



Speech characteristics after articulation therapy in children with cleft palate and velopharyngeal dysfunction – A single case experimental design



Fatemeh Derakhshandeh ^{a,b}, Mohammadreza Nikmaram ^c,
Hedieh Hashemi Hosseinabad ^d, Mehrdad Memarzadeh ^e, Masoud Taheri ^f,
Mohammadreza Omrani ^g, Shohreh Jalaie ^h, Mahmood Bijankhan ⁱ, Debbie Sell ^{j,*}

^a Department of Speech Therapy, School of Rehabilitation, Iran University of Medical Sciences, Tehran, Iran

^b Craniofacial Anomalies and Cleft Palate Research Center, Isfahan university of Medical Sciences, Isfahan, Iran

^c Department of Physiology, School of Rehabilitation, Iran University of Medical Sciences, Tehran, Iran

^d Department of Communication Sciences and Disorders, College of Allied Health Sciences, University of Cincinnati, Cincinnati, USA

^e Department of Surgery, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran

^f C.T Scan Fellowship, Social Security Organization, Isfahan, Iran

^g IBCFPRS, Department of Otorhinology, Alzahra Hospital, Isfahan University of Medical Sciences, Isfahan, Iran

^h Department of physiotherapy, Biostatistics, School of rehabilitation, Tehran University of Medical sciences, Tehran, Iran

ⁱ Laboratory of Linguistics Department, University of Tehran, Tehran, Iran

^j North Thames Regional Cleft Centre, Speech and Language Therapy Department, Great Ormond Street Hospital Trust for Children, London, UK

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ABSTRACT

Objective: The aim of this study was to investigate the impact of an intensive 10-week course of articulation therapy on articulation errors in cleft lip and palate patients who have Velopharyngeal Insufficiency (VPI), non-oral and passive cleft speech characteristics.

Methods: Five children with cleft palate (+/-cleft lip) with VPI and non-oral and passive cleft speech characteristics underwent 40 intensive articulation therapies over 10 weeks in a single case experimental design. The percentage of non-oral CSCs (NCSCs), passive CSCs (PCSCs), stimulative consonants (SC), correct consonants in word imitation (CCI), and correct consonants in picture naming (CCN) were captured at baseline, during intervention and in follow up phases. Visual analysis and two effect size indexes of Percentage of Nonoverlapping Data and Percentage of Improvement Rate Difference were analyzed.

Results: Articulation therapy resulted in visible decrease in NCSCs for all 5 participants across the intervention phases. Intervention was effective in changing percentage of passive CSCs in two different ways; it reduced the PCSCs in three cases and resulted in an increase in PCSCs in the other two cases. This was interpreted as intervention having changed the non-oral CSCs to consonants produced within the oral cavity but with passive characteristics affecting manner of production including weakness, nasalized plosives and nasal realizations of plosives and fricatives. Percent SC increased throughout the intervention period in all five patients. All participants demonstrated an increase in percentage of CCI and CCN suggesting an increase in the consonant inventory. Follow-up data showed that all the subjects were able to maintain their ability to articulate learned phonemes correctly even after a 4-week break from intervention.

Conclusion: This single case experimental study supports the hypothesis that speech intervention in patients with VPI can result in an improvement in oral placements and passive CSCs.

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1. Introduction

Development of normal speech is one of the most important objectives in the management of children with cleft lip and palate [1]. Speech development in individuals with cleft palate is affected by many factors, the primary one being persistent velopharyngeal

* Corresponding author at: North Thames Regional Cleft Centre, Speech and Language Therapy Department, Great Ormond Street Hospital Trust for Children, London, UK. Tel.: 0044 207 813 8110; fax: 0044 207 813 8278.

E-mail address: Debbie.Sell@gosh.nhs.uk (D. Sell).

insufficiency (VPI). VPI is known to occur following primary palate surgery which is expected in around 30% of children [2]. In a retrospective study of children referred to the Isfahan Cleft team between 2005 and 2009, the prevalence of post-surgical moderate – severe VPI was estimated at 66.5% based on 131 file reviews [3]. VPI results in excessive nasal resonance (hypernasality) on vowels and vocalic consonants, nasal emission, nasal turbulence, and/or reduced intraoral air pressure on obstruent consonants [2]. There are two main ways in which consonant production is affected. One is a process reflecting the structural defect; in this, the patient cannot lessen the unavoidable consequences of oral–nasal coupling, resulting in nasalized plosives and fricatives, weak and/or nasalized consonants, nasal realization of plosives and/or suspected passive nasal fricative and gliding [4,5]. In contrast, there is an active strategy, where the individual tries to prevent the effects of oral–nasal coupling and changes the place and/or manner of articulation [4]. In this strategy, the placement of oral pressure consonants (e.g., plosives, fricatives and affricates) is made further back in the vocal tract often valving below the velopharyngeal (VP) port at the pharyngeal and glottal places of articulation [6]. These backed productions were referred to as *compensatory articulations* (CA) by Trost [7] in her landmark paper, and as non-oral *cleft speech characteristics* (CSCs) by a number of European researchers [5,8–10].

Resonance disorders associated with VPI are usually corrected by physical management of the VP sphincter, either through secondary surgery or prosthetic appliances [2]. However, successful surgical correction of VPI does not eliminate non-oral CSCs [2,11]. Therefore, identification and intervention of these articulation errors is frequently essential for effective treatment planning [12].

A review of the literature suggests that although clinicians need to know the optimal timing for undertaking speech therapy in patients with non-oral CSCs and VPI, there are few studies specifically addressing this. There is much controversy in the literature regarding the timing of intervention for these errors [13]. There are different rationales for providing or not providing treatment. Some investigators have advocated that speech therapy prior to VP surgery is not only possible but advisable [7,14–18]. This is because non-oral CSCs such as glottal stops, active nasal fricatives, pharyngeal stops and fricatives may adversely affect the full mobility of the VP sphincter. Ysunza reported that therapy for these types of articulation disorders prior to surgery reduces the extent of the VP gap by increasing VP movements [19]. Shprintzen and Golding-Kushner extended this concept further, proposing that an increase in movements of the VP structures through correcting articulation preoperatively leads to a reduction in the width of pharyngeal flaps used for surgery [20]. It has also been proposed that pre-operative therapy not only facilitates oral consonant production, but also may serve to reduce hypernasal resonance and may avoid unnecessary surgery [21]. These studies therefore recommend that patients with non-oral placement of consonants should receive articulation therapy before instrumental assessments of VP function, with the aim of obtaining a minimum of one or two oral consonants, in order to be able to demonstrate the maximum potential of the VP mechanism during these investigations [22]. Henningson and Isberg also pointed out that when surgery is undertaken in the absence of articulation errors, the postoperative results can be appreciated immediately [23].

On the other hand, there are studies proposing that surgery for VPI should be undertaken before speech intervention. In other words, correcting articulatory placements should be delayed until after the surgical correction of VPI [24–26]. Riski and DeLong reported that although significant gains in articulation therapy can be made in children with cleft palate and adequate VP closure, minimal or no gain is made in children with cleft palate and VPI [25]. Sell and Pereira (2011) stated that it is ill-advised to struggle for many months or even years to eliminate the articulatory errors, as this is very de-

motivating both for therapist and child [13]. Kummer reflected on the burden of care and the costs of this approach [2]. In addition, more recently there has been a trend for performing secondary VPI surgeries involving the palate (e.g. palate re-repair, furrow palatoplasty, and buccinator flaps) in which case lateral pharyngeal wall movements associated with articulatory errors and the need for pre-operative articulation therapy is less relevant [27].

It is worth noting that some of the controversy may stem from the lack of detail, limited data points and information on individual performance, which are masked in the averaged results typical of group designs. Carter et al proposed that the difference between pre-intervention and post-intervention phases in group designs could be a result of natural fluctuation rather than the effect of the independent variable [28]. Group design lacks external validity; the intervention might be found to be efficient for a group of patients reflecting the average performance; however, study at an individual level gives more accurate information in terms of changes in the dependent and independent variables [28]. Indeed, one of the initial steps in establishing a credible evidence base is to provide treatment and carefully measure performance. The single subject experimental design (SSEDs) is ideal for this purpose in clinical applications [29]. In this design, a baseline and an intervention condition are included and each patient serves as his or own control. The dependent variable is targeted repeatedly and measured across different conditions to confirm the internal validity [30].

It is suggested that the generalizability of the results in group designs is “sample-to-population”, while in SSEDs it is “case-to-case”. The literature suggests that this type of generalizability is key for clinicians working in an evidence-based framework [28]. Vance and Clegg have suggested that “SSEDs are ideal tools for establishing the viability of treatments in real-life settings before attempts are made to implement them in large scale studies needed for RCTs. Interventions can be tailored to the individual’s needs and individual responses to the intervention can be analyzed” [31]. The underlying goal of this design is most often to determine which intervention is effective for which case or cases.

Using SSED, this study was designed to provide a detailed examination of the effects of intensive speech therapy on the speech performance of children with concurrent non-oral and passive CSCs and VPI. Specifically the following research questions were posed:

1. Does articulation therapy reduce the number of non-oral CSCs (glottal articulation, pharyngeal articulation, active nasal fricative, double articulation) over a 10 week intensive articulation therapy period in individuals with co-existent VPI and non-oral CSCs?
2. What is the effect of articulation therapy on the number of passive CSCs (weak and/or nasalized consonants, nasal realization of plosives, and gliding of fricatives/affricates) in the presence of VPI?
3. Does articulation therapy increase the number of stimulative consonants (SC)?
4. Does articulation therapy increase the number of correct consonants in word imitation (CCI)?
5. Does articulation therapy increase the number of correct consonants in picture naming (CCN)?

2. Material and methods

2.1. Participants

The clinical records of 1200 patients with a history of cleft palate (with or without cleft lip) or submucous cleft palate under the care of the Isfahan Cleft Palate Team were reviewed by the lead researcher (F.D). The inclusion criteria were patients whose speech had been evaluated by the team, between the ages of 3 and 12 years

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