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· 综述 ·

唇腭裂患儿早期语音干预的研究进展

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【摘要】 唇腭裂患儿早期发生语音相关障碍风险较高, 这些障碍可能会持续到患儿语音发展后期并进展为代偿性发音错误, 且早期发音过程中的这些缺陷将直接影响到患儿随后的语音和词汇发展阶段。研究表明, 在唇腭裂患儿3岁以前提供聚焦刺激和环境教学等自然干预方式对患儿语言的发展有积极影响, 并且能增加患儿音量和提高辅音正确率。除了语音治疗师进行系统的语音治疗外, 家长也可以通过接受语音治疗培训为患儿进行早期语音干预。辅音正确率与辅音表、语音清晰度等其他指标相结合可以用来评估语音干预结果。然而, 迄今为止, 该领域的许多研究都集中在年龄较大的患儿上。因此, 有必要确定在早期语音典型发展阶段提供干预是否会带来更好的结果。未来关于语音干预方面的研究应因注重使用更系统性的语音干预方法, 探讨早期语音干预是否对唇腭裂患儿的语音以及今后的身心发展带来积极影响。

【关键词】 唇腭裂; 早期干预; 语音; 自然干预; 发音; 语音清晰度; 辅音正确率; 评估; 生活质量

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Research progress on early speech interventions for children with cleft lip and palate GULIZIBA · Aihaiti¹, YAERKEN · Aji², XU Hui³. 1. Postgraduate College of Xinjiang Medical University, Urumqi 830011, China; 2. Department of Stomatology, People's Hospital of Xinjiang Uygur Autonomous Region, Urumqi 830011, China; 3. Department of Oral and Maxillofacial Surgery, People's Hospital of Xinjiang Uygur Autonomous Region, Urumqi 830011, China

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【Abstract】 Children born with cleft lip and palate (CLP) are at high risk of disorders involving early speech production, and these problems can persist into later childhood, leading to the emergence of compensatory articulation errors. These difficulties in early vocalization directly impact the subsequent development of children's speech and vocabulary. Studies have shown that providing naturalistic interventions, such as milieu teaching and focused stimulation, for children with CLP in the first three years of life have positive impacts on the speech development of children, such as the potential to increase phonemic inventories and the percentage of correct consonants. In addition to speech and language therapists who perform systematic speech therapy, parents can be trained and supervised to deliver early speech intervention. The percentage of correct consonants can be used to assess outcome measures of speech intervention when combined with other measures, such as consonant inventory and speech intelligibility. However, much recent research in the field has focused on older children. Therefore, it must be determined if intervening during the early phase of typical speech development leads to better results. Future research should use more robust methodological designs to determine whether early speech intervention exhibits a positive impact on the speech and future physical and mental development of children with CLP.

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颅颌面裂是最常见的先天性缺陷之一,唇腭裂是最为常见的先天性颅颌面畸形^[1],我国非综合征型新生儿颅颌面裂的发生率为1.67:100^[2]。目前腭裂治疗的最基本方法是通过外科手术修复腭部裂隙。然而,尽管进行腭裂修复手术,术后仍有25.6%~32%的患者存在腭咽闭合不全(velopharyngeal insufficiency, VPI)^[3-5]。患者由于VPI出现高鼻音、鼻漏气和口腔压力弱,进而出现辅音省略、弱化等一系列腭裂语音障碍^[6-8]。语音障碍进而影响了患儿的语音发展过程。关于先天性唇腭裂患儿的大型横断面队列研究发现,5岁以上的患儿只有48%语音在正常范围内,而在唇腭裂患儿中,20%患儿在5岁以内的语音被认为是不可理解或几乎不可理解的,其严重影响了患儿与他人的交流^[9-10]。

先天性唇腭裂患儿在语音习得发展过程中存在的差异。在婴儿早期学习发音过程中,这些患者的语音就更能表现出延期习得,并只能发出非常有限的音位^[11]。并且,这些患儿能正确发出的音位数量也很少,尤其是无法发出压力性辅音(塞音、爆破音)等^[12]。许多唇腭裂患儿有一种典型的发音习惯,即试图使用口腔中更靠后的位置来代偿性发出爆破音,继而形成了代偿性发音错误^[13]。有证据表明,早期发音过程中的这些缺陷直接影响到患儿随后的语音和词汇发展阶段^[11]。鉴于先天性唇腭裂患儿会早期呈现语音发育障碍,对于这些患儿的语音干预也应该尽早开始^[14]。本文将回顾针对唇腭裂患儿3岁以内进行的语音干预的研究,并且对干预方法、干预者、干预年龄以及干预结果的评估等因素进行概述。

1 唇腭裂患儿的语音干预方法

目前国内外关于唇腭裂患儿3岁以内进行语音干预的研究较少。对于3岁以下的患儿来说,将某种独立语音设为治疗目标并包含发音重复训练的干预措施实施起来是很困难的,因为他们可能还没有达到该层次所需的意识和注意力^[12]。自然

干预(naturalistic interventions)是一种将目标音位针对性地嵌入到言语与日常对话中,并主要注重词汇量增加的早期干预方法。在干预过程中,当所选的干预词汇包括目标音位时,该方法可以有效促进发音和语言的发展^[15-16]。聚焦刺激(focused stimulation)^[17]和环境教学(milieu teaching)^[14]是自然干预方式的案例,这两种方法并不强调独立语音的发音,而更关注于语音与语言的形式。聚焦刺激重点包括对一种或多种具体的语音形式进行建模,例如对词汇、语法、句子结构或语音进行建模。儿童不需要模仿目标语音,但有机会在自然对话中,根据说话者发出的声音和单词等多个模型中学习并说出目标语音^[17]。环境教学是一种为了形成建模,促进儿童模仿声音和单词,鼓励他们自发表达,多说话的一种干预技巧^[14]。研究发现,强化环境教学能早期有效地提高患有语言障碍患儿的词汇量^[18]。

有多项研究表明,自然干预方法对患儿语音发展有积极的影响。在3岁以内进行早期自然干预,如包含或不包含音韵学的环境教学方法,或者聚焦刺激方法,可以促进先天性唇腭裂患儿音位量增长和辅音正确率(percentage of consonants correct, PCC)的增加^[14-15]。然而,由于干预程序的不一致性与干预结果存在差异,使得很难就3岁以下唇腭裂患儿的最佳干预方法达成一致^[18]。

2 早期语音干预的引导者

唇腭裂患儿语音治疗干预通常是直接由语音治疗师进行治疗,并由家长在家完成后续的治疗活动^[19]。家长参与语音干预很常见,它是唇腭裂患儿语音干预治疗的特点^[20]。研究表明,家长可以先接受培训,在家中为患有唇腭裂的患儿进行语音清晰度治疗,并建议家长为患儿进行语音清晰度治疗可以作为语音治疗服务的替代方法^[21]。有文献报道,3例21~27月龄唇腭裂患儿的母亲通过视频远程培训学习了强调音韵学的环境教学干预方法,并作为早期语音干预的引导者辅导患

儿语音治疗,语音治疗师发现所有患儿在词汇量、发音准确性和语音清晰度方面均有明显的提高^[22]。然而,家长实施干预的效果对家庭背景和家庭发展情况要求较高,因此该方法有一定局限性^[23]。

近年来,自动语音识别计算机技术的不断发展,计算机语音处理系统和频谱分析逐渐被应用到唇腭裂语音评价与治疗^[24]。目前国内外已有远程语音治疗平台用于评估病理语音,在线平台能够使医患双方进行便捷的实时交互,保证医师密切掌握患者治疗进度,以便制定个体精准、适应患者身心发育的治疗计划。

3 早期语音干预频率以及干预开始年龄

Kaiser等^[12]使用强化环境教学,对19例15~36月龄非综合征性唇腭裂患儿进行48节语音治疗课程,课程设置为1周2次,每节课时常为30 min。治疗接受后,干预组和对照组的PCC评分存在统计学差异,即接受语音干预后患儿的语音清晰度、辅音正确率明显有提高。另一项研究中,29例平均月龄为24.5个月的唇腭裂患儿接受了1周2次,每节课时长30~45 min,持续12周的强化环境教学干预方法。干预接受后,患儿的词汇量以及辅音正确率也显著提高^[14]。然而,也有针对患有语音障碍患儿的干预研究报告称,干预频率并不会影响最终语音效果。Cummings等^[25]研究者发现,无论干预的强度和频率如何,患有语音障碍的患儿在干预结束时都能获得类似的收益。然而,该作者认为,提供更高的干预频率可能是一种更有效的干预方式,因为儿童更有可能记住最后一次治疗中练习的技能,定期语音治疗对患儿语音改善有更好的效果。

目前的语音干预研究中,患儿年龄、评估时间和干预开始时间等信息不一致,难以确定干预开始时的年龄对语音结果的影响^[18]。2018年,美国颅面-腭裂协会(American Cleft Palate-Craniofacial Association)的指南规定,出生后患有唇腭裂的患儿语音发育应在婴幼儿早期进行监测,对语音障碍问题尽早干预^[18]。

4 语音评估方法

研究者对3岁以下唇腭裂患儿使用聚焦刺激干预方法,干预结束后,患儿的PCC评分显著提高。研究者使用PCC评分证明聚焦刺激干预方法能有效提高患儿的发音准确性以及减少患儿使用

声门塞音^[26]。PCC通常用于语音干预试验,但在使用它作为单一语音结果指标时应谨慎,因为它用于5岁以下患儿的语音评估时,可能会产生误导性的结果。这是因为使用PCC为指标的评估结果没能考虑到语音正常发展过程中的不成熟性,并且,当患者说出目标音位的准确度提高,但还未到正常范围内时,它不能准确地反映干预效果^[27]。然而,即使存在局限性,PCC与辅音表、语音清晰度等其他指标相结合,也是一个有价值的评估指标^[12,26]。唇腭裂患儿存在的沟通障碍对他们们的生活质量(quality of life, QOL)有明显影响,尤其是对患儿心理发育以及与同龄患儿的社交关系方面影响较大^[28-29]。在一项探讨3~18岁腭裂术后患者QOL的变化的研究中,家长填写的VELO量表结果表明,当患儿的发音越接近正常语音时,患儿的QOL得分越高^[29]。目前,中国版本的VELO量表也用于评估腭裂患儿的QOL^[30]。关于对唇腭裂患儿早期语音干预的结果,笔者建议使用中国版本的由家长填写VELO量表进行评估。

5 总结与展望

目前对唇腭裂患儿3岁以内提供的语音干预措施中,聚焦刺激和环境教学等自然干预方式有可能增加唇腭裂患儿的音位量和辅音正确率。除了语音治疗师系统地进行语音治疗外,家长也可以通过接受语音治疗培训为患儿进行早期语音干预。然而,由于干预使用方法和干预结果评估方面存在巨大差异,关于最有效的干预模式、干预者和语音持续时间以及建议的开始干预年龄仍未确定。未来的关于语音干预方面的研究应因注重使用更系统性的语音干预方法。

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参考文献

- [1] Worley ML, Patel KG, Kilpatrick LA. Cleft lip and palate[J]. Clin Perinatol, 2018, 45(4): 661-678. doi: 10.1016/j.clp.2018.07.006.
- [2] Fan D, Wu S, Liu L, et al. Prevalence of non-syndromic orofacial clefts: based on 15, 094, 978 Chinese perinatal infants[J]. Oncotarget, 2018, 9(17): 13981-13990. doi: 10.18632/oncotarget.24238.
- [3] Bruce MK, Zhang C, Vodovotz L, et al. Revision pharyngoplasty in cleft palate and velopharyngeal insufficiency: management and outcomes[J]. Ann Plast Surg, 2022, 88(3 Suppl 3): S152-S155. doi: 10.1097/SAP.0000000000003198.
- [4] McCrary H, Pollard SH, Torrecillas V, et al. Increased risk of velopharyngeal insufficiency in patients undergoing staged palate re-

- pair[J]. *Cleft Palate Craniofac J*, 2020, 57(8): 975 - 983. doi: 10.1177/1055665620913440.
- [5] Sakran KA, Al-Rokhami RK, Wu M, et al. Correlation of the Chinese velopharyngeal insufficiency - related quality of life instrument and speech in subjects with cleft palate [J]. *Laryngoscope Investig Otolaryngol*, 2022, 7(1): 180-189. doi: 10.1002/liv.2.705.
- [6] Bennun RD. Cleft palate repair: predictive factors of difficulty and planned strategies to solve it[J]. *J Craniofac Surg*, 2020, 31(6): 1664-1667. doi: 10.1097/SCS.0000000000006606.
- [7] Dubey AK, Prasanna SRM, Dandapat S. Detection and assessment of hypernasality in repaired cleft palate speech using vocal tract and residual features[J]. *J Acoust Soc Am*, 2019, 146(6): 4211. doi: 10.1121/1.5134433.
- [8] Lou Q, Wang X, Chen Y. Speech outcomes comparison between adult velopharyngeal insufficiency and patients with unrepaired cleft palate[J]. *J Craniofac Surg*, 2021, 32(2): 655 - 659. doi: 10.1097/SCS.0000000000006994.
- [9] Britton L, Albery L, Bowden M, et al. A cross - sectional cohort study of speech in five-year-olds with cleft palate ± lip to support development of national audit standards: benchmarking speech standards in the United Kingdom[J]. *Cleft Palate Craniofac J*, 2014, 51(4): 431-451. doi: 10.1597/13-121.
- [10] Larsson A, Miniscalco C, Mark H, et al. Persisting speech difficulties at 7-8 years of age - a longitudinal study of speech production in internationally adopted children with cleft lip and palate[J]. *Logoped Phoniatr Vocol*, 2022, 7(13): 1 - 10. doi: 10.1080/14015439.2022.2083673.
- [11] Chapman KL, Hardin-Jones M, Halter KA. The relationship between early speech and later speech and language performance for children with cleft lip and palate[J]. *Clin Linguist Phon*, 2003, 17(3): 173-197. doi: 10.1080/0269920021000047864.
- [12] Kaiser AP, Scherer NJ, Frey JR, et al. The effects of enhanced milieu teaching with phonological emphasis on the speech and language skills of young children with cleft palate: a pilot study[J]. *Am J Speech Lang Pathol*, 2017, 26(3): 806 - 818. doi: 10.1044/2016_AJSLP-16-0008.
- [13] Govathi Nikhila K. A study on patterns of compensatory articulation errors with reference to age of surgery in children with repaired cleft lip and palate[J]. *Glob J Otolaryngol*, 2017, 7(2): 15-30. doi: 10.19080/gjo.2017.07.555706.
- [14] Scherer NJ, Yamashita R, de Oliveira DN, et al. Early speech and language intervention in Brazilian-Portuguese toddlers with cleft lip and/or palate[J]. *Clin Linguist Phon*, 2022, 36(1): 34-53. doi: 10.1080/02699206.2021.1912187.
- [15] Scherer NJ. The speech and language status of toddlers with cleft lip and/or palate following early vocabulary intervention[J]. *Am J Speech Lang Pathol*, 1999, 8(1): 81 - 93. doi: 10.1044/1058 - 0360.0801.81.
- [16] Scherer NJ, Kaiser AP, Frey JR, et al. Effects of a naturalistic intervention on the speech outcomes of young children with cleft palate[J]. *Int J Speech Lang Pathol*, 2020, 22(5): 549 - 558. doi: 10.1080/17549507.2019.1702719.
- [18] Lane H, Harding S, Wren Y. A systematic review of early speech interventions for children with cleft palate[J]. *Int J Lang Commun Disord*, 2022, 57(1): 226-245. doi: 10.1111/1460-6984.12683.
- [19] DeVeeney SL, Hagaman JL, Bjornsen AL. Parent-implemented versus clinician-directed interventions for late-talking toddlers: a systematic review of the literature[J]. *Commun Disord Q*, 2017, 39(1): 293-302. doi: 10.1177/1525740117705116.
- [20] Sweeney T, Hegarty F, Powell K, et al. Randomized controlled trial comparing parent led therapist supervised articulation therapy (PLAT) with routine intervention for children with speech disorders associated with cleft palate[J]. *Int J Lang Commun Disord*, 2020, 55(5): 639-660. doi: 10.1111/1460-6984.12542.
- [21] Sugden E, Munro N, Trivette CM, et al. Parents' experiences of completing home practice for speech sound disorders[J]. *J Early Intervention*, 2019, 41(2): 159-181. doi: 10.1177/1053815119828409.
- [22] Philp J, Ellis PK, Scherer NJ, et al. Enhanced milieu teaching with phonological emphasis: a pilot telepractice parent training study for toddlers with clefts[J]. *Children*, 2021, 8(9): 736. doi: 10.3390/children8090736.
- [23] Sugden E, Baker E, Munro N, et al. Involvement of parents in intervention for childhood speech sound disorders: a review of the evidence[J]. *Int J Lang Commun Disord*, 2016, 51(6): 597 - 625. doi: 10.1111/1460-6984.12247.
- [24] 郭毅波, 蔡鸣. 计算机科学应用于唇腭裂语音诊疗的研究进展[J]. *口腔疾病防治*, 2022, 30(6): 453-456. doi: 10.12016/j.issn.2096-1456.2022.06.012.
- Guo YB, Cai M. Research progress of computer science in cleft lip and palate speech therapy[J]. *J Prev Treat Stomatol Dis*, 2022, 30(6): 453-456. doi: 10.12016/j.issn.2096-1456.2022.06.012.
- [25] Cummings A, Giesbrecht K, Hallgrimson J. Intervention dose frequency: phonological generalization is similar regardless of schedule[J]. *Child Lang Teach Ther*, 2021, 37(1): 99-115. doi: 10.1177/0265659020960766.
- [26] Scherer NJ, Williams AL, Proctor-Williams K. Early and later vocalization skills in children with and without cleft palate[J]. *Int J Pediatr Otorhinolaryngol*, 2008, 72(6): 827 - 840. doi: 10.1016/j.ijporl.2008.02.010.
- [27] Sell D, Sweeney T. Percent consonant correct as an outcome measure for cleft speech in an intervention study[J]. *Folia Phoniatr Logop*, 2020, 72(2): 143-151. doi: 10.1159/000501095.
- [28] Vuletić M, Gabrić D, Sušić M, et al. Development of a valid and reliable instrument for the assessment of quality of life in parents of children with clefts[J]. *J Stomatol Oral Maxillofac Surg*, 2020, 121(5): 527-533. doi: 10.1016/j.jormas.2019.11.002.
- [29] Pedersen HM, Goodie PA, Braden MN, et al. Comparing quality of life and perceptual speech ratings in children with cleft palate[J]. *Cleft Palate Craniofac J*, 2021, 58(2): 139 - 145. doi: 10.1177/1055665620949435.
- [30] Lu L, Yakupu A, Wu Y, et al. Quality of life in patients with velopharyngeal insufficiency in West China[J]. *Cleft Palate Craniofac J*, 2022, 59(8): 1024-1029. doi: 10.1177/10556656211034107.

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