Evaluation of consonant development following cleft palate surgery: Comparison of Isolated Cleft Palate Versus Pierre Robin sequence.

Malka Jayathilake[1], Deepthi De Silva[2], Romesh Gunasekera[3]

[1]Speech and Language Unit , Lady Ridgeway Hospital for Children
[2]Faculty of Medicine, Kelaniya University
[3]Multi Disciplinary Cleft Clinic, Lady Ridgeway Hospital for Children

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Abstract: The optimal management of Cleft palate patient from birth to completion of the treatment continue to present a formidable challenge the professionals of the multidisciplinary cleft clinic (MDCC) specially children with Pierre Robin Sequence (PRS) than the children with Isolated cleft palate (ICP). The children with PRS have more challenge to develop normal articulation of consonants (Speech sounds except vowels) than ICP due to they are associated with congenital sequence of multiple anomalies, which cause to occur delayed surgery procedures.

Consonant development of fifty children with PRS, compared with fifty children with ICP by the age at 5 years. All of them have registered in MDCC and Speech and Language unit (SLTU) at Lady Ridgeway hospital (LRH). They have undergone the surgical procedures and same technique has used. According to the results, 94% of children with PRS and 90% children with ICP have developed normal articulation for all the consonants in spoken Sinhala language. There is no significant difference in development of normal consonants in between children with PRS and ICP according to the results of statistical analysis. In further according to the statistical analysis, there is no association of sex, consanguinity, family history of cleft lip and/or palate (CL and/ or P) and speech and language delay mainly in expressive language. However, age at surgery, ethnic group and hearing problems before the surgery are associated with the development of consonants in both children with PRS and ICP. This study is worthy to optimize the services of proper, well establish management, and prioritize the surgeries for the children with PRS as well as ICP.

Key words: Pierre Robin Sequence, Isolated cleft palate, consonant development, place of articulation

Introduction

The children with Pierre Robin sequence are at considerable risk to develop delayed or disordered communication development than children with isolated cleft palate. This study investigated the effectiveness of early communication intervention and surgical intervention with fifty children with PRS and fifty children with ICP by assessing their speech outcome measuring with their speech consonant production at the age of five years.

The most important mode of communication among human being is Speech. For a better communication process, speech should be intelligible to understand. Therefore, the normal production of speech sound articulation is essential. However, most of the time, the development of normal production of speech consonants is more often challenging for the children with repaired cleft lip and/ or palate (CL and/or P). Successful surgical procedures in palate repair and performances of Speech and Language Therapy (SLT) prior to palate repair and post surgery help the child to develop normal articulation of consonants. While comparing the children with Isolated Cleft Palate (ICP) and children with Pierre Robin Sequence (PRS), the group of children with PRS has more challenges to develop normal articulation of consonants than group of children with ICP. PRS originally reported retrognathia and glossoptosis in infants with airway obstruction and later they were found cleft palate as an associated symptom, (Robin 1934)[41]. According to the studies, children with PRS have shown to have worse phonological outcomes than children with ICP although no strong prognostic

factors have been identified. (Hardwicke et al, 2016)[26]. The children with PRS present with many other problems, such as micrognathia (small jaw or mandible), glossoptosis (more posterior position of tongue), 'U' shaped cleft palate and present of airway obstructions.

Intelligibility of speech is an essential factor for a successful communication. After palate repair, the speech and language therapist (SLT) has an important role in improving quality of speech sound production of children with CL and/or P by correcting their place of articulation. According to Schuster M.(2006), "the type and dimension of the cleft determine the functional limitations of respiration, swallowing, speech, articulation and hearing. In addition cosmetic and communicative restrictions influence the cognitive, social and educational progress and the wellbeing of the children affected"[44]. Development of consonants mainly effected by the cleft palate deformity. According to Debbie Sell et al (2008), "unlike normal development, in cleft palate speech, voiceless stops frequently appear before voiced stops" [18].

1.1 Surgical timing

The concern of early closure of cleft palate is recommended to conduct at the age of 9 months to 11 months prior they are starting their speech production. In PRS, the palatal surgery tends to be performed few months later depending on severity of breathing difficulties and due to their structural issues in the oral cavity.

1.2 Speech outcome at the age of 5 years

The most distinctive speech disorders associated with CL and/or P are deviant consonant production and hypernasal resonance.

2. Material and Methods

2.1 Participants

A consecutive series of fifty children with PRS and fifty children with ICP from 2006 to 2010 who presented with PRS and ICP were selected as the study population. The children with PRS

and ICP who had an association with syndromic condition were excluded. The children presented with hearing impairment with standard hearing tests after palatal surgery were excluded. The children presented with a speech and language delay by the age of five years were excluded after conducting formal speech and language assessments. The children who could not follow the cleft speech assessment sentences and procedures were excluded. Children below 5 years and the children of parents who did not give consent to audio and video recording were excluded.

2.2 Surgical procedure

One experienced surgeon in multidisciplinary cleft clinic in this particular hospital, applied an unique investigations and decision making protocol with all these children and same surgical procedure has been used in repairing the cleft palate in both groups of children with PRS and ICP .

2.3 Speech outcome measures

All the children with PRS and ICP had received different amount of Speech and Language therapy according to their requirement based on the formal and informal speech and language assessments and information collected from medical reports. All the children with PRS and ICP were assessed for their speech and language development with adopted Derbyshire assessment for Sinhala language. Production of Speech consonants were assessed with modified sentences consists of words to assess different consonants in word initial, middle and final position.

2.4 Audiological data

All the children's were undergone with standard hearing tests. In PRS group, 80% of children presented with normal hearing levels prior to palatal surgery and 20% were presented with mild to moderate conductive hearing loss. In ICP group, 98% of children with ICP presented with normal hearing levels and 2% were indicated mild to moderate conductive hearing loss.

2.5 Material

Audio and video recordings were collected from all the children with PRS and ICP at the age of five years. Perceptual analysis conducted for all the audio and video recordings. The speech samples consisted of number counting 1-10, repeated standard sentences

and connected speech. There were 19 sentences to find out the consonant production in word initial, middle and final position in spoken Sinhala language.

2.6 Speech analysis

All the speech samples were analyzed independently to find out their cleft type characteristics. Speech samples were assessed with unlimited number of times to find out the consonant production in word initial, middle and final position. The following speech parameters were evaluated: with an overall assessment of velopharyngeal insufficiency, retracted oral articulation and cleft type characteristics such as glottal, double articulation, and pharyngeal fricatives etc. Then found out the relationship of following dependent variables sex, consanguinity, family history of cleft lip and/ or palate , age at surgery, ethnic group, speech and language delay before surgery and present with hearing problems before surgery with the speech consonant production in children with PRS and ICP.

2.7 Statistical analysis

Statistics were performed with standard deviation test to analyze the results. Data were given numerical variables, percentages, mean +/- standard deviation or median. P values < 0.05 were considered as the significant value.

3. Results

Table 1: Following is the distribution of Demographic data.

Demographic data	No of PRS /ICP %	p-value
Sex:		
PRS : Male / Female	44/56	NS
ICP : Male / Female	42/48	NS
Consanguinity:		
PRS : Non Consanguinity / consanguinity	98/2	NS
ICP : Non Consanguinity / consanguinity	96/4	NS
Family History:		
PRS : No family history /Present family history	98/2	NS
ICP : No family history /Present family history	96/4	NS
Age at surgery:		
PRS less than 18 months/ more than 18 months	88/12	NS
ICP less than 18 months/ more than 18 months	82/18	NS
Ethics:		
PRS Sinhalese /other	92/8	NS
ICP Sinhalese /other	94/6	NS
Speech and Language development before surgery:		
PRS:Normal speech and Lang development/Speech and Language delay	60/40	NS
ICP: Normal speech and Lang development/Speech and Language delay	78/22	NS
Hearing problems before surgery:		
PRS: normal hearing/ present hearing problems	80/20	NS
ICP : normal hearing/ present hearing problems	98/2	NS

S- Significant , NS – not significant

Table 2: Distribution of consonant development in children with PRS and ICP at the age of 5 years.

Place of articulation	No of PRS/ICP %	p-value
PRS/ICP		

Bilabials	98/98	P=1	NS
Labio dentals	96/90	P=0.764 NS	
Dentals	94/92	P=0.697 NS	
Alveolar	94/90	P=0271 NS	
Palatal	94/92	P=0.697 NS	
Velar	100/94	P=0.075 NS	
Glottal	98/94	P=0.309 NS	

S- Significant , NS – not significant

There is no significant association between consonant production in children with PRS and children with ICP.

Table 3: Relationship between consonant development in both study groups (Children with PRS and ICP) and Sex.

Place of articulation	No of PRS/ICP %	p-value	
PRS ;Sex			
Bilabials Male/ Female	44/54	P=0.317 NS	
Labio dentals	44/52	P=0.764 NS	
Dentals	44/50	P=0.067 NS	
Alveolar	44/50	P=0.067 NS	
Palatal	42/52	P=0.757 NS	
Velar	44/56	P=1	NS
Glottal	44/54	P=0.317 NS	
ICP			
Bilabials	40/48	P=0.308 NS	
Labio dentals	40/50	P=0.254 NS	
Dentals	40/52	P=0.447 NS	
Alveolar	40/50	P=0.254 NS	
Palatal	40/52	P=0.254 NS	
Velar	40/54	P=0.749 NS	
Glottal	40/54	P=0.749 NS	

S- Significant , NS – not significant

There is no significant association between consonant development in either PRS or ICP with sex.

Table 4: Relationship between consonant development in both study groups (Children with PRS and ICP) and consanguinity.

There is no significant association in between consonant development in both PRS and ICP groups with consanguinity.

Place of articulation	No of PRS/ICP %	p-value	
PRS			
Bilabials none consanguinity / consanguinity	88/4	P=0.271 NS	
Labio dentals none consanguinity / consanguinity	86/4	P=0.150 NS	
Dentals none consanguinity / consanguinity	84/4	P=0.073 NS	
Alveolar none consanguinity / consanguinity	84/4	P=0.073 NS	
Palatal none consanguinity / consanguinity	84/4	P=0.073 NS	
Velar none consanguinity / consanguinity	90/4	P=1	NS
glottal none consanguinity / consanguinity	88/4	P=0.271 NS	

ICP		
Bilabials none consanguinity / consanguinity	90/8	P=0.312 NS
Labio dentals none consanguinity / consanguinity	84/6	P=0.465 NS
Dentals none consanguinity / consanguinity	86/6	P=0.401 NS
Alveolar none consanguinity / consanguinity	84/6	P=0.465 NS
Palatal none consanguinity / consanguinity	86/6	P=0.401 NS
Velar none consanguinity / consanguinity	86/8	P=0.073 NS
Glottal none consanguinity / consanguinity	86/8	P=0.073 NS

S- Significant , NS – not significant

Table 5: Relationship between consonant development in both study groups, (Children with PRS and ICP) and family history of cleft lip and/ or Palate.

Place of articulation	No of PRS/ICP %	p-value	
PRS			
Bilabial: no family history/ present family history	96/2	P=0.317 NS	
Labio dentals: no family history/ present family history	96/2	P=0.317 NS	
Dental : no family history/ present family history	92/2	P=0.075 NS	
Alveolar: no family history/ present family history	92/2	P=0.075 NS	
Palatal: no family history/ present family history	92/2	P=0.075 NS	
Velar: no family history/ present family history	98/2	P=1	NS
Glottal : no family history/ present family history	96/2	P=0.317 NS	
ICP			
Bilabial: no family history/ present family history	94/4	P=0.920	NS
Labio dentals: no family history/ present family history	88/2	P=0.242 NS	
Dental : no family history/ present family history	90/2	P=0.219 NS	
Alveolar: no family history/ present family history	88/2	P=0.242 NS	
Palatal: no family history/ present family history	90/2	P=0.219 NS	
Velar: no family history/ present family history	90/4	P=0.075 NS	
Glottal:no family history/ present family history	92/2	P=0.197 NS	

S- Significant , NS – not significant

There is no significant association between consonant development in children with PRS or ICP and family history of cleft lip and/or palate.

Table 6: Relationship between consonant development in both study groups, (Children with PRS and ICP) and age at of surgery.

Place of articulation	No of PRS/ICP %	p-value
PRS		
Bilabial : >18 months/ < 18 months	86/12	P=0.271 NS
Labio dentals : >18 months/ < 18 months	88/8	P=0.084 NS
Dentals : >18 months/ < 18 months	88/6	P=0.015 S
Alveolar : >18 months/ < 18 months	88/6	P=0.015 S
Palatal: >18 months/ < 18 months	88/6	P=0.015 S

Velar : >18 months/ < 18 months	88/12	P=1	NS
Glottal: >18 months/ < 18 months	86/12	P=0.071	NS
ICP			
Bilabial : >18 months/ < 18 months	84/16	P=0.904	NS
Labio dentals : >18 months/ < 18 months	84/8	P=0.001	S
Dentals : >18 months/ < 18 months	84/10	P=0.007	S
Alveolar : >18 months/ < 18 months	84/8	P=0.001	S
Palatal:>18 months/ < 18 months	84/10	P=0.007	S
Velar :>18 months/ < 18 months	78/16	P=0.575	NS
Glottal:>18 months/ < 18 months	78/16	P=0.575	NS

S- Significant , NS – not significant

There is no association between age at surgery and consonant development in bilabial, labiodental, velar and glottal consonants in PRS group. In ICP group, there is no association between age at surgery and consonant development in bilabial, velar and glottal consonants. There is a significant association between age at surgery and development of dental, alveolar and palatal consonants in both PRS and ICP groups. In ICP group there is a highly significant association between age at surgery and development of labio dental consonants.

Table 7: Relationship between consonant development in both study groups, (Children with PRS and ICP) and ethnic groups.

Place of articulation	No of PRS/ICP %	p-value	
PRS			
Bilabial :Sinhalese/other	92/6	P=0.250	NS
Labio dental :Sinhalese/other	90/6	P=0.298	NS
Dental :Sinhalese/other	92/2	P=0.001	S
Alveolar :Sinhalese/other	92/2	P=0.001	S
Palatal :Sinhalese/other	92/2	P=0.001	S
Velar :Sinhalese/other	92/8	P=1	NS
Glottal :Sinhalese/other	92/6	P=0.250	NS
ICP			
Bilabial :Sinhalese/other	92/6	P=0.337	NS
Labio dental :Sinhalese/other	86/4	P=0.001	S
Dental :Sinhalese/other	88/4	P=0.001	S
Alveolar :Sinhalese/other	86/4	P=0.001	S
Palatal :Sinhalese/other	88/4	P=0.001	S
Velar :Sinhalese/other	88/6	P=0.073	NS
Glottal :Sinhalese/other	88/6	P=0.073	NS

S- Significant , NS – not significant

There is no significant association between ethnic group and consonant development in bilabial, labiodentals, velar and glottal consonants in PRS group. In the same time, there is no significant association between ethnic group and consonant development in bilabial, velar and glottal consonants in ICP group. There is a highly significant association between ethnic group and consonant development in dental, alveolar and palatal consonants in PRS group. In addition, there is a highly significant association between ethnic group and consonant development in labio dental, dental, alveolar and palatal consonants in ICP group.

Table 8: Relationship between consonant development in both study groups, (Children with PRS and ICP) and hearing problems before surgery.

Place of articulation	No of PRS/ICP %	p-value
PRS		
Bilabials: normal hearing/ hearing problems present	78/20	P=0.312 NS
Labio dentals: normal hearing/ hearing problems present	76/20	P=0.624 NS
Dentals: normal hearing/ hearing problems present	76/18	P=0.038 S
Alveolar : normal hearing/ hearing problems present	76/18	P=0.038 S
Palatal : normal hearing/ hearing problems present	76/18	P=0.038 S
Velar: normal hearing/ hearing problems present	80/20	P=1 NS
Glottal: normal hearing/ hearing problems present	78/20	P=0.312 NS
ICP		
Bilabials: normal hearing/hearing problems present	96/2	P=0.992 NS
Labio dentals: normal hearing/hearing problems present	90/0	P=0.001 S
Dentals: normal hearing/hearing problems present	92/0	P=0.001 S
Alveolar : normal hearing/hearing problems present	90/0	P=0.001 S
Palatal : normal hearing/hearing problems present	92/0	P=0.001 S
Velar: normal hearing/hearing problems present	94/0	P=0.031 S
Glottal: normal hearing/hearing problems present	92/0	P=0.075 NS

S- Significant , NS – not significant

There is no significant association hearing problems before surgery and consonant development in bilabial, labiodentals, velar and glottal consonants in PRS group. In the same time, there is no significant association between hearing problems before surgery and consonant development in bilabial and glottal consonants in ICP group. There is a highly significant association between hearing problems before surgery and consonant development in dental, alveolar and palatal consonants in PRS group. In addition, there is a highly significant association between hearing problems before surgery and consonant development in labio dental, dental, alveolar, palatal velar consonants in ICP group.

In PRS group there is a significant association with consonants anomalies such as affected mainly dental, alveolar and palatal sounds at the age of surgery (surgery after 19 months) the P value was 0.015 and hearing impairment prior to surgery (P=0.038). There is no differences with sex, ethnicity, consanguinity, family history, speech and language delay before surgery. In ICP group, there is a significant association with consonants anomalies such as affected mainly labio-dental, dental, alveolar and palatal sounds at the age of surgery (surgery after 19 months) the P value was 0.007, Abnormal articulation prior to surgery (P=0), develop normal expressive language development before surgery (P= 0.005) and occurring hearing impairment prior to surgery (P=0.031). There is no association with sex, ethnicity, consanguinity, family history.

4. Discussion

The focus of the study was to search for predictors of consonant production in children with PRS and ICP at the age of five years. According to the results among isolated cleft palate cases, 10% had developed abnormal articulation. Delay in surgery (>19 months), normal expressive language development and hearing loss before surgery predictors of abnormal articulation. Among Pierre Robin sequence cases, 6% had developed abnormal articulation. Delay in surgery (>19 months) and hearing loss before surgery were the predictors of articulation abnormalities. Our results showed the absence of significant association between the consonant production in children with Pierre Robin sequence and isolated cleft palate even though the children with PRS have a possible of poor prognosis with their presented sequence of problems such as airway obstruction problems, micrognatia and glossoptosis in neonatal period. There is no significant difference in development of normal consonants in between children with PRS and ICP according to the results of statistical analysis. In further according to the statistical analysis, there is no association of sex, consanguinity, family history of cleft lip and/or palate (CL and/ or P) and speech and language delay mainly in expressive language. However, age at

surgery, ethnic group and hearing problems before the surgery are associated with the development of consonants in both children with PRS and ICP.

5. Conclusion

Even though there are lot of commitments and concerns with the poor prognosis in development of the children with PRS in breathing and swallowing issues, it is worthy to prioritize their palate repair surgeries as other control group (ICP) because 94% of children with PRS have developed normal articulation for all the consonants in spoken Sinhala language rather than 90% children with ICP have developed normal articulation for all the consonants in spoken Sinhala language. This study is worthy to optimize the services of proper, well establish management, and prioritize the surgeries for the children with PRS as well as ICP.

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