Korean language specific dysarthria associated with idiopathic peripheral facial palsy

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Abstract
To investigate the patterns of dysarthria in Korean patients with idiopathic peripheral facial palsy. Seventy-eight patients diagnosed with idiopathic peripheral facial palsy within the onset of symptom to 7 days were prospectively enrolled. The initial symptom of facial palsy was examined by the House-Brackmann scale. All patients were tested by Urimal-Test of Articulation and Phonology-2 (U-TAP-2), which is specialized for the evaluation of dysarthria in Korean language - Hangeul - when the patients first visited and were followed up at 4 weeks after the onset, respectively. The facial electromyography was performed after 7 days, since the presentation of the first symptom. Electric stimulation therapy and simple face exercise education were performed in all patients as routine treatments for facial palsy with or without dysarthria. The patterns of dysarthria were analyzed by initial and follow-up U-TAP-2 results, respectively.

Among 78 patients, 50 patients (64.1%) had dysarthria in the first assessment. The 6 consonants and 3 vowels were errored in U-TAP-2 test. The bilabial consonants “[p]” or “[b]” were substituted with labiodental consonant “[f]”, and palato-alveolar consonants were replaced by alveolar consonants - “[k]” to “[t]”. Bilabial consonant of “[m]” was replaced by velar nasal consonant of “[n]”. Liquid consonant was altered to nasal sound. For example, “[r]” is replaced by “[l]”. The velar consonant of “[k]” was pronounced as “[g]”. The diphthong vowels “[wa]” were pronounced as monothong “[a]”, “[i]”, or “[e]”, or “[i]”. The most common error was substitution. Among 78 patients with idiopathic peripheral facial palsy, 50 patients had dysarthria and 14 out of 50 patients with dysarthria lasted more than 4 weeks. Five consonants (“[p]”, “[k]”, “[t]”, “[w]”, “[n]”) and 3 vowels (“[a]”, “[u]”, “[e]”) were still mispronounced after 4 weeks and most common error was substitution. Therefore, speech evaluation and speech therapy specialized for errors in high frequency of consonants and vowels are needed in patients with idiopathic peripheral facial palsy, in Korea.

Abbreviations: ENoG = electroneurography, HBS = House-Brackmann scale, U-TAP-2 = Urimal-Test of Articulation and Phonology-2.

Keywords: Bell palsy, dysarthria, facial palsy, idiopathic facial palsy, Korean alphabet, Korean language, rehabilitation

1. Introduction
Dysarthria is defined as motor speech disorder resulting from central or peripheral nervous system abnormalities, including impairment to articulation, phonation, resonation, and respiration.[1] Dysarthria is common in various conditions that cause disturbance in controlling speech production, such as Parkinson disease, amyotrophic lateral sclerosis, multiple sclerosis, cerebral palsy, stroke, and peripheral neuropathy.[2]

Bell palsy, or idiopathic form, is one of the peripheral facial palsy, which is characterized by acute onset, unilateral facial muscle weakness.[3] The incidence is reported to be 20–35 per 100,000 per year.[4,5] Patients with facial palsy suffer from not only motor dysfunction but also non-motor disabilities such as social, communicative, and psychological difficulties because of cosmetic problems, speech disturbance, and lack of emotional expressions.[6–8] Therefore, evaluation of facial palsy should be accompanied by evaluation of speech assessment. However, despite the fact that patients with idiopathic peripheral facial palsy are known to have problems with specific pronunciation, there is less research on specific patterns of dysarthria in idiopathic peripheral facial palsy.[9,10]
Therefore, in this research, we investigated the speech pattern and factors influencing the persistence of dysarthria in Korean patients with idiopathic peripheral facial palsy.

2. Methods

The initial symptom of facial palsy was examined by the House-Brackmann scale (HBS). All patients were tested by Urimal-Test of Articulation and Phonology-2 (U-TAP-2), which is specialized for evaluation of dysarthria in Korean language - Hangeul - when the patients first visited and were followed up at 4 weeks after the onset, respectively. The facial electromyography was performed after 7 days since the presentation of the first symptom. Electric stimulation therapy and simple facial exercise education were performed in all patients as routine treatments for facial palsy with or without dysarthria. The patterns of dysarthria were analyzed by initial and follow-up U-TAP-2 results, respectively.

2.1. Subject

Seventy-eight patients with idiopathic peripheral facial palsy were recruited from rehabilitation department of single tertiary institution in Korea.

Inclusion criteria were

1. patients clinically diagnosed with idiopathic peripheral facial palsy,
2. time since onset of facial palsy at or less than 7 days,
3. age between 10 and 90,
4. patients with cognitive function enough to participate assessment (≥28 points on Korean version of Mini-Mental State Examination; MMSE-K).

Diagnostic criteria of idiopathic peripheral facial palsy were

1. peripheral cranial nerve VII palsy;
2. absence of cranial neuropathy of other than cranial nerve VII or neurologic deficit, and
3. absence of identifiable causes other than herpes simplex virus infection, which is presumed to be a causative agent.

Exclusion criteria were

1. facial palsy with other etiologies - stroke, head trauma, Parkinson disease, neurologic disorders such as polyneuropathy, Guillain-Barré syndrome, myasthenia gravis, multiple sclerosis, muscular disorders such as muscular dystrophy, otitis media, or malignancies;
2. premorbid language disorders including dysarthria, apraxia of speech, and aphasia.

Our Institutional Review Board approved the study design (No. OC15OISI0151). Written informed consent was obtained from the patients for this research after they had been briefed about the study.

2.2. Speech evaluation

2.2.1. Hangeul (Korean alphabet). The Korean alphabet, Hangeul, is composed of 24 characters, with 14 consonants (ㄱ, ㄴ, ㄷ, ㄹ, ㅁ, ㅂ, ㅅ, ㅈ, ㅊ, ㅋ, ㅌ, ㅍ, ㄹ, ㅎ) and 10 vowels (ㅏ, ㅑ, ㅓ, ㅕ, ㅗ, ㅛ, ㅜ, ㅠ, ㅡ, ㅣ). Double consonants made up of 2 consonants (eg, ㅗ, ㅜ) and diphthongs made up of 2 vowels (eg, ㅐ, ㅒ) are pronounced as one sound. Korean syllables are constructed with a consonant, a vowel, and an optional consonant. Syllables are typically started with initial consonant (cho-seong), followed by a vowel (jung-seong), and an optional consonant (jong-seong). A syllable ending with a consonant is called a closed syllable, whereas a syllable ending with a vowel an open syllable. For example, the closed syllable “ㅌ” is written and pronounced in the order of “L, /-n/”, “ㅏ”, “ㅏ, /-m/”, whereas the open syllable “ㅌ” is written and pronounced in the order of “L, /-n/”, “ㅏ, /-m/”.

2.2.2. Urimal-Test of Articulation and Phonology-2 test. All patients were tested by U-TAP-2 when they first visited the department of rehabilitation and followed up after 4 weeks. U-TAP-2 is a standardized assessment tool for evaluation of dysarthria in Korean language, Hangeul.[12] It is originally designed for children aged between 2 and 12 and commonly used for evaluating dysarthria in children with intellectual disorder, hearing impairment, behavioral disorder, or cerebral palsy. It detects the abnormalities of speech production in level of words and sentences by instructing subjects to read written language and to name objects or explain situation described in pictures. U-TAP-2 test is composed of 43 consonants and 10 vowels. In U-TAP-2 test, the phonemes of each position were considered to be independent, and a total of 43 phonemes were examined for consonant evaluation. For example, “ㄸ ㄸ” is composed of 2 different consonant “ㄸ” in the initial and the last syllable.

In our research, we omitted the courses of describing pictures or reading sentences and only instructed patients to read prepared words in an attempt to focus on detecting articulation patterns as our patients did not have cognitive dysfunction or aphasia. Patients were instructed to read 30 words in two or three times. One speech-language pathologist tested the patients, recorded the speech sample, and analyzed the articulation patterns to minimize measurement bias.

The number of words with error was counted, and the error patterns were analyzed (such as omission, substitution, addition, etc.). Articulatory accuracy, which is an indicator of how accurately the speaker pronounces the words, calculated by the number of phonemes correctly pronounced divided by the number of whole phonemes, was assessed.[13] In addition, the percentage of consonant correct and the percentage of vowels correct calculated by the number of correctly pronounced consonants/vowels divided by the number of whole consonants/vowels were calculated.[14,15]

2.3. Severity evaluation of facial palsy

All patients were classified by severity of facial palsy using HBS from grade I to VI at initial evaluation and within 7 days after the onset of facial palsy. House-Brackmann grade I means normal facial function. Grade VI describes total paralysis, in which no movement is observed in any area.[16]

All patients underwent electrodiagnostic studies within 7 days after the onset of facial palsy. Side-to-side differences in compound action potential of facial motor nerve on bilateral orbicularis oris muscle were assessed. Rehabilitation therapy including electrical stimulation therapy and simple facial expression exercise education were performed in all patients.

2.4. Statistics

We conducted Student t-test to see if there was a difference in the age and electroneurography (ENoG) in orbicularis oris muscle between 2 groups. Also, chi-square test and Fisher exact test were used to identify association between the groups with and without dysarthria according to gender or House-Brackmann grade,
respectively. Finally, we performed logistic regression analysis to find the factors affecting the presence or the absence of persistent dysarthria. Data analyses were performed with SPSS ver. 21.0 for Windows (SPSS Inc., Chicago, IL, USA). The significance level for all statistical analyses was set at p < 0.05.

3. Results

Among 78 patients, 50 patients (64.1%) had impaired articulation. Dysarthria was checked in 6 consonants and 3 vowels at initial evaluation. The baseline characteristic of patient with or without dysarthria is described in Table 1. The patients with dysarthria was older than the patients without dysarthria (56.66 ± 14.17 vs 39.86 ± 15.33, P = .000). Also, there was significant difference of ENoG in orbicularis oris muscle between the patients with and without dysarthria (43.93 ± 26.35 vs 61.95 ± 15.33, P = .002). On the other hand, the patients with dysarthria significantly tend to show higher House-Brackmann grade than the patients without dysarthria (P = .015).

3.1. Analysis of dysarthria patterns

Velar consonant “TI”[k] in “꼬리”[kori] was pronounced as velar consonant “TI”[k] in the initial syllable (Fig. 1A). This is the only pattern of dysarthria that disappeared in follow-up test, and all other patterns were still observed after 4 weeks. Jong-seong bilabial consonant “로”[l] in “역마”[lyemma] was substituted with velar nasal “O”[n] in “명마” [myema] (Fig. 1B). Palate-alveolar “F”[θ] was pronounced as alveolar plosive “F”[θ], (Fig. 1C). Bilabial consonants “WP”[p] from “뽀뽀”[popo] were replaced by labiodental [f] in both initial and last syllable (Fig. 1D). “Ψ”[pʰ] in “풍선”[pʰwun] and “연필” [janpʰ] also were pronounced as [f] (Fig. 1D). Liquid consonant “M”[r] in “로봇”[robot] was pronounced as alveolar nasal consonant “LEE”[n] in initial syllable (Fig. 1E). Double vowel “A”[u] in “귀”[gui] was altered to “I”[i] as “GI”[ki] and double vowel “A”[a] in “괴물” [kumul] was substituted with “AI”[ai] as “아이”[ai] (Fig. 2A). Diphthong “ sợ”[wa] in “전화”[jeonwa] was altered to monophthong “AI”[ai] as “전 화”[jeonha] (Fig. 2B), and “옷”[mot] were segmented to 2 syllable “으옷”[muot], due to slow speech problem (Fig. 2C).

The pronunciations of phonemes and words were described based on International Phonetic Alphabet. As mentioned earlier, all patterns of how consonant changes by dysarthria in U-TAP-2 test can be explained by substitution. In addition, most variations in vowels are also explained by substitution. Moreover, the most change of consonants occurred in the “cho-seong”, not the “jong-seong”, except for one word. After 4 weeks, 14 out of 50 patients (28%) showed persistent dysarthria and still showed pronunciation errors in 3 consonants and 3 vowels. (단주, 뽀뽀, 연필, 엽마, 로 몫, 괴물, 전화).

3.2. Affecting factors

After following up after 4 weeks since the onset, we presented the affecting factors of patients with or without persistent dysarthria (Table 2). The results showed House Brackmann grade was the only significant factor affecting persistent dysarthria (P = .030). On the other hand, age, gender, and ENoG in orbicularis oris muscle had no significant effect on persistence of dysarthria.

4. Discussion

In the present study, 64.1% (50/78) of patients with idiopathic peripheral facial palsy developed dysarthria within 7 days from the onset of symptom. Dysarthria continued to be present in 28.0% (14/78) of patients after 4 weeks. The most common error was substitution. Dysarthria is classified by its characteristics into flaccid, spastic, ataxic, hypokinetic, hyperkinetic, unilateral upper motor neuron, and mixed types.[17] Patients with idiopathic peripheral facial palsy present flaccid type of dysarthria, which accounts for about 8% of primary communication disorder.[17] Flaccid type of dysarthria is due to unilateral or bilateral pathology of lower motor neuron, neuromuscular junction, or muscle, which causes weakness and impaired coordination in muscles responsible for respiration, phonation, resonance, and articulation.[13] Imprecise consonants and distorted vowels in patients with flaccid dysarthria are correlated with poor speech intelligibility, which is the concept of understandability of the speech.[1] As impaired movement due to facial neuropathy is the main problem in patients with idiopathic peripheral facial palsy, components

| Table 1 |
| Baseline characteristics of patients (n=78). |
| Total patients (n=78) | Patients with dysarthria (n=50) | Patients without dysarthria (n=28) | P value |
| --- | --- | --- | --- |
| Average age (years) | 50.63 ± 16.71 | 56.66 ± 14.17 | 39.86 ± 15.33 | 0.000* |
| Gender (male: female), n | 36:42 | 22:28 | 14:14 | 0.610* |
| Initial HB grade, n | 3 | 3 | 1 | 0.015* |
| III | 14 | 8 | 6 | 0.030* |
| II | 31 | 18 | 13 | 0.030* |
| IV | 18 | 12 | 6 | 0.030* |
| V | 10 | 10 | 0 | 0.030* |
| VI | 2 | 2 | 0 | 0.030* |
| ENoG in orbicularis oris muscle | 49.76% ± 26.48 | 43.93% ± 26.35 | 61.95% ± 21.93 | 0.002* |
| PCC | 97.49% (±3.53%) | 96.09% (±3.53%) | 100% | 0.000* |
| PVC | 91.26% (±10.41%) | 86.36% (±10.00%) | 100% | 0.000* |
| Articulatory accuracy | 96.25% (±4.21%) | 94.15% (±3.87%) | 100% | 0.000* |

ENoG = Electroneurography, HB grade = House-Brackmann grade, PCC = percentage of consonant correct, PVC = percentage of vowel correct.

* P < .05 for differences between patients with and without dysarthria by Student’s t-test.

† P < .05 for differences between patients with and without dysarthria by Chi-square test.

‡ P < .05 for differences between patients with and without dysarthria by Fisher exact test.

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regarding respiration, phonation, and resonance are generally not affected and patients mainly present impaired articulation. In pronouncing consonants, substitution of bilabial consonants with labiodental consonants such as pronouncing /ㅍ/, /ㅃ/ as /f/ was the most common error observed in patients in our study. Patients had difficulty closing lips together completely because of unilateral weakness of orbicularis oris muscle. On the other hand, patients had difficulty pronouncing vowels. Instead of pronouncing double vowels as one sound, patients pronounced 2 vowels separately. For example, they pronounced "귀" as "/ku-I/", not "/kwi/". This finding is supposed to be due to inappropriate tongue movement with delayed transition time of moving the tongue from posterior to anterior position as described in previous research. To pronounce double vowels, tongue and mandible need to be fixed, which is difficult for patients with idiopathic peripheral facial palsy to perform. Prolonged phonemes were observed in 5 patients, for they pronounced "못"[mot] as "므옷"[mɯot]. Weakness in orbicularis oris muscle producing slow, weak, inaccurate, and irregular movement is speculated to be responsible for prolonged phonemes.

In this study, we were able to identify frequent patterns that may appear in patients with idiopathic peripheral facial palsy, which would allow more efficient treatment by presenting an error to be corrected intensively when performing speech therapy.
From the perspective of clinical assessment, HBS III or higher suggests poor outcomes. Our results demonstrated significant association between HBS and presence of persistent dysarthria. Patients who have assessed to be severe by HBS presented persistent dysarthria. Therefore, we could confirm a significant connection between ENoG and dysarthria at the beginning of this study. As our statistical analysis revealed, however, ENoG was not a significant factor affecting the presence of persistent dysarthria after 4 weeks. From this perspective, speech therapy for patients with severe facial palsy needs to be provided especially because patients with higher grade of HBS are more likely to have persistent dysarthria. Further researches would be beneficial to see if the long-term treatment effect in idiopathic peripheral facial palsy differs according to HBS.

Dysarthria has a negative impact on the quality of life not only by inducing difficulty in communicating with other people but also by negatively changing psychological aspects such as self-identity and emotional disruptions. Patients with facial palsy are confronted with psychosocial problems due to discomfort in daily life and their own appearance, which can be aggravated by dysarthria. Therefore, speech training to improve dysarthria as well as neuromuscular retraining for restoring facial motor strength are needed for patients with facial palsy. Flaccid dysarthria can benefit from speech rehabilitation targeting strengthening of lip and perioral muscles. Strengthening such as pressing lips together hard and providing auditory feedback by surface electromyography from orbicularis oris muscle has shown to be effective in improving flaccid dysarthria in previous research.

This study has some limitations such that the patient with mild and subjective dysarthria who was diagnosed as negative on the test was overlooked. Also, the effect of dysarthria on the quality of life of the patients was unknown. In addition, all patients in this study were planned for follow-up only after 4 weeks, which suggests a possibility of long-term follow-up in future studies. Furthermore, the subject of the present study was limited to Korean, however, it is significant that our study is the first to focus on the dysarthria of idiopathic facial palsy patients and its patterns. Therefore, we are looking forward to further developing speech therapy in idiopathic facial palsy patients, with subsequent studies based on various languages, such as English, which is used by a much larger population.

In summary, among 78 patients with idiopathic peripheral facial palsy, 50 (64.1%) patients initially had dysarthria, of which 14 (28%) patients still showed persistent dysarthria after 4 weeks. Error was shown in 6 consonants and 3 vowels, and 5 consonants and 3 vowels were still mispronounced after 4 weeks. Therefore, speech evaluation and speech therapy specialized for these errors of consonants and vowels in high frequency are necessary in patients with idiopathic peripheral facial palsy.

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### Author contributions
Dae-Hyun Jang, MD, PhD takes responsibility for the study as a whole.
Dong-Woo Lee and Dae-Hyun Jang wrote the article and critically revised articles.
Ja-Young Oh and Dae-Hyun Jang contributed to study design and reviewed articles.
Ja-Young Oh, Mi-Hyang Han, and Dae-Hyun Jang contributed to data analysis.
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