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CHINA CLINICAL STUDIES V
ORIGINAL ARTICLE

Application and Development Of Non-Invasive Method Of Congenital Ear Deformity Correction

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Abstract

Objective: Congenital ear malformation may cause abnormal appearance, and affect the development of hearing and mental. It is a disease which demand great attention and active treatment. Congenital malformations have a complex etiology, congenital genetic factors, biological factors, chemical factors, physical factors has been found to be the cause of human malformations. Infant auricular deformities are classified eitheras malformation or deformation. Malformations are characterized by a partial absence ofthe skin or cartilage resulting in a constricted or underdeveloped pinna, whereas deformations are characterized by a misshaped but fully developed pinnawhich was caused by external force acts on the normal structure. There are several different classifications according to ear morphology and abnormal areas. There are still no complete data which include incidence and outcome of malformations. In Japan, America and other countries, non-surgical treatment have been widely used to correct congenital ear malformations, and made a very good therapeutic effect. This treatment has been used in some cities in China which is worthy of further research and application.

Key words: Ear Malformation, Congenital, Non-Invasive, Typing.

INTRODUCTION

The ear is merely a small part of the body, but it acts as an important organ in the facial appearance, which possesses a complex 3-dimensional anatomical structure. Congenital ear malformation may not be as influential as other facial organs on its aesthetic perception, but it is one of the key influential factors on the hearing ability of children [1]. Moreover, patients' psychological development would be affected as well, which would influence children's social activities. Therefore, attention and active treatments should be given on the congenital ear malformation. With the development of perinatal medicine, embryology and neonatology, neonatal congenital malformations attract an increasing-quantity of focus, and risk factors leading to congenital malformation and early interventions have become the research focus. Therefore, we need to achieve further understanding of the occurrence, treatment, transformation and medical development on congenital ear malformation.

PATHOGENIES AND NOSOGENESIS

The pathogenies of congenital malformation are complex. Currently, it has been recognized that the causes of congenital malformation include genetic factors, biological factors, chemical factors and physical factors, which can be categorized into three classes: Genetical Factors, Environmental Factors and the Interaction Between These Two Factors. Among these factors, 25% of them are genetical, 10% are environmental and the remaining 65% belongs to the interactions and unknown factors. The occurrence of congenital ear malformations is not a single process, but a result affected by many factors [2]. At present, there is no final verdict on the exact teratogenic factors of auricle development among various genetic and environmental factors. The congenital malformation in the external ear and middle ear is related to branchial apparatus developmental disorder. The malformation of the external ear can reflect the level of middle ear malformation. During the 5th to 9th week of pregnancy, the congenital malformations caused by various fetal dysplasia, such as microtia, usually appears as ectrogen of some important anatomical structures [3]. Additionally, some occurrences of auricular malformations are after this period of ear embryo development, and they are caused by the structural abnormity of normally-developed subcutaneous auricular cartilage, such as lop-ears, cup ears and protruding ears, etc. The pathogenies of these ear malformations are related to defects and fissions of mono or multiple hills. For example, the defects of the third hill on embryo can lead to the cup-ear of Tanzer I and Tanzer II. The impact of antenatal intrauterine pressure and resistance of the birth canal cannot be ignored to the ear malformation.

CLASSIFICATION OF CONGENITAL AURICLE MALFORMATION

Neonatal ear malformation can be divided into ear transformation and ear dysplasia. Ear dysplasia is usually caused by the dysplasia of the pinna skin and the cartilage. Ear transformation is the malformation caused by the external force on normal structures, and the cartilage development is normal. Saphenous ears, anotia and microtia belong to dysplasia, while protruding ears and cup ears belong to ear formation[3].

For the convenience of treatment selection, Wei Wang[5] classified congenital auricle malfunction into 4 categories: The whole auricular deformity, upper auricular deformity, lower auricle deformity and etc. The auricular malformation is characterized by malformations or absences

of the entire auricle, such as the microtia syndrome, which manifests as a whole auricular deformity with external auditory canal and jaw deformity. The upper auricular deformity is mainly the deformity of the upper half of the auricle, and may also be accompanied by other abnormalities of the auricle, such as the protruding ear deformity, the cup ear deformity, and the crypt deformity. The lower auricle deformity is mainly the lower semi-auricular deformity, but it can also be accompanied by the upper auricle deformity, such as the split ear deformity, the earlobe deformity, etc. Other deformities include the ear, the front ear tube, and etc.

Schonauer, et al, [6] proposed the following classification method based on mild auricular morphological malformations caused by internal/external pressure exerted on the auricle during pregnancy. Type 1: Vertically deformed ears, the auricle is mainly deformed by forces in the direction of the vertical axis (this type includes Type I and Type II A constricted ears classified by Tanzer). Type 2: Horizontally deformed ears, the force along the horizontal axis limits the development of the auricle. Type 3: Focal deformed ear, divided into two subtypes: Type 3A —local reverse folding of the antihelix caused by single-focal pressure; Type 3B—multiple deformities of the antihelix caused by multi-focal pressure (such as spiral kinking or squeezing pressure).

Byrd, et al, [7] divided the neonatal auricle deformities into the following nine types: Protruding ears, lop-ears, cup ears, Stahl's ears, constricted ears, cryptotia, horizontally-protruding cristae helices, helix deformity and compound deformity.

1. Protruding ear means that the angle between the concha auriculae and the fossae helices is greater than 90 ° (normally 90 °), usually above 150 °. The clinical manifestation is that the auricle is flat, 90° to the skull, and the auricle is large and not completely symmetrical. The helix is underdeveloped, and the fossae helices are deep. The normal anatomy of the ear canal and the helix disappears. The upper part of the auricle is flat, and the angle of the stern is greater than 150° or completely disappears. The distance between the upper end of the auricle and the cranial wall is greater than 1.2 cm.
2. The lop-ear is characterized by the fact that the helix folds itself, and the upper part of the auricle hangs down in a curtain-like shape, covering the upper angle of the helix, and the height of the auricle is lowered.
3. Cup ear is characterized by the tightening of the flange, the curling and adhesion of the auricle and auricular cartilage, and the position of the helix is shifted forward and downward, often combined with the malformation of the auricle transverse process; the auricle becomes smaller, mainly because the length becomes shorter; the position is lower; the auricle is tilted forward, similar to the protruding ear, but the fossae helices and the triangular cavity are narrowed but do not disappear. When the patient is lying down, the auricle is an upward with the shape similar to a cup.
4. Stahl's ear is characterized by an abnormal protrusion between the upper angle of the helix and the helix, forming an abnormal third protrusion of the helix; the fossae helices disappears; the helix of the protruding part is not curled; the upper angle of the ear wheel disappears.
5. The constricted ear is characterized by the adhesion of the flange to the helix, and the fossae helices disappear.
6. Cryptic ear, also known as pocket ear, can be unilateral or bilateral, which shows the upper end of the auricular cartilage hidden under the skin of the scalp, and the upper cranial groove becomes shallow or disappear, showing no obvious posterior sulcus. The whole auricle can be seen by pulling the upper part of the auricle with fingers, but when released, it is restored to its original shape due to the tension of the skin and the elasticity of the cartilage. For mild patients, only the upper part of the auricle is short of skin; in severe cases, in addition to severe skin shortage, the cartilage in the upper part of the auricle is also obviously poorly developed, which is characterized by the forward curling of the helix, the deformation of the fossae helices socket, and the buckling deformation of the helix.
7. The horizontal protruding crura helices are characterized by abnormal protrusion of the helix in the concha auriculae, and some protrusions can be extended to the antihelix.
8. Helix deformity is characterized by the uncurled flange, the flat and even disappeared helix. The helix can be pressed curl and against the anthelix due to the compression, causing the disappearance of the fossae helices.
9. Compound malformations are combined 2 or more of the above deformities.

INCIDENCE STUDY

The incidence of congenital auricular malformations is unknown and varies by ethnicity. Foreign literature reports that the incidence of microtia malformations is (0.11 ~ 6.40)/10,000 newborns. According to the resource from the International Birth Defects Monitoring Information Exchange, the incidence of anotia/microtia varies greatly, with a minimum of 0.11/10,000 in Hungary and a maximum of 6.40/10,000 in Mexico. The incidence of whites is lower than in Spain and Asia [8,9]. The total incidence rate in China is 1.4/10,000 newborns, including 0.55/10,000 in the anotia and 0.85/10,000 in the microtia, and the overall prevalence and the prevalence of the microtia show a downward trend in 5 years. Except that the prevalence of microtia was lower than that of anotia in 1992, the prevalence of microtia was higher than that of anotia. The incidence of urban areas is higher than that of rural areas, and there is no gender difference in the incidence. The Xinjiang province has the highest incidence rate, and the lowest is Inner Mongolia, which is 0.33/10,000 [10]. The cryptotia is common in the East. In Japan, it is reported that the incidence of newborns is 2%. The deformity is mostly male, and the ratio of male to female is about 2:1. The bilateral deformity accounts for about 40% [11]. Unilateral microtia malformation is common on the right side.

The study of Qi Xiangdong, et al, [12] in parts of the Pearl River Delta showed that Chinese newborns also have eight kinds of auricular deformities: Protruding ears, cup ears, lop-ear, Stahl's ear, ring ear, Conchal Crus, helix deformity and compound deformity. The incidence of auricular malformation at birth was 43.46%, which was less than the 55.2% reported abroad. Among the morphological types, the incidence of ring ear is the highest, which is 13.40%. When the newborn is born, the incidence of lop-ear is 5.61%. The incidence of Stahl's ear is similar to that of lop-ear, and the incidence decreases with age and has the self-healing feature. The incidence decreases from 8.7% at birth to 1.2% at 1 month, and 1.3% at 1 year, Stahl's ear was 2.49% at birth, and the incidence of protruding ears was 3.74%, higher than 0.4% reported abroad. Unlike lop-ear and Stahl's ear, the incidence of protruding ear increases with age, 4.4% at 1 month and 5.5% at 1 year. However, due to the lack of knowledge popularization of such malformations, the relevant medical personnel have not enough knowledge, there is no complete morbidity statistics and abnormal outcome data in China currently.

ORTHODONTIC TREATMENT OF AURICULAR MALFORMATION

The traditional treatment of congenital auricular deformities in neonates is surgical treatment, and different auricular malformations have different surgical methods. Physiologically, the ear of a 3-year-old child has reached 85% of the adults' ear. The auricle growth is rapid in childhood and is slow in adulthood. After 10 years of age, the width of the auricle almost stopped growing, and the distance from the ear to the mastoid remained unchanged after this. From the perspective of the development of costal cartilage, it is generally believed that the cos-

tal cartilage of children aged 6 years or older has been satisfied with the auricular cartilage stent. Tanzer [13] and Brent [14] both believe that the age of surgery should be around 6 years old. There are more than 200 methods for protruding ear plastic surgery, and the most classic and popular ones are Stenstrom and Heftner [15] ear cartilage scratching method and Mustarde [16]. There are many surgical methods for cup-shaped ear correction. For patients with mild to moderate cup-shaped ear malformation, local auricle can be reconstructed. In severe cases, partial auricle reconstruction is often required due to severe tissue defects. Tanzer [17] divided the ring ear into three types, and there are many corrective surgical methods for ring ear deformities, but there is still no suitable procedure.

Non-surgical treatment of congenital ear malformations was first proposed by Japanese scholars Matsuo, et al, [18] and Kurozumi, et al, [19] in the 1980s. Later, Japan, Europe and the United States gradually carried out experimental research on non-surgical treatment of congenital ear malformations in neonates, and achieved significant therapeutic effects (effectiveness 100%) and widely used. Studies have shown that ear morphology is constantly changing with age, and some congenital ear malformations have self-corrective features, reducing their incidence. However, the conditions for self-healing are still unknown.

The neonatal circulatory system contains a large amount of maternal estrogen. The hormone reaches a distinct peak within 72 hours after birth, increasing the concentration of hyaluronic acid in the cartilage, thereby increasing the ductility and plasticity of the cartilage. The maternal estrogen in the neonate will gradually return to normal levels in 6 weeks after birth, after which the cartilage plasticity and ductility will also decrease [20]. Non-surgical treatment of auricular deformities is based on this mechanism to mechanically correct the deformed auricle of the newborn to avoid the risk and trauma of school-aged surgical correction.

Since the 1980s, various shape correction materials have been used in clinical applications, and the application of plastic shaping materials and clinical non-surgical treatment methods have been greatly developed. The combined shaping materials can be roughly divided into four categories: The first category is surgical tape or bandages. Matuso, et al, used bandage therapy to correct the neonatal ear to achieve certain effects. The second category is plastic composition + tape or bandage. The plasticity compound is combined with tape and bandage external fixation to make the shape to correct the ear deformity last longer. Matuso, et al, first applied Aluwax with tape external fixation. Yotsuyanagi, et al, used Thermoplastic material to successfully correct various types of ear malformations, and made new progress in non-surgical correction of cryptotia. Ullmann, et al, putty soft material can transform from soft to semi-solid traits in 3 minutes, which advances one step further into the accurate shaping correction. The third category is wire splint + tape or bandage, referred to as splint therapy, which is a variety of plastics in the early stage. For the upgrading of the application of shaped materials, many scholars often use a bendable and flexible wire-like bracket, and a silicone rubber tube or suction tube is positioned in the boat-shaped socket, which can respectively position the helix and the antihelix, and can also be used as the upward support force to correct the lop-ear and cup ear deformity. Tan, et al, developed it into Ear Buddies shaping material with commercial perspective. Later, various types of brackets such as lead-free filaments, silver-welded wire, and other splint materials with polyethylene pipes or other suction pipes continued to develop. The design of the silk-like stent is ingenious and easy to operate. The length of application depends on the degree of deformity of the auricle. A short stent material can be used to correct local deformities. Complex deformities often require long stents to expand and balance. The fourth category is a new method innovatively devised by Byrd, et al,— the EarWell Infant Ear Correction System. The material consists mainly of a bracket (including the chassis and cover), a lug tractor and an ear armor. EarWell has the advantages of the previous shaping material, can restore 1/3 of the important anatomical structure of the ear, and can rely on special ear armor to reshape the normal ear cavity/mastoid angle, and reduce the recurrence rate of deformity. It is a new type of effective and comprehensive ear orthodontic device [21].

Earmold treatment has a significant effect on ear deformation and is also effective for mild ear developmental abnormalities. The material used to make the earmold should meet the following characteristics: 1. Fine enough; 2. Non-irritating; 3. Extendable; 4. Easy to obtain and not expensive; 5. Strong. At present, several kinds of correction materials for auricular deformities have their own advantages and disadvantages. The fixation of the wire stent requires the use of a metal material, which is likely to cause skin damage in newborns. The method of fixing the sliver is not sufficient, and it is ineffective for the protruding ear, the ear narrowing and the Stahl's ear. Silica gel fixing requires the use of a plaster mold, which is time-consuming and labor-intensive. The EarWell device works well and is easy to use, but expensive. All methods were clinically proven to be effective and should be treated promptly after the detection of neonatal auricular deformity.

The earmold is recommended to be worn 5 to 7 days after the birth of the newborn. The wearing time depends on the time when the ear mold is started. In the study of Melissa, et al, they believed that the treatment should be as early as possible after the birth of the newborn, and the estrogen drops rapidly (72 hours). Before the ear mold is worn, the treatment can be completed within 1 week [22]. Neonatal ear deformation has a self-healing rate of 30%, but there is no screening method at present. Byrd, et al, [7] think about waiting for 5 to 7 days and expecting some newborns to heal themselves. For children who cannot heal after 1 week of birth, treatments are needed. At present, it is generally believed that treatment should be carried out within 1 week after birth, and treatment after more than 3 weeks is not easy to reach normal ear morphology and requires a longer duration of treatment [23]. The duration of treatment varies according to the start time of the treatment and the type and extent of the deformity. Correction begins within 72 hours after birth, and the treatment time can be shortened to 1 week. The neonates treated within 3 weeks require 6 to 8 weeks of treatment (6 after the week estrogen drops to the baseline level) and neonates who start treatment more than 3 weeks require treatment for 3 months or longer.

At present, there are few clinical epidemiological studies on congenital auricle malformations in neonates in China, and reports related to this are rare. Due to the lack of knowledge in this field, in addition to plastic surgeons, there are fewer pediatricians and obstetricians who can clearly diagnose neonatal congenital auricular deformities. Most medical workers only pay attention to dysfunction when analyzing the results of the current birth defect census, and often ignore morphological malformations. Parents who did not have a correct understanding of the ear deformation caused the medical treatment to be untimely and the children missed the best treatment time, which caused very regrettable results. This illustrates the necessity and urgency of popularizing the work of ear shape malformation and its treatment-related knowledge for domestic medical workers and the general public. The cooperation of the relevant departments and the cooperation of parents can help overcome this dilemma.

The author summarizes the experience and lessons of applying non-invasive treatment in the correction of neonatal auricular deformity in clinical practice in recent years: 1. Early diagnosis and treatment timing is the key; 2. Correct classification of auricular malformation, different

malformation types and regions require different treatment methods; 3. For Chinese newborns, the frequent encountered situation is that the treatment is too late, often more than 6 weeks, the neonatal cartilage extensibility and plasticity decline, on the one hand, the treatment effect may not be satisfying, and treatment duration may be prolonged, on the other hand, it is easy to cause local skin compression and damage due to the close hardness of cartilage and ear mold; 4. Some auricular deformities, such as cryptotia, need to be treated in different stages, in an attempt to completely correct the ear shape at one time may lead to the consequence that the mold is loose or the skin of the child is damaged; 5. Once the skin lesion of the auricle occurs, remove the mold and wait for 3 to 5 days after the skin of the child heals, then continue treatment; 6. It is essential to educate the parents of the child, guiding them to know the disease correctly, understand the treatment methods and principles, which will benefit the prognosis of the disease.

Non-invasive correction technology is to use the principle of the softness of children's ears and the capableness of their cartilages to correct the shape of the auricle. It has valid curative effects, simple operations, short treatment periods and no obvious adverse reactions. It is urgent to promote its application. Early application of non-invasive, practical and reliable technology can avoid long-term surgical treatment and psychological damage.

For children with auricular deformities that are older than 3 months or have failed corrective treatment and recurrence, the current treatment is usually the surgical treatment of school age [24], and there are reports of using lasers with tape to correct adults' ears [25]. Although it is not widely used, it is still a solid research direction. The training of relevant medical workers and the knowledge of the public can help promote the application of noninvasive orthodontic techniques in the treatment of congenital auricular deformities.

REFERENCES

[1] 戚以胜. 新生儿婴幼儿和小龄儿童听损伤危险指标// 韩德民，主编. 新生儿及婴幼儿听力筛查. 北京：人民卫生出版社，2003; 162-165.

[2] 杨萍. 婴儿先天畸形临床分析. 新医学，2002; 33: 732-733.

[3] Tan, S.T.; Abramson, D.L.; MacDonald, D.M.; et al. *Molding Therapy For Infants With Deformational Auricular Anomalies.* Ann Plast Surg, 1997; 38: 263-268.

[4] Porter C.J.; Tan, S.T. *Congenital Auricular Anomalies: Topographic Anatomy, Embryology, Classification and Treatment Strategies.* Plast Reconstr Surg, 2005; 115: 1701-1712.

[5] 王炜. 整形外科学. 杭州：浙江科学技术出版社，1999.

[6] Schonauer, F.; LaRusca, I.; Molea, G. *Non-Surgical Correction Of Deformational Auricular Anomalies.* J Plast Reconstr Aesthet Surg, 2009; 62: 876-883.

[7] Byrd, H.S.; Langevin, C.J.; Ghidoni, L.A. *Ear Molding In Newborn Infants With Auricular Deformities.* Plast Reconstr Surg, 2010; 126: 1191-1200.

[8] Harris, J.; Källén, B.; Robert, E. *The Epidemiology Of Anotia and Microtia.* JMed Genet, 1996; 33: 809-813.

[9] Mastroiacovo, P.; Corchia, C.; Botto, L.D.; et al. *Epidemiology and Genetics Of Microtia-Anotia: A Registry-Based Study On Over One Million Births.* J MedGenet, 1995; 32: 453-457.

[10] 朱军，王艳萍，梁娟，等. 1988-1992 年全国先天性无耳和小耳畸形发病率的抽样调查. 中华耳鼻喉咽科杂志，2000; 35: 62-65.

[11] Matsuo, K.; Hayashi, R.; Kiyono, M.; et al. *Nons-Surgical Correction Of Congenitalauricular Deformities.* Clin Pla Surg, 1990; 17: 383-395.

[12] 吴胜林，齐向东，赵卉，等. 新生儿耳廓形态学分型的初步研究.中国临床解剖学杂志，2013; 31: 384-388.

[13] Tanzer, R.C. *Microtia: A Long-Term Follow-Up Of 44 Reconstructed Auricles.* Plast Reconstr Surg, 1978; 61: 161-166.

[14] Brent, B. *Microtia Repair With Rib Cartilage Grafts: A Review Of Personal Experience With 1000 Cases.* Clin Plast Surg, 2002; 29: 257-271.

[15] Stenstrom, S.; Heftner, J. *The Stenstrom Otoplasty.* Clin Plast Surg, 1978; 5: 465-470.

[16] Mustarde, J.C. *Correction Of Prominent Ears Using Buried Mattress Sutures.* Clin Plast Surg, 1978; 5: 459-464.

[17] Tanzer, R.C. *The Constricted Cup and Lop Ear.* Plast Reconstr Surg, 1975; 55: 406-415.

[18] Matsuo, K.; Hirose, T.; Tomono, T.; et al. *Non-Surgical Correction Of Congenital Auricular Deformities In the Early Neonate: A Preliminary Report.* Plast Reconstr Surg, 1984; 73: 38-51.

[19] Kurozumi, N.; Ono, S.; Ishida, H. *Non-Surgical Correction Of A Congenital Lop Ear Deformity By Splinting With Reston Foam.* Br J Plast Surg, 1982; 35: 181-182.

[20] Leonardi, A.; Bianca, C.; Basile, E.; et al. *Neonatal Molding In Deformational Auricolar Anomalies.* Eur Rev Med Pharmacol Sci, 2012; 16: 1554-1558.

[21] 吴荣薇，潘博. 耳郭畸形的非手术治疗研究进展. 中国美容整形外科杂志，2012; 23: 757-759.

[22] Doft, M.A.; Goodkind, A.B.; Diamond, S.; et al. *The Newborn Butterfly Project: A Shortened Treatment Protocol For Ear Molding.* Plast Reconstr Surg, 2015; 135: 577e-583e.

[23] Anstadt, E.E.; Johns, D.N.; Kwok, A.C.; et al. *Neonatal Ear Molding: Timing and Technique.* Pediatrics, 2016; 137: e2015-2831.

[24] 张天宇. 先天性外中耳畸形分期联合手术进展. 中国医学文摘耳鼻咽喉科学，2012; 27: 8-10.

[25] Leclère, F.M.; Mordon, S.; Alcolea, J.; et al. 1064-nm Nd: *YAG Laser-Assisted Cartilage Reshaping For Treating Ear Protrusions.* Laryngoscope, 2015; 125: 2461-2467.