2 The Value of Longitudinal Facial and Dental Casts Records in Clinical Research and Treatment Analysis

Samuel Berkowitz

After 40 years of treating children with various types of clefts, this author has concluded that the success or failure of a surgical procedure depends on the degree of palatal cleft defect at the time of surgery and the resulting facial growth pattern as well as the surgical skills and the surgical procedure utilized. This conclusion will not be new to the experienced orthodontist who in all probability recognizes that the progress recorded in treatment depends, for the most part on the skeletal and facial growth patterns inherent in the patient and the interaction of surgery with facial and palatal growth.

S. Berkowitz, DDS, M.S., FICD
Adjunct Professor,
Department of Orthodontics,
College of Dentistry, University of Illinois,
Chicago, IL, USA

Clinical Professor of Surgery and Pediatrics (Ret),
Director of Research (Ret),
South Florida Cleft Palate Clinic,
University of Miami School of Medicine,
Miami, FL, USA

Consultant (Ret),
Craniofacial Anomalies Program,
Miami Children's Hospital,
Miami, FL, USA
e-mail: sberk3140@aol.com

2.1 Serial Cephaloradiographs and Casts of the Maxillary and Mandibular Dentition and Occlusion

To properly assess the results of treatment, there is a fundamental need for serial casts, lateral cephalometric films, and photographs in individual case reports.

Pruzansky (1953, 1955) often stated that it is unfortunate that plastic surgeons' training in the realm of clefts and their variations tends to be totally inadequate because their first encounters with patients usually occur in the clinic or operating room. Furthermore, there is seldom recourse to anatomical specimens to better appreciate the nature of the cleft deformity. The trainee is dependent on the empirical experience of his preceptor for knowledge of the natural history of the defect and long-term response to therapy. In most cases, other than before and after facial photographs, there are no objective records to determine why the outcome was a success or failure.

The collected serial data to be shown in this text will provide the clinician in training with an overview of the variations that can be encountered in each cleft type, the significance of genotype differences that influence growth and response to surgery, and the natural history of each cleft entity.

Over the years certain cephalometric measurements have become standardized and have

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been applied to selected population samples to develop statistical means or averages. In the treatment of cleft lip and/or palate, this approach has provided useful data in studying morphologic growth changes in the head, evaluating dentofacial abnormalities, and assessing responses to surgical and orthodontic treatment. The data has been particularly useful in determining the timing and type of procedure selected to treat individual problems. The measurements and analyses utilized are primarily profile-oriented and reveal both anteroposterior and vertical relationships of the various parts of the dentofacial complex.

To assess changes during the course of general growth and treatment, head radiographs of the same individual taken at separate times are traced and the tracings superimposed to ascertain the changes that have occurred. A common method is to register the two tracings at the point sella with the sella-nasion lines superimposed (Fig. 2.2a, b). This method provides a gross overview of changes in the dentofacial complex and in soft tissue but is useful only in evaluating what has already occurred. In this text, we also use the Coben superimposition procedure (basion horizontal) because it more accurately reflects actual craniofacial growth direction (Figs. 2.1d and 2.2c).

The use of "landmark," or baseline, images associated with the basicranium to show the composite results of facial growth can provide meaningful information because it is the enlargement of the face relative to the cranial base that is being evaluated. In the child, further growth changes in the anterior part of the cranial base slow considerably at about 5–6 years of age, whereas facial growth continues actively through adolescence or beyond. Comparing the relative growth between these two regions, rather than simply focusing on a single fixed point, provides clinically useful information when cephalometrically evaluated.

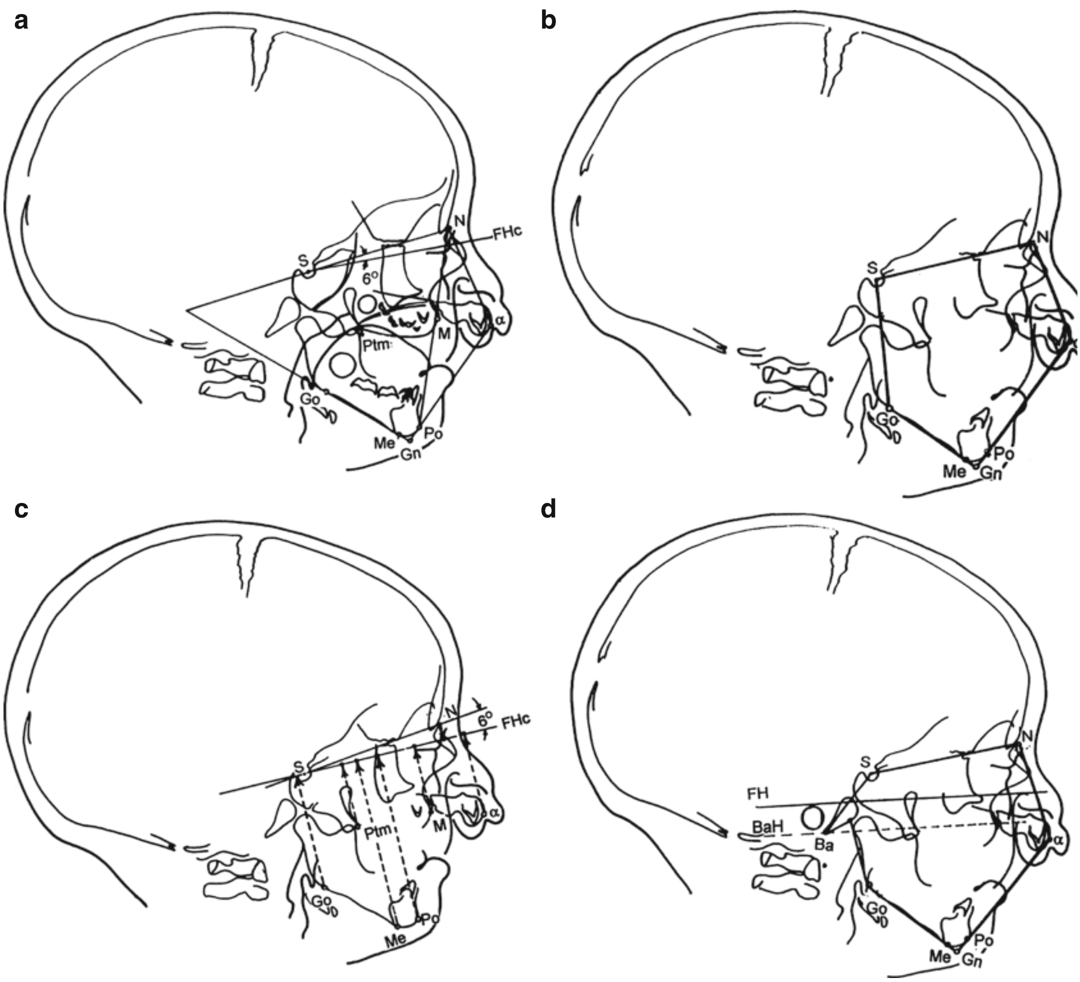

Fig. 2.1 (**a**–**d**) Various methods used to demonstrate facial changes using lateral cephalometrics. (**a**) Facial angles. These are just a few of the angles which describe changes in the skeletal profile. There are many more angles and linear measurements which can be used to relate the maxilla to the mandible and both jaws to the cranial base. (**b**) Facial polygon. This is a graphic method used to describe the boundaries of the skeletal face. (Pogonion constructed, Po', is the same point as gnathion.) Facial growth changes can be shown by superimposing each succeeding polygon on the anterior cranial base (*SN*) and registering on sella turcica (*S*). (**c**) Projecting facial landmarks to a constructed Frankfort horizontal line which is arbitrarily drawn 6° off the SN line. This angle can vary with steepness of the anterior cranial base. This graphic method will show the relative contribution of various structures within the maxilla and the mandible to the profile. (**d**) Basion horizontal created facial polygon (Coben 1986). This method of superimposing tracings graphically reflects his overall concept of fixed growth. A plane at the level of the anterior border of foramen magnum (basion) parallel to Frankfort horizontal where Basion is the point of reference for the analyses of craniofacial growth

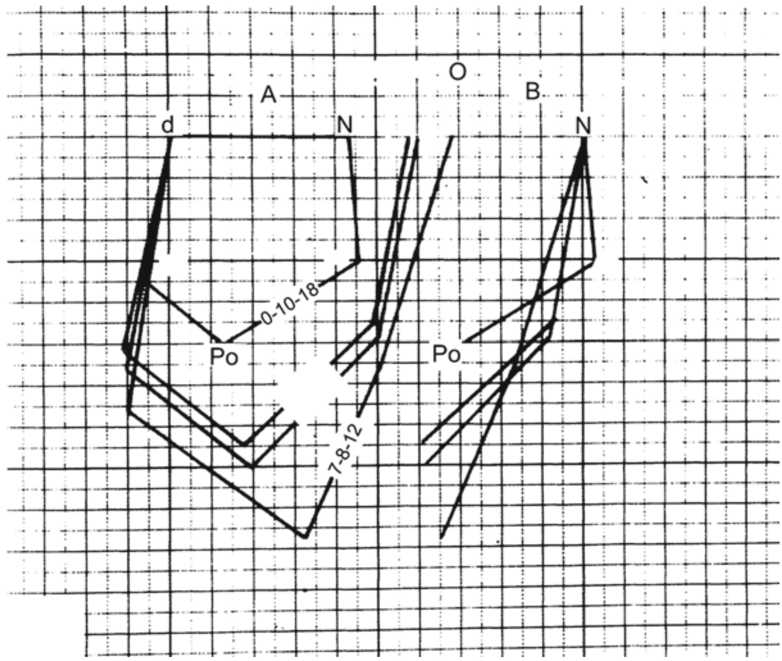

Fig. 2.2 (**a**) Case CP #127 (CPCLP). Superimposed facial polygons from 1 month, 18 days of age to 7 years, 8 months, and 12 days of age. A result of the mandible's downward growth increments exceeding its horizontal growth increments, this is an example of "poor" facial growth in that the profile fails to flatten as the mandible remains retrognathic. Note that in this and the following illustration, the forward projection of the premaxilla does not increase after 1 year, 2 months, and 22 days

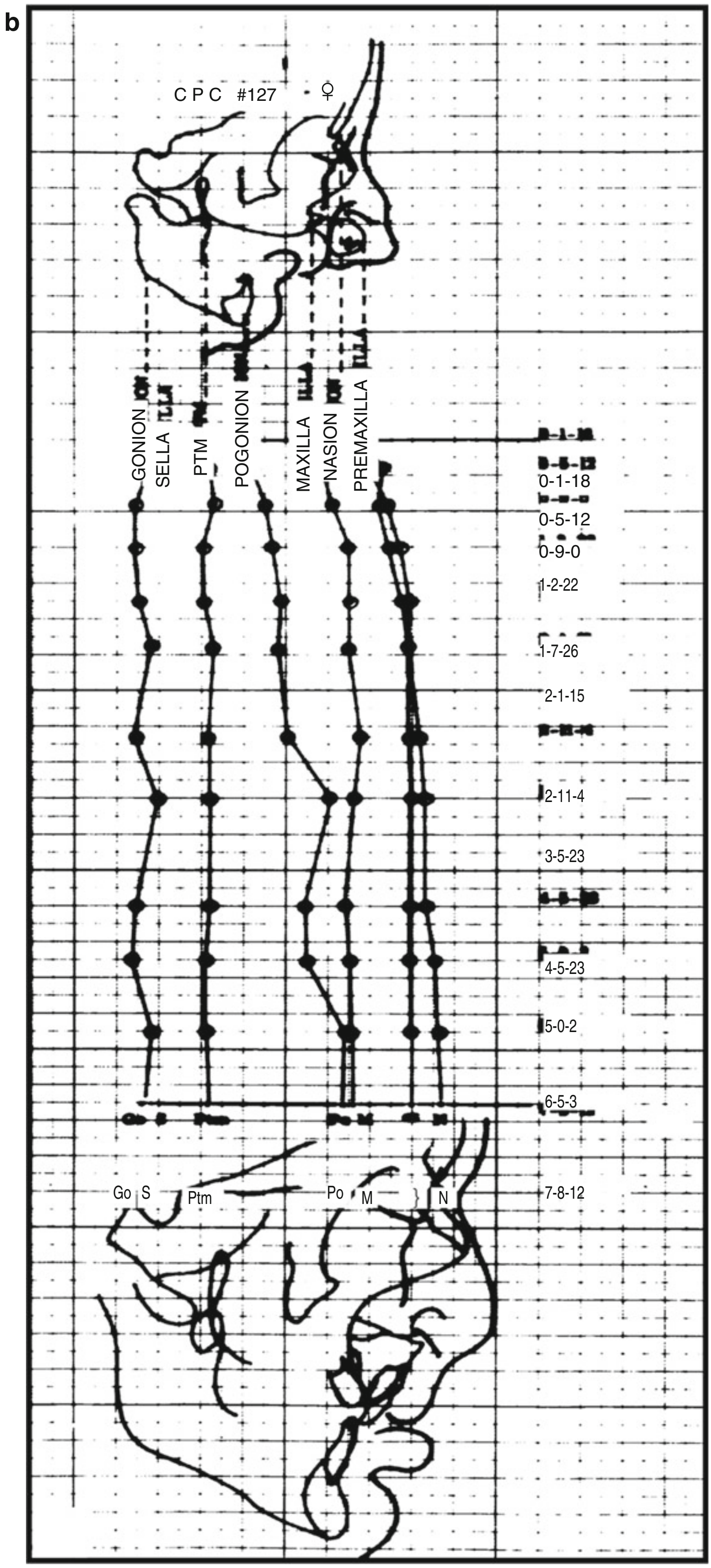

Fig. 2.2 (*continued*) (**b**) Case CP #127. Projecting facial landmarks to a constructed Frankfort horizontal 6° off the SN line. Although each of the skeletal structures except for the mandible has increased in size, the relative position of midfacial structures to the anterior cranial base has remained relatively stable

c

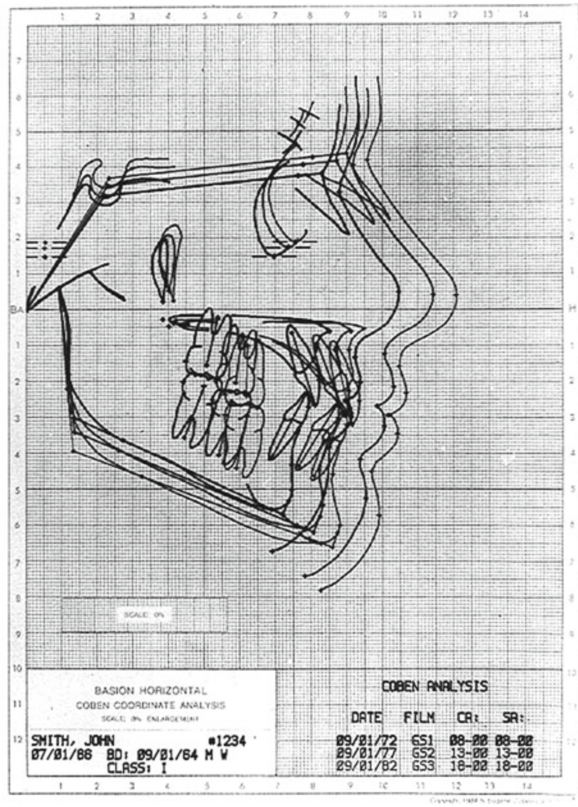

Fig. 2.2 (*continued*) (**c**) Case CP #127. Basion horizontal coordinate computer craniofacial serial tracings at ages 8, 13, and 18 years. Tracings are registered at Basion and oriented in Frankfort horizontal. Serial tracings maintain a constant S-N/FH relationship. S-N and FH planes are parallel. Tracings depict Coben's growth philosophy, which states that craniofacial growth is reflected away from the foramen magnum (basion) and the vertebral column (Reprinted from Coben (1986))

2.2 The Beginning of Longitudinal Cleft: Palate Research Studies

Two major research problems were common in cleft palate surgical studies prior to the 1950s. Pruzansky (1953, 1955) commented on the surgeon's tendency to group all types of clefts together in research and clinical treatment. He also stated that surgeons were limited in their study of pathologic anatomy of clefts due to the unavailability of serial dental casts, cephaloradiographs, and photographic records.

The need for clinical records was apparent to many researchers, and within a decade, many retrospective clinical data sets were developed. These data sets spawned many investigators to determine the long-term influences of surgical and neonatal maxillary orthopedic procedures on palatal and facial growth and development. As a result of these early studies, useful diagnostic and prognostic information was obtained that provided a rationale for the management of individual cleft cases. These clinical records offered an accurate means for measuring and recording individual variation and for plotting the progress of each case in terms of growth and response to various treatments. As a result of these findings, the quality of care improved, resulting in more aesthetic and functional outcomes. Proper documentation, using objective records and individual treatment outcomes, has extended to many more modifications where it is possible to perform multicenter retrospective studies.

2.3 **Research Methods** (Atkins 1966; Byse et al. 1983)

2.3.1 Retrospective Studies

In a retrospective study, the nature of the study group must be delineated precisely. Definite criteria should be established so that there is no ambiguity about types of cases and stages of growth development to be included in, or excluded from, the study. The choice of the case and control groups should be guided by concerns of validity. The advantages of retrospective studies are that they can be conducted relatively rapidly because the records of patients whose treatment is already complete can be used. The investigator is protected against the circumstance of "subject dropout" during the course of treatment, and they are relatively economical.

2.3.2 Prospective Studies

The advantages and disadvantages of prospective studies are in essence the inverse of those of retrospective studies. Provided that ethically and logistically satisfactory plans for random assignment to treatment can be developed, prospective trials afford an opportunity to control selection bias and to define and control the records acquisition process.

The main disadvantages of prospective trials are that they are expensive and a great deal of time must inevitably elapse between project initiation and the point at which data on most of the main outcome variables become available for analysis.

Multicenter comparisons of surgical orthodontic treatment outcomes are an efficient way of testing the effectiveness of various treatment philosophies and surgical techniques. Differences among surgeons, variances in performance by the same surgeon over the years, and differences in techniques are difficult to identify and compare in isolation. However, in multicenter clinical studies, differences in clinical procedures among operators can, within defined limits, be compared and evaluated successfully without arousing criticism.

2.3.3 Clinical Trials

A clinical trial may be defined as a carefully designed prospective study that attempts to answer a precisely defined set of questions with respect to the effects of a particular treatment. A clinical trial is a major undertaking which requires considerable money, personnel, facilities, time, and effort.

The simplest design for a clinical trial involves randomization between two different surgical treatment regimens to answer one specific question; for example, which of two surgical procedures is the most beneficial. To add a larger number of surgical procedures makes the trials more difficult to manage.

There are two reasons for not using a randomized clinical trial (RCT) method for surgical evaluation of cleft closure procedures whether done as a multicenter or single-center trial. The first is the need for the surgeon to disregard the unique nature of the individual cleft defect and perform a standard surgical treatment being tested, the presumption being that clefts of all sizes and shapes will react the same way to the same surgical procedure. The second reason concerns the ethical questions involving the sequencing of surgical procedures and the use of the surgeon's skills.

2.3.3.1 Randomization of Surgical Procedures

In proposed multicenter RCT, it is expected that each surgeon will randomly utilize various surgical procedures sequentially for each type of cleft to determine the relative differences in outcome between procedures.

With the present restraints on certain types of human research, Human Subject Research Review Committees in most settings would be reluctant to permit the use of various elective surgical procedures in a research setting if there is a possibility that a surgical procedure might lead to facial disfigurement. Most surgeons would reject participating in a study employing a particular procedure they already have used and found to be inadequate. Many surgeons see the choice and timing of cleft surgical procedures as varying with the geometric characteristics of the palatal defect; therefore, the concept of randomization cannot be considered as an alternative to what they are already doing. The factor of surgical skill in a randomized trial must be considered as a variable in determining the effectiveness of a procedure. Can all participating surgeons be equally skilled in all procedures?

2.3.3.2 The Ethics of Surgical Retrospective Clinical Trials (RCT)

It is impossible to disassociate scientific from ethical considerations when dealing with cleft palate research (Gifford 1986; Hellman and Hellman 1991; Israel 1978; Kukafka 1989). Different research protocols and evaluation methods carry different ethical problems, the more so when life or death issues are not being considered.

2.3.3.3 Informed Consent

When a patient is deemed appropriate for a particular clinical trial, a first step is often to obtain informed consent. This is a legal requirement in the USA, but not in all countries. In some European countries, each participating hospital decides on whether and how to handle informed consent.

Informed consent is a social construct based on ethical guidelines and supported by legal precedents. In order for consent to be legally valid, it must be obtained voluntarily from a mentally competent person of legal age.

The greater the seriousness of the potential injury, even if the risk is minimal, the greater the obligation to inform the patient (or parent). The greater the chance of a risk occurring, even if the injury would be minimal, the greater the obligation to inform the patient (parent). The more elective the proposed treatment, the more serious injury will be perceived.

Sheldon Baumrind (1993), summarizing the role of clinical research in orthodontics which is also applicable to cleft palate research, states:

> Cogent arguments can be made concerning the ethics of conducting structured clinical experiments in the kinds of long-term therapeutic situations which interest orthodontists. One telling argument is that since therapists have an absolute and transcendent obligation as professionals to deliver for each patient the treatment which they believe best for that patient, no subject can ethically be randomized to one of two possible treatments unless there is true uncertainty as to which of the two treatments is in the patient's best interest. For the same reason any experimental design that asks a clinician to treat a patient against the clinician's own professional bias is inappropriate at best. And even if ethical reservations could be overcome, it would clearly be of only minimal scientific value to accumulate data on how patients fare under treatments not considered optimal at the time they are delivered.

Baumrind (1993) concludes:

> Except in special and very limited circumstances, clinical studies in orthodontics cannot and should not be expected to reveal categorically which of two or more treatments is better in a global sense. They can and should be expected to supply valid and reliable information about the mean effects of different treatments. But more important, they should supply information about the usual individual variability of human growth, development, and response to therapeutic intervention.

Retrospective studies have permitted clinical investigators to evaluate the palatal and facial growth and development responses within a particular cleft type according to the type and timing of the surgical procedures employed. Such studies have shown that the degree of palatal scarring is directly related to the areas of denuded bone resulting from the displacement of the palates mucoperiosteum during cleft closure.

Roentgencephalometry has aided in the elucidation of the nature of the craniofacial malformation associated with facial clefts as it affects the mandible, maxilla, orbits, nasopharyngeal area, and the base of the skull and cervical vertebrae. Moreover, current studies on the variable growth and involution of tonsils and adenoids have raised a number of questions of interest to immunologists.

2.4 The Need for Geometric and Quantitative Analysis of Cleft Palate Casts

Treatment planning in cleft lip and palate habilitation is contingent upon understanding the natural history of the palatal cleft defect and the face in which it exists. Longitudinal dental cast studies ultimately helped explain many cause and effect relationships which existed between palatal surgery and subsequent facial development. However, there still remains an important need for our understanding of palatal development, which will further refine and improve rehabilitative procedures. The purpose is to consider previously posed questions in light of newer biostereometric techniques. Specially, the following questions have been asked:

1. Are the palatal shelves intrinsically deficient, adequate, or excessive in mass?
2. To what extent does the geometric relationship of the palatal shelves in one cleft compare with that of another in the same type of cleft? With other types of clefts? With normal palates?
3. To what extent are the palatal shelves displaced in space?
4. How are these parameters altered as a consequence of growth and surgical reconstruction? The advent of advanced biostereometric technique and 3D digital cameras with computer made it possible to analyze the size and shape of the palate in greater detail through intensive geometric survey. The data collected by these systems can be reduced to a mathematical format and subjected to analysis by high-speed computers.

In accordance with these objectives, we undertook a series of phased studies utilizing stereophotogrammetric electromechanical 3D digital analysis of serial casts of infants with cleft lip and palate with the following specific aims:

1. Test the reliability of the method for selecting the proper anatomical landmarks when extrapolating data from the stereophotographs.
2. Compare and contrast 2D and 3D surface area measurements with other descriptive measurements to determine if there are significant differences in their interpretive values.
3. Perform 3D analysis of serial casts in order to describe the changing geometry of the palatal vault in mathematical terms.
4. Determine whether the descriptive analysis revealed additional information relative to the geometric changes that follow in the course of time.
5. Determine by differential analysis if a constant geometrical relationship might exist between the size and shape of the lesser to greater palatal segment in a complete unilateral cleft lip and palate.
6. The best time to determine palatal surgery based on the size of the cleft relative to the size of the body palate.

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