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The Cleft Palate-Craniofacial Journal

Speech outcomes of a national cohort of children with orofacial cleft at 5 and 10 years of age

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Keywords:	Epidemiology, Nasality, Speech development, Speech disorders, Speech perception, Speech production
Abstract:	<p>Objective: To assess speech outcomes at five and ten years of age in a nationwide study of children with orofacial cleft</p> <p>Design: Prospective study</p> <p>Participants: Children born with orofacial cleft and having primary surgery in New Zealand. Speech samples were available for 151 five-year-old, and 163 ten-year-old children.</p> <p>Main outcome measures: Intelligibility, Acceptability, Velopharyngeal function, Hypernasality, Hyponasality, severity of airflow evaluated by perceptual speech assessment (using the standardised Rhinocleft assessment), and overall assessment of requirement for clinical intervention.</p> <p>Results: A large proportion of five-year-old children had speech that was considered to be not completely intelligible, was not acceptable, and had inadequate velopharyngeal function. The noted deficiencies led to a clinical judgement that further speech and/or surgical intervention was required in 85% with cleft lip and palate, 65% with cleft palate and 26% with cleft lip. The proportion of children with poor speech outcomes in the ten-year-old children was lower, though of clinical importance, further intervention required for 25% with CLP, 15% with CP and 3% with CL. The number of sound production errors in both age groups followed the same pattern with fewest in those with CL and most in those with CLP.</p> <p>Conclusions: A significant proportion of children with orofacial cleft were found to have poor speech outcomes requiring further treatment. The outcomes are poor compared to centres reported in the UK and Scandinavia. New Zealand requires a review of the current services for individuals born with cleft to improve speech outcomes and interdisciplinary care.</p>

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Speech outcomes of a national cohort of children with orofacial cleft at 5 and 10 years of age

For Peer Review

Abstract

Objective: To assess speech outcomes at five and ten years of age in a nationwide study of children with orofacial cleft

Design: Prospective study

Participants: Children born with orofacial cleft and having primary surgery in New Zealand. Speech samples were available for 151 five-year-old, and 163 ten-year-old children.

Main outcome measures: Intelligibility, Acceptability, Velopharyngeal function, Hypernasality, Hyponasality, severity of airflow evaluated by perceptual speech assessment (using the standardised Rhinocleft assessment), and overall assessment of requirement for clinical intervention.

Results: A large proportion of five-year-old children had speech that was considered to be not completely intelligible, was not acceptable, and had inadequate velopharyngeal function. The noted deficiencies led to a clinical judgement that further speech and/or surgical intervention was required in 85% with cleft lip and palate, 65% with cleft palate and 26% with cleft lip. The proportion of children with poor speech outcomes in the ten-year-old children was lower, though of clinical importance, further intervention required for 25% with CLP, 15% with CP and 3% with CL. The number of sound production errors in both age groups followed the same pattern with fewest in those with CL and most in those with CLP.

Conclusions: A significant proportion of children with orofacial cleft were found to have poor speech outcomes requiring further treatment. The outcomes are poor compared to centres reported in the UK and Scandinavia. New Zealand requires a review of the current services for individuals born with cleft to improve speech outcomes and interdisciplinary care.

Key words: Epidemiology, Nasality, Speech development, Speech disorders, Speech Perception, Speech production

Introduction

Children with a history of orofacial cleft require the services of an interdisciplinary team. The teams include a wide range of professionals responsible for a range of assessments and management interventions at various points in the treatment course. One of the key members of this team throughout the treatment journey is the speech-language therapist (SLT). Speech is considered one of the primary outcome measures in the management of individuals with orofacial cleft (Jones et al., 2014). The goal for most children with non-syndromic orofacial cleft is intelligible and acceptable speech by school entry (Kuehn & Moller, 2000), which is five years of age in New Zealand (NZ). Following primary surgical repair, children with orofacial cleft are at risk of developing speech problems, many of whom will require speech language therapy. Some of these children will also require videofluoroscopy or nasendoscopy to investigate the need for secondary surgery for velopharyngeal insufficiency (Sommerlad, 2005; Speake & Harding-Bell, 2019). The typical primary surgical protocol in NZ is for children to have initial repair (cleft lip +/- hard palate) at 3 to 6 months and repair of the soft palate +/- hard palate at 9 to 12 months, which would normally involve an Intravelar veloplasty Sommerlad style (Sommerlad, 2003). Surgery for cleft palate involves realignment of the palatal muscles to create a functional palate of adequate length to enable adequate velopharyngeal closure. However, this is not always achieved with the primary surgery. Structural anomalies such as velopharyngeal insufficiency (VPI) and residual palatal fistulae are known complications that may affect speech and resonance following primary surgery, thus children may require secondary surgery to manage structural deficits (Lohmander, 2011).

Speech disorders in children following repair can be structural or articulatory which can have an impact on their phonological development (Chapman, 1993). However, there is the potential for developmental errors in children with orofacial cleft, as there is for all children (Sell et al., 1999). SLTs often use the term 'cleft palate speech' to cover speech patterns that are associated with a history of cleft palate and/or VPI (McWilliams et al., 1990; Sell et al., 1999; Peterson-Falzone et al., 2016; Harding-Bell, 2019). These speech patterns are characterised by hypernasality, abnormal nasal airflow (nasal emission and nasal turbulence), and articulation disorders mostly affecting consonants requiring intraoral pressure (Sell and Harding-Bell, 2019). Consonant disorders reported in association with cleft palate are commonly termed cleft speech characteristics (CSCs) (Sell et al., 1999; John et al., 2006; Howard et al., 2019). CSCs which were originally known as cleft-type characteristics (CTCs) are categorised as either active or passive as defined by Harding and Grunwell (1998). In passive cleft speech characteristics, the child makes the correct articulatory gesture for the target sound but it does not match the target (Howard et al., 2019). Such speech is

characterised by nasal air emission, weakened and/or nasalised pressure consonants and/or nasal substitutions. Active CSCs occur when the child’s articulatory gestures appear to compensate for the structural deficit (Harding and Grunwell, 1998; John et al., 2006; Harding-Bell, 2019; Howard et al., 2019). When there is a lack of intraoral pressure due to VPI, the child may substitute non oral productions such as glottal stops for plosives and pharyngeal or glottal fricatives for fricatives. If there is an anterior or mid palatal fistula the child may compensate by articulating with a more retracted tongue contact for some or all alveolar consonant targets /t d n l s z sh ch j/ which is referred to as a ‘backing pattern’ (Harding and Grunwell, 1998; Sell et al., 1999; John et al., 2006; Fitzsimons, 2018). These changes in sound production can make it difficult to understand the child’s speech and adversely affect speech acceptability. Active CSCs, also known as compensatory speech errors, may persist even after secondary surgery to rectify structural issues of the palate and need to be addressed by speech-language therapy, whereas passive CSCs are usually resolved by surgery to correct the abnormal structure.

Children with an orofacial cleft are also at increased risk of developmental speech and language disorders compared with their peers (Pamplona and Ysunza, 2008; Lancaster et al., 2020). In the preschool years they may present with poorer receptive and expressive language skills as well as immature articulation and limited consonant inventories (Chapman et al., 2003; Scherer et al., 2008). Developmental speech and language disorders require treatment with speech-language therapy and not further surgical intervention (Peterson-Falzone, et al., 2016).

In addition to developmental speech and language disorders, children with an orofacial cleft are more susceptible to conductive hearing loss, due to poor eustachian tube function, resulting in a build-up of fluid in the middle ear leading to persistent or recurring otitis media with effusion (OME). This can result in hearing loss (Purdy et al., 2019), which can impact on developing speech and language, therefore children with orofacial cleft are at risk of developmental speech and language delays/disorders (Sheahan et al., 2003; Purdy et al., 2019).

The gold standard for speech assessment in children with an orofacial cleft is perceptual assessment of speech by a cleft specialist SLT (Sell, 2005) and the gold standard for clinical audit is independent analysis of standardised speech recordings by at least two cleft specialist SLTs (Sell et al., 1999). Following perceptual speech assessment, if VPI is suspected, further investigation of palatal function by instrumental assessment is indicated. The two most commonly used instrumental assessments of VPI are videofluoroscopy and nasendoscopy (Sommerlad, 2005; Speake and Harding-Bell, 2019). If VPI is

confirmed by instrumental assessment, this can determine if secondary surgery is generally required to improve palatal function for speech, and also which surgical procedure is indicated (Sommerlad, 2005).

Speech results following primary palatal surgery that have been reported in the literature vary considerably with regard to the incidence and prevalence as well as the speech parameters that are measured. One of the most comprehensive early studies of speech outcomes was from the Clinical Standards Advisory Group (CSAG) (Sell et al., 2001). This study reported the speech outcomes of children with repaired unilateral cleft lip and palate (UCLP) as assessed at age 5 (n=235) and 12 (n=218) years of age. They found that only 19% of the 5-year-olds had speech that was considered entirely normal and intelligible to strangers. In the 12-year-old group 81% were judged to have speech that was intelligible and unlikely to provoke comment. Hypernasality was present in 32% of 5-year-olds, in over half of these children, the hypernasality was considered to be appreciable, irrespective of whether velopharyngeal surgery had been undertaken. In the 12-year-old group, 26% had hypernasality, and this was found to be more likely to be considered appreciable in those who had not undergone velopharyngeal surgery.

In a review of the literature on speech outcomes of 5-year-old children with an orofacial cleft history, it was noted that most studies reported 50% to 80% of children achieve normal speech by the age of 5. This is in comparison to 85% to 90% of children without a cleft (Lohmander, 2011). Following development of national standards, one of the recommended standards in the United Kingdom (UK) was that by 5 years of age over 70% of children with a cleft palate with or without lip involvement should have speech with no evidence of a structurally related problem, and should not have had velopharyngeal surgery or a fistula repair (Britton et al., 2014). Within this project they then benchmarked 1110 5-year-old children with cleft lip and palate and cleft palate only, and found that 48% had speech and resonance judged to be within the normal range. Additionally, 66% had speech and resonance with no evidence of structurally related speech communication problems or history of speech related secondary surgery (Britton, et al., 2014). A longitudinal study of 391 five year-old English children with CLP and CP, all of whom had been operated on by a single surgeon over a 13-year period (1993-2006), using a robust speech methodology, reported that 68% of children had speech in the normal range. Adequate velopharyngeal function was reported in 97% of children with only 2.6% having previously undergone secondary velopharyngeal surgery (Baillie et al., 2020).

Following re-organisation of the cleft services in the UK, a subsequent study of 5-year-olds with UCLP Cleft Care UK (CCUK), reported significant improvements in speech and resonance outcomes (Sell et al., 2015).

Compared to 19% described as having ‘normal’ speech in CSAG, 56% were reported to have ‘normal’ speech (acceptable and intelligible speech) in CCUK, however, there remained 19% of 5-year-olds judged to be unintelligible. This persistent speech disorder in approximately a fifth of children with orofacial cleft is consistent with other studies (Hall and Golding-Kushner, 1989; Sell et al., 2001; Brunnegård and Lohmander, 2007). Additionally, the presence of hypernasality had reduced from 32% of children in the CSAG study to 10.2% in CCUK.

Scandcleft, a multicentre study involving three trials was established to evaluate the outcomes of a standard surgical procedure in one trial arm against three different surgical procedures in independent arms (Lohmander et al., 2017; Willadsen et al., 2017). Speech outcomes in this study of children aged 5 with UCLP were found to be no different in relation to the surgical protocols (Lohmander et al., 2017, Willadsen et al., 2019). This study included a composite measure of perceived velopharyngeal closure (the listeners overall impression of the degree of velopharyngeal insufficiency (Lohmander, et al., 2017)) as well as measures of hypernasality, nasal air emission and articulation errors. They concluded the composite measure of velopharyngeal function showed good agreement between the listeners and should be included in cleft speech assessment. They also recommended adding intelligibility as a measure of communicative activity and participation (Lohmander et al., 2009). Scandcleft trial 1 found an overall VPI rate of 38.5% in UCLP at five years of age, and no difference was seen in the VPI rate across treatment arms (Willadsen et al., 2019). Consistent with this, the full Scandcleft study (n=391) of three trials across 10 centres found no differences in the prevalence of VPI or hypernasality across trial or arms of trials. The rate of hypernasality was 22-45% and the VPI rate was 39-65% (Lohmander et al., 2017).

Similar results were reported in a further study from a single Swedish centre, that studied 69 children with UCLP recruited between 1987 and 1997 using a standardised surgical protocol. Approximately 40% had hypernasality at 5 years of age, and the prevalence was lower, 10% in a 10-year-old group. Audible nasal emission was similarly present in approximately 40% of the children at 5 years of age and was not notably different (32%) in the 10-year-old group. VPI was present in 60% of the children at 5 and 7% at 10 years of age. In terms of general impression of speech, 57% were considered normal in the 5-year-old group, and 89% in the 10-year-old group. The improvement in VPI and the general impression of speech at 10 was attributed to a high rate of secondary surgical repair of the pharyngeal flap (Nyberg et al., 2014).

A similar study at the same institution, but in children with isolated CP, found that at 5 years of age 50% of those with a cleft of the soft palate only were hypernasal, while 25% of those with a cleft of the hard

and soft palate were hypernasal. At 10 years of age, the rate of hypernasality was lower, with 33% in the soft palate group and 21% in the hard and soft palate group. Audible nasal emission was similar in both groups, 41% and 36% respectively at 5 years of age and was higher in the 10-year-old group, 67% and 46% respectively. The prevalence of VPI was lower in this study than those reported above with only 14% and 11% at 5 years, and 10% and 8% respectively at 10 years of age, however the reasons for this are unclear (Nyberg et al., 2018).

In summary, results from the largest comprehensive studies indicate that a group of approximately 20% of children with orofacial cleft at the age of five years have significant speech issues requiring further significant treatment. The results of nationwide studies are relatively limited with the majority of data coming from Scandinavia and the UK. The results from Scandinavia have improved slightly over time. Meanwhile, outcomes in the UK have improved considerably since centralisation and standardisation of services, where large numbers of children are being treated in fewer cleft centres by a specialised workforce.

To date there has been no comprehensive study of speech outcomes for children with orofacial cleft in NZ, a country without a centralised cleft service, and as such the primary aim of the current study was to determine the speech status of children with orofacial cleft in NZ at approximately 5 and 10 years of age. In addition, we aimed to investigate whether the speech outcomes differed by cleft phenotype (cleft lip, cleft lip and palate and cleft palate), and how outcomes varied by age group (5 years and 10 years) in children who were born and had their primary surgical repair carried out in New Zealand.

Methods

This analysis is part of a broad national investigation of outcomes including surgical, orthodontic, dental, craniofacial form, facial aesthetics and quality of life. The study was undertaken primarily because there was no national data on outcomes in New Zealand, and little data published from local units. The study was thus designed to use measures similar to other national studies performed elsewhere in the world, in particular those carried out in the UK, and Scandinavia. These studies have provided important baseline data to which other countries can compare. All treatment for children with orofacial cleft in NZ is provided free of charge within the public health system, and this is provided by five centres that range in the number seen from 3 to 47 newborns per year. The details of the methodology of the study as a whole and other outcomes are included in previous publications relating to bone grafting (Fowler, Al-Ani, et al.,

2019), dental arch relationships (Fowler, Keall, et al., 2019), craniofacial morphology (Fowler, Wholley, et al., 2019), nasolabial aesthetics (Thompson et al., 2019) quality of life (Fowler et al., 2020; Thompson et al., 2020b) and behaviour (Thompson et al., 2020a).

Participants

All children with cleft lip, cleft lip and palate, and cleft palate (including submucous cleft palate if surgically repaired) who were aged 4.5 years to 6 years (5-year-old group, n=151) or 9 to 11 years (10-year-old group, n=163) and seen at a clinic appointment between October 2014 and September 2017 were assessed for inclusion. We excluded children younger than 4.5 years due to the increased likelihood of developmental speech immaturities at this age (Bowen, 2015). Children were excluded from the study if they had a cleft repair in another country or if they were unable to provide a speech sample or had a syndromic condition that would be considered to have an impact on the child’s speech from either a functional or developmental aspect. In addition, sex and ethnicity of the children were collected to ensure that the sample was generally representative of the population of children with an orofacial cleft.

Speech sample collection

Clinical speech recordings were collected from children by the cleft SLT from each of the five cleft centres in NZ as part of an agreed standard clinical record. Speech stimuli included a connected/conversational speech sample of approximately 2 minutes, rote speech involving counting from 1 to 20, and repetition of the recently validated cleft speech assessment Rhinocleft® short sentence sample (Fitzsimons, 2018) (Supplemental table 1). Speech samples were collected during appointments associated with aspects of children’s normal cleft care by the SLT with a parent/caregiver present. All samples were collected by six SLTs using the same standardised protocol and all using a Zoom Q3 digital video camera with an external lapel microphone. Five of the six SLTs who collected the samples had previously attended a workshop day focused on standardisation of the elicitation and recording of the clinical speech sample. The other SLT was provided with the same information and had the same speech collection protocol to follow. Unedited copies of the children’s speech recordings were uploaded to Seafire, a secure cloud-based platform hosted by the centre for e-Research at the University of Auckland. Digital audio recordings were extracted from the uploaded video recordings and de-identified for subsequent blind assessment.

Speech sample assessment

This study utilised an adapted version of the Rhinocleft® Perceptual Cleft Palate Speech Assessment (Rhinocleft®)(Fitzsimons, 2018). The Rhinocleft® has been formally evaluated and found to be a valid and reliable clinical assessment tool for the evaluation of speech disorders related to cleft palate speech. (Fitzsimons, 2018). The speech stimuli as well as the inclusion of global measures of speech are consistent with the recommendations made by an international working group (Henningsson et al., 2008). Two cleft SLTs independently scored perceptual ratings of speech from the recordings. The two SLTs have extensive experience (25 and 13 years) of working in cleft teams, including using the Rhinocleft® assessment on a regular basis. One of the SLTs had considerable experience with consensus listening using cleft speech assessments on a frequent basis with both local cleft specialist SLTs and international cleft speech specialists. The cleft speech assessment provided ratings of both global (intelligibility, acceptability and velopharyngeal function) and specific (hypernasality, hyponasality, abnormal nasal airflow, cleft speech characteristics, and developmental speech errors) measures of speech performance. Prior to scoring all the samples the two raters completed a pilot reliability study of 29 randomly selected samples from the study recordings, and then discussed differences in their ratings. Due to the discrepancies in inter-rater reliability, we decided that both SLTs would score all samples independently first, and where differences existed consensus listening would be used to determine the final rating. Additionally, after a meeting of SLTs at an Australasian Cleft Lip and Palate Association meeting in Brisbane, twelve other SLTs also independently scored these same samples to provide a consensus view of the ratings of these samples.

Both raters independently listened to the entire speech sample up to three times using high quality headphones connected to a computer before making ordinal, categorical and visual analogue scale (VAS) ratings of the following speech parameters: speech intelligibility (after listening to the conversational sample once, at the start of the assessment), speech acceptability, velopharyngeal function, hypernasality, hyponasality and abnormal nasal airflow (nasal emission and/or turbulence). Scoring of hypernasality was rated on a 5-point scale while intelligibility, acceptability, velopharyngeal function, hyponasality and abnormal nasal airflow were rated on a 4-point scale. The responses to each of the ordinal scales are identifiable in tables 3 and 4. The scales range from what would be expected in normal speech e.g. essentially normal sounding speech, no hypernasality to outcomes associated with severe differences in speech, though some groups with lower frequencies have been combined. Perceived voice quality, presence of any cleft speech characteristics, developmental speech errors, language skills and perceived need for treatment (speech language therapy and/or consideration for further surgery) were rated using ordinal scales only. Assessment was carried out as to whether the child presented with any of

the 21 articulation errors (12 cleft speech characteristics and 9 developmental processes (see supplementary table)). These errors were rated from automatic speech and sentence repetition and considered to be present if the characteristic was heard three or more times. The number of articulation error types was then collated for each child. Each SLT was permitted to make notes (including narrow phonetic transcription) for each speech sample. Completed ratings were recorded using Qualtrics^{XM} software (Qualtrics, Provo, UT).

Reliability of speech scoring

Assessment of the agreement between the SLT raters' independent ratings was carried out for each speech parameter. A lack of agreement between raters was defined as "not in the same category" for the ordinal and categorical scales, and a difference of more than 20mm on the 100mm line of the visual analogue scales. The change of 20mm was used to approximate a change in category on a nominal scale, with the assumption that differences between categories would be relatively equal in size.

Following identification of disagreement, the SLTs met in person for three days over a four-week period to review all the speech samples where consensus had not been reached. The raters utilised a consensus listening procedure based on that described by Shriberg et al. (1984) which has also been replicated by other authors (Chapman et al., 2016; Ahl and Harding-Bell, 2018) to address any disputed ratings. Some instances of disagreement were identified as being due to data entry errors which were easily rectified. The consensus data was used in all analyses.

Statistical analysis

Inter-rater reliability was determined using Cohen's kappa coefficient (k) for the ordinal scales, we also report the percentage of samples where there was agreement between scorers, and Pearson's correlation co-efficient for the VAS scales. Interpretation of the kappa statistics was carried out using the standards published by Landis and Koch (<0 Poor, 0.0-0.2 Slight, 0.2-0.4 Fair, 0.4-0.6 Moderate, 0.6-0.8 Substantial, >0.8-1.0 Almost perfect) (Landis and Koch, 1977). Results are presented as the proportion of subjects in each group for categorical outcomes. Comparisons of the ordinal scales in relation to cleft phenotype (CL, CP, CLP) was assessed using Chi-square statistics. An overall statistic (comparing across all three groups) was calculated, and then pairwise Chi-square statistics were calculated for the CLP and CP groups individually in comparison to the CL group.

Ethics approval

██████████ along with locality approval from each of the sites where cleft patients were assessed. Additionally, for speech samples the parent/caregiver of each child signed a consent that their clinical speech sample could be used for the purposes of this outcomes project.

Results

Of the 397 speech recordings that were collected, 151 met the criteria for collection between 4.5 and 6 years of age (the 5-year-old group, mean=5.2 years, s.d.=0.42) and 163 met the criteria of having been collected when children were between 9 and 11 years of age (the 10-year-old group, mean=10.1 years, s.d.=0.50). There were 80 recordings that were outside the specified age ranges and excluded from analyses. A small number of recordings (n=3) were excluded due to poor quality, limited amplitude or an inadequate speech sample. The distribution of samples according to cleft phenotype, sex and ethnicity is provided in table 1, and is similar to the distribution of cleft phenotype previously reported in a 10-year audit of orofacial cleft cases in NZ (Thompson et al., 2016).

Interrater reliability:

Following individual rating, lack of agreement between the two raters was identified in one or more parameters across approximately 90% of the samples. The inter-rater agreement was moderate for intelligibility ($k=0.56$, 95%CI=(0.48, 0.64)), acceptability ($k=0.49$, 95%CI=(0.42, 0.56)), and velopharyngeal function ($k=0.39$, 95%CI=(0.28, 0.49)), was fair for hypernasality ($k=0.37$, 95%CI=(0.28, 0.46)), hyponasality ($k=0.35$, 95%CI=(0.16, 0.55)) and severity of airflow ($k=0.30$, 95%CI=(0.19, 0.42)). For assessment as to whether the scorers recommended clinical management of speech, agreement was also moderate ($k=0.5$, 95%CI=(0.41, 0.58)). The percentage that were in agreement is also displayed ranging from just over half for acceptability to nearly three quarters for nasal emission (Table 2). Assessment of agreement using the visual analogue scale showed the same pattern, with the correlations between the two scorers were highest for intelligibility and acceptability, followed by velopharyngeal function, hypernasality, and severity of airflow (Table 2).

Speech Outcomes:

Outcome measures using the consensus scoring were assessed in relation to cleft phenotype by age group. Tables 3 and 4 demonstrate the outcomes for each of the speech parameters in the two age groups and are described in detail below.

Intelligibility and acceptability

At five years of age, 20.0% (8/40) of children in the CLP group (27.6% UCLP), 31.3% (25/80) in the CP group and 64.5% (20/31) in the CL group had speech that was considered completely intelligible, while 12.5% (5/40) of the CLP group (13.8% UCLP), 16.3% (13/80) of the CP group and 51.6% (16/31) of the CL group were considered to have acceptable speech that sounded essentially normal. At the other end of the scale, 37.5% (15/40) of children in the CLP group (24.1% UCLP), 32.5% (26/80) of children in the CP group and 12.9% (4/31) of children in the CL group were rated as unintelligible at five years of age. In relation to acceptability 27.5% (11/40) of the CLP group (17.2 UCLP), 23.7% (19/80) of the CP group and 9.7% (3/31) of the CL group had speech that was considered to be unacceptable, i.e., speech that would be considered very different by an unfamiliar listener.

For the 10-year-olds the levels of intelligibility were considerably higher, with 78.6% (44/56) of the CLP group (87.9% UCLP), (vs 20.0% at 5 years; $\chi^2=33.65$, $p<0.0001$), 93.2% (69/74) of the CP group (vs 31.3% at 5 years, $\chi^2=62.15$, $p<0.0001$) and 94.0% (31/33) of the CL group considered intelligible (vs 64.5% at 5 years; $\chi^2=9.10$, $p=0.01$), while the level of acceptability was 41.1% (23/56) in the CLP group (51.5% UCLP), (vs 12.5% at 5 years; $\chi^2=27.02$, $p<0.0001$), 73.0% (54/74) in the CP group (vs 16.3 at 5 years; $\chi^2=57.17$, $p<0.0001$) and 90.9% (30/33) in the CL group (vs 51.6% at 5 years; $\chi^2=13.21$, $p=0.004$). At 10 years of age 5.4% (3/56) of the CLP group (3.0% UCLP), 2.7% (2/74) of the CP group and none of the CL group had unintelligible speech. Similarly, 3.6% (2/56) of the CLP group, 2.7% (2/74) of the CP group and none of the CL group had speech that was “different enough to provoke comment most of the time”.

Hypernasality, abnormal nasal airflow and velopharyngeal insufficiency (VPI)

At 5 years of age hypernasality was present in more than half of both the CLP and CP groups (65.0%; 26/40) CLP (58.6% UCLP) and 59.5% (47/80) CP); of these children 22.5% (9/40) of the CLP group (13.8% UCLP) and 22.8% (18/80) of the CP group had hypernasality that was considered to be moderate to severe. Abnormal nasal airflow was present in 42.5% (17/40) of the CLP group (34.5% UCLP) and 37.5% (30/80) CP group. Using the composite global measure of velopharyngeal function, VPI was reported in 65.0% (26/40) of the CLP group (55.2% UCLP), 57.5% (46/80) of the CP group and 9.7% (3/31) in the CL group. In the 10-year-old group these proportions were lower, with 35.7% (20/56) of the CLP group (33.1% UCLP), (vs 65.0% at 5 years; $\chi^2=8.52$, $p=0.01$) and 19.0% (14/74) of the CP group (vs 59.5% at 5 years; $\chi^2=29.70$, $p<0.0001$) presenting with some level of hypernasality. Abnormal nasal airflow was described in 16.1% (9/56) of the CLP group (18.2% UCLP), (vs 42.5% at 5 years; $\chi^2=8.25$, $p=0.0004$) and 8.1% (6/74) of the CP group (vs 37.5% at 5 years; $\chi^2=18.54$, $p<0.0001$). VPI was reported in 33.9% (19/56) of the CLP group (30.3% UCLP), (vs 65.0% at 5 years; $\chi^2=9.05$, $p=0.003$), 14.9% (11/74) of the CP group (vs 57.5% at 5 years; $\chi^2=29.97$, $p<0.0001$) and none of the CL group. There were no children in the study who had a voice disorder such that hypernasality was unable to be rated. Of all recordings there were 7 that were noted to have reduced volume, in terms of quality 16 were breathy, 5 hoarse, and 1 that had high pitch. Hyponasality was not remarkable in either of the age groups (Table 3).

Management

At age 5, 85.0% (34/40) of children with CLP (82.8% UCLP), 65.0% (52/80) of children with CP and 25.8% (8/31) of children with CL were considered to require further management (speech language therapy and/or consideration for further surgery). The majority, 82.5% (33/40) of children with CLP, 62.5% (50/80) of children with CP and 25.8% (8/31) children with CL were considered to require speech language therapy. Of the children requiring speech language therapy, 57.6% (14/33) of the CLP group and 50.0% (25/50) of the CP group were also recommended for instrumental assessment. Notably two of the eight (25%) CL children requiring speech language therapy were also recommended for instrumental assessment to assess for VPI. A small proportion of the CLP and CP children were recommended for instrumental assessment of velopharyngeal function without a requirement for speech language therapy.

In the 10-year-old group, the proportion of children recommended for treatment was lower in both CLP and CP groups. In the CLP group 25.0% (14/56) (18.2% UCLP), (vs 85.0% at 5 years; $\chi^2=36.17$, $p<0.0001$) and in the CP group 14.9% (11/74) (vs 65.0% at 5 years; $\chi^2=44.11$, $p<0.0001$) were still considered to require treatment. Of these children, 85.7% (12/14) of the CLP group and 72.7% (8/11) CP needed speech language therapy, of which 58.3% (7/12) and 37.5% (5/8) also required instrumental investigation. In the CL group just 3% were considered to need SLT and none required instrumental investigation.

Speech errors

Speech sound production errors (cleft speech characteristics and/or developmental speech processes) were identified in 80.8% (122/151) of children at five years of age; 90.0% (36/40) of those with CLP and 83.7% (67/80) with CP. Five or more errors were seen in 45.0% (18/40) of five-year-olds with CLP and 35.0% (28/80) of those with CP. In the five-year-old group over half of children (61.3%; 19/31) with CL had at least one speech error, most had only had one or two errors, though 16.1% (5/31) had three or more errors.

At 10 years of age there was an improvement in the proportion of children who presented with speech production errors and the number of errors present. Notably, 64.3% (36/56) of the CLP group had remaining speech production errors, with 35.7% (20/56) having three or more errors. Of the CP group, 43.2% (32/74) had remaining speech production errors, with 17.6% (13/74) presenting with more than one error. In the CL group 12.1% (4/33) continued to present with speech production errors (Table 5).

Discussion

This analysis is a comprehensive study of speech outcomes for children with orofacial cleft in NZ assessed as part of a wider NZ cleft outcomes study. A large proportion of children in this study were considered to have speech that was unintelligible to some degree or sounded different from their peers. This is particularly the case for those assessed at five years of age; a critical juncture in their lives as they start school. This has the potential to interfere with academic performance as well as having an impact on the child's ability to develop social relationships (Scherer et al., 2008; Illingworth et al., 2019; Lancaster et al., 2020). We have reported that in our population of children with an orofacial cleft a considerable proportion still require treatment for their speech at five years of age. The requirement is least in those with CL as would be expected, and higher in those with CLP compared to those with CP. This need for

treatment was lower in the 10-year-olds but was still a notable proportion and followed the same pattern in relation to cleft phenotype as at five years of age.

Most international outcome studies to date have focused on the phenotype of UCLP. This study has collected speech samples from all cleft phenotypes, enabling comparison of speech outcomes across cleft phenotypes in NZ, whilst allowing international comparison for the CLP group. Of the 5-year-olds in the UCLP group in NZ, as they start school, only 27.6% had speech that was considered mostly intelligible and only 13.8% had speech that was acceptable (that did not provoke comment). This is very similar to the 19% reported in the UK CSAG study, however, is poor compared to the 56.3 % in the CCUK study whose speech was considered to be in the normal range at age 5 (Sell et al., 2015). These data were collected for children born after centralisation of cleft services in response to CSAG. The CCUK results are also in the same range as the results from Scandinavian studies which report 50%-80% achieve normal speech by 5 years of age (Nyberg et al., 2014; Lohmander et al., 2017; Willadsen et al., 2019). The proportion of children assessed at 10 years of age with intelligible and acceptable speech was higher than observed in the 5 year old samples. There remained though, 18% of children with UCLP whose speech and resonance skills indicated a need for further surgery. This is in keeping with some of the international studies that report persistent speech and resonance disorders in approximately 20% children with UCLP repair (Hall and Golding-Kushner, 1989; Sell et al., 2001; Brunnegård and Lohmander, 2007).

An unexpected finding was that 25% of 5-year-olds with cleft lip (without cleft palate) required speech language therapy which was higher than expected in the general population. Having only a cleft lip should not result in cleft palate speech. While the speech disorder was likely to be developmental in origin for most of these children, two were noted to also have suspected VPI and were recommended for further instrumental assessment. This could suggest an occult submucous cleft palate. Therefore, children with an initial diagnosis of cleft lip should be monitored by the interdisciplinary team for cleft palate speech disorders as well as developmental speech and language disorders and managed appropriately throughout their childhood years (Vallino et al., 2008).

The incidence of VPI in both CLP and CP groups was higher than in most other reported studies. We found that 55% of the 5-year-old NZ UCLP group were considered to have VPI compared to 39-65% in the Scandcleft studies. The lower VPI rates in the Scandcleft studies were found in the centres with surgeons performing higher volumes of primary surgeries. In the NZ CP group 57.5% had VPI compared to under

15% in the Swedish study (Nyberg et al., 2018). Of note, the lowest incidence of inadequate velopharyngeal function that has been reported is from a study involving a single high volume surgeon operating across two sites in England, where the rate of VPI in children with CLP or CP was 3% (Baillie and Sell, 2020).

It is of concern that in NZ approximately 15% of the 10-year-olds in the CLP and CP groups are entering adolescence who still need speech-language therapy. This is slightly greater than reported in the CSAG study prior to centralisation in the UK (Sell et al., 2001). By this age therapy for speech is very limited through the public system in NZ which has significant implications for these children in terms of their learning, self-esteem and peer relationships (Illingworth et al., 2019). Speech-language therapy is available free of charge through the public health and education systems in NZ. Access to these services can be difficult due to a large demand on the services for numerous conditions, some of which have higher priority for service. Private speech-language therapy is available however this is a parental choice, and no data exists on the proportion of families that follow this treatment route.

The speech outcomes from this study are more comparable to the CSAG results from the UK in 1998 prior to centralisation of services, which reported that in children with UCLP normal speech was present in only 19% of the 5-year-olds and in 47% of the 12-year-olds (Sell et al., 2001). They concluded that primary cleft surgery in the UK was producing poor speech outcomes and that the provision for speech therapy was not adequate. In NZ, we are in a similar position 20 years later, with poor speech outcomes and a significant need for speech-language therapy. Centralisation of cleft services in the UK and subsequent reassessment 15 years later (Sell et al., 2015) reported considerable improvements in speech amongst other outcomes. Higher volumes of patients having treatment from expert clinicians working in interdisciplinary teams has shown significant improvements in speech outcomes in the UK where a minimum of 40 primary surgical cases per surgeon are expected to be performed in a year. The expertise gained through multiple surgeries is highlighted by the excellent speech outcomes from a single surgeon over a period of more than a decade performing on average of over 60 primary palatal surgeries per year (Baillie and Sell, 2020).

Study Strengths and validity of observations

All NZ cleft centres were provided with the same recording equipment before speech data collection commenced, ensuring consistency in the quality of speech samples. These standard recordings are

continuing as part of standard clinical practice and will provide comparable data to measures changes in speech outcomes over time.

The inclusion of all phenotypes of orofacial cleft in this study not only provides baseline data for future comparison of speech outcomes in NZ, but also provides data for those children with CLP and CP for comparison to those centres considered to be performing best practice care. Additionally, the study has provided valuable data of speech outcomes for those children with a CL or CP alone. CP is the most common cleft phenotype and is of particular importance in NZ as the indigenous Māori population has the highest rate of CP internationally of 1.54 per 1,000 live births (Thompson et al., 2016). The findings in relation to speech issues in the group with CL alone highlight the need for continued screening of speech in all children with orofacial cleft so any speech issues no matter what phenotype can be managed appropriately. Children with orofacial cleft remain at more risk of speech disorder than their non-cleft peers.

We used a shortened adapted version of the recently validated cleft speech assessment, Rhinocleft® (Fitzsimons, 2018) in this study. Rhinocleft® is now the preferred assessment tool used to assess individuals in cleft centres across NZ. Rhinocleft® includes the global measures of intelligibility and acceptability, a composite measure of velopharyngeal function and speech stimuli all consistent with international recommendations.

Study Limitations

As with all observational studies, there are some limitations in regards to the findings of the current study. Interrater reliability of the two SLTs involved in speech assessment and analysis was fair to moderate but not dissimilar to inter-rater agreement reported in other studies (Sell et al., 2001; Sell et al., 2015; Baillie and Sell, 2020). One of the SLTs has previously been shown to also have good agreement with other SLTs in an Australian study (Fitzsimons, 2018). The two raters were however able to re-listen to samples where there was disagreement and reach consensus decisions.

In regards to the generalisability of the results, some children had been lost to follow-up or did not attend for regular review in the 3-year period of data collection, though the exact numbers are unknown. It was not possible to determine the proportion of children for whom we gained speech samples due to the fact that the research team does not have access to clinic appointment lists and thus does not have a denominator to calculate the response rate.

In this study, we have not been able to report on the number of children who previously had secondary surgery for speech following their primary repair (fistula repair or surgery for VPI) prior to sample collection. Neither did we have information on the number of surgeries each child had undergone to date in their treatment journey. Neither did we have information on those who were due for further surgery or being considered for further surgery at the time of data collection. Therefore, it is difficult to determine the burden of care for children of unsuccessful primary surgery. Nevertheless, the fact that such a large proportion of children, particularly those with CLP and CP, are starting school with unintelligible or unacceptable speech is concerning. This is part of the longer-term outcomes of the study for which data is yet to be obtained.

Information on hearing status at the time of recording and previous history of hearing loss was not available when it is known hearing loss can influence speech and language development, and collection of such data should be part of future protocols.

In this study we only used the audio recording and not the video component of the recording for analysis. We therefore would not have noted facial or nasal grimace which some children use to reduce nasal airflow. However, audio was selected over video as the scorer may have known the child and this could have biased their assessment.

Changes clearly need to be made to enable an improvement in speech outcomes particularly when considering contemporary outcomes reported elsewhere. Future work on more detailed speech outcomes will be essential to better understand speech deficiencies in these children. From the data collected further differentiation between developmental speech errors and cleft speech characteristics need to be assessed. Cleft speech characteristics will also be further analysed into active and passive CSCs and this will provide more detailed information in regard to the need for speech management.

Conclusion

This clinical outcome study provides baseline data for children in New Zealand across all cleft phenotypes (cleft lip, cleft palate and cleft lip and palate) at five and ten years of age. A considerable proportion of children with orofacial cleft were found to have poor speech outcomes. These data allow for benchmarking standards to be set for future audits of cleft services. It also provides health professionals and policy managers from both health and education sectors in NZ with information that can help with

the planning and improvement of future services. It is clear NZ needs a review of the current services for individuals born with orofacial cleft to improve speech outcomes and interdisciplinary care.

For Peer Review

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Table 1. Distribution of speech samples according to cleft phenotype by age group and published data

Descriptive Statistics	5 Years (n=151)	10 Years (n=163)	NZ orofacial cases 2000-2009
	n (%)	n (%)	n (%)
Cleft Lip and Palate	40 (26.5%)	56 (34.4%)	288 (27.9%)
Cleft Palate	80 (53.0%)	74 (45.4%)	540 (52.2%)
Cleft Lip	31 (20.5%)	33 (20.2%)	206 (19.9%)
Sex			
Male	75 (49.7%)	89 (54.6%)	
Female	76 (50.3%)	74 (45.4%)	
Ethnicity			
Maori	35 (23.3%)	39 (24.1%)	
Pacific Island	17 (11.3%)	21 (13.0%)	
European	77 (51.3%)	91 (56.2%)	
Other*	21 (14.0%)	11 (6.8%)	
	Mean (s.d.)	Mean (s.d.)	
Age at sample	5.2 (0.42)	10.1 (0.50)	

*Other ethnicities are those where the individual numbers are small and include Chinese, Indian, Other South East Asian, Middle Eastern, Latin American and African.

Table 2: Interrater reliability of speech parameters according to categorical and continuous scales

	All Age Groups	Pilot study (n=29)	Ordinal Scales		VAS
		% agreement	% agreement	Kappa*	Correlation
Global Measures	Speech Intelligibility	62.1%	69.1%	0.56	0.76
	Speech Acceptability	24.1%	50.3%	0.49	0.72
	Velopharyngeal Function	72.4%	72.9%	0.39	0.63
Specific Measures	Hypernasality	55.2%	61.3%	0.37	0.61
	Hyponasality	69.0%	92.0%	0.35	
	Abnormal Nasal Emission	65.5%	76.1%	0.30	0.43
	Speech errors		70.7%	0.36	N/A
Clinical Measures	Need for Speech Therapy /Instrumental investigation	69.0%	66.2%	0.50	N/A
Voice	Volume		94.0%	Undefined	N/A
	Quality		85.5%	0.12	N/A
	Pitch		98.5%	0.00	N/A
Language	Expressive language skills	62.1%	79.4%	Undefined	N/A

*Undefined kappa statistics are the result of unequal categories between scorers i.e. one scorer has scored at least one sample in a category than the other scorer did not score any samples in.

Table 3: Speech outcomes for 5-year-olds

	Cleft Lip and Palate	Cleft Palate	Cleft Lip
	n=40	n=80	n=31
Intelligibility	$\chi^2=14.77$, p=0.0006	$\chi^2=10.56$, p=0.0051	$\chi^2=16.58$, p=0.0023
Essentially all speech able to be understood	8 (20.0%)	25 (31.3%)	20 (64.5%)
Most speech able to be understood	17 (42.5%)	29 (36.2%)	7 (22.6%)
Little to no/only some speech able to be understood	15 (37.5%)	26 (32.5%)	4 (12.9%)
Acceptability	$\chi^2=22.33$, p<0.0001	$\chi^2=17.78$, p=0.0005	$\chi^2=28.19$, p<0.0001
Essentially normal sounding speech	5 (12.5%)	13 (16.3%)	16 (51.6%)
Speech is different, but not enough to provoke a comment	5 (12.5%)	21 (26.3%)	9 (29.0%)
Speech is different enough to provoke a comment some of the time	19 (47.5%)	27 (33.7%)	3 (9.7%)
Speech is different enough to provoke a comment most of the time	11 (27.5%)	19 (23.7%)	3 (9.7%)
Velopharyngeal function	$\chi^2=22.12$, p<0.0001	$\chi^2=20.72$, p<0.0001	$\chi^2=25.56$, p<0.0001
Appear adequate	14 (35.0%)	34 (42.5%)	28 (90.3%)
Marginal/moderate	26 (65.0%)	46 (57.5%)	3 (9.7%)
Hypernasality	$\chi^2=22.12$, p<0.0001	$\chi^2=22.29$, p<0.0001	$\chi^2=13.91$, p=0.0076
None	14 (35.0%)	32 (40.5%)	28 (90.3%)
Mild	17 (42.5%)	29 (36.7%)	2 (6.5%)
Moderate/Severe	9 (22.5%)	18 (22.8%)	1 (3.2%)
Hyponasality	$\chi^2=1.41$, p=0.24	$\chi^2=2.09$, p=0.15	$\chi^2=2.49$, p=0.28
None	38 (95.0%)	76 (95.0%)	27 (87.1%)
Mild/Moderate	2 (5.0%)	4 (5.0%)	4 (12.9%)
Severity of airflow	$\chi^2=14.23$, p=0.0002	$\chi^2=13.04$, p=0.0003	$\chi^2=14.98$, p=0.0006
None	23 (57.5%)	50 (62.5%)	30 (96.8%)
Mild/Moderate	17 (42.5%)	30 (37.5%)	1 (3.2%)
Treatment	$\chi^2=26.00$, p<0.0001	$\chi^2=15.03$, p=0.0018	$\chi^2=27.98$, p<0.0001
No treatment required	6 (15.0%)	28 (35.0%)	23 (74.2%)
Speech therapy only	19 (47.5%)	25 (31.3%)	6 (19.3%)
Instrumental investigations to consider physical management (surgery) ONLY	1 (2.5%)	2 (2.5%)	0 (0.0%)
Speech therapy AND instrumental investigations	14 (35.0%)	25 (31.2%)	2 (6.5%)

*Chi-square and p-values presented in CL column are the overall statistic across the 3 cleft phenotypes. Those in the CLP and CP columns test the difference of each of these phenotypes compared to the CL phenotype.

Table 4: Speech outcomes for 10-year-olds

	Cleft Lip and Palate	Cleft Palate only	Cleft Lip only
	n=56	n=74	n=33
Intelligibility	$X^2=4.03$, p=0.13	$X^2=1.09$, p=0.58	$X^2=8.59$, p=0.07
Essentially all speech able to be understood	44 (78.6%)	69 (93.2%)	31 (93.9%)
Most speech able to be understood	9 (16.1%)	3 (4.1%)	2 (6.1%)
Little to no/only some speech able to be understood	3 (5.4%)	2 (2.7%)	0 (0.0%)
Acceptability	$X^2=22.02$, p<0.0001	$X^2=5.00$, p=0.17	$X^2=28.25$, p<0.0001
Essentially normal sounding speech	23 (41.1%)	54 (73.0%)	30 (90.9%)
Speech is different, but not enough to provoke a comment	20 (35.7%)	14 (18.9%)	3 (9.1%)
Speech is different enough to provoke a comment some of the time	11 (19.6%)	4 (5.4%)	0 (0.0%)
Speech is different enough to provoke a comment most of the time	2 (3.6%)	2 (2.7%)	0 (0.0%)
Velopharyngeal function	$X^2=14.24$, p=0.0002	$X^2=5.47$, p=0.02	$X^2=17.05$, p=0.0002
Appear adequate	37 (66.1%)	63 (85.1%)	33 (100.0%)
Marginal/moderate	19 (33.9%)	11 (14.9%)	0 (0.0%)
Hypernasality	$X^2=10.04$, p=0.0066	$X^2=3.05$, p=0.22	$X^2=13.91$, p=0.0076
None	36 (64.3%)	60 (81.1%)	31 (93.9%)
Mild	15 (26.8%)	13 (17.6%)	2 (6.1%)
Moderate/Severe	5 (8.9%)	1 (1.4%)	0 (0.0%)
Hyponasality	$X^2=3.12$, p=0.08	$X^2=1.38$, p=0.24	$X^2=3.76$, p=0.15
None	51 (91.1%)	71 (96.0%)	33 (100.0%)
Mild/Moderate	5 (8.9%)	3 (4.0%)	0 (0.0%)
Severity of airflow	$X^2=5.90$, p=0.02	$X^2=2.83$, p=0.09	$X^2=6.61$, p=0.04
None	47 (83.9%)	68 (91.9%)	33 (100.0%)
Mild/Moderate	9 (16.1%)	6 (8.1%)	0 (0.0%)
Treatment	$X^2=7.58$, p=0.06	$X^2=15.03$, p=0.0018	$X^2=9.82$, p=0.13
No treatment required	42 (75.0%)	63 (85.1%)	32 (97.0%)
Speech therapy only	5 (8.9%)	5 (6.8%)	1 (3.0%)
Instrumental investigations to consider physical management (surgery) ONLY	2 (3.6%)	3 (4.1%)	0 (0.0%)
Speech therapy AND instrumental investigations	7 (12.5%)	3 (4.1%)	0 (0.0%)

*Chi-square and p-values presented in CL column are the overall statistic across the 3 cleft phenotypes. Those in the CLP and CP columns test the difference of each of these phenotypes compared to the CL phenotype.

Table 5: Speech sound production errors by cleft phenotype

Number of speech errors	Age					
	5 years			10 years		
	CLP	CP	CL	CLP	CP	CL
0	10.00%	16.30%	38.70%	35.70%	56.80%	87.90%
1	7.30%	3.70%	29.00%	21.40%	25.70%	0.00%
2	12.50%	12.50%	16.10%	7.10%	2.70%	12.10%
3	2.50%	16.30%	6.40%	10.70%	4.10%	0.00%
4	22.50%	16.30%	3.20%	14.30%	5.40%	0.00%
5+	45.00%	35.00%	6.50%	10.70%	5.40%	0.00%

Supplementary Table 1: Rhinocleft® short sentence sample

Pressure consonants without nasals

A puppy will play up
A baby will blow a bubble
A turtle ate a tree
Daddy drilled a hole
I like your creaky car
A girl will wiggle her leg
Fluffy fell off a wall
I have a very heavy whale
Lisa saw a slow horse
Zoey is really lazy
Ashley will wash a shell
A witch will chew her itchy hair
George will hear Roger
The other three were healthy

Pressure consonants adjacent to nasals

The umpire will remember my number
I went into a snowman’s tent
A snail has no hands
Your angry uncle will yell

Low pressure consonants

A yo-yo will roll away
How will Harry hear a whale

Nasal consonants

My lamb is yummy
Nanna knew I won
Mum rang in the morning

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Consonant Errors

Cleft Speech Characteristics

Dentalisation
Lateralisation
Palatalisation
Double articulation
Backing to velar
Backing to uvular
Pharyngeal articulation
Glottal articulation
Nasal fricatives
Weak articulation
Nasal substitutions
Gliding of fricatives

Phonological processes

Stopping
Fronting
Cluster reduction
Weak syllable deletion
Final consonant deletion
Labialisation
Assimilation/Consonant harmony
Gliding
other