Effects of Behavioural and Prosthetic Management in a Case with Ankyloglossia and High Arched Palate: Pre and Post Operative Comparison

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Abstract: *Purpose*: This single subject research design across multiple treatments was aimed towards determining the efficacy of behavioral and prosthetic management in congenital ankyloglossia class IV and high arched palate; pre and post lingual frenectomy.

Method: The study was conducted over four phases: two pre operative and two post operative. The first phase was started with behavioral management solely followed by the second phase, where a combination of behavioral and prosthetic management simultaneously, was tried out. The participant then underwent lingual frenectomy. Post operatively, the third phase was initiated with a combinatorial use of behavioral and prosthetic management. However noticing the simultaneous improvement in the range of motion of tongue, a fourth trial phase was conducted, by seizing the prosthetic management and continuing only with the behavioral management.

Results: The first two pre operative trial phases resulted in an insignificant shift in percentage of correct consonants (PCC) from 68.59% to 70.93%. However the post operative phases yielded interesting results. Although in the third phase the PCC shifted to 72.7%, but a remarkable improvement was visible in the fourth phase of the study, as the PCC shifted to 84.57%.

Conclusion: The results highlighted that in such classical cases, professionals should shed their dilemma of whether to clip the tie or not? This decision will actually help in reducing the unnecessary clinical trials along with better prognosis of client's speech.

Keywords: Ankyloglossia, High-arched Palate, Frenectomy, Behavioral Management, Prosthetic Management.

INTRODUCTION

Tongue is a mobile mass of muscular tissue that is covered with mucous membrane, occupies much of the cavity of the mouth, forms part of its floor, bears the organ of taste; and assists in chewing, swallowing and speech [1]. All the speech sounds (consonants and vowels) are produced by precise and complex movements of this structure. During consonant production, it channelizes the breath stream, while during vowel production; it forms a resonance vessel by upward and downward movement within the oral cavity [2]. The oral peripheral mechanism encloses another noteworthy structure known as "Hard Palate", which is responsible for feeding as well as speech. The tongue in interaction with the hard palate produces speech sounds like /t/, /d/, /j/ and /n/ [3].

Ankyloglossia often referred to as 'Tongue tie'; is a congenital anomaly that is usually detected soon after birth and is caused by an abnormally tight lingual frenulum [4]. The frenum is actually a left over tissue of the developing fetus which is expected to reduce to insignificance before birth. A normal frenum is generally elastic and does not interfere with the primary and secondary function of the tongue. On the contrary, when it is short, thick, tight or broad it has an adverse effect on oro-muscular function, feeding and speech [5].

Ankyloglossia, though an uncommon disorder, is more prevalent in males (with a male-to-female ratio of 3:1) and is mostly manifested as an isolated anatomic variation [6]. However some malformations of the tongue that happen during embryogenesis are seen in combination with a high arched palate, recessed chin and/or other congenital defects and syndromes [7].

Identifying tongue tie is somewhat difficult-as all the variants of the disorder are not identical by phenotype. It varies from being thick and white, thin and membranous, short, long, or wide, extending from the margin of the tongue all the way to the lower front teeth, or so short and tight that they make a web connecting the tongue to the floor of the mouth. When the "tie" extends towards the margin of the tongue, it produces a heart-shaped look at the front of the tongue, thus reducing the tongue tip visibility to nil. While, when the "tie" extends across the floor of the

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mouth, it causes pain when the tongue is elevated, and may also cause separation or inward tilting of the incisors. A baby with a tongue tie will look different from an older child with the same condition [5].

There are various classification systems available to differentiate the tongue tie. The classification system given by Kotlow is as follows: "Normal Range of Motion: > 1.5 inches or 16mm, Class I (Mild): 12-16mm, Class II (Moderate): 8-12mm, Class III (Severe): 4-8mm, and Class IV (Complete): 0-4mm" [8].

Tongue tie generally has a familial background, and its manifestation may range from either milder effect or no apparent symptoms to a severe impact on structure and function. Similarities can be observed in between older child and their relatives with tongue tie, in terms of postures of lips and tongue, habits of speech, and shapes of the nose and face. Several congenital conditions like cleft lip or palate, severe hearing loss or cerebral palsy can also be present simultaneously with ankyloglossia [5].

Ankyloglossia does not have a pronounce effect on speech, as minimal tongue tip excursion is required for normal speech production [9]. In fact, the farthest that the tongue needs to protrude is against the back of the maxillary incisors for a /th/ sound, and the most it has to elevate is to the alveolar ridge for /l/ sound. Since ankyloglossia rarely causes problems with speech, unless there is also oral-motor dysfunction, frenectomy is usually not indicated for speech purposes. However, it may be indicated for feeding purposes and to improve aesthetics.

Children usually adapt adequately to a short lingual frenum and make tongue tip contact against the lingual surface of the maxillary incisors. Examination of the tongue tip for speech purposes can be accomplished by having the patient bite on a tongue blade placed between the first molars. With this amount of obligatory mouth opening, if the patient can produce acceptable /th/, /t/, /d/, /n/, and /l/ sound by contacts with the maxillary incisors, there is no need to recommend a frenectomy to free the tongue tip. The consequences of an untreated tongue tie can be many and varied, which depend largely on variables like the age of the subject and the severity of the condition. Delay in treatment can have very negative consequences for infants, children as well as adults; because in today's scenario, neither any mistake is being forgiven nor any care is being taken regarding placement of appropriate demand in contrast to an individual's capacity [5].

Adults with unrepaired tongue tie generally exhibit old habits of compensation against restricted tongue movement. These habits extend to affect the individual's social and domestic situations, self-esteem, work environment, and dental health; thus depicting the fact that the consequences of unrepaired tongue tie do not reduce with time, rather more difficulties are experienced as time passes. The specific challenges that an adult with a tongue tie may face with time include; inability to open the mouth widely (affecting speech and eating habits), inability to speak clearly when talking fast/loud/soft, difficulty talking even after moderate amounts of alcohol, clicky jaws, pain in the jaws, protrusion of the lower jaws, inferior prognathism, multiple effects in work situations, effects on social situations, eating out, dental health, a tendency to have inflamed gums, increased need for fillings and extractions, sensitivity about personal appearance, emotional factors resulting in rising levels of stress, and difficulty in fitting a denture in place.

Literature doesn't clearly define the effect of ankyloglossia on speech. Historically, the layman population has correlated speech problems directly with tongue-tie; however tongue-tie has actually comparatively slight effects on speech. Subjective evidence indicates that some children with ankyloglossia develop normal speech and compensate for limited tongue tip mobility, even without surgical repair or speech therapy [10, 11]; while in some cases tongue-tie contributes to articulation errors. Speech sounds; specially the linguals and the sibilants like /t/, /d/, /z/, /s/, /t^h/, /n/ and /l/ may be affected [12]. Various compensatory techniques like restricted mouth opening while speaking, and alternating tongue placement for sounds requiring tongue-tip elevation, are being used by children with ankyloglossia. These compensatory techniques are however affected by factors like dentures, missing incisors, tongue size, sensory and motor function of the tongue, as well as the degree of ankyloglossia [13].

The articulation problem may vary in severity and can be manifested either at single word level or in a milder form so as to be noticed only in connected speech.

A mandatory point to remember is that ankyloglossia cannot be a causative agent for speech delay. Such children are expected to acquire speech and language skills at a normal rate, although articulation difficulties for certain speech sounds may be experienced by them. Those with delayed speech

should be marked as "Red Flag" cases and must be critically directed for other evaluations [15].

AIM OF THE STUDY

To determine the efficacy of behavioral and prosthetic management in a twenty eight years old male participant with ankyloglossia and high arched palate; pre and post lingual frenectomy, and to decide whether to "clip the tie" was a correct clinical decision or not.

METHODOLOGY

Subject

A twenty eight year old male, self reported the clinic with the complaint of misarticulating various speech sounds resulting in poor intelligibility in connected speech, and affecting his social and emotional life. On intraoral examination, it was found that the individual had ankyloglossia (tongue-tie) and was classified as Class IV by utilizing Kotlow's assessment, along with high arched palate without any malocclusion (Appendix I) [8]. Normal findings were obtained in ENT and general physical examination.

Tools Used

Formal Assessment

Hindi version of Photo Articulation Test (H-PAT) was administered, in order to track phoneme specific improvements at initial, medial and final positions in word level [16].

Informal Assessment

A narrative speech sample of approximately 200 utterances (devoid of significant pausing) was recorded during the initiation and termination of each phase. Each of the recorded samples was subjected to both quantitative and qualitative analysis. Under quantitative analysis, the Percentage of Consonants Correct (PCC) was calculated through narrow phonetic transcription in order to determine the phonetic accuracy, while on the other hand qualitative judgment was made through a 7 point Likert scale in order to analyze the speech intelligibility [17].

All the speech sample recordings of both formal and informal assessments were performed in an anechoic studio-chamber of the Speech Science Laboratory of Sri Aurobindo Institute of Medical Sciences, Indore (India), using a Sony ICD-UX523F voice recorder.

Materials Used

Tongue depressor, gloves, mirror, Hindi reading materials, Sony ICD-UX523F voice recorder and artificial palate (Appendix I).

Procedure

The participant received one hour of therapy (thrice a week), during each phase of intervention. Formal assessment (Hindi version of Photo Articulation Test; H-PAT) and informal assessment (Percentage of Consonants Correct; PCC and Speech Intelligibility) were used for both baseline evaluation and progress monitoring in the following two phases of pre and post operative intervention. Repeated baseline measurements were done in each phase of pre and post operative intervention, so that validity of intervention of each phase can be established.

Pre Operative

Phase I (2 Months): Behavioral Therapy without Prosthetic Management.

Phase II (3 Months): Behavioral Therapy with Prosthetic Management.

Post Operative

Phase III (3 Months): Behavioral Therapy with Prosthetic Management.

Phase IV (3 Months): Behavioral Therapy without Prosthetic Management.

RESULTS

In the present study, the rationale of both the pre operative phases of intervention was to teach the client to articulate properly through compensatory strategies along with eliminating the obligatory errors. This decision was in support of the view that in spite of having limited range of tongue mobility, individuals with ankyloglossia can compensate their speech and do not require surgical intervention [9,13].

Both formal and informal analysis of speech sample in these phases revealed insignificant progress. The severity of phonetic inaccuracy and the speech intelligibility retained in mild-moderate degree and at Level 5 respectively in both the phases, though quantitatively the PCC shifted from 58.59% in phase one to 60.93% in phase two (Figure **1**). Apart from that there was no significant change in terms of accuracy of phoneme production (since still 44.5% and 40% of

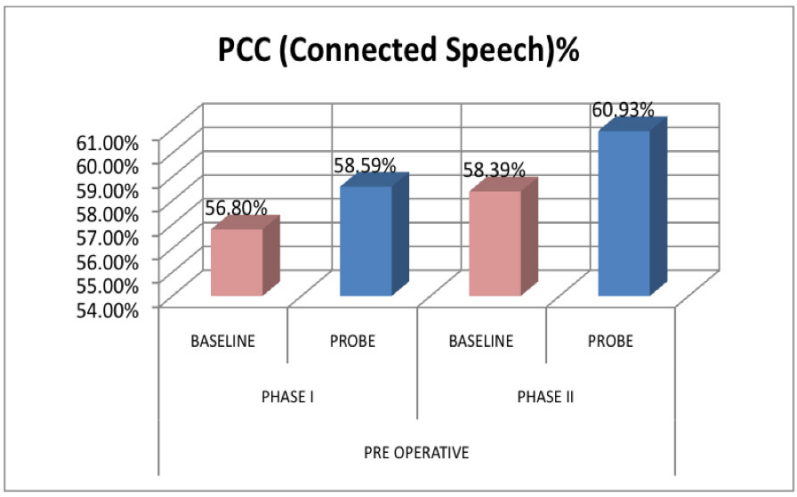

Figure 1: A comparison of change in PCC over the two pre operative phases of management.

consonants were produced erroneously in phase I and II respectively), as revealed by formal analysis through H-PAT administration (Figure **2**).

Followed by the preoperative phases of failure, the client was counseled for undergoing lingual frenectomy, followed by prosthetic and behavioral management in two post operative phases. The rationales of these phases were to improve lingual proficiency along with imparting placement training.

On formal analysis, H-PAT suggested that there is a significant change in accuracy of phoneme production at single word level, as only 13.3 % of consonants were produced inaccurately in phase four as compared to 26.6 % in phase three (Figure **2**). On the other side informal quantitative analysis revealed a shift towards milder form of phonetic inaccuracy, as the PCC shifted significantly from 72.7% in phase three to 85.57% in phase four (Figure **3**). Qualitatively speech intelligibility also shifted from Level 4 to Level 2 in the two post operative phases respectively (Figure **4**). However, a remarkable post operative finding is that the release of the tie, lead Speech Language Pathologists to seize prosthetic management and continue behavioral

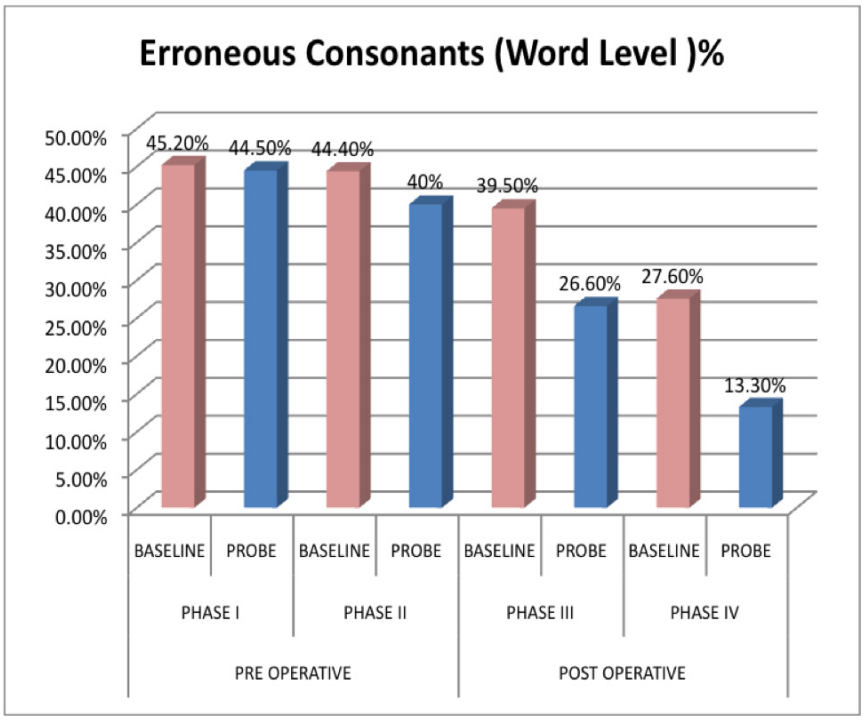

Figure 2: A comparison of change in percentage of erroneous consonants over two pre operative and two post operative phases of management.

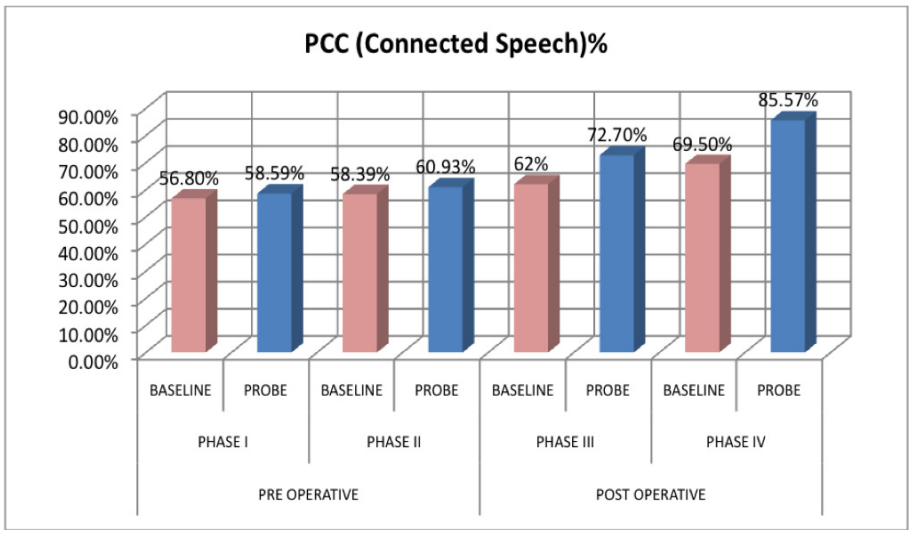

Figure 3: A comparison of change in Percentage of Consonants Correct (PCC) over two pre operative and two post operative phases of management.

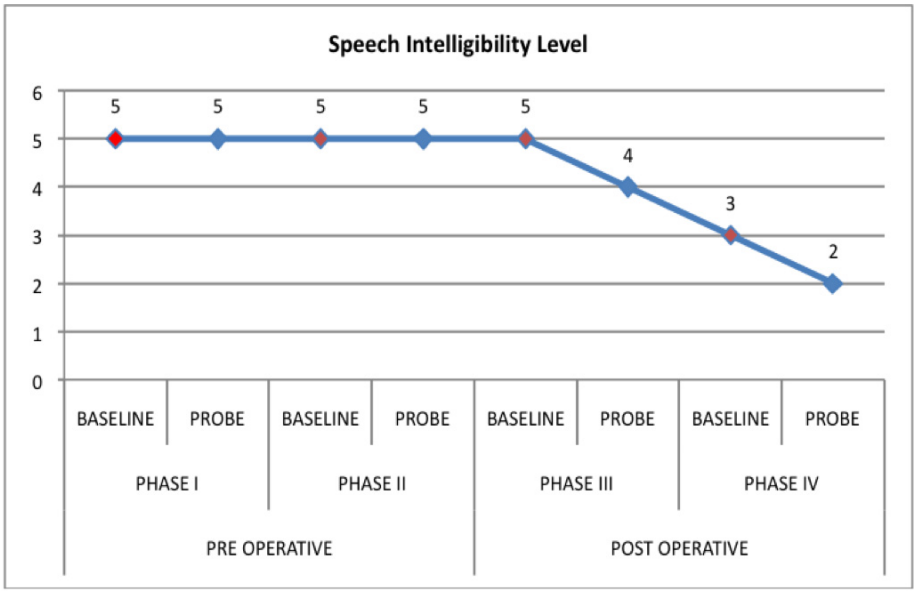

Figure 4: A comparison of change in Speech Intelligibility over two pre operative and two post operative phases of management.

management in isolation, while the reverse was the situation in pre operative phase.

DISCUSSION

Literature doesn't clearly define the effect of ankyloglossia on speech. Although speech problems historically have been viewed as the hallmark manifestation of symptomatic tongue tie, particularly among the layman, others in the literature have vociferously denied that any such relationships exist [18]. Current opinion among speech therapists is clearly divided; 49% of those responding to a survey on this topic believed that ankyloglossia was never or rarely associated with speech problems and an equal number believed that it was sometimes or frequently associated with speech problems [19].

The findings of the pre operative phase of the present study is in accordance to Fernando [5] who said that speech therapy without surgical intervention in cases with ankyloglossia is at best a lengthy process, and at worst, expensive, frustrating for patient and therapist, and unsuccessful. Apart from that, one disadvantage of pre operative speech therapy is that it may require a lengthy period, during which time the individual may be self-conscious about his or her impairment [15]. The benefit of speech therapy must be weighed against surgical treatment that, although

invasive, it is simple, quick and effective. Another disadvantage is that although speech therapy may allow the individual to improve his or her articulation over time by using various compensatory mechanisms, it may still leave him or her vulnerable to manifesting mechanical problems associated with restricted lingual mobility.

The post operative findings are remarkable, and show a significant improvement in terms of PCC, speech intelligibility and reduction in erroneous consonants. This is correlated with a study by Johnson [8], who highlighted that an individual who has tongue-tie and difficulties with speech sounds may well benefit from tongue-tie division. At least by optimizing tongue movement, speech therapy can be maximally beneficial.

Though literature review highlights that the coexistence of oral anomalies and disorders of speech production do not automatically mean that there is a cause-effect relationship, and hence it's not mandatory that speech therapy will result in improved speech, but there are some individuals who will not be able to produce sounds correctly until the oral anomaly is treated and speech therapy is initiated [20].

CONCLUSION

Ankyloglossia with high arched palate is a rare oral anomaly leading to speech and articulation errors. However there is virtually no evidence in the literature to establish a causal relationship between ankyloglossia and speech disorders.

The present longitudinal single case study, lead us to think that in classical cases of Ankyloglossia and high arched palate, professionals should shed their dilemma of whether to clip the tie or not? Literature highlights that interdisciplinary team members often initiate management on a trial basis to determine prognosis, which actually increases the intervention span unnecessarily. This should be prevented, and the possible organic cause should be eliminated prior to behavioral and prosthetic management.

A question was also raised regarding whether prosthetic management should be incorporated in such cases where in spite of lowering the palatal height, efficacy was not significant. Parental education (while their child is below 1 year of age) regarding the possible long-term effects of tongue-tie, should be considered as a priority task; so that they may make an appropriate choice regarding the possible treatments.

As a final note, the findings of present study cannot be generalized as whole. In order to generalize the finding, similar studies can be taken up on a larger sample, across various age ranges.

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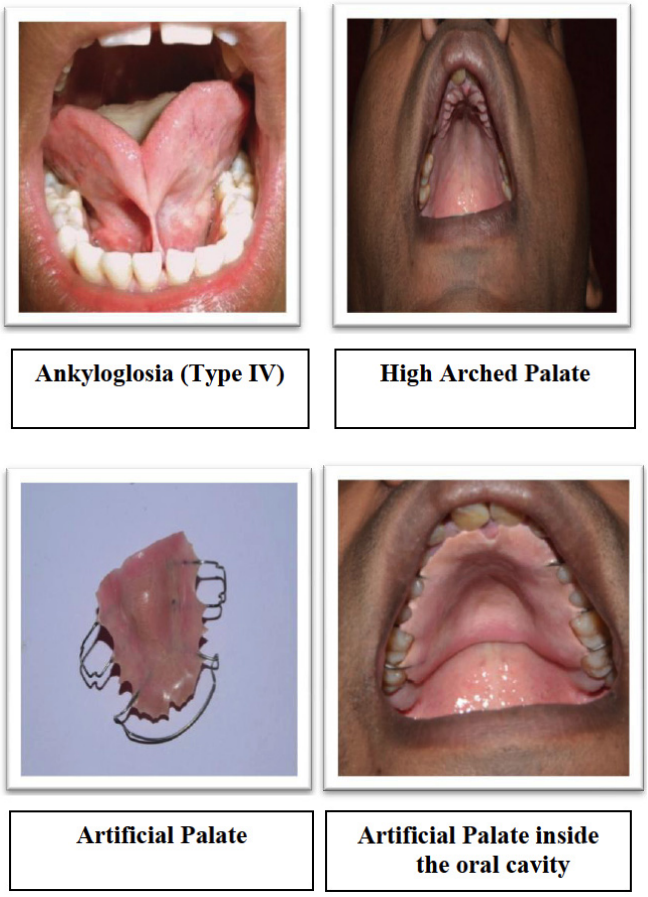
APPENDIX I



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