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Behavioral assessment of auditory processing disorder in children with non-syndromic cleft lip and/or palate

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ABSTRACT

Objective: Peripheral hearing disorders have been frequently described in children with non-syndromic cleft lip and/or palate (NSCL/P). However, auditory processing problems are rarely considered for children with NSCL/P despite their poor academic performance in general compared to their craniofacially normal peers. This study aimed to compare auditory processing skills, using behavioral assessment techniques, in school age children with and without NSCL/P.
Methods: One hundred and forty one Mandarin-speaking children with NSCL/P aged from 6.00 to 15.67 years, and 60 age-matched, craniofacially normal children, were recruited. Standard hearing health tests were conducted to evaluate peripheral hearing. Behavioral auditory processing assessment included adaptive tests of temporal resolution (ATTR), and the Mandarin pediatric lexical tone and disyllabic-word picture identification test in noise (MAPPID-N).
Results: Age effects were found in children with cleft disorder but not in the control group for gap detection thresholds with ATTR narrow band noise in the across-channel stimuli condition, with a significant difference in test performance between the 6 to 8 year group and 12 to 15 year group of children with NSCL/P. For MAPPID-N, the bilateral cleft lip and palate subgroup showed significantly poorer SNR-50% scores than the control group in the condition where speech was spatially separated from noise. Also, the cleft palate participants showed a significantly smaller spatial separation advantage for speech recognition in noise compared to the control group children.
Conclusion: ATTR gap detection test results indicated that maturation for temporal resolution abilities was not achieved in children with NSCL/P until approximately 8 years of age compared to approximately 6 years for craniofacially normal children. For speech recognition in noisy environments, poorer abilities to use timing and intensity cues were found in children with cleft palate and children with bilateral cleft lip and palate compared to craniofacially normal children. Consequently, it is worthwhile to consider the potential for auditory processing disorder in when assessing the auditory status of children with NSCL/P.

Introduction

Clefts of the lip and/or palate (CL/P) are recognized as common congenital craniofacial malformations. A recent study [1] indicated the overall global prevalence of CL/P was 0.992/1000 live births, and the prevalence of cleft lip and palate (CLP) was significantly greater than cleft lip (CL), with prevalence of 0.664/1000 and 0.328/1000, respectively. Among children with cleft disorders, approximately 70% occur as isolated CL/P not associated with other cognitive or craniofacial structural abnormalities, and these cases are regarded as non-syndromic cleft lip and/or palate (NSCL/P) [2]. In China, the incidence of CL/P is of concern. A survey in Shanghai showed the incidence of NSCL/P in the 1980s was 1.11/1000 [3]. At present there are at least 30,000 babies with CL/P born in China every year, and the prevalence rate of all types of cleft is 1.30/1000, with 1.20/1000 being non-syndromic CL/P [4].

For children with NSCL/P, considerable emphasis has been given their high prevalence of middle ear deficits that are most likely due to Eustachian tube dysfunction [5]. Inner ear disorder is rare in children with NSCL/P compared to children with syndromic clefts. However, little is known concerning the prevalence of auditory processing disorder (APD) in this population. A study of young adult men with cleft lip and/or palate, by Nopoulos and colleagues [6], revealed patients with orofacial clefts had radiological indications of abnormal cortical regions, including significant differences in tissue volume and thickness,

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with the most significant differences in the left temporal lobe, which may imply differences in auditory processing abilities. More recently, Yang and colleagues [7] also found significantly decreased volume and thickness for the left superior temporal plane, and other cortical developmental abnormalities, in 6 to 24 month infants with NSCL/P. Individuals with APD typically show poor performance with one or more of the following auditory behaviors: sound localization and lateralization; auditory discrimination; auditory pattern recognition; temporal aspects of audition; auditory performance decrements with competing acoustic signals and degraded acoustic signals [8]. It is more effective to estimate auditory processing deficits and the functional impact of these deficits in older children and adults using behavioral tests rather than through electrophysiological assessment [9]. Clinically, behavioral assessment plays an important role in determining the auditory abilities of children with APD.

It has been strongly recommended that behavioral test batteries should contain both verbal and non-verbal test stimuli to explore different auditory processing skills [10]. Temporal resolution, which refers to the ability to follow rapid changes in intensity and frequency over time, is considered an important processing factor for accurate understanding of speech in noisy environments [11]. Gap detection tests are often used clinically for nonspeech evaluation of temporal resolution abilities. Verbal tests, using speech sounds as test stimuli to assess auditory function, aim to measure how well listeners understand speech in a controlled listening environment, thereby reflecting their likely performance in actual communication conditions.

Yang [12] evaluated a behavioral assessment battery for auditory processing abilities in Chinese children with NSCL/P. Eighteen school age children with NSCL/P (and normal peripheral hearing) and 65 craniofacially normal children were recruited. For the hearing in noise test (HINT) [13], children with NSCL/P were found to be significantly more adversely affected by noise than the control participants. In addition, performance on the gaps in noise test [14] in children with NSCL/P was significantly poorer than in the control children. Although of limited sample size, this study indicated that children with NSCL/P may exhibit abnormal auditory processing abilities compared to their craniofacially normal peers.

The current study aimed to utilize both verbal and non-verbal auditory behavioral tests for auditory processing skills in a large sample of school age children with NSCL/P, and to compare their performance to that of craniofacially normal children, to determine whether auditory processing difficulties are more prevalent in the pediatric population with cleft disorders.

Materials and methods

Participants

One hundred and forty one children (96 males; 45 females) with NSCL/P aged from 6.00 to 15.67 years (mean = 10.16) were recruited into the current study. All of the children were native Mandarin speakers and attended regular schools. They were divided into three subgroups by cleft type: 37 children with CL, 24 children with CP, and 80 children with CLP, who were further divided into two categories: BCLP and UCLP. They were classified into three age groups: 47 children aged from 6 to 8 years, 57 children aged from 9 to 11 years, and 37 children aged from 12 to 15 years. The children were visiting the outpatient department of the Cleft Lip and Palate Clinic Center, Beijing Stomatology Hospital for further consultation, as recommended by their primary care doctors. The hospital is a large polyclinic and patients visit the Cleft Lip and Palate Center from all regions of China. The control group comprised 60 participants who were craniofacially normal children (25 boys and 35 girls) aged from 6.00 to 15.50 years (mean = 10.16). Among them were 20 children aged from 6 to 8 years; 20 children aged from 9 to 11 years; and 20 children aged from 12 to 15 years. Participants provided their informed consent before joining the research program by completing parent consent and student assent forms. The study was approved by the Human Research Ethics Committee for Non-Clinical Faculties, The University of Hong Kong (reference number EA140811).

Hearing health tests

All participant children with NSCL/P and their craniofacially normal peers undertook auditory screening tests to assess their peripheral hearing. The screening protocol included otoscopic examination, pure tone audiometry, 226 Hz probe tone tympanometry and ipsilateral 1 kHz acoustic reflex threshold measures. A diagnostic otoscope, a SA 204 diagnostic audiometer (Entomed, Sweden), and a GSI 39 automatic tympanometry middle ear analyzer (GSI Corp., USA) were utilized to perform the assessment. Screening tests were conducted in a quiet research room, and the background noise was no more than 35 dBA at any time. To be considered to have normal hearing, the external ear canal was visualized as unobstructed, with any cerumen impaction removed prior to testing. Each participant pure tone audiometric air-conduction threshold level was required to be 25 dBHL or less at a four frequency average (0.5, 1, 2, and 4 kHz) in both ears. Furthermore, all recruited children were required to have type A tympanograms [15], and acoustic reflex thresholds $\leq$100 dB SPL. If these screening criteria for bilateral normal peripheral auditory function were met, the child then completed the following behavioral assessment.

Behavioral auditory processing tests

Two behavioral tests were utilized to evaluate the auditory processing ability of the subjects: (a) adaptive tests of temporal resolution (ATTR) [16], and (b) Mandarin pediatric lexical tone and disyllabic-word picture identification test in noise (MAPPID-N) [17].

ATTR is a gap detection test procedure that assesses temporal resolution function by both bursts of broadband noise and narrowband noise. The software produces an adaptive two-interval, two-alternative forced-choice (2I/2AFC) psychophysical paradigm targeting 70.7% correct gap detection. Listeners hear three sounds with different intervals. The first sound has the standard interval with two noise bursts separated by a silent period of known gap duration. This standard stimulus is presented to provide patients with an immediate reference for comparison to the second and third sounds. After the first sound, the second and third sounds are presented, one of which has the same gap duration as the standard sound, and the gap duration of the other (the target interval) is adaptively varied, based on the patient's previous response. The stimuli presentation level used was the highest comfortable loudness level, which was obtained by presenting the practice items and adjusting the volume in a standard manner. The task of the listener is to select the odd stimulus—the one that is different from the other two. If the listener selects the correct answer (the target interval), the gap duration in the target interval is decreased. Conversely, if the listener selects an incorrect answer, the gap duration in the target interval will be increased. The whole test protocol includes four conditions, narrow-band noise within-channel gap detection (NBN-WC) and narrow-band noise across-channel falling gap detection (NBN AC-F), narrow-band noise across-channel rising gap detection (NBN AC-R), and broad-band noise within-channel