomodeum narrows as the maxillary and mandibular prominences fuse. A primitive septum forms from the FNP, and vertically oriented palatal shelves form from the maxillary prominence. During weeks 7 to 8, primitive eyes migrate medially, the medial nasal processes fully form the philtrum and primary palate, palatal shelves elevate, and the primary palate begins to fuse with the secondary palate. By week 10, primitive ears have formed from the first and second branchial arches and migrate superolaterally and the palatal shelves fuse with each other and the developing septum in an anteroposterior direction. By week 12, the secondary palate has fully fused.

FNP and paired lateral palatine processes originating on the maxillary prominences (see Figure 1–1). The palatine processes grow inferiorly, lateral to the developing tongue. As the maxilla and mandible develop and enlarge, the tongue moves caudally, permitting the palatine processes to elevate into a horizontal position (known as "palatal shelves") during the 7th and 8th weeks. The palatal shelves fuse with the primary palate in a Y-shaped configuration, with the apex forming the incisive foramen in the 6th and 7th weeks. Posterior to the incisive foramen, the palatal shelves fuse with each other and with the nasal septum starting anteriorly and extending posteriorly beginning around the 8th week and ending by the 12th week. Ossification of the primary palate and anterior secondary palate forms the hard palate. The posterior secondary palate does not ossify and forms the soft palate and uvula (Sperber et al., 2001). Failure of the palatal shelves to elevate and fuse at the midline results in a cleft palate (Larrabee et al., 2004).

## Anatomy

### Normal Lip and Nasal Anatomy

Upper lip surface anatomy reflects its embryologic formation and underlying muscular anatomy. Understanding key surface landmarks is critical for properly designing a cleft lip repair (Figure 1–2). The columellar-labial junction is the intersection between the columella and superior part of the upper lip. Inferior to this is the philtral dimple, a cutaneous indentation in the center of the upper lip. The philtral ridges are raised areas of skin flanking the philtral dimple on either side. They extend inferiorly to elevated areas of vermilion called the peaks of Cupid's bow. Between these two elevated areas is a vermillion depression, the low point or nadir of Cupid's bow. The white roll is a raised area of skin just superior to the vermillion. The red line represents the transition from dry,

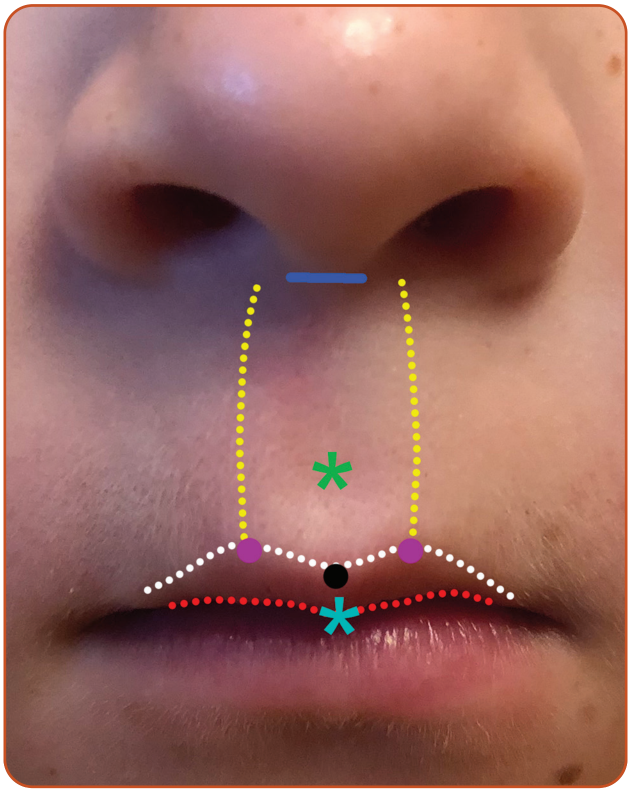


**FIGURE 1–2.** Surface anatomy of the lip. Key landmarks include columellar-labial junction (blue line), philtral dimple (green asterisk), philtral ridges (yellow dotted line), high points of Cupid's bow (purple dots), low point of Cupid's bow (black dot), white roll (white dotted line), red line (red dotted line), and median tubercle (teal asterisk).

keratinized vermillion to labial mucosa. Finally, the median tubercle is the prominence at the most inferior extent of the upper lip.

In a normal upper and lower lip, the orbicularis oris muscle (OOM) extends from modiolus to modiolus (the confluence of facial muscles lateral to the oral commissure), creating a muscular sphincter. There are two major OOM subdivisions in the upper lip, the pars marginalis (inferior) and the pars peripheralis (superior). Pars marginalis is a continuous band of muscle fibers while pars peripheralis fibers decussate at the midline with fibers from the contralateral side inserting deep to dermis of the contralateral philtral ridge (Figure 1–3). This forms the underlying muscular architecture of the philtral ridges and dimple but is not the only factor contributing to philtral shape. Thickened dermis and connective tissue are present at the ridges, and there is a paucity of connective tissue at the dimple. In cross section, the configuration of the OOM is a subtle "J" shape with the distal tip of the "J" lying deep to the white roll and contributing to its raised appearance (Rogers et al., 2014).

In the normal nose, three-dimensional shape is primarily supported by the paired lower and upper lateral cartilages as well as the single midline cartilaginous septum (see Figure 1–3). The arch-shaped lower lateral cartilages are conceptualized as three sections. Inferiorly, the medial crura are narrow portions of the arch beginning superficial to the anterior nasal spine and extending along the inferior border

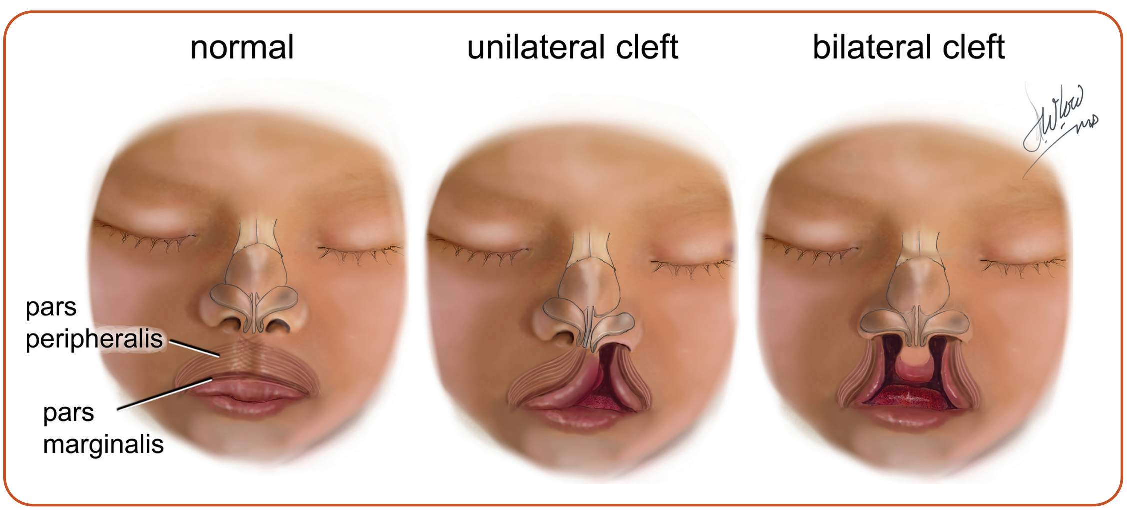



**FIGURE 1–3.** Orbicularis oris muscle and nasal cartilage anatomy. (Left) Normal anatomy. (Center) Unilateral cleft lip. (Right) Bilateral cleft lip.

of the septum, before bending laterally. This bend, or genu, is called the middle crus and supports the nasal tip. The cartilages widen, extending superolaterally from the genua as the lateral crura, providing soft tissue support to the lateral nasal tip and alae. The upper lateral cartilages lie superior to the lower lateral cartilages and extend laterally from the septum supporting the middle vault of the nose. The midline nasal septum supports the dorsum of the nose and separates the two nasal vaults. It intersects posteriorly with the perpendicular plate of the ethmoid bone (superior) and the vomer (inferior), which merges with the hard palate (Fisher & Mann, 1998).

The major blood supply to the upper lip and nose arises from branches of the paired facial arteries (Figure 1–4). The upper lip is primarily supplied by the superior labial branches. Beyond the superior labial branch point, the facial artery continues as the lateral nasal artery to supply to the nasal tip. The superior labial arteries anastomose with each other at the midline and give off septal and columellar branches. The columellar branches anastomose with the dorsal and lateral nasal arteries. The clinical significance of this rich anastomotic network is that the lip and nose remain well perfused despite disruption of major vascular branches.

Sensory innervation of the lip and nose comes from branches of the trigeminal nerve (cranial nerve V), primarily the infraorbital branch of the maxillary nerve, but with contribution from the external nasal branch of the ophthalmic nerve. These branches are amenable to regional anesthetic block during cleft lip repair (see Chapter 12).

The skeletal structures most relevant to a cleft lip are portions of the maxilla (Figure 1–5). These form the inferior borders of the piriform aperture that encompasses the nasal cavity. Anteriorly, a midline outcropping called the anterior nasal spine supports the nasal tip, columella, and superior upper lip. The maxilla also comprises the tooth-bearing alveolus and anterior hard palate.

## Cleft Lip/Nasal Anatomy

Cleft lip involves structures of the lip, nose, and primary palate. Nasolabial tissue is malformed, rather than deficient. In a unilateral cleft, the nasal tip and ala on the cleft side are displaced inferiorly and posterolaterally, corresponding to deformity of the underlying lower lateral cartilage and maxilla. The arch of the genu is widened and lateral crus inferiorly and posterolaterally positioned (see Figure 1–3). In more severe unilateral clefts, the lateral crus has a downward deflection or "recurvatum." The anterior nasal spine and caudal septum deviate away from the cleft and the mid-septum bows toward the cleft (Fisher & Sommerlad, 2011). The lip is divided where the philtral ridge should be, extending through the peak of Cupid's bow and vermillion. This division may be incomplete, including only the inferior margin of the lip, or complete, extending all the way

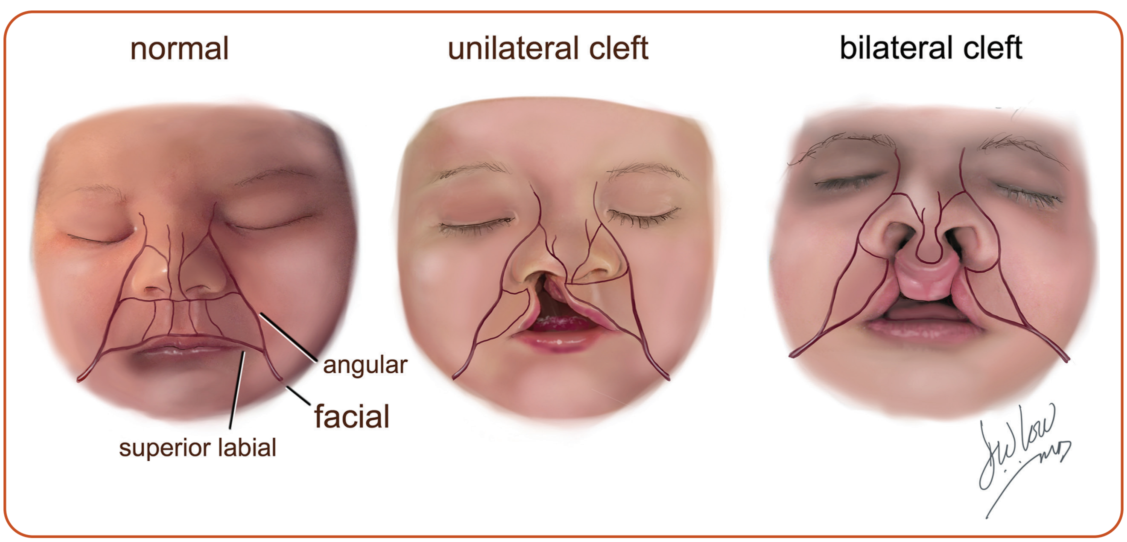



**FIGURE 1–4.** Lip/nasal vascular anatomy. (Left) Normal anatomy. (Center) Unilateral cleft lip. (Right) Bilateral cleft lip.

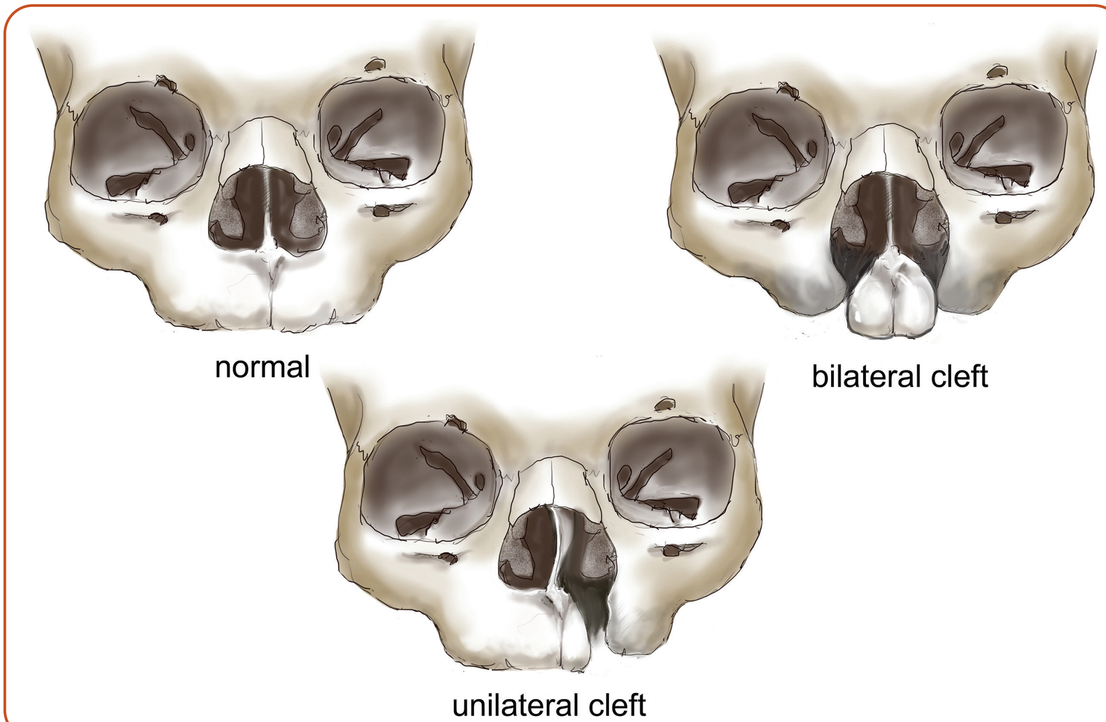



**FIGURE 1–5.** Skeletal anatomy. (Top left) Normal anatomy. (Bottom center) Unilateral cleft lip. (Top right) Bilateral cleft lip.

into the floor of the nose. On the labial surface, the vermillion narrows beyond where the peak of Cupid's bow should be, with the vermillion-cutaneous junction and vermillion-mucosal junction eventually converging along the margin of the cleft. In a complete cleft, the OOM is discontinuous, whereas there may be superior bridging fibers in an incomplete cleft. Orbicularis oris muscle fibers are oriented toward the alar base lateral to the cleft and columella medial to the cleft (see Figure 1–3). With increasing cleft severity, the philtral dimple effaces but remains present, in contrast to a bilateral cleft. The philtral ridge remains present on the noncleft side (Rogers et al., 2014). The superior labial artery is discontinuous, oriented along the cleft margin (see Figure 1–4). The underlying maxillary alveolus is protuberant on the noncleft side and may be collapsed on the cleft side (see Figure 1–5). The lateral incisor tooth is often absent on the cleft side, but particularly in the setting of an incomplete cleft, there may be an abnormal lateral incisor or supernumerary tooth.

In a bilateral cleft lip, the nasal tip is broad and flat, with both lower lateral cartilages having the abnormal configuration described for the cleft side in a unilateral cleft lip (see Figure 1–3). In a symmetric bilateral cleft lip, the septum and anterior nasal spine are midline, but in some cases, a bilateral cleft is asymmetric with corresponding deviation. The lip is divided at the area of the philtral ridge bilaterally. The central soft tissue element, the prolabium, has narrow vermillion and lacks a white roll in a complete cleft. The lateral lip vermillion deformity is analogous to the cleft side in a unilateral cleft lip. Orbicularis oris muscle fibers are oriented toward both alar bases laterally (see Figure 1–3). In a bilateral complete cleft lip, the prolabium lacks muscle tissue and the philtral dimple is completely effaced, whereas in a bilateral incomplete cleft lip, a small amount of OOM may bridge the prolabium and a subtle philtral dimple may be present. The superior labial arteries supply lateral lip elements, while the columellar and posterior septal arteries supply the prolabium (see Figure 1–4). Prolabial blood supply is critical to bear in mind when designing a bilateral cleft lip repair. When there is a complete alveolar cleft, the premaxilla is protuberant due to excessive anterior growth at the premaxillary-vomerine suture and the lateral alveolar segments collapsed (see Figure 1–5). Blood supply to the premaxilla comes primarily from branches of the posterior septal and anterior ethmoidal arteries, an important consideration during premaxillary setback. The lateral incisors may be absent, abnormal, or supernumerary teeth may be present bilaterally.

## Normal Palatal Anatomy

The anterior two thirds of the hard palate is formed by paired palatine processes of the maxilla. The posterior third, mesial to the tooth-bearing alveolus, is formed by the paired palatine bones. Superiorly, the hard palate and vomer articulate supporting the nasal floor. The incisive foramen is located at the midline, just posterior to the alveolus at the junction of the primary and secondary palate. Structures anterior to the incisive foramen are considered the primary palate, and structures posterior to it, the secondary palate.

The soft palate, or velum, is located posterior to the hard palate (Figure 1–6). It is a muscular sling formed by the paired levator veli palatini, palatopharyngeus, palatoglossus, and musculus uvuli, reinforced by the aponeurosis of the tensor veli palatini muscles. The levator in particular extends from the temporal bone into the velum and acts as the primary elevator of the soft palate during speech, whereas the palatoglossus and palatopharyngeus muscles elevate the posterior tongue and lateral pharyngeal walls while drawing the soft palate inferiorly to propel the food bolus during deglutition. Together with the superior constrictor muscle in the pharynx, these muscles form the velopharyngeal sphincter that provides dynamic separation of the oro- and nasopharynx during deglutition and speech. In addition, the tensor veli palatini acts as the principal dilator of the Eustachian tube, with further input from the levator veli palati, salpingopharyngeus, and tensor tympani muscles (Cho et al., 2013).

Palatal and velopharyngeal blood supply comes from branches of the paired maxillary arteries, ascending pharyngeal arteries, and ascending pala-

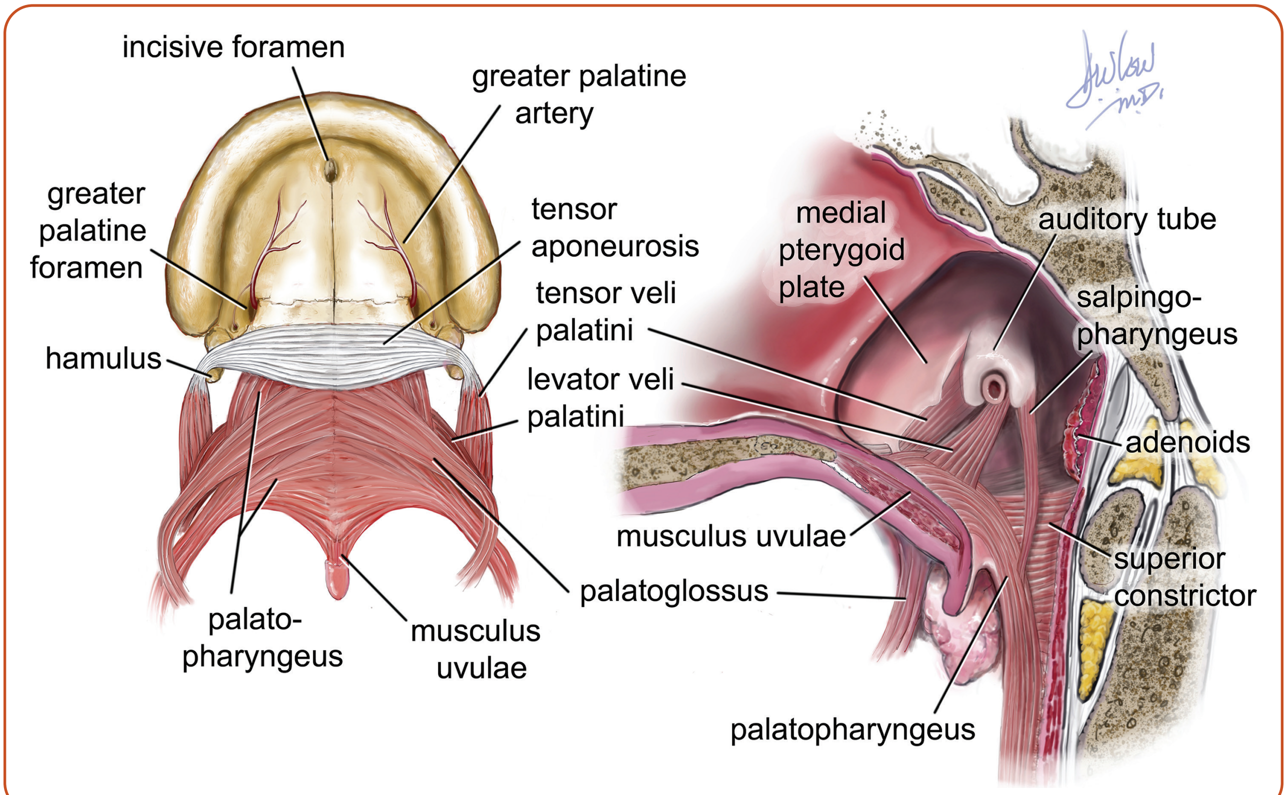



**FIGURE 1–6.** Palatal anatomy—normal anatomy from intraoral (left) and sagittal (right) views. Deep to the glandular submucosa of the soft palate, the palatoglossus and palatopharyngeus muscles enter. The levator enters deep to these muscles but interdigitates with them, approaching the midline to form the velopharyngeal sling. The tensor tendon wraps around the pterygoid hamulus, spreading into a broad aponeurosis approaching the midline. The musculus uvula is primarily located on the deep/nasal surface of the velopharyngeal sling, extending from the tensor aponeurosis to the uvula.

tine arteries. The soft palate has a rich blood supply from branches of these arteries. The hard palate mucoperiosteum is primarily supplied by the greater palatine branches of the maxillary artery, an important consideration during cleft palate repair.

Hard palate sensory innervation comes from the greater palatine and nasopalatine branches of the maxillary nerve while soft palate sensation is from the lesser palatine branch of the maxillary nerve. The maxillary nerve is a branch of the trigeminal nerve amenable to regional anesthetic block at the pterygopalatine fossa (see Chapter 10).

### Cleft Palate Anatomy

Cleft palate involves structures of the secondary palate. When the hard palate is cleft, the maxillary and palatine bones are separated both from the contralateral side and from the vomer, resulting in communication between the oro- and nasopharynx. When the soft palate is cleft, there is a midline separation of the velum (Figure 1–7). The tensor veli palatini aponeuroses, levator veli palatini, palatoglossus, and palatopharyngeus muscles are discontinuous with each other and disoriented anteriorly. The tensor aponeuroses inserts on the posterolateral hard palate, while the palatoglossus and palatopharyngeus muscles insert on the posteromedial hard palate and cleft edge. The levator runs parallel to the other velar muscles but fails to reach the midline (Fisher & Sommerlad, 2011). Thus, these muscles fail to interdigitate and form the dynamic sling needed to control oronasal airflow and facilitate Eustachian tube function.

Submucous cleft palate, a lesser form of cleft palate, involves muscular diastasis of the soft palate with intact mucosa. The diastasis may be visualized as a bluish "zona pellucida," or furrow, in the soft palate. Other findings may include bifid uvula and/or notch in the posterior hard palate (Calnan, 1954). This type of cleft is often diagnosed during evaluation of abnormal speech.

## Phenotypic Spectrum and Classification

The wide phenotypic variation of CL/P has inspired numerous systems for describing the extent of the defect. The classifications described here are not all-inclusive but are some of the most commonly used ways to document clefting.

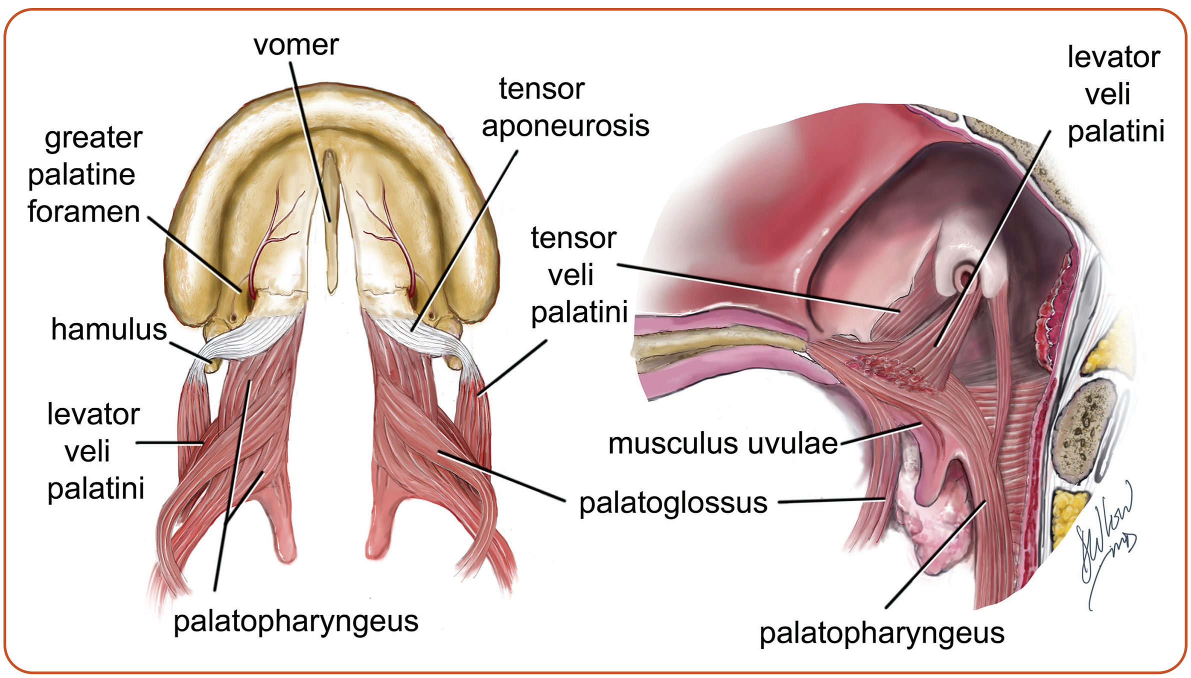



**FIGURE 1–7.** Palatal anatomy—cleft palate anatomy from intraoral (left) and sagittal (right) views. In addition to the cleft itself, key findings are anteromedial disorientation of the levator veli palatini, palatoglossus, and palatopharyngeus musles and tensor veli palatini aponeurosis without medial interdigitation. The musculus uvulae are small or absent.

## Cleft Lip

Cleft lip is broadly classified as unilateral (one-sided) or bilateral (two-sided) and further categorized by the extent of the discontinuity. Complete cleft lip involves a separation extending from the lip margin through the nasal floor. In an incomplete cleft lip, there is separation at the free margin of the lip with a superior connection, ranging from a thin skin bridge or band to a substantial musculocutaneous connection. Lesser forms of incomplete cleft lip involve smaller degrees of disruption of the vermillion-cutaneous junction at the peak of Cupid's bow: mini-microform (notch of the vermilion-cutaneous junction without elevation of the peak of Cupid's bow), microform (disruption of the free margin of the lip with <3 mm elevation of the peak of Cupid's bow), and minor form (disruption of the free margin of the lip with 3 to 5 mm elevation of the peak of Cupid's bow). Classifying a labial cleft according to these descriptive terms is useful for both verbally communicating the degree of clefting and determining extent of the lip repair (Yuzuriha & Mulliken, 2008).

## Cleft Palate

Cleft palate is often described using the Veau classification (Figure 1–8), allowing clinicians to communicate the anatomic nature of the defect. A Veau I cleft involves the soft palate only, a Veau II cleft includes the hard and soft palate, a Veau III extends unilaterally along the junction of the primary and secondary palate (i.e., a unilateral complete cleft lip and palate),

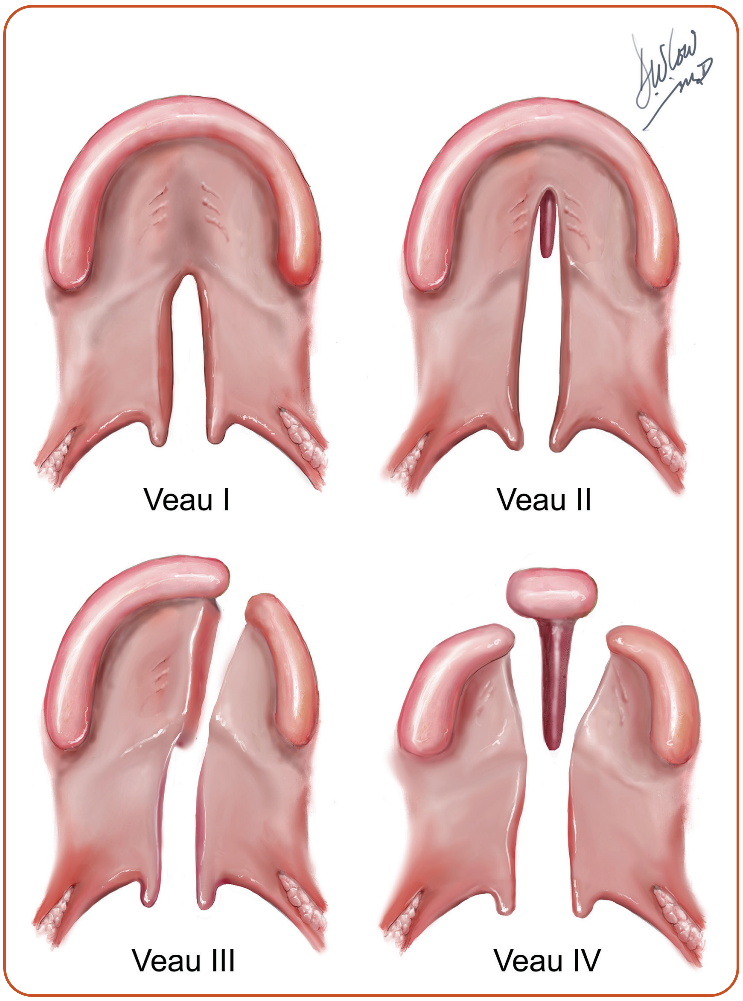



**FIGURE 1–8.** Veau classification of cleft palate. (Top left) Veau I. (Top right) Veau II. (Bottom left) Veau III. (Bottom right) Veau IV.