

Review

Recent Researches in Repair of Unilateral Cleft Lip Nasal Deformity

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ABSTRACT

The cleft lip nasal deformity accounts for 84% of nasal deformities, which is one of the most common diseases in plastic surgery. The Unilateral cleft lip nasal deformity, which has a high incidence, had many treatments but not very effective. In this article, we will focus on the repair of unilateral cleft lip nasal deformity, embryology and etiology, anatomy and pathology, classification, preoperative evaluation and repair timing strategies, surgical goals and surgical techniques, postoperative care and effect evaluation. I hope to bring some useful information for clinicians to further guide clinical work.

KEY WORDS

unilateral cleft lip; nasal deformity; treatment methods; effect evaluation; research progress

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Cleft lip (with or without cleft palate) is the most common congenital malformation, the incidence of which is 0.2 to 2.3 per thousand newborns^[1] and most common in boys. Incidence of left cleft lip: right cleft lip: bilateral cleft lip is approximately 6:3:1^[2]. Since 390 B.C., deformity correction for cleft lip patients has been a formidable challenge for plastic surgeons. In the past decades, many disciplines, including plastic surgery, have jointly explored and made efforts. In spite of many literatures on how to correct cleft lip and cleft lip nasal deformity, none of these techniques has been widely accepted as the best practice at present.

EMBRYOLOGY AND ETIOLOGY

Embryology

Lip formation occurs between the 4th and 7th weeks of pregnancy^[3-4]. The primary mouth is the basis of the oral cavity and nasal cavity. There are five protrusions around the primary mouth, with the top protrusion being frontonasal process and protrusions on lateral and tail sides being the maxillary process and the mandibular process formed by the first parotid arch. The maxillary and nasal structures evolve from these structures: the two globular processes form the original palate, then gradually develop to separate the olfactory sac from the primary

mouth, and form the columella, the middle part of the upper lip and the maxillary incisors in the future; the lateral nasal process forms the nasal ala and the bilateral part of the external nose; the maxillary process and the median nasal process eventually fuse to form the upper lip^[5]; philtrum is formed by the cells of the maxillary mesoderm on both sides of the midline. When the maxillary process and the median nasal process (globular process) are fused, only one or both sides are partially connected or disconnected, then unilateral or bilateral incomplete or complete cleft lip appears^[1,5].

Etiology

The pathogenesis of cleft lip has not been fully understood. It is generally believed that there are two causes of cleft lip. One is genetic factors. If an immediate family member has cleft lip, the probability of a mother giving birth to a cleft lip baby is higher than that of a mother without family history. On the other hand, non-genetic factors are generally considered to be associated with nutritional deficiency, drug effects, viral infection, maternal negative emotion and exposure to radiation during early pregnancy^[5]. Smoking has long been considered to be closely related to cleft lip and has a dose-effect relationship^[6]. Drinking during pregnancy has been considered as a risk of cleft lip and palate, but the increase of incidence is not statistically significant^[7].

ANATOMY AND PATHOLOGY

The difficulties in repairing cleft lip and nasal deformities lie in the congenital anatomical deformities of cleft lip patients, scars left by early cleft lip repair and growth-related changes, which increase the difficulty of repairing cleft lip^[8]. Successful repair of cleft lip deformity requires a detailed understanding of normal and pathological anatomy, as well as the biomechanics of muscles.

Applied Anatomy of Lip and Nose

The upper lip is lined with non-keratinized oral mucosa. The front is keratinized vermilion mucosa and hair skin. The nodular protuberances in the middle of the vermilion mucosa are called vermilion tubercle. The two highest points symmetric in the lip arch are called labial peaks. The central depression of the upper lip is called philtrum concave. Philtrum is composed of symmetrically distributed philtrum crests and central depression between crests^[9]. The orbicularis oris muscle is the main muscle of the lip. In the upper lip, the surface fibers of the orbicularis oris muscle intersect at the midline and insert into the skin of philtrum area. The deeper fibers anchor the muscles on the anterior spine of the lower nose, mainly playing the role of lip retraction and oral closure^[10-12]. The upper lip mainly receives blood supply through the branch of facial artery, which travels in the orbicularis oris muscle and forms a pair of columnar arteries^[13].

Pathological Anatomy and Manifestation of Unilateral Cleft Lip Nasal Deformity

The main characteristics of unilateral cleft lip nasal deformity are asymmetry of nasal tip and alar base [14], which is mainly caused by the discontinuity of upper lip tissues, loss of maxilla and abnormal pulling of lip and nasal muscles. Owing to the existence of cleft lip, the orbicularis oris muscle loses its normal circular structure, and the broken muscle fibers change from the original horizontal direction to the vertical direction, attaching to the root of the nasal columella on the affected side and the nasal base on both sides of the cleft. The orbicularis oris muscle and the periosteum of the pyriform foramen on the cleft side are abnormally inserted along the base of nose ala, forming a transverse pulling force to make the nostril more horizontal^[15], which are often accompanied by the curvature of the nasal septum. The nasal septum was deviated from the caudal side to the affected side and from the posterior side to the healthy side. Unilateral cleft lip nasal deformity corresponds to the severity of cleft lip^[16-17]. Common deformities are: round nose tip and deflection to the affected side, existence of bilateral alar cartilage "separation phenomenon", collapse of the affected alar, flat nostril,

lateral foot of the alar moving outward and downward, short and oblique columella of the affected side to the healthy side, and depression of the affected nasal base resulting in no obvious nasal sill^[8,18,19].

Biomechanics of Muscle Fibers in Unilateral Cleft Lip Nasal Deformity

For a long time, many disciplines have never stopped exploring how to better correct cleft lip nasal deformity. In recent years, for repairing cleft lip nasal deformity from the perspective of muscle biomechanics, a new biomimetic repair theory of muscle biomechanics has been established, and a certain therapeutic effect achieved. According to the latest theory, lip and nose muscles mechanical group is composed of four groups of muscle fibers: orbicularis orifice, descending nasal septum, alar nasal muscles and alar lip and nose muscle. These four groups of muscles and fibers interweave with each other, interact and inseparable in the mechanical relationship, forming a biomechanical complex of lip and nose muscles. The tension of muscle fibers forms three groups of tension bands. Among them, the primary tension band and the first auxiliary tension band directly affect the width of ala nasi, the protrusion of nasal tip, the location of ala nasi, the shape of columella nasi and the basal profile of nostril^[20-23]. From a biomechanical point of view, unilateral cleft lip nasal deformity is actually the rupture of the tension band of the lip and nose muscles, the absence of the first auxiliary tension band on the affected side, the absence of a supporting structure at the nasal base, the ectopic nasal muscles and orbicularis oris muscles along the nasal base to the bottom of the cleft and the root of the columella, and the lack of internal traction on the lateral alar of the affected side of the nose, resulting in the outward movement of the alar foot and the transverse nostril; at the edge of the foramen, the cartilage of the pterygoid cartilage is pulled, resulting in the collapse of the affected alar and deformity of the nasal tip^[21].

CLINICAL CLASSIFICATION

The clinical classification of unilateral cleft lip is of little practical value. According to the degree of unilateral cleft lip, it can be divided into unilateral complete cleft lip (the cleft of the lip reaches the nasal base) and unilateral incomplete cleft lip (the cleft of the lip does not reach the nasal base). Complete cleft lip has greater influence on nasal morphology than incomplete cleft lip. Unilateral cleft lip should also include harelip, which is characterized by continuous and complete skin of the whole lip, but a groove depression can be seen on the upper lip, usually without nasal deformity^[5]. We should pay attention to distinguishing unilateral cleft lip from median cleft lip. The median cleft lip is manifested by the middle cleft of

upper lip, which is rare. The occurrence of median cleft lip seems to be inconsistent with the classical theory of failure of fusion in normal combined facial process^[24,25]. There are few reports about median cleft lip and related nasal abnormalities, and the degree of median cleft with nasal deformities varies greatly. Personalized treatment is needed^[26].

PREOPERATIVE ASSESSMENT AND REPAIR TIMING STRATEGIES

Preoperative Assessment

Foreign scholars pointed out that infants born with cleft lip should be assessed as soon as possible after birth in order to provide counseling for family members and evaluate the feeding and growth of infants. The assessment should involve multiple disciplines, including plastic surgeons, language pathologists, feeding experts, otorhinolaryngologists, geneticists and pediatricians, etc.^[26]. Preliminary assessment should be aimed at determining whether infants have associated syndromes. The anatomical structure of the cleft should be evaluated and imaging data be retained. For infants who are in need and suitable for preoperative rhinoplasty, preoperative rhinoplasty should be recommended. Before surgical correction, the width of the cleft should be significantly reduced and nasal asymmetry improved by repositioning the dislocation structure. Minimizing malformations will help us to reduce trauma, improve surgical outcomes and achieve minimally invasive treatment^[27]. Informed consent is required before operation to inform preoperative complications of nasal cavity plasty^[28]. The most common complications are tissue irritation or ulcer. Re-evaluation should focus on assessing the general condition of the patient, surgical tolerance and planning for the forthcoming surgery. For corrective surgery for definite lip nasal deformity, apart from assessing the general situation and surgical tolerance before operation, emphasis should be placed at the characteristics of nasal deformity, whether there are obvious skeletal deformity, severe nasal septum deviation, obstructive ventilation symptoms, etc.^[28], which will affect the preparation of surgical instruments and the choice of surgical methods, and will also affect the final effect of surgery and patient satisfaction.

Repairing Timing

Repair of cleft lip nasal deformity includes primary repair of cleft lip and secondary repair of nasal deformity. The latter includes intermediate augmentation rhinoplasty and definite augmentation rhinoplasty. Intermediate rhinoplasty refers to the operation between lip repair and definite rhinoplasty, and should be done before nose is

fully grown. Most surgeons choose to repair unilateral cleft lip and nose for the first time when the patient is 3 to 6 months old. The reason for choosing this time is to allow the baby to gain weight, achieve more accurate alignment, and bypass the period of high anesthesia risk to increase the tolerance of surgery^[29]. Haddock et al.^[30] reported that patients undergoing primary cleft lip rhinoplasty had a higher symmetry of nasal tip, and needed fewer operations with less complexity in secondary rhinoplasty. Moreover, maximizing nasal outcomes during the first repair operation can help achieve better secondary rhinoplasty results and improve patient satisfaction^[31]. Nasal septal plasty in infants is still a controversial field. Brian K. Hall^[32] and Aditi Bhuskute^[33] summarized the literature. Previous animal and clinical studies have shown that nasal septal cartilage is the center of facial growth. Periosteum preservation plays an important role in the development of nasal septal cartilage. The main stage of nasal growth occurs in adolescence. If there are obvious functional or social problems, conservative surgery to preserve the integrity of nasal septum as much as possible may be safe before puberty. Intermediate rhinoplasty has two different timing strategies^[34]. One is when the patient is 4 to 6 years old, it may alleviate the psychological pressure from peers of the same age; the other is when the patient is 8 to 12 years old or after orthodontic and alveolar bone transplantation, it can obtain better skeletal base, so that serious nasal deformity can be corrected for a longer time. When possible, delaying definitive rhinoplasty to the end of adolescence is the best option. Girls' facial growth is finished at about 16 years old and boys' at about 18 years old. It is considered safe to perform definitive rhinoplasty at this age and later, when more active septoplasty, osteotomy and cartilage transplantation can be performed without worrying about affecting the growth of nose and middle part of the face^[8]. However, the choice of specific surgical methods should be personalized, and there is no recognized surgical method suitable for all patients.

SURGICAL GOALS AND TECHNIQUES

Surgical Goals

The ultimate goal of repairing unilateral cleft lip nasal deformity is to restore the anatomical features of the lip and nose, lip function and/or nasal function, to restore normal mid-facial anatomical relationship and to obtain long-term symmetry. The primary repair technique of cleft lip mainly restores lip function, maximizes nasal results as far as possible, produces favorable scar shape, reduces the number of secondary rhinoplasty operations and the complexity of surgery^[35,36]. The main purpose of

intermediate rhinoplasty is to improve symmetry, improve ventilation dysfunction caused by severe nasal deformity, alleviate patients' psychological pressure and improve patients' quality of life [35]. The goal of definite rhinoplasty is to restore the aesthetics and function of the nose.

Surgical Technologies

The primary repair of unilateral cleft lip nasal deformity is mainly focused on lip repair, which is composed of closure suture, rectangular flap, triangular flap, Millard operation and modified Noordhoff operation [37,38]. Lip repair technology will not be discussed here. Over the past decades, more and more doctors have acknowledged the primary cleft lip repair and nasal deformity repair at the same time. Most surgeons choose the open surgical method [1,8,32,39], which is convenient to expose the tissue structure to the maximum extent and easy to suture and implant. To avoid the transverse incision of nasal columella, the incision along the medial edge of nasal alar is usually designed; the skin of nasal alar, nasal tip, nasal dorsum and medial nasal mucosa is fully separated along the nasal alar cartilage on the affected side, and the abnormal muscle tissue attachment of nasal alar cartilage on the affected side is relieved or the nasal alar collapse on the affected side is corrected by suspending the nasal alar to the contralateral nasal cartilage on the affected side. Abnormal fibrous tissue attachment of cartilage, suture of orbicularis oris muscle of affected side to anterior nasal crest and caudal edge of nasal septum (horizontal mattress suture is recommended [40]) and suture of alar cartilage fornix of healthy side after release of alar cartilage of affected side are important for improving columella deflection, lack of columellar length and nasal tip deflection of affected side. The bone defect caused by dysplasia of jaw and pyriform foramen on the affected side was shown as the improvement of depression at the base of nasal alar on the affected side. Unlike the final definitive rhinoplasty, most scholars are in favor of conservative surgery for the primary correction of unilateral cleft lip nasal deformity in the initial stage, and do not recommend destructive surgery to improve the deformity. Of course, some scholars believe that operation method should be chosen according to the degree of deformity. Those with severe deformity may consider improving the deformity through transplantation to alleviate the psychological pressure. Unilateral cleft lip rhinoplasty is similar to the primary repair, and conservative surgery is the main method to solve severe nasal obstruction or unsolved deformity caused by deviation of nasal septum or severe emotional distress caused by peer pressure in children [8]. Definite rhinoplasty for unilateral cleft lip nasal deformity (Phase II surgery) is performed in different ways [41-44].

Autogenous cartilage and prosthesis are often used to help correct the deformity in order to obtain perfect nasal shape such as nasal dorsum, tip, columella and even nasal sill. In order to improve the shape of nasal tip and columella deformity, ear cartilage or nasal septum cartilage should be cut to improve the shape of nasal tip and columella deformity. If the shape of nasal dorsum is not good enough and the patient has strong desire, the deformity can be corrected by autologous costal cartilage or prosthesis plasticity. According to the degree of nasal alar basal depression, the nasal sill should be filled with upper lip scar tissue or autologous cartilage or prosthesis transplantation to improve the patient's satisfaction. In recent years, the concepts of biomimetics, mechanics and anatomical subunit are very common in literatures. Using biomechanics to reconstruct the biomechanical complex of lip and nose muscles can achieve the effect of anatomical repair without autologous cartilage and prosthesis transplantation. This may become a hotspot in the following repair of cleft lip nasal deformity, but its effect still lacks long-term efficacy evaluation from large sample random control group test.

POSTOPERATIVE NURSING

Postoperative use of nasal model is preferred for at least 6 weeks, ideally 3 months. Inform patients and family members of the effectiveness and use of nasal model in advance before operation. The appropriate size should be worn immediately after leaving the operation room, and the stent should be close to the mucosa, which can exert the effect of compression hemostasis to a certain extent. Instruct patients or family members to remove nasal model every day for cleaning, and teach them how to replace the mold. If possible, when the swelling subsides around 7 days after surgery, replacing a nasal model of more suitable size can help to shape the nostril and scar contracture [1,46].

EVALUATION OF SURGICAL EFFECTS

Anthropometry is the measurement of individual human beings. Dr. Leslie Farkas, father of medical anthropometry, and his colleagues [47,48] provide standard measurements of lips and noses. Indicators can be measured directly or indirectly. In the existing literatures, the objective evaluation mainly centers on symmetry. The evaluation of unilateral cleft lip nasal deformity involves such measurement indexes as lip height, lip length, nasal width, columella width, nasal tip height, bilateral alar width, nostril width, columella length, as well as the oblique angle of nasal dorsal middle axis, columella oblique

angle, nasal alar basal depression degree and nostril area [39,49,50]; the subjective evaluation is mainly about the satisfaction of surgery, the improvement of quality of life, the improvement of nasal ventilation obstruction and so on by questionnaire or scale sheet [6,7,29].

Objective Evaluation

Objective evaluation-Direct anthropometry: it is a direct measurement of surface size. The measurement of unilateral cleft lip nasal deformity is usually carried out by using millimeter scale alone or combined with sub-rules. It is the most accurate measurement in the operating room under general anesthesia, but it needs experience and patient's cooperation very much, and it is more suitable for measurement with skeletal markers on the body surface and yet difficult to be identified in photos. Because of the deformability of soft tissues, the direct measurement error is increased. This method is simple, non-invasive, low equipment cost and easy to be widely accepted. Its shortcoming is that it needs training and experience, and data is not easy to copy [50].

Objective evaluation-Indirect anthropometry: it includes two-dimensional measurement and three-dimensional measurement. It is a two-dimensional measurement method to measure the shape characteristics of nose by plane photographs, and to compare them by measuring tools or computer and application software. The photographs are easy to be obtained and recorded permanently. This method needs to fix the head position, standardize the image and set the linear standard measurement and calibration. Unfortunately, there may be errors in defining bone markers, and fewer measurement results can be obtained from photographs than from direct methods. Two-dimensional photography is suitable for distinguishing scale and angle, but it is difficult to measure the distance and angle accurately. This problem can be eliminated mainly by constructing the ratio of measurement [50]. Three-dimensional anthropometry includes stereo photogrammetry [53], laser surface scanning [54] and three-dimensional photogrammetry [55]. This technology has been favored as an alternative to direct anthropometry because images can be captured in only 3.5 milliseconds with high accuracy. Wong and his colleagues [56] used three-dimensional photography to evaluate the effectiveness and reliability of nasolabial groove anthropometry compared with direct anthropometry. It is found that the linear measurements are highly correlated and the overall accuracy of 3D measurements is within 1mm of the direct measurements.

Subjective Evaluation

The subjective evaluation of unilateral cleft lip nasal

deformity in the existing literature mainly focuses on the following four points [57-59]: (1) nasal morphology satisfaction: both doctors and patients, even the third party, participate in the evaluation of nasal morphology, and score the nasal morphology before and after operation by naked eye observation. Generally, the percentile system is adopted with three grades, namely, satisfied "80-100 points", basically satisfied "60-80 points", and dissatisfied "lower than 60 points". (2) Quality of life: adolescent quality of life-facial difference questionnaire, pediatric voice-related quality of life survey, children's oral health impact profile and children's oral health quality of life scale were used to assess the quality of life of patients. The influence of cleft lip nasal deformity on patients' life was also discussed. According to the attention to the appearance of the nose, patients' quality of life was assessed, which was divided into five grades: very frequent "0", frequent "1", medium "2", slightly "3", and no "4". (3) Ventilation function: for the evaluation of the ventilation function of unilateral cleft lip nasal deformity, the "NOSE" score was often used clinically, and also through the objective evaluation of cavity. (4) Psychological impact: Generally, scoring scale is used to assess the psychological impact of the disease on patients.

Significance of Surgical Effect Evaluation

The surgical effect is an important means to evaluate the technique of repairing unilateral cleft lip nasal deformity. It is of great significance for preoperative treatment, design of operation plans and postoperative effect observation. In anthropometric measurement, nose provides objective data on the shape and size of the external nose, and objective evaluation is helpful to answer important clinical questions in the study. For example, many surgeons initially believed that primary rhinoplasty would interfere with nasal growth, but now anthropometric studies have shown that primary rhinoplasty does not interfere with nasal growth, but rather promote nasal growth balance. It is also an objective tool for investigating whether reconstruction of the nasal cavity and sinus has any harmful effects on the nose and the middle part of face in pediatric operations. It also plays an important role in objective assessment and description of malformation syndrome [60,61]. Improvement of patient's facial appearance is the common goal of all kinds of clinicians. Improvement of plans requires guidelines or some universally recognized "ideal" facial ratio [62]. However, these values should not be interpreted as treatment objectives, but should only contribute to treatment planning. According to the needs and expectations of each patient, the surgeon's assessment of the face is very important in planning plastic surgery, but clinical impressions alone can be

misleading^[63]. Therefore, it is advocated that objective evaluation combined with subjective evaluation should be used as far as possible to guide the diagnosis and treatment of diseases.

CONCLUSIONS AND PROSPECTS

Due to the potential complexity of the shape and function of the nose and the residual scar, the repair of cleft lip nasal deformity is still a challenging problem. The diversity of the surgical methods of cleft lip nasal deformity further verifies the difficulty of this kind of repair. Complete correction of all nasal defects is still a difficult goal for many people to achieve. A thorough understanding of deformities and their correction methods is the basis for successful reconstruction. Future research needs more subjective and objective results measurements. Consistent anthropometry can accurately assess and report long-term results. Such reports are needed to guide perioperative care and validate the best repair principles, and to resolve persistent disputes.

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