CLINICAL APPLICATION OF USAGE-BASED PHONOLOGY: TREATMENT OF CLEFT PALATE SPEECH USING USAGE-BASED ELECTROPALATOGRAPHY

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Abstract

Background: Electropalatography (EPG) is an instrumental technique which has been used to treat persistent speech sound disorders in school-aged children and adults born with cleft palate+/- lip. The effectiveness of EPG therapy with this client group has been reported in multiple case studies and small group studies (e.g. Gibbon et al., 2001). Findings show many individuals can change their speech with EPG therapy. However, patients commonly have difficulty with generalisation and maintenance of improvements following therapy, leading to the conclusion that further development of the EPG treatment technique is needed. This thesis describes and evaluates patients' response to a novel EPG therapeutic technique which uses usage-based phonological theory as its theoretical underpinning. In comparison to other phonological theories, which provide economical descriptions of sound systems, usage-based phonology presents an explanatory account of speech sound development (e.g. Menn et al, 2013). The resultant therapy technique focuses on high volume production of words and connected speech.

Method: Six consecutively treated school-aged children and adults with long-standing lingual speech errors secondary to cleft palate enrolled on a multiple baseline (ABA) within-participant case series. Speech was assessed on three baselines prior to intervention, during weekly intervention at a regional hospital, at completion of therapy, and then 3-months post-therapy. Two speech and language therapists unrelated to treatment blindly transcribed and rated participants' speech.

Results: Large treatment effect sizes were shown for all participants. Percentage of targets correct for treated and untreated words improved from near 0% pre-therapy, to near 100% post-therapy, for most target speech sounds. Generalisation of target sounds to spontaneous connected speech occurred for all participants and ranged from 78.95 - 100% (mean = 90.66) on maintenance assessment, 3-months post-therapy.

Conclusions: Clinically relevant speech change occurred for all participants following intervention. EPG feedback was central for acquisition of speech sounds, while other aspects of the treatment approach appeared important for functional generalisation. EPG provided objective speech data and was an important complement to acoustic phonetic transcription. This therapy technique shows promise and further research is indicated.

Key words: Cleft palate, Speech, Electropalatography, Visual biofeedback, Usage-based phonology theory.

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List of acronyms and abbreviations used in this thesis

A5	Assessment at the end of five sessions
A10	Assessment at the end of 10 sessions
A15	Assessment at the end of 15 sessions
A20	Assessment at the end of 20 sessions
AC	Alveolar Closure
ACPA	American Cleft Palate Association
AT	Alveolar Total
B1	Baseline 1
B2	Baseline 2
B3	Baseline 3
BCLP	Bilateral Cleft Lip and Palate
CL	Cleft Lip
CoG	Centre of Gravity
CI	Confidence Intervals
CP	Cleft Palate
CPO	Cleft Palate Only
CV	Consonant Vowel Sequence
CVC	Consonant Vowel Consonant Sequence
CVCV	Consonant Vowel Consonant Vowel sequence
CVCVC	Consonant Vowel Consonant Vowel Consonant Sequence
D	Effect Size Statistic
DEAP	Diagnostic Assessment of Articulation and Phonology
EPG	Electropalatography
EPG CLEFTNET	Electropalatography Cleft Network
ES	Effect Size
GMP	Generalised Motor Programmes
GOS.SP.ASS	Great Ormond Street Speech Assessment
IA	Initial Assessment
ICC	Interclass Correlation Coefficient
ICS	Intelligibility in Context Scale
IPA	International Phonetic Alphabet
κ	Cohen's' Kappa Statistic
KP	Knowledge of Performance
KR	Knowledge of Results
M	Maintenance Assessment
RCT	Randomised Control Trial
SLT	Speech and Language Therapist
SSD	Speech Sound Disorder
SMCP	Submucous Cleft Palate
SPAA-C	Speech Participation and Activities of Children
STARS	Speech Training Aid and Recording System

P	Post-therapy Assessment
PCC	Percentage of Consonants Correct
QoL-Dys	Quality of Life Instrument - Dysarthria
UCLP	Unilateral Cleft Lip and Palate
UK	United Kingdom
VC	Vowel Consonant sequence
VPD	Velopharyngeal Dysfunction
VPI	Velopharyngeal Insufficiency
Win EPG	Windows Electropalatography system
WT	Whole Total
%CC	Percentage of Consonants Correct
%TC	Percentage of targets correct

1 INTRODUCTION

Cleft lip and palate is one of the most frequently occurring birth anomalies with an incidence of 1 in 700 births (CRANE Database, 2019). The negative consequences of a cleft palate +/- lip on speech is well recognised, and maximising speech outcomes is a key goal in the treatment of this client group. In the United Kingdom (UK), individuals born with this condition are managed by a specialist, multidisciplinary cleft team from birth through to adulthood. Although the structural deficit is typically repaired in infancy, many individuals born with this condition in the UK experience significant, on-going speech sound disorders (SSD), which for some can extend into adulthood (Britton et al., 2014; Sell et al., 2001; Whitehill et al., 1996). Delivery of effective therapy for SSD secondary to cleft palate +/- lip can be very challenging, especially for school aged children and adults with very entrenched, long-standing SSD. Treatment of residual/persistent SSD, whatever the aetiology, is very difficult and represents a much neglected area (Flipsen, 2015; Gibbon & Paterson, 2006; Pascoe, 2006).

Electropalatography (EPG) is an instrumental treatment technique that has been used by speech and language therapists (SLTs) to treat school aged children and adults with persistent SSD secondary to cleft palate +/- lip. EPG is one of a small number of visual biofeedback techniques used in the treatment of SSD. EPG and other visual biofeedback techniques, such as ultrasound and acoustic biofeedback, are usually “last resort” treatments, used when standard speech therapy has been unsuccessful. At present the evidence base for EPG therapy with SSD secondary to cleft consists of clinical consensus, within-participant studies, and small group studies (Lee et al., 2009). While improvements with speech sound production have been shown for many patients, generalisation and maintenance of gains to everyday speech has proven more problematic (Cleland & Preston, 2021; Gibbon & Paterson, 2006; Lee et al., 2009).

Like many areas of speech and language therapy practice, understanding of, and intervention for, SSD has been heavily influenced by rule-based, generative linguistic theory (Chomsky, 1957 onwards). However, in recent times, rule-based linguistic theory has been widely challenged in a number of areas, such as child phonology, child language and sociophonetics (e.g. Goldinger, 1998; Jannedy & Hay, 2006; Menn et al., 2013; Vihman & Croft, 2007; Vihman & Keren-Portnoy, 2013), leading to development of alternative linguistic theories. Usage-based linguistic theory is one such alternative theory that has been growing in significance (Menn et al., 2013; Vihman & Keren-Portnoy, 2013). This research details and evaluates six cleft patients’ response to an EPG therapeutic technique which uses usage-based phonology theory as its theoretical underpinning. This thesis is among the first to consider usage-based phonology with regard to SSD, and to our knowledge, the first to apply usage-based phonology to SSD associated with cleft palate +/-

lip. Participants' response to usage-based EPG intervention is examined and evaluated using a multiple baseline (ABA) within-participant design.

Chapter Two of this thesis provides a brief overview of the cleft lip and palate condition. This short overview chapter goes on to describe the impact of a cleft of the palate on speech. Chapter Three provides an overview of EPG and reviews previous EPG intervention studies with this client group. The chapter then considers the theoretical basis of previous and current EPG therapy. Chapter Four presents usage-based phonology theory as an alternative theory to underpin therapy utilising EPG. In this chapter, usage-based models are discussed and the implication of these models for the treatment of SSD secondary to cleft palate is considered. Chapter Four concludes with the study's aim and research questions. Chapter Five provides details of the method used in this research. The results of the study are given in Chapters Six and Seven. Chapter Six presents impressionistic perceptual speech results, while Chapter Seven gives EPG results. Chapter Eight discusses the study's findings, and includes strengths and weaknesses of the study, implications for further research, and clinical and theoretical implications. Chapter Nine provides final conclusions.

Note: In this work, unless otherwise indicated, the term "phonology" is used broadly to encompass the sub-discipline of linguistics concerned with the sounds of a language (Lass, 1984).

2 CLEFT LIP AND PALATE AND ASSOCIATED SPEECH

The first section of this overview chapter provides an outline of the cleft lip and palate condition, including embryological development, anatomy, classification and incidence. The second section of this chapter describes the impact of a cleft of the palate on speech.

2.1 Cleft lip and palate

Cleft lip and palate occurs when the lip and roof of the mouth do not fuse together during early foetal development, leaving a structural deficit involving a gap or a split (Atkinson & Howard, 2011; Peterson-Falzone et al., 2010; Watson, 2001).

2.1.1 Embryological development and anatomy

The face begins to develop from the fifth week of pregnancy when neural folds at the head end of the embryo begin to fuse and form a tube (Mossey et al., 2009; Watson, 2001). For the lip and the anterior part of the hard palate, known as the primary palate (i.e. all tissue anterior to incisive foramen), this fusion occurs between five to seven weeks of pregnancy. For the remainder of the plate, known as the secondary palate (i.e. all tissue posterior to the incisive foramen), this fusing occurs between eight and nine weeks of pregnancy (Atkinson & Howard, 2011; Mossey et al., 2009). Anything which interferes with this fusion may cause a cleft of the lip and/or palate (Atkinson & Howard, 2011; Peterson-Falzone et al., 2010; Watson, 2001). Clefts of the primary palate are distinct from clefts of the secondary palate (cleft palate only) in terms of incidence, genetic basis and sex distribution (Mossey et al., 2009). Clefts of primary palate can cause a cleft of the secondary palate due to the tongue tip becoming trapped in the cleft so that the secondary palate cannot fuse (Watson, 2001). Clefts of the primary and secondary palate may be complete or incomplete, and may be unilateral, bilateral or median (Mossey et al., 2009). A submucous cleft palate (SMCP) is a type of cleft of the secondary palate and occurs when the oral surface mucosa is intact, but the underlying musculature and structure is impaired (Peterson-Falzone et al., 2010). Since the skin layer of the roof of the mouth is intact, often SMCPs are not identified at birth, and commonly are not diagnosed until the affected individual's speech fails to develop normally (Peterson-Falzone et al., 2010).

2.1.2 Classification and incidence

Cleft lip +/- palate has an incidence of 1 in 700 births in the UK (CRANE Database, 2019). Over the years orofacial clefts have been described using many different classification systems (Jones & Jones, 2016; Mossey et al., 2009; Watson, 2001). Classification is of importance for research, as it provides a basis for inquiry, be it fundamental, epidemiological or clinical (Mossey et al., 2009; Mossey & Modell, 2012). The UK Cleft Registry and Audit Network (CRANE) group non-syndromic orofacial clefts into the following types (CRANE Database, 2019):

- Cleft lip (CL);
- Unilateral cleft lip and palate (UCLP);

- Bilateral cleft lip and palate (BCLP);
- Cleft palate only (CPO).

The reader is referred to Mossey et al. (2009) for illustrations and descriptions of these different types of clefts. For babies born in the UK in 2018 (n = 1,002), 39.3% were CPO, 21% UCLP, 19.6% CL, 10.6% BCLP, and 9.5% were unregistered (CRANE Database, 2019).

Oro-facial clefts can also be categorised as non-syndromic or syndromic (Jones & Jones, 2016; Lees, 2001). A cleft is described as "non-syndromic", when the affected individual does not have any additional structural and/or cognitive anomalies. The cause of non-syndromic clefts is thought to be multifactorial and is surmised to occur when a genetic disposition is paired with environmental factors, such as smoking, alcohol, maternal illness, or drugs (Jones & Jones, 2016; Mossey et al., 2009). A syndromic cleft is where other anomalies occur alongside the cleft, such as learning disability and other physical problems (Jones & Jones, 2016; Lees, 2001). Multiple anomalies in syndromic clefts have the same cause; these are teratogenic, genetic or a mixture of the two (Lees, 2001; Stoll et al., 2000). Genetic conditions include gene alterations or changes to entire chromosomes (Stoll et al., 2000). Reports of the frequency of syndromic clefts vary between 20 – 60%, with a higher incidence in clefts of the secondary palate (Mossey et al., 2009; Stoll et al., 2000). Over 400 syndromes include cleft lip +/- palate as a feature, including, for example, 22q1.1 deletion syndrome, Stickler's syndrome, and Treacher Collin's syndrome (Lees, 2001; Stoll et al., 2000).

2.2 Speech in individuals with cleft palate

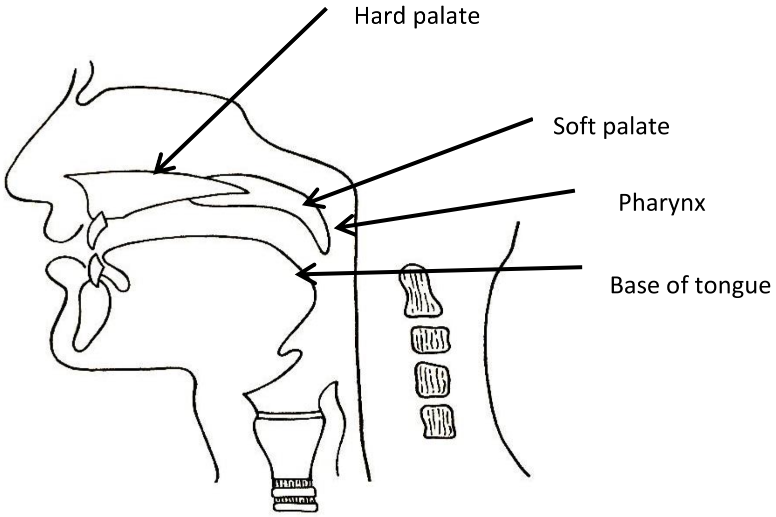
The negative consequences of a cleft of the palate on speech are well recognised. An intact hard palate and a moving soft palate are needed to make a range of speech sounds in all languages. The speech of individuals with unrepaired cleft palate +/- lip is typically highly unintelligible (Sell & Grunwell, 1993). Consequently, maximising speech outcomes is a key goal in the management of this client group. This section begins with a description of soft palate function for speech. The section goes on to describe the effect of a cleft palate on speech development. This is followed by discussion of the speech features associated with cleft palate.

2.2.1 Soft palate function

The soft palate (velum) is attached to the hard palate anteriorly. The soft palate's free edge sits above the base of the tongue in the oro-pharynx (see Figure 1). When a person speaks, the soft palate lifts and extends towards the pharynx through the action of a sling of velar muscles (levator, palatopharyngeus and palatoglossus) (Watson, 2001). Velopharyngeal closure (i.e. closure of the velum against the pharynx) typically involves lifting and extension of the soft palate, together with varying degrees of lateral and posterior pharyngeal wall movement (Watson, 2001). Velopharyngeal closure seals off the nasal cavity from the oro-pharngeal cavity. This closure is needed for the production of a range of English consonants, including /p/, /b/, /t/, /d/, /f/, /v/, /s/, /z/, /ʃ/, /ʒ/, /tʃ/, /dʒ/, /k/, and /g/. To make these sounds, air from the lungs is forced into the mouth, the velopharyngeal sphincter closes and different lip and tongue configurations contribute to different sound productions. As such, these speech sounds involve a higher level of oral pressure and are known as oral pressure consonants and accurate production, in part, depends on an intact hard and soft palate, together with a soft palate capable of achieving velopharyngeal closure. Velopharyngeal dysfunction (VPD) occurs when the velum fails to close off with the pharynx, resulting in air and sound going up into the nasal cavity and a loss of pressure with the production of oral pressure consonants. VPD can be due to structural problems with the velopharyngeal mechanism, commonly known as velopharyngeal insufficiency (VPI), motor difficulties with the velopharyngeal mechanism, known as velopharyngeal incompetency, and/or velopharyngeal mislearning (Peterson-Falzone et al., 2006; Trost-Cardamone, 1989). The latter is where the velophayngeal mechanism is capable of closure, but where air/sound is deliberately forced up into the nose to make a sound.

Figure 1

Sagittal view of the face showing hard and soft palate



(adapted from Peterson-Falzone et al. 2010; original figure copyright free)

2.2.2 Effect of cleft palate on speech development

For most individuals, the effects of an unrepaired cleft palate first become apparent at 6-months of age when the infant begins to babble using single syllables. If the cleft palate is unrepaired, syllables containing oral pressure consonants, such as /b/ and /d/ will normally be unachievable (Russell & Grunwell, 1993). However, sounds not requiring oro-nasal closure are possible, such as /m/ and /n/ or glottal sounds, /h/ and /ʔ/. If the palate is not repaired, such atypical speech patterns continue. Studies of children and adults with unrepaired cleft palate in the developing world show these individuals' speech is highly unintelligible and, in the main, is characterised by excessive hypernasality (too much sound resonating in the nose) and difficulty with the production of oral pressure consonants (Peterson-Falzone et al., 2006; Rees et al., 2016; Sell & Grunwell, 1993).

In order to prevent speech problems, in the UK, the cleft condition is typically repaired in the individual's first year of life. However, even with palate repair at an early age, some children are slower to develop speech, or speech is characterised by features shown in those with unrepaired cleft palate. For some of these children, the cause of these problems lies with on-going difficulties with velopharyngeal closure, the result of partial or unsuccessful palate repair (Grunwell & Sell, 2001). In the UK, the incidence of VPD due to structural difficulties (VPI) is estimated at around 30% for individuals born with a non-syndromic cleft palate +/- lip (CRANE Database 2019). For some children, speech difficulties are associated with hearing difficulties (Sell et al., 2017). For many children the cause of on-going speech difficulties subsequent to palate repair is not clear, and is likely to be multifactorial and complex (Grunwell & Sell, 2001).

2.2.3 Incidence of speech problems

Around two-thirds of children born with cleft palate require speech and language therapy intervention (Albery & P Grunwell, 1993; Dalston, 1990; Hardin-Jones & Jones, 2005; Sell et al., 2001). According to the UK CRANE Database, at age 5-years, 39% of children with non-syndromic cleft born between 2010 and 2012 in the UK, had on-going speech difficulties (n = 1,346) (CRANE database 2019). It is estimated that around 20% of individuals born with the cleft condition will go onto show significant persistent and intransient speech difficulties requiring long term therapy (Peterson-Falzone, 1995; Sell et al., 2001; Sell et al., 2017). The focus of this research is with this 20% of patients with persistent SSD.

2.2.4 Characteristics of cleft palate speech

The term "cleft palate speech" is used to describe the characteristics of speech commonly seen and described with individuals born with a cleft palate (Grunwell & Sell, 2001; McWilliams et al., 1990). The most common features of cleft palate speech include abnormal nasal resonance, abnormal nasal airflow and atypical consonant production. Each of these features is discussed further below.

2.2.4.1 Abnormal nasal resonance

In the context of speech, resonance may be defined as the vibration of sound in the pharyngeal, oral and nasal chambers as a person speaks. Too much sound vibrating in the nasal chambers is known as hypernasality, too little is referred to as hyponasality (Sell et al., 1999). Hypernasality is commonly seen in individuals born with cleft palate (Peterson-Falzone, 2006). For these individuals, hypernasality occurs if the velum fails to close off with the pharynx during production of speech sounds involving a higher level of oral pressure. In addition, hypernasality can occur in the present of an oro-nasal fistula (Peterson-Falzone, 2006). Hyponasality can also be a feature of cleft palate speech due to, for example, nasal defects, nasal congestion, and obstructive pharyngoplasties (Sell et al., 1999). At times mixed hyper- hypo-nasality is seen in individuals with cleft palate (Peterson-Falzone, 2006). Some individuals can also present with cul-de-sac resonance (Sweeney, 2011). This is where sound resonates in a cavity, usually the nose, but where an obstruction results in the sound being "trapped". Perceptually, this resonance produces a muffled tone (Sweeney, 2011).

2.2.4.2 Abnormal nasal airflow

Abnormal nasal airflow occurs when production of oral pressure consonants is accompanied with nasal airflow (Sell et al., 1999). Nasal airflow errors include nasal emission or nasal turbulence (Sweeney, 2011). Nasal emission is described as having a frictional quality, while nasal turbulence is defined as a "snorting" or turbulent noise (Sweeney, 2011). Nasal turbulence commonly occurs when air is forced through a small velopharngeal gap or small oro-nasal fistula, while nasal emission is associated with a larger velophangeal gap or larger oro-nasal fistula (Sell et al., 1999; Sweeney, 2011). Nasal airflow errors are another indication that the velopharyngeal sphincter is not closing adequately for production of oral pressure consonants, or that the individual has an oro-nasal fistula, so that air escapes into the nose during production of oral pressure consonants (Peterson-Falzone, 2006).

2.2.4.3 Consonant production

Errors with consonant production seen in cleft palate speech largely lie with production of oral pressure consonants. In the UK, most academics and clinicians describe atypical consonant production using the descriptive system outlined in the Great Ormond Street Speech Assessment (GOS.SP.ASS) (Sell et al., 1994; Sell et al., 1999), including: **anterior oral cleft speech characteristics; posterior oral cleft speech characteristics; non-oral cleft speech characteristics; and passive cleft speech characteristics. Anterior oral cleft speech characteristics** commonly seen in cleft palate speech include: dentalisation of sounds involving alveolar placement; lateralisation/lateral production of lingual sibilant sounds, and occasionally lingual stop sounds; palatalisation/palatal production of lingual sibilant and stop sounds; and double articulation of stop sounds (e.g. /t/ →[t͡k], /p/ → [p͡k]) (Sell et al., 2001; Sell et al., 2015). Anterior cleft speech characteristics are commonly associated with malocclusion, though in some cases may be linked with oro-nasal fistulas (Albery & Grunwell, 1993). Double articulation often occurs following speech and language therapy, where correct placement is achieved, but this placement is paired with habitual, in error, placement (Grunwell & Sell, 2001). **Posterior oral cleft speech characteristics** commonly seen in cleft palate speech include backing of bilabial, alveolar and post-alveolar sounds to velar or uvular placement. Backing to velar or uvular placement is commonly associated with oro-nasal fistulas and VPI (Sell et al., 2001; Sell et al., 2015) .

Non-oral cleft speech characteristics include: glottal articulation, where oral stop sounds are replaced by glottal stops and lingual sibilant sounds are replaced by [h]; pharyngeal articulation, where typically lingual sibilant sounds are replaced by [ħ], and use of active nasal fricatives (Sell et al., 2001; Sell et al., 2015). Active nasal fricatives is the term used to describe the deliberate, noisy, blowing of air down the nose, usually to mark a sibilant sound (Grunwell & Sell, 2001). Non-oral speech characteristics are usually associated with significant VPI. **Passive cleft speech characteristics** include weak/nasalised productions of oral pressure consonants, nasal realisations of sibilants, nasal realisations of stops, absent oral pressure consonants, and gliding of sibilants (Sell et al., 2001; Sell et al., 2015). Passive speech characteristics are again associated with VPI.

Posterior and non-oral cleft speech characteristics are often termed as "active" or "compensatory" (Grunwell & Sell, 2001; Peterson-Falzone, 2006). It is surmised that, typically, these characteristics occur in response to current or previous velopharyngeal deficits/oro-nasal fistula: the individual is unable to produce the oral pressure sound correctly because of difficulties with palate closure, so they produce the sound in another way, within the constraints of their defective speech mechanism. Should the velopharyngeal deficit be fixed, for example through primary or secondary speech surgery, this will not necessarily resolve the problem, since the speech sound errors have become habitualised. These individuals are likely to need speech therapy to remediate these speech sounds. In comparison, with "passive" or "obligatory" speech characteristics, the individual is targeting the oral pressure speech sound accurately, however VPI or an oro-nasal fistula results in hypernasality and/or abnormal airflow. If the velopharyngeal mechanism can be normalised, for example through secondary speech surgery, then the passive or obligatory speech characteristics will be remediated, without the need for speech therapy (Grunwell & Sell, 2001; Peterson-Falzone, 2006). Active and passive cleft speech characteristics frequently co-occur (Grunwell & Sell, 2001).

As described above, at age 5-years 39% (n = 1,346) of children with non-syndromic cleft born in the UK between 2010 and 2012 had on-going speech difficulties (CRANE Database 2019). The most common speech errors (involving one of more consonants) included: palatalisation/palatal production (23%), backing to velar/uvular (12.1%), use of active nasal fricatives (10.2%) and lateralisation/lateral production (8.1%).

The consonant errors associated with cleft palate speech may be primarily viewed as articulatory in nature, that is, they occur because of difficulty with the physical production of speech sounds due to the cleft condition. However, in a seminal review paper, Grunwell and Harding (1996) point out that some of the errors seen in cleft palate speech result in loss of contrast between speech sounds, resulting in inability to signal differences in meaning. In addition, these authors also identify a number of "cleft type developmental processes" commonly seen in cleft palate speech. Developmental phonological processes describe regular speech patterns observed in children developing speech, such as where all sibilant sounds are "stopped" and substituted with stop sounds, or where all sounds with velar placement are "fronted" to alveolar placement (McLeod & Baker, 2017). Such developmental processes can be:

- Typical, i.e. the speech patterns shown in a particular child are the same as those seen in children developing normal speech;

- Delayed, i.e. the speech patterns shown in a particular child are typical of younger children and should have resolved;
- Disordered, i.e. the speech patterns shown in a particular child are not typical of the speech of younger or older children (McLeod & Baker, 2017).

With regard to cleft palate speech, Harding and Grunwell (1996) identify both delayed and disordered developmental patterns. For example, children born with cleft palate often present with the delayed pattern of persistent “stopping” (Chapman, 1993; Russell & Grunwell, 1993). Atypical patterns seen with this client group include, for example, “backing” patterns, where sounds with anterior placement are substituted with sounds with more posterior placement, and non-oral substitution patterns, where oral pressure sounds are replaced by glottal, pharyngeal or active nasal fricative sounds (Chapman, 1993; Russell & Grunwell, 1993). A number of researchers speculate that developmental patterns shown in cleft palate speech are a consequence of the impact of a defective phonetic mechanism of the child’s phonological learning over time (Chapman, 1993; Grunwell & Dive, 1988; Harding & Grunwell, 1996; Russell & Grunwell, 1993). Harding-Bell and Howard (2011) also suggest cleft developmental patterns may be in part due to fluctuating hearing loss.

3 ELECTROPALATOGRAPHY (EPG) THERAPY

As identified in the previous chapter, at age 5-years, close to 40% of children with non-syndromic cleft palate +/- lip in the UK have on-going SSD requiring intervention (CRANE database, 2019). Articulation, or "motor-phonetic" therapy, is the most common treatment technique for cleft palate speech (Bessell et al., 2013; Golding-Kushner, 2001; Peterson-Falzone, 2006). Articulation therapy, as first described by Van Riper (1939), involves facilitation of the physical production of speech sounds through demonstration and phonetic cueing (i.e. prompts to achieve correct physical production of a speech sound). Speech production is taught in a hierarchical way including, production of a target sound in isolation, production of the target sound in consonant vowel (CV) and vowel consonant (VC) sequences, practice of the target sound at word level, phrase level and then sentence level. Finally the target sound will be practiced in connected speech (Preston & Leece, 2021). This type of therapy, used commonly in the treatment of all SSDs (Brumbaugh & Smit, 2013; Joffe & Pring, 2008), is often adapted for the cleft client group. For example, Harding and Grunwell (1998) suggest adjustments such as, targeting voiceless sounds and working on syllable final targets first. These adjustments account for difficulties with production due to on-going VPI, i.e. where VPI is present, production of voiceless sounds and sounds in word final position will be easiest. Articulation therapy is described further in 3.3.1.

In addition to motor-phonetic therapy, "phonological" approaches have been used to treat cleft palate speech (Pamplona et al., 1999; Pamplona et al., 2004; Pamplona et al., 1996; Williams et al., 2021). Phonological therapy seeks to increase the individual's awareness or knowledge of the structure of their ambient speech sound system (McLeod & Baker, 2017). Many phonological therapies strive to increase this awareness and knowledge through contrastive speech practice, such as homophony confrontation (Baker, 2010; Baker & Williams, 2010). For example, if a child is producing [k] for /t/, they will be confronted with homophones such as "key" and "tea" to highlight the importance of [t] versus [k] production to signal differences in meaning.

More recently, psycholinguistic-based therapy and early intervention therapy have been used with this client group (Calladine, 2019; Scherer & Kaiser, 2010; Williams et al., 2021). Psycholinguisitic models conceptualise how speech is processed and represented in the brain. Psycholinguistic theory used in the treatment of SSDs typically conceptualises multiple levels of speech processing and this processing is presented using box and arrow diagrams. For example, Stackhouse and Wells (1997) and Dodd (2005) propose models with three core elements: input processes (where the auditory speech signal is detected and processed in a series of stages), cognitive-linguistic storage and processing (involving the creation, storage and access of lexical representations of words, including semantic, phonological, syntactic, orthographic and motor information) and output processes (where speech sounds are articulated through a series of stages). Psycholinguistic management typically involves identification of a child's strengths and weaknesses with speech processing; subsequent therapy aims to build on strengths or address weaknesses, usually through techniques used in articulation and phonological therapy (Pascoe, 2006). Early intervention therapy seeks to modify deviant and restricted speech output in the babble and first words of infants with cleft palate (Scherer & Kaiser, 2010). It does this through techniques such as providing multiple models of target speech sounds, and through shaping output using behavioural modification strategies (Scherer & Kaiser, 2010).

Bessell et al. (2013) carried out a systematic review of published group studies involving the treatment of cleft palate speech. These reviewers concluded that, up to this point in time (2013), little statistical evidence existed to support use of any treatment approach, including motor-phonetic, linguistic-phonological, psycholinguistic and early intervention approaches. Therefore, these authors argue adequately powered, methodologically robust, intervention studies are needed with this client group. Sand et al. (2022) reported on a further systematic review of published studies involving treatment of cleft palate speech. These authors suggested small group studies involving heterogeneous participants are potentially flawed. They pointed out that inconsistent pre-post speech differences for individual participants receiving intervention (i.e. where some individual show large improvements, while other individuals show small improvements) can lead to group findings that do not achieve statistical significance. This lack of statistical significance can occur despite clinically important speech change for some individual participants. These authors thus re-examined published studies involving the treatment of cleft palate speech and considered improvements for individual participants. This review included participants in both single case studies and group studies (n = 343). The conclusion was that 75% of all participants improved to a clinically relevant degree following motor-phonetic and phonological therapy.

Visual biofeedback therapy, including EPG and more recently ultrasound, has also been used with school-aged children and adults with cleft palate +/- lip (e.g. Lohmander et al., 2010; Roxburgh, 2018). Use of visual feedback through EPG or ultrasound is typically done using motor-phonetic therapy (e.g. Fujiwara, 2007; Lohmander et al., 2010; Roxburgh, 2018). Section one of this chapter gives a general overview of EPG therapy. Section two reviews previous EPG intervention studies with individuals with SSD secondary to cleft palate. Section three considers the theoretical underpinnings for current EPG therapy. The chapter concludes with a summary and conclusions.

3.1 Overview of EPG

EPG is an instrumental technique used to assess and treat individuals with SSD. EPG detects tongue contact with the hard palate during speech. This contact produces a visual display of patterns of tongue-palate contact, viewed on a screen in real time (Gibbon, 2004; Gibbon & Wood, 2010; Lee, 2021). EPG is one of a small number of visual biofeedback interventions in which instruments are used to provide the patient with visual feedback on their speech sound production in real-time. Other biofeedback techniques include ultrasound and acoustic biofeedback (Cleland & Preston, 2021). Biofeedback techniques, such as EPG, are usually “second-line” intervention techniques and are typically used with school aged children and adults with complex, long-standing SSD that have not responded to more conventional speech therapy interventions, such as traditional articulation therapy (Gibbon & Wood, 2010). Instrumental interventions are commonly used with SSD involving difficulties with the physical production of speech sounds, for example, where /s/ is produced as a lateral “s”, [ɬ], or where /t/ is produced as [k]. Individuals put forward for instrumental treatment are typically those who have never or rarely been able to articulate lingual speech sounds accurately. Many individuals with long-standing, cleft-related SSDs fall into this group. Lee et al. (2009) suggests biofeedback techniques work by giving the patient specific, real-time information on physiological systems, which the patient normally cannot observe directly, such as tongue-palate contact. This real-time information is used by the individual to facilitate control of a particular physiological system (Lee et al., 2009). EPG identifies and helps treat SSDs involving tongue-palate contact such as /t/, /d/, /s/, /z/, /n/, /l/, /ʃ/, /ʒ/, /tʃ/, and /dʒ/ (Gibbon & Patterson, 2006). Since these speech sounds are particularly vulnerable to the cleft condition (Britton et al., 2014; Sell et al., 2015), EPG may be viewed as particularly suited to intervention with this patient group.

EPG visual information on tongue-palate contact for lingual speech sounds is provided by way of a removable plate that fits to the roof of the mouth (Gibbon 2004; Gibbon & Wood, 2010). This plate, known as an artificial palate, contains sensors that detect tongue-palate contact. The artificial palate connects to processing units and a screen. The screen provides the patient and the SLT with a real-time visual display of the patient’s tongue-palate contact during speech by means of a visual grid (corresponding squares light up when the tongue makes contact with the artificial palate, see Figure 4 for an example of images seen) (Lee, 2021). Alongside auditory-perceptual judgement of speech, the SLT uses information on tongue-palate contact to examine placement and timing of the patient’s lingual speech sounds. Following assessment, the EPG visual display is used during therapy to facilitate and monitor more typical production of speech sounds, usually in real time (Gibbon, 2004; Gibbon & Wood, 2010) (see 3.1.4 for description of EPG therapy). Most EPG systems have the facility to record and store EPG data. This is done using specialist software, such as Articulate Assistant software (Wrench, 2007). EPG data recorded and stored before, during and after therapy permits detailed analysis of the timing and tongue-palate placement for speech sounds (Gibbon & Wood, 2010; Hardcastle et al., 1991). A description of types of EPG analysis is discussed in 3.1.2 of this chapter. As observed by Cleland and Preston (2021) and Lee (2021), many articulatory gestures are hidden within the vocal tract and can be difficult to identify and describe. Further, production of speech sounds in words and continuous connected speech is fleeting. EPG permits capture, visualisation and, with some systems, the facility for detailed evaluation of articulatory gestures. Thus, EPG provides information not available to auditory-perceptual analysis alone (Lee, 2021). Figure 2 shows a picture of an EPG system in use.

Figure 2

An EPG system (WinEPG) in use: Person wearing artificial palate connected to processing units and a screen

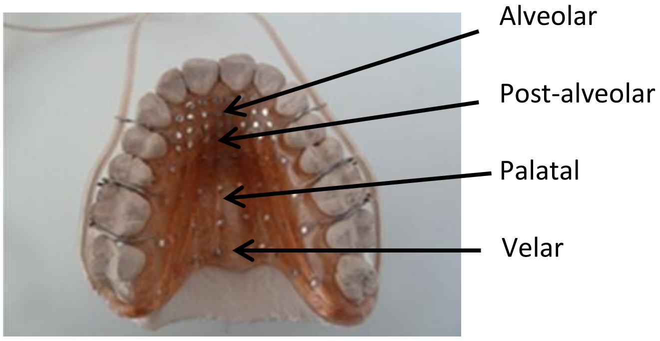
EPG systems have been used for clinical and research purposes over the past four decades (Hardcastle et al., 1991). In this time, EPG has been used to treat a variety of groups with speech impairment, including, for example, functional articulation disorders (e.g. Carter & Edwards, 2004; Dent et al., 1995), hearing impairment (e.g. Bacsfalvi et al., 2007; Darelid et al., 2016), cerebral palsy (e.g. Gibbon & Wood, 2003; Nordberg et al., 2011), developmental dyspraxia (e.g. Leniston & Ebbels, 2021; Lundeborg & McAllister, 2007), and Down's syndrome (e.g. Cleland et al., 2009; Gibbon et al., 2003; Wood et al., 2019). It has also been used to treat individuals with SSD secondary to cleft palate (e.g. Fujiwara, 2007; Lohmander et al., 2010; Whitehill et al., 1996). Gibbon (2013) provides a comprehensive bibliography of EPG studies in English from 1957-2013. In the UK, the majority of regional cleft centres have EPG systems and are able to offer this type of visual feedback therapy to their patients (Patrick, 2013).

3.1.1 Artificial palates

Artificial palates detecting tongue-palate contact are custom-made to fit snuggly onto the patient's palate. The plate covers the entire hard palate, extending lengthways from the alveolar region to the junction of the hard and soft palate and laterally to the gingival border (Wrench, 2007). Artificial palates are made by specialist dental technicians using a dental cast of the patient's upper teeth and palate. The dental technician embeds sensors into a thin layer of acrylic to form the plate (Gibbon & Wood, 2010). A number of different artificial palates are in use world-wide, including the Reading EPG plate, the Articulate EPG plate and the CompleteSpeech EPG plate (see Lee, 2021, for a detailed description of these different plates). The Reading EPG plate is used in this research. The Reading EPG plate contains 62 electrodes in total. These sensors are positioned in eight horizontal rows in the areas where the tongue typically touches the palate for speech sounds. Since the majority of lingual speech sounds are made by making contact with the anterior part of the palate, four of the eight horizontal rows are concentrated in this anterior region (Hardcastle et al., 1989). Figure 3 shows the underside of an EPG palate and the positioning of the silver sensors according to anatomical landmarks.

Figure 3

Underside of a Reading-style EPG palate showing positioning of silver sensors according to anatomical landmarks

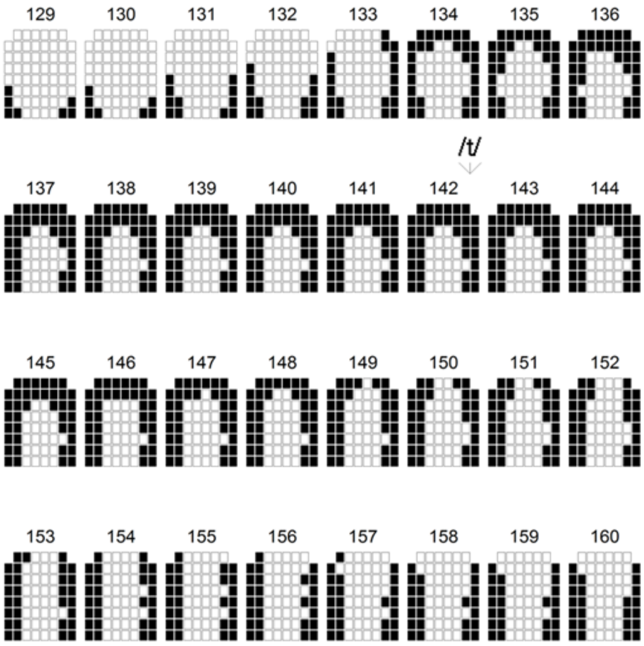


3.1.2 Visual display and analysis of EPG data using the WinEPG system

A number of EPG systems are in use across the world, including the WinEPG system (Articulate Instruments Ltd, 2010), the Rose system (https://www.rose.medical.com), the Speech Training Aid and Recording (STARS) system (Yamamoto, 2020), and the Smart Palate palatometer system (http://completespeech.com). The WinEPG EPG system is the system used in this research. This system displays tongue-palate contact dynamically in two-dimensional pictures that in appear every 10 milliseconds (though this rate can be adjusted). Individual pictures are known as EPG frames or palatograms. Figure 3 shows the EPG frames for a /t/ sound in the phrase “a tea” in an adult speaker with typical speech. The black squares represent tongue-palate contact; the white squares no contact. The frames show contact over time and are read from left to right. The EPG frame identified by an arrow represents the frame of maximum tongue contact before the /t/ speech sound is released. Sound, waveform and spectrographic data facilitate the segmentation of the speech stream (not shown in Figure 3). In this way, EPG provides spatial and temporal information on tongue-palate contact during continuous speech. EPG does not give any information on tongue shape or the part of the tongue involved in articulation of lingual speech sounds. A further limitation of the system is that tongue-palate contact is limited to the area covered by the artificial plate and placement of the sensors. For example, uvula contact will not be shown. In addition, the transfer of contacts to a standard grid potentially leads to further data reduction (Gibbon & Wood, 2010; Hardcastle et al., 1989; Hardcastle et al., 1991; Lee, 2021; Wrench, 2007). For example, if the patient has misaligned teeth or a narrow, misshapen hard palate, the dental technician may have difficulty positioning sensors, so that the number of sensors may be reduced and/or sensors may be positioned in slightly different areas compared to a typical EPG palate (Wrench, 2007). In these circumstances, the patient's EPG frames may be similar or identical to other individuals' EPG frames for a particular sound, and yet exact tongue-palate placement may differ.

Figure 4

EPG frames for the phrase "a tea". Frames 129 – 133 show tongue-palate placement for onset of /t/, frames 134 – 153 show tongue-palate contact for the closure phase of /t/, with frame 142 representing the frame of maximum contact and frame 149 showing release of /t/.



Despite some individual variation within and across speakers (Gibbon & Lee, 2011), all lingual-palatal speech sounds in mature speakers with typical hard palates and speech have visually recognisable and distinct palatograms. For example, the horseshoe-shape pattern shown in Figure 4 is the broad EPG pattern shown in typical speakers for production of /t/, /d/, and /n/ (McLeod & Singh, 2009). Examples of EPG palatograms for all English consonants and vowels (adult speakers with typically speech) are illustrated in McLeod and Singh (2009) and can be found in a number of research papers (Cheng et al, 2007; Dagenais &Critz-Crosby, 1991; Fletcher, 1989).

Palatograms can be described qualitatively and quantitatively. Qualitative analysis consists of descriptive statements relating to EPG frames, for example, "EPG frames showed" "anterior groove formation" (Gibbon et al., 2001) and "complete closure" (Cleland et al., 2009). Gibbon (2004) presents a descriptive classification system to label the types of EPG error patterns commonly seen in cleft palate speech. This classification system was developed from data collected by the EPG research team at Queen Margaret University, Edinburgh, together with published EPG studies, and is used in this research. The following patterns are described in this descriptive classification:

- Increased contact: This error pattern, which can affect all lingual speech sounds including consonants and vowels, is used to describe the pattern where there is activation of a large number of sensors across the palate. Gibbon observes that this is one of the most frequently observed error patterns of individuals with cleft palate +/- lip.
- Retracted to palatal or velar placement: This pattern typically affects speech sounds with alveolar/post alveolar placement, such as /s/, /t/, /ʃ/, /tʃ/, and visually is shown by activation of sensors in the palatal/velar regions.
- Fronted placement: A fronted pattern affects velar speech sounds, such as /k/, /g/ and /ŋ/ and is shown by activation of sensors in the palatal or alveolar region. Gibbon observes that this pattern is less common in cleft palate speech.
- Complete closure: This pattern affects speech sounds such as /s/, /ʃ/, and /tʃ/, which are produced with an anterior tongue "groove" in typical speech. This groove permits airflow over the tongue and out of the mouth in a central airstream and is shown by a white channel on palatograms. In the "complete closure" error pattern, no channel/groove is seen and air either escapes around the lateral margins of the tongue (resulting in lateral production), or is directed into the nose (resulting in active nasal production). It should be noted assessment of lateral and active nasal production is also based on auditory-perceptual evaluation and nose-holding (Harding & Grunwell, 1998), since EPG does not detect direction of air flow.
- Open pattern: This pattern is shown when lingual targets shown no or little tongue-palate contact. Constriction for speech sounds typically occurs further back in the uvular region or in the pharynx (resulting in pharyngeal or glottal speech sounds).
- Double articulation: Double articulation occurs when constriction of the articulators occurs in two regions. This error most commonly affects /t/, /d/, and /n/ speech targets, and sensor contact is shown in both the alveolar and velar regions. Gibbon (2004) reports that speech sounds involving bilabial closure (e.g. /p/, /b/) can also be accompanied by tongue contact in the velar region.
- Increased variability: Gibbon (2004) observes that EPG traces in individuals with cleft palate speech can be highly variable, i.e. EPG patterns can vary over repetitions of the same word or utterance.
- Abnormal timing: Although less common, Gibbon (2004) reported two studies showing abnormal timing of articulations by two speakers with speech disorders secondary to cleft palate. The first study involved a 6-year-old girl with cleft palate speech (Howard, 1993). The stop targets /t/, /d/, /k/ and /g/ were phonetically transcribed as [ʔ]. However, instrumental analysis showed duration of stop closure differed with voiced and voiceless stop target production, with the voiceless stop targets, /t/ and /k/ having a significantly longer closure phase than their voiced counterparts, /d/ and /g/. Howard (1993) suggested this difference was intended by the speaker and served to produce a voicing distinction. In the second study, which involved an adult with cleft palate speech (Gibbon & Crampin, 2001), abnormal

timing with the acoustic burst on the release phase of /t/ and /k/ was interpreted by the authors as relating to lateral release.

In addition to qualitative data analysis, numerical measures or indices are used by some clinicians and researchers (Hardcastle et al., 1989; Lee, 2021). These indices reduce EPG data to single numerical values, and these values can be compared across different test points and used in statistical analysis (Hardcastle et al., 1989). Lee (2021) gives a comprehensive description of EPG indices used or proposed in the literature, including formula for calculation of these indices. Articulate Assistant software (Wrench, 2007) automatically calculates a range of numerical indices. EPG indices were used in this research and these were computed using Articulate Assistant software. Indices reported in the present study include: Alveolar Total; Alveolar Total, Alveolar Closure; and Centre of Gravity. A detailed description of these indices, including their formula, is provided in the Articulate Assistant User Guide, pp. 32-42 (http://materials.articulateinstruments.com). What follows is an overview of each of the indices used in this research from this user guide:

- Alveolar Total (AT) – AT is the number of electrodes contacted in the first three rows of the EPG palate per EPG frame. This analysis is used where anterior-posterior change with speech sound production is the focus of therapy. An increase in the numerical value of AT across test points represents progress with, for example, a /t/ to [k] speech error, where /t/ is the focus of therapy (i.e. the value for a velar stop will be zero; the value rises with increased contact in the first three rows of the EPG palate per EPG frame for an alveolar stop target).
- Whole Total (WT) – WT represents the total number of electrodes contacted per EPG frame. High WT numbers (compared to reference data) is considered a sign of undifferentiated tongue contact patterns (Gibbon, 2004). A reduction in WT across test points indicates increasingly differentiated tongue contact.
- Alveolar Closure (AC) – AC provides a score for the connectivity between the right and the left sides of the first three rows of the EPG plate. The higher the AC score, the more contact between the two sides of the palate. This analysis is used, for example where /s/ is produced as [ɬ], and where therapy is aiming for a groove formation for /s/ (i.e. air flowing out of the mouth with central airstream).
- Centre of Gravity (CoG) – The CoG index indicates the position of the main concentration of electrodes across the palate per EPG frame. Small CoG values relate to posterior tongue-palate contact. CoG values rise with increases in anterior tongue-palate contact. An increase in CoG values across test points represents progress with, for example, a /n/ to [ŋ] speech error, where /n/ is the focus of therapy.

A range of EPG indices have been used in previous EPG intervention research with individuals with cleft palate speech. Lohmander et al. (2010) used AT, WT and CoG to measure change with a posteriorly placed /s/ and /t/ in a 11-year old child with cleft palate. Examination of these indices showed considerable change in values relating to word initial /t/ following treatment, compared to before treatment (i.e. increases in AT, CoG and decreases in WT, all in the direction of improvement). Further, these differences were statistically significant. Word final /t/ and /s/ in words and sentences following treatment showed a trend towards improvement using these indices. However, these trends were not confirmed statistically. In this study, improvement in EPG indexes for word initial /t/ and trends towards improvement for word final /t/ and /s/ corresponded to improved accuracy of production of /t/ and /s/ on auditory-perceptual assessment following treatment, compared to before treatment. Fujiwara (2007) used CoG to examine change from posterior tongue-palate placement to more anterior placement for anterior stop and sibilant speech sounds for five participants with cleft palate speech. Following treatment, CoG values increased for 4/5 participants, i.e. in the direction of improvement. However, no statistic statistical testing was completed. In this study, improved CoG scores corresponded to subjective reports of improved auditory-perceptual judgements following treatment. Gibbon et al (2001) also used CoG to examine change from posterior tongue-palate placement to more anterior placement for anterior stop and sibilant speech sounds for participants with the cleft condition (n = 12). Two of the authors of this paper made judgements on improvement with CoG values, and results were presented descriptively in terms of: “positive change”; “no change”, and “negative change”. Specific CoG values were not provided. Analysis showed that 75% of participants had more normal articulatory patterns following EPG therapy. No auditory-perceptual outcomes were included in this paper.

In some previous studies, the EPG frames of individuals with speech difficulties have been compared to the EPG frames of people with typical speech (e.g. Gibbon et al., 1998; Gibbon & Hardcastle, 1989; Stokes et al., 1995; Whitehill et al., 1996). However, in some cases this may not be a valid comparison. As discussed in 3.1.2, in individuals with cleft lip and palate, the configuration of the hard palate will be affected. Therefore, it may be difficult to position the sensors in a standard way (Wrench, 2007). For example, if the individual has a very narrow dental arch, then the dental technician may not be able to place all sensors. Consequently, in these cases, direct comparison with EPG palatograms of typical speakers is not possible. In addition, though all speakers appear to have the same broad spatial patterns for target sounds, for example, horseshoe shape for /t/ (McLeod & Singh, 2009), research (Gibbon et al., 2010; Liker & Gibbon, 2008) suggests variation in amount of tongue-palate contact between typical speakers. For example, Gibbon et al. (2010) found considerable inter-speaker variation in percentage of tongue-palate contact for high vowels in ten typical English-speaking adults. Likewise, Liker and Gibbon (2008) reported extensive inter-speaker variability in percentage of tongue-palate contact for velar stops in seven normally speaking adults. Thus, caution is needed in using EPG indices from typical speakers as comparators, even with patients with the cleft condition who have more typical dentopalatal morphology. In other studies, individual's EPG traces are compared pre- and post-therapy (Cleland et al., 2009; Lohmander et al., 2010). The shortcoming of this approach is that, although changes may be seen on EPG data, this change may not be evident perceptually (i.e. no or minimal change on speech sound accuracy and no change with ratings of intelligibility of speech). For this reason, EPG analysis is best done alongside impressionistic phonetic transcription (Lee et al., 2009; Lee, 2021).

3.1.3 Patient selection

Not all patients will be appropriate for EPG intervention. EPG therapy assists with speech sounds involving tongue-palate contact (i.e. /t/, /d/, /s/, /z/, /j/, /ʃ/, /ʒ/, /tʃ/, /dʒ/, /k/, /g/, /ŋ/, /r/, /l/, /i/). It can also be used to eliminate incorrect lingual gestures, such as, for example, when /p/ is produced with double articulation, [p͡k] (Gibbon & Wood, 2010). Consequently, perceptual speech assessment by the SLT is needed to determine suitability for this intervention. Obligatory to this technique is a custom-made artificial palate that fits snuggly to the roof of the mouth. Patients therefore need to have relatively stable dentition so that the plate can be fitted and worn for a period of at least several months (Gibbon & Wood, 2010). Children and young people whose dentition is changing through loss of deciduous teeth and eruption of adult teeth will not be eligible for this treatment at different points in time. Likewise, patients undergoing orthodontic treatment will be ineligible while orthodontics is on-going. Patients also need to be able to tolerate having a plate in the mouth and to be able to talk normally without excessive salivation (Lee, 2021). To check tolerance, potential patients are typically fitted with a training plate (acrylic plate without sensors), before proceeding to an EPG palate (Hardcastle et al., 1991). Those who cannot tolerate a plate in the mouth will not eligible for this type of therapy. Dental assessment by a dentist or orthodontist is therefore needed to assess dental suitability and tolerance.

Since EPG involves visualisation of tongue-palate contact, visual acuity is required. In addition, in order to benefit from this type of therapy, patients need to be able to integrate what they see (i.e. visualisation of palatograms) with speech movements. Patients therefore need to have the cognitive ability to achieve this integration, though Cleland et al. (2009) reports that this integration is relatively intuitive, even for patients with notable cognitive difficulties.

EPG palates are relatively expensive to make. Consequently, typically, this technique is only offered to patients with lingual speech difficulties who are able to commit to the therapeutic process, who can attend for regular therapy appointments and who can carry out regular practice at home (Patrick, 2013). As discussed previously, EPG therapy is also usually only offered to patients who have not responded to several episodes of conventional articulation therapy and who are unable to achieve correct articulatory gestures for lingual speech sounds (Lee et al., 2007; Lee, 2021).

Even given these patent selection criteria, research shows that some patients benefit more than others from EPG therapy, while some show no response (Gibbon et al., 2001; Gibbon & Paterson, 2006). Over the years, different researchers (Carter & Edwards, 2004; Gibbon et al., 1998; Gibbon & Wood, 2010; Hardcastle et al., 1991) have attempted to ascertain additional critical factors for success with EPG therapy. Hardcastle et al. (1991) identify patient-related factors which, from the authors' clinical experience, appear to influence success / progress with EPG therapy. These include:

- Age and cognitive ability of the patient, with increasing age and more advanced cognitive ability resulting in better outcomes;
- Severity of the patient's SSD, with more severe SSD affecting a large number of sound classes requiring longer treatment;
- Awareness and motivation, with patients with higher levels of awareness and motivation resulting in more rapid progress;
- Concomitant difficulties, with patients with additional problems, such as profound hearing loss, significant VPI, and significantly impaired dentition progressing more slowly and perhaps being unable to achieve "normal" speech.

3.1.4 Description of EPG treatment

If the patient is viewed as appropriate for EPG therapy, an EPG palate is made, EPG assessment, either through qualitative or quantitative analysis is carried out and therapy commences. EPG therapy is clinician-led and normally consists of individual sessions lasting 30 – 60 minutes (Lohmander et al., 2010; Stokes et al., 1995). EPG therapy for all patient groups, including cleft, typically follows the following sequence (Gibbon & Wood, 2010; Gibbon & Lee, 2015; Lohmander et al., 2010; Stokes et al., 1995): With the EPG palate in-situ, the SLT demonstrates how different movements of the tongue result in different displays on the monitor, e.g. front tongue movement results in squares being lit up at the front of the palatogram, while back tongue movement lights up square at the back of the palatogram. If appropriate, the therapist may talk about and show typical and atypical tongue-palate contact patterns. The therapist will then attempt to elicit individual speech sounds by showing the visual pattern required for the speech sound. This is often done by using static palatograms of typical speakers as a reference, by giving instructions for tongue placement, and by giving live demonstrations. Sounds may be elicited by using existing motor skills in different speech sound contexts (e.g. using the release phase of a /t/ sound to elicit a /s/), or by adaption of existing motor skills (e.g. eliciting a /ʃ/ by producing a /s/ sound and then dragging the tongue back for /ʃ/). EPG vocabulary terms such as "horseshoe shape", "groove" may be used to describe palatograms. In some instances, the therapist will first instruct the patient to produce silent posturing for the sound (i.e. get the patient to hold the tongue in a particular way without releasing the sound, such as bracing the tongue for an unreleased /t/ sound).

Once single sounds are elicited in isolation, therapy typically progresses in a hierarchical fashion, in the manner of articulation therapy, as described in 3.3.1. Normally patients need to achieve 80% accuracy before proceeding to the next level of complexity (Van Riper & Emerick, 1990). Speech production at each stage typically involves repetitive drill of speech targets. As patients progress with therapy the visual feedback provided by EPG is gradually reduced.

More recently, EPG therapy has been influenced by motor learning theory (e.g. Cleland & Preston, 2021; Gibbon & Wood, 2010). Motor learning theories (Adams, 1971; Schmidt, 1975) relate to motor skills in humans in general (i.e. not just speech) and consider how simple and complex motor skills are learnt. Application of motor learning theory has become increasingly popular in the treatment of a range of speech sound disorders, such as dysarthria, and apraxia of speech, where the main problem is seen to arise from impairment with the motor execution of speech (e.g. Caruso & Strand, 1999; Heng et al., 2016; Preston et al., 2016). Two key principles of motor learning theory in relation to the treatment of SSD include repetitive practice and provision of augmented feedback (i.e. feedback in addition to the individual's own intrinsic feedback systems). Thus, key to the motor learning EPG technique is a high number of production trials in each EPG session, together with provision of visual feedback through examination of real-time EPG frames (Cleland & Preston, 2021; Maas et al., 2008). Motor learning theory is discussed further in 3.3.2.

3.2 Previous EPG intervention studies with cleft palate speech

A number of studies have looked at the effectiveness of EPG therapy with individuals with SSD secondary to cleft palate +/-lip. Most studies consist of within-participant studies (e.g. Fujiwara, 2007; Lohmander, 2010; Michi, 1986; Stokes, 1996; Whitehill, 1996). To date, few group studies have examined the effectiveness of EPG therapy with this client group (Lee et al., 2009). This situation is mirrored in EPG intervention studies for other client groups with SSD and with biofeedback interventions as a whole, i.e. concentration of within-participant studies and fewer group studies (Cleland & Preston, 2020). Lee, Gibbon and Law (2009) carried out a Cochrane systematic review of EPG intervention for articulation difficulties secondary to the cleft condition (review updated in 2017). Only one study met the criteria for this Cochrane review (i.e. randomised control trial) and that was a study by Michi et al. (1993) (see Table 1 for further details of study). However, Lee et al.'s examination of the Michi et al. (1993) study identified a number of shortcomings, including:

- Small number of participants and no attempt to calculate the size of the sample required to produce statistically significant results;
- Method for random allocation into treatment groups was not described;
- Unclear blinding of outcome assessors;
- Few quantitative outcome measures were used.

Lee et al. (2009) therefore concluded: "the current evidence supporting the efficacy of EPG therapy is not strong and there remains a need for high-quality randomised controlled trial to be undertaken in this area" (Lee et al., 2009, p2). More recent reviews of the visual biofeedback speech intervention literature found similar findings and reached the same conclusion, i.e. a concentration of within-participant designs and the need for larger scale studies for all types of biofeedback techniques, including EPG, with all client groups (Cleland & Preston, 2021; Sugden et al., 2019). As discussed earlier in this chapter, Bessell et al. (2013) carried out a systematic review of all speech and language therapy intervention for children with cleft palate. Only studies that included at least 10 participants were included in the review. The only EPG study meeting this criterion was that of Gibbon et al. (2001) (see Table 1 for further details regarding this study). Bessell et al. found wide Confidence Intervals (CI) with this intervention study due to its small size (n = 12) and therefore advocated caution in generalising findings from this study to wider populations. This wider review of intervention for SSD secondary to cleft palate found little evidence to support any type of intervention with cleft palate speech and concluded adequately powered, methodologically robust, intervention studies were needed with this client group. As discussed earlier, in a more recent review, Sand et al. (2022) examined individual participant data from speech and language therapy intervention studies with cleft palate speech (n = 343). This review included both single case and group studies. Sand et al. (2022) concluded that 75% of individuals improved to a clinically relevant degree following therapy. However, none of the studies examined in this systematic review included EPG therapy, as visual feedback therapies were excluded from this investigation. Following this review, Sand et al. (2022) suggest studies should include information of whether or not intervention produces clinically significant change for individual participants. They also argue for studies that better control for confounding factors, such as maturation, and for studies where less heterogeneity amongst participants occurs.

While more methodologically robust EPG intervention studies are clearly needed, before undertaking more extensive research, Lee et al. (2009) suggest more work is needed on developing the EPG treatment technique. The reason for this is two-fold. Firstly, not all patients benefit from EPG therapy (Gibbon et al., 2001; Gibbon & Paterson, 2006). More information is needed on which individuals are most likely to benefit from this type of therapy. Secondly, even if patients do achieve some improvements, clinical experience suggests generalisation and maintenance of speech gains following EPG intervention can be particularly difficult (Cleland & Preston, 2021; Gibbon & Patterson, 2006). Gibbon and Patterson (2006) surveyed SLTs' views on EPG therapy outcomes for patients with SSD in Scotland over a 10-year period. Speech and language therapists returned questionnaires for 95% of the patients who had EPG palates made for them in Scotland over this time period (n = 71). Of those undergoing EPG treatment (n = 60), 12.5% of patients achieved total success, 41.1% showed "moderate success where some or all sounds targeted in therapy showed improvement and were used in some speaking contexts" (p 283), 19%

showed “slight improvement in some speaking contexts” (p 283), while 12.5% showed no change in any speaking contexts. Respondents attributed difficulties with generalisation and maintenance to “poor motivation, immaturities, lack of concern about speech and little awareness of speech problems” (Gibbon & Paterson, 2006, p. 284). Cleland and Preston (2021) also reach the same conclusion for visual feedback therapies in general, i.e. need for further development of treatment paradigms, to a). identify which patients do best with these types of therapy, and b). to better facilitate transfer and retention of learning.

In many ways difficulty with generalisation and maintenance with individuals undergoing EPG therapy is unsurprising. As discussed in 3.1, those patients put forward for EPG therapy, and other visual biofeedback techniques, are generally patients with very entrenched SSD who have not responded to more standard speech therapy. Consequently, any intervention study for this group of hard-to-treat patients must be considered in this context. Nevertheless, if these therapies are to be used, significant clinical change must be shown to justify their use in non-research settings.

One way of developing the EPG therapy paradigm is through well designed within-participant studies, as is done in this research. Within-participant designs allow for in-depth examination of participants difficulties, together with detailed analysis of participant’s response to the intervention. In this way, such studies can indicate if a treatment works for an individual patient and can give a detailed indication of why therapy was, or was not effective (Ebbels, 2017; Pring, 2005). A series of within-participant studies can further enhance knowledge of common key ingredients for effective EPG therapy (Pring, 2005), as done in this research.

Table 1 gives an overview and review of all published EPG intervention studies with individuals with speech difficulties secondary to cleft palate up to the present date. Only studies with an element of experimental control are included (i.e. general case descriptions are not listed) resulting in seven studies in total. Five of these utilise within-participant designs. The remaining two are small group studies. These studies are critiqued in terms of: treatment intensity/dosage; outcome measures used; reliability measures and; results. These areas have been selected as they are of current interest in the cleft palate intervention literature and the SSD literature as a whole. This critique informs the within-participant methodology used in this research. Each of these topics is discussed in the following subsections. The subsections begin with a general discussion of the topic under critique, followed by an appraisal of the seven studies accordingly.

Table 1

Overview of published experimental treatment studies using EPG with individuals with speech sound difficulties related to cleft palate

First author, year	No of participants	Age in years	Language	Lingual speech sound difficulties	Design	Intervention intensity / Dosage	Outcome: EPG	Outcome: Auditory perceptions	Outcome: Holistic	Outcome: Comment	Reliability	Results
Michi 1986	1	6	Japanese	Palatalisation or lateral mis-articulation	Within-subject before and after	/p/,/b/,/m/,/t//, /d/, /n/, /s/, / ʃ/, /t ʃ/, /dʒ/, /j/,/k/,/g/, adapted articulation therapy approach 49 x 1-hour sessions, weekly, over 12-months, therapy to discharge	Visual analysis	Judgement of nonsense syllables; ratings of articulation with conversational speech	-	No repeated baseline pre-treatment; use of SLT and naïve listeners to judge speech sound accuracy	Inter-rater agreement for phonetic transcript-ion on initial assessment only; not blind	"Almost" normal production of target sounds in conversation
Michi 1993	6	4-6	Japanese	Misarticulated /s/	Group comparison of three types of treatment	/s/, three types of therapy: no visual feedback, visual feedback using EPG, visual feedback using EPG and friction display, adapted articulation therapy approach 1-hour-session/week 8 sessions (2-months)	-	% /s/ correct in isolation	-	Therapy progress was assessed by progression through treatment levels	Inter-rater for judging treatment progress	Improved speech production at word level for all types of therapy, but more progress with visual feedback techniques
Whitehill 1996	1	18	Canton-ese	Posteriorly placed alveolars	Within-subject multiple baseline	/s/ and /t/, articulation therapy approach 23 x 1-hour sessions over 4-months, therapy to	Visual analysis of EPG frames pre and post, comparison with normal EPG frames	Phonetic transcription of single words and sample of conversational	-	No repeated baseline pre-treatment	Consensus for phonetic transcript-ion	Improved /s/ at word level, accurate /t/ at word level, /ts/ accurate 90% of time in

						discharge	% of lingual-palatal contact	speech; % CC in single words and conversational speech				conversation
Stokes 1996	2	5,7	Canton-ese	Misarticulated /s/ and /ts/	Within-subject multiple baseline	/s/, 1-hour/week, articulation therapy approach 7- weeks, therapy to discharge	Visual anaylsis of frame of maximum constriction;	% target accuracy isolation, word, phrase, story retell, conversation	-	No repeated baseline pre-treatment; maint. assessment carried out for one participant	Consensus, two raters	Generalisation to connected speech for one participant, use of speech targets in story retell for the second participant
Gibbon 2001	12	5-18	English	Compensat-ory misarticulat-ion	Randomised group study, cross over design	/t/ and /s/, four sessions (30-40 min) EPG-therapy vs articulation therapy, treatment in clinical setting	CoG at frame of maximum constriction; visual analysis; ratings of positive change in EPG patterns following therapy at three time-points	-	-	Two raters rated change in EPG patterns	-	75% of subjects had more normal tongue-palate contact patterns following EPG, 25% showed no change
Fujiwara 2007	5	8-13	Japanese	Palatalisation or lateral misarticulat-ion	Case series	Alveolars, monthly sessions, traditional articulation therapy, home training with PTU 30-minutes/day 10 – 15- months, treatment in clinical setting	CoG at frame of maximum constriction for /t/ words at eight time points during therapy; Visual analysis	-	-	General comments made re. changes to speech production	95% agreement for EPG annotation	Four participants showed perceptually acceptable realisation of all target sounds, one participant showed accurate production of

												targets
Lohmand-er 2010	1	11	Swedish	Posteriorly placed /s/ and /t/	Within-subject multiple baseline	/s/ and /t/, monthly sessions, articulation therapy, home training with PTU 10-minutes/day over 8-months Estimated 20 hours of training	CoG, AT, WT from frame of maximum contact for words and sentences; statistical analysis of change to CoG, AT, WT; Visual analysis	Phonetic transcription of standardised articulation test; Comparison of pre- and post-speech (words and sentences) by three naïve listeners, blind to time point	Judgement of speech intelligibil-ity by three naïve listeners , blind to time point using a four point ordinal scale developed for the study	Phonetic transcript-ion completed by one SLT	Intra and inter-rater agreement for the naïve listeners (>90% and 100%)	Both EPG and perceptual analysis showed improvement. No change to intelligibility of speech

Note: PTU = Portable training unit, CoG = Centre of gravity, AT= Alveolar total, WT = Whole total, SLT = Speech and language therapist

(adapted from Lohmander et al., 2010)

3.2.1 Review of EPG cleft studies: Intervention intensity/dosage

Dosage has been a topic of growing interest in the SSD literature in recent times since the total amount of therapy children need, to result in discharge with intelligible speech, is largely unknown. Sugden et al. (2018) define dosage as how much, and what kind of, practice is needed to cure SSD in a child. Warren et al. (2007) suggest a description of intervention dosage should include: dose frequency, total intervention duration, dose form, dose, and cumulative intervention intensity. Dose frequency relates to how often a child receives therapy (e.g. once per week for 60-minutes). Total intervention duration is how long intervention lasts (e.g. 6-weeks; 6-months). Dose form relates to what is conceptualised as the active ingredient of intervention, and relates to the theoretical rationale of the particular treatment approach (e.g. making a conceptual judgement of a target sound in a /k/ - /t/ minimal pair task, where the person has to point to all the words beginning with /k/). Dose relates to how many times that active ingredient or "teaching episode" (Warren et al., 2007, p 720) is delivered per session (e.g. child asked to make a conceptual judgement 20 times over the course of a 30-minute session). Cumulative intervention consists of the following formula: dose x dose frequency x total intervention duration. Sugden et al (2018) further categorises dose form as production dose (number of times the individual produces a speech target/s), perception dose (number of times the individual completes a listening task) and conceptual dose (number of times the individual carries out a conceptual task, such as making a phonemic judgement).

Information on dosage represents a relative strength of the studies reviewed in Table 1, compared with wider SSD intervention literature, where dosage information is often lacking (Sugden et al., 2018) . All studies reported on dose frequency (sessions occurred either weekly or monthly, and lasted from 30- to 60-minutes in duration). All reported on total duration (7-weeks to 15-months; four reported on therapy to discharge). All gave some information on dose form. Five studies employed an EPG therapy technique involving practice of speech sounds in a hierarchy of stages, i.e. isolation, syllable, and so on, in the manner of articulation therapy. The remaining two studies (Michi et al., 1986; Michi et al., 1993) described an adaption of articulation therapy, with the addition of extra levels of learning and some flexibility in applying the hierarchy of stages. As such, dose form for all studies involved production trials (as opposed to perceptual or conceptual trials). However, none of the studies provide information on the number of production trials per session, and none report on cumulative intervention.

3.2.2 Review of EPG cleft studies: Measurement of auditory perceptual and EPG outcomes

When evaluating the effectiveness of EPG therapy, Lee et al. (2009) suggest that both EPG analysis and perceptual speech analysis should be included. As discussed in 3.1.2, two types of EPG measurement are available: visual analysis of palatograms and computation of EPG indices. In terms of perceptual speech analysis, the most common method for reporting outcomes in SSD intervention research is percentage of consonants correct (%CC) and the related index of percentage of targets correct (%TC) (Cleland et al., 2019; Heng et al., 2016; Roxburgh, 2018). To calculate this speech measure, speech samples are phonetically transcribed by trained SLTs or phoneticians using symbols from the International Phonetic Alphabet (IPA). Transcription of cleft palate speech is typically narrow and includes use of diacritics developed specifically for transcribing cleft palate speech (Sell et al., 2009). From this transcription, each consonant is examined and identified as correct or incorrect. The percentage of consonants correct is the number of consonants correct divided by the total number of consonants. The percentage of targets correct is the number of targets correct divided by the total number of instances of the target sounds.

Since perceptual speech assessment is impressionistic and therefore susceptible to a number of biases (e.g. skill and consistency of the persons transcribing, quality of the speech sample etc.), a number of researchers (Dalston et al., 1988; Lohmander & Olsson, 2004; Sell, 2005; Sell & Pereira, 2016) advocate the following standards for perceptual speech assessment of cleft palate speech:

- The speech sample should be rated by more than one listener and inter- and intra-agreement/reliability measures reported;
- Listeners should make their perceptual assessment from high quality speech recordings, rather than live transcription;
- Listeners should undergo training prior to perceptual speech assessment;
- Speech samples should be standardised as much as possible (i.e. the same words/sentences used by all patients, elicited in the same way, and recorded in the same way);
- Transcription/analysis should be carried out by independent listeners who are blind to the test time point, thereby countering any potential bias regarding knowledge of intervention.

In terms of EPG analysis, all of the studies listed in Table 1, apart from Michi et al. (1993), provide pictures of palatograms for visual analysis by the reader. Four out of seven studies also report EPG indices. The two most recent studies (Lohmander et al., 2010; Fujiwara, 2007) used the WinEPG system (Articulate Instruments Ltd., 2010) and the frame of maximum contact to compute indices.

Two out of seven EPG intervention studies do not include perceptual speech outcomes in any detailed way (Gibbon et al., 2001; Fujiwara, 2007). Phonetic transcription was used for the other five studies. In addition, one study (Lohmander et al., 2010) included a comparative method for reporting speech accuracy: Pairs of words (from before and after treatment) were presented to listeners. Raters were asked to make a judgement of which word sounded more accurate. If no difference was perceived this was also reported. Only one study (Lohmander et al., 2010) meet the standards for recording and reporting perceptual speech outcomes, as listed above, and this represents a particular weakness with the reviewed studies.

3.2.3 Review of EPG cleft studies: Reliability

Researchers and clinicians reporting impressionistic perceptual speech ratings are obliged to show reproducibility of ratings, so that readers can judge results (Chapman et al., 2016). This requires a report of agreement and/or reliability scores. Agreement scores relate to the degree to which ratings are identical. These scores do not take account of chance agreement. Reliability scores identify the degree to which ratings are identical, but also take into account measurement errors such as chance agreement (de Vet et al., 2006). A number of cleft researchers (Dalston et al., 1988; Lohmander & Olsson, 2004; Sell, 2005) argue that both inter-rater (agreement between different raters, independently rating the same material) and intra-rater (agreement by a single rater, rating the same material on two separate occasions) agreement/reliability should be included when reporting intervention outcomes (Dalston et al., 1988; Lohmander & Olsson, 2004; Sell, 2005). High inter-rater agreement/reliability indicates different listeners are applying the same criterion when rating the material. High intra-rater agreement/reliability indicates the rating is reproducible under similar conditions. Levels of agreement/reliability are important for confidence in study results: High agreement may produce high confidence in results, while low agreement may produce uncertainty in results (see 8.1.1 for level of agreement benchmarks)

Reliability reporting was limited in the cleft EPG intervention studies included in this review and this represents a significant weakness with existing studies. Three out of seven studies reported on inter-rater agreement for perceptual speech assessment (Michi et al. 1986; Michi et al. 1993; Lohmander 2010). Only one study reported on intra-rater agreement for perceptual speech assessment (Lohmander et al., 2010).

3.2.4 Review of cleft studies: Holistic outcomes

In addition to evaluation of errors with consonant production, in recent times a number of authors have also argued strongly for greater use of more holistic outcomes when reporting cleft palate speech outcomes (Howard & Lohmander, 2011; Sell & Pereira, 2016). These authors suggest that in addition to degree of speech impairment (e.g. percentage of consonants correct), outcome indices should also include measures which assess an individual's functional speech performance, together with measures which consider the social consequences of the SSD. Function speech performance is typically measured by assessment of speech understandability, also known as speech comprehensibility (Sell & Pereira, 2016). Speech understandibility is defined as how well the individual can make themselves understood within a particular communicative context (Yorkson et al., 1996). Measures of the social consequences of SSD include patient-reported quality of life outcomes and measurement of speech acceptability. Speech acceptability is defined as the degree to which a person's speech draws attention to itself, outside of the content of the spoken message (Henningsson et al., 2008). As identified by Miller (2013), even when speech understandability is not particularly affected, identifiable differences with speech sound production may be associated with marked psychosocial consequences for the speaker.

Again, measurement of holistic outcomes is a significant weakness of existing cleft EPG intervention studies. Only one study (Lohmander et al. 2010) reported on changes to speech understandability following EPG, and that was for no change. No study has considered changes to quality of life and speech acceptability following intervention.

3.2.5 Review of cleft studies: Results

Following a review of the visual biofeedback intervention literature, Cleland and Preston (2021) suggest that biofeedback intervention studies carried out in the 1980s and 1990s may have been subject to publication bias, in that non-responders may have been under-reported. They point out that more recent visual biofeedback intervention studies show a greater number of non-responders (i.e. children not responding to biofeedback intervention), compared with earlier studies. However, these most recent studies largely report on ultrasound intervention, rather than EPG therapy.

Of the studies examined in this review, 4/7 occurred in 1980s and 1990s. All participants in all four studies showed a degree of improvement with perceptual speech assessment and EPG assessment. In the three more contemporary studies, all showed a degree of improvement with perceptual speech and/or EPG outcomes, apart from three of the 12 participants from the Gibbon et al. study (2001) where no change occurred with EPG analysis. For those participants showing improvement in the seven studies (n = 28), this improvement consisted of: increased accuracy of single word naming; more exact contacts on EPG; and/or improved accuracy with connected speech. Generalisation to everyday speech was reported in 2/28 participants. Normal production occurred in one participant (Stokes et al., 1995), while the other achieved "almost" normal production (Michi et al., 1986).

3.2.6 Section summary and conclusions

The current evidence base for EPG intervention with cleft palate speech consists of within-participant case studies and two small group studies. A review of the literature in this section identified some methodological shortcomings of existing studies, in particular, lack of reliability measures and absence of more holistic outcomes. Despite these weaknesses, studies report positive speech improvement for many patients receiving this type of instrumental treatment. However, not all patients respond to EPG therapy, and earlier studies may have under-reported non-responders. In addition, fewer studies have examined and reported on transfer to, and maintenance of, the treatment effect to everyday speech. Clinical experience indicates that generalisation and maintenance can be difficult for many patients. Consequently, it appears further development of the EPG therapy paradigm is needed.

3.3 Theoretical basis of current EPG therapy

Crucial to any therapy paradigm, including EPG, is the theoretical basis of the treatment technique (Kamhi, 1992). Teaching episodes (i.e. dose form) and frequency of these teaching episodes stem directly from whatever theory underpins the treatment. In addition, should a client not respond in the predicted way, a robust theory can direct the therapist to other teaching episodes. Likewise, a client's response to therapy can contribute to further theoretical development, either by providing support for the theory or by identifying apparent theoretical shortcomings. If the aims of therapy are to improve or normalise speech sound production and to integrate this change into everyday talking in a lasting way, it follows that key to any therapy paradigm is a theoretical basis which explains how typical phonology develops over time, together with an explanation of how atypical phonological development may arise. Snowling and Hulme (2011) describe such explanatory theories as "causal" models, and argue for the importance of such models in the remediation of speech, language and reading difficulties. This section will consider the existing theoretical basis of EPG therapy. In particular, facilitation of generalisation and maintenance within these existing theories will be considered, as this has been identified as a problem with EPG therapy for persistent cleft palate speech.

3.3.1 Traditional articulation therapy

As discussed in section 3.1.4, to date, much of the EPG therapy described in the past has been embedded in articulation therapy. Articulation therapy, as described by Van Riper (Van Riper, 1963, 1978; Van Riper & Emerick, 1990) follows the following process:

1. Sensory- perceptual "ear" training, where the patient learns to identify a target sound perceptually in the speech of others and then identify their own inaccurate production.
2. Teaching production of the new sound in isolation.
3. Teaching production of the new sound in nonsense syllables, including consonant vowel (CV), vowel consonant (VC) and consonant vowel consonant (CVC) combinations.
4. Teaching production at word level. Van Riper suggests that initially the therapist should focus on a small number of key words. These words should include the target speech sound in word initial, medial and word final positions.
5. Teaching production of the target sound in sentences.

In each of the levels (sound, syllable, word, and sentence) drill production practice (i.e. producing the target sound many times) is a crucial component of this treatment approach. Positive reinforcement of correct productions is also important with this technique. Van Riper suggested one or two speech sounds should be worked on at a time. The visual feedback provided by EPG is normally provided in the early stages of articulation therapy. In relation to generalisation and maintenance, Van Riper (1963) describes three methods for facilitating generalisation and maintenance: Practice in non-therapy settings, such as school and home; use of speech for different communicative functions (e.g. to instruct, to ask a question etc.); focus on development of proprioceptive feedback (defined as perception of speech contacts, movements and postures).

3.3.1.1 Theoretical basis of traditional articulation therapy

Van Riper (1963) does not specifically state the theory/concepts underpinning his articulation therapy. Rather, his therapy approach appears to have derived largely from his practical experience with treating SSD. However, some principles can be gleaned from his texts, and reflect dominant behavioural and linguistic theory prevalent at the time Van Riper was developing his technique in the mid-twentieth century:

- Behavioural principles of operant learning (Skinner, 1938): behaviours are elicited via an antecedent stimulus (e.g. therapist to the child: "Name this picture") and the resulting behaviour (e.g. child: "sun") is then shaped (increased/ strengthened, decreased or eliminated) by consequences such as positive reinforcement or punishment (e.g. therapist: "Yes, that is a sun, well done you used a fantastic "s" sound at the beginning of the word").
- Focus on individual speech sounds, and the idea that words are made up of a series of individual sounds.
- Emphasis on the motor movements of sounds and the importance of drill practice of new movements: Van Riper writes "The changes which must occur must be new patterns of muscular movement which produces new sounds" (Van Riper, 1963 p 245)
- Metalinguistic awareness: Although not labelled by Van Riper as metalinguistic awareness, a strong theme running throughout the treatment technique is having the patient to think about and reflect on their own speech and the speech of others. In particular, segmentation of syllables or words into component sounds is a key component of both "ear" and speech production training.

While these principles give the clinician clear direction on ways to teach specific speech behaviours, these principles provide little direction for how to achieve transfer and retention of new speech behaviours should Van Riper's general techniques for generalisation and maintenance, as described above, prove less successful. In discussing traditional articulation approaches to treatment, Fey (1992) also observes: "generally there are no a priori reasons to predict any system-wide generalisations patterns" (p 277).

More recently, articulation therapy has been reinterpreted and developed using motor learning theory (Preston & Leece, 2021).

3.3.2 Motor learning theory-based speech intervention

Motor learning theories (e.g. Adams, 1971; Schmidt, 1975) relate to motor skills in humans in general (i.e. not just speech) and consider how simple and complex motor skills are learnt. Two theories of motor learning commonly discussed in the motor learning literature include Adam's closed loop theory (1971) and Schmidt's Schema theory (1975). In particular, Schema theory has been used as a theoretical rationale for treatment of SSD involving difficulties with the physical production of speech sounds (Baker & McLeod, 2011).

3.3.2.1 Schmidt's Schema Theory

Schmidt's schema theory (Schmidt, 1975; Schmidt & Lee, 2005) suggests that motor movement arises from generalised motor programmes (GMP). GMPs are abstract frameworks, stored in memory, that capture the invariant aspects of a particular set of motor movements (e.g. a golf swing). GMP are adapted to meet specific task demands, including novel movements (e.g. getting a gold ball into a hole from a particular distance) through recall and recognition schema. These schemas may be conceived as "rules" which operate to provide detailed instructions to the musculature (Schmidt, 2003). These schemas are refined by a positive and negative feedback system whereby errors in particular are used to develop and improve the schemas. This updating occurs through information on the outcome of the movement, held in short term memory (Schmidt, 1975). This theory suggests that learning is enhanced by variable practice since this leads to schemas than are more comprehensive and stronger.

3.3.2.2 Principles of motor learning in relation to the treatment of speech sound disorders

Drawing on schema theory, Maas et al. (2008) detail what they consider to be key principles of motor learning in relation to the treatment of SSD:

- Pre-practice: Before new motor learning can occur, the learner needs to meet three requirements. Firstly, they must be motivated to learn. Secondly, they must have good understanding of the task at hand. Finally, they need to be stimulable for accepted responses.
- Repetitive practice: With regard to learning motor skills, in general, a large amount of practice is important.
- Feedback: Augmented feedback (i.e. feedback in addition to the individuals own intrinsic feedback systems such as tactile and proprioceptive feedback) facilitates learning. Two types of augmented feedback are described in the motor learning literature: knowledge of results (KR) and knowledge of performance (KP). In the context of speech production, knowledge of results relates to feedback on the auditory outcome of the speech production task (e.g. "Yes, you said that word correctly"), while knowledge of performance relates to specific information relating to how the motor speech task has been executed (e.g. feedback on placement of the lips to achieve a bilabial speech sound).

Generally, these principles are adopted by other SSD intervention researchers using motor learning theory (Cleland & Preston, 2021; Preston & Leece, 2021).

3.3.2.3 Facilitation of generalisation and maintenance

According to motor learning theory, key to transfer and retention of motor skills is type of practice and type of feedback used (Schmidt & Lee, 2005). In the motor learning literature, "generalisation" is defined more narrowly as transfer of learning from treated to untreated words in the clinic setting (Cleland & Preston, 2021).

Practice: Traditional articulation therapy and conventional EPG therapy involves massed (less time between sessions), blocked (predicted order of practice) constant (same speech skills in same way in same context) practice. While schema theory predicts constant practice may facilitate GMP, according to this theory variable practice best promotes transfer and retention of motor skills. Variable practice includes, for example: practice at sentence level, then word level, i.e. not in a hierarchical way; practice in different word positions; practice at different times during a week. Variable practice leads to schemas that are more comprehensive and stronger and therefore better able to inform novel motor movements.

Feedback: Feedback is intrinsic or augmented. Non-speech motor learning studies suggest that provision of knowledge of performance (i.e. augmented feedback) is useful when a motor task is novel or unclear (Newell et al., 1990). However, as the person's skills level increases, knowledge of performance feedback can be detrimental as it blocks intrinsic feedback systems (Hodges & Franks, 2001). Non-speech motor learning studies also suggest that reduced frequency of augmented feedback (knowledge of results and knowledge of performance) can enhance motor learning, as the individual is forced to develop intrinsic feedback mechanisms (Winstein & Schmidt, 1990). Likewise a small delay of a few seconds before giving augmented feedback may assist self-regulatory feedback systems (Swinnen et al., 1990). A delay allows the individual to evaluate their motor behaviour, and then compare their evaluation with extrinsic feedback.

Within schema theory, errors are important in the development of recall and recognition schema (Schmidt, 1995). Therefore, performance which includes errors will enhance transfer and retention of learning.

3.3.2.4 Motor learning theory and EPG

As stated, Gibbon and Wood (2010) and Cleland and Preston (2021) identify motor learning theory as the predominant theory underpinning their enactment of EPG therapy. More recently motor learning theory has been used as the theoretical underpinning for articulation therapy in general (Preston & Leece, 2021). Gibbon and Wood (2010) do not identify which specific motor theory/theories direct their EPG therapy. However, these authors do list three principles of motor learning which they consider integral in the EPG therapy technique: repetitive and intensive practice; grading of motor complexity; feedback on knowledge of performance. According to Gibbon and Wood (2010), intensive practice should be carried out within EPG therapy sessions. In addition, portable EPG training units permit continued practice of speech targets outside of EPG sessions. Their therapy does not contain variable, random practice. Rather, motor complexity gradually increases as therapy progresses and moves from production of single sounds, to production of syllables, words, phrases and sentences, to finally practice of speech in connected sentences, in the manner of articulation therapy. Feedback on performance is facilitated by way of the visual feedback of tongue-palate contact using EPG pictures. The visual feedback provided by EPG is gradually withdrawn during therapy, so that be the end of therapy the patient is able to rely on their own internal monitoring/feedback mechanisms.

Cleland and Preston (2021) identify schemata theory as the dominant motor learning theory underpinning for biofeedback interventions, including EPG. In particular they focus on the concept of knowledge of performance and the benefits visualisation can provide to increasing the patient's knowledge of their own performance during the acquisition of a speech sound. A key component of their enactment of biofeedback therapy is dosage, i.e. the number of production trials per session. These authors suggest a large number of production trials are important for motor speech acquisition and learning. They also advocate the requirement of some error in learning, consistent with schema theory, i.e. they state that a patient does not need to achieve 100% accuracy with production before before moving on to more difficult tasks. However, Cleland and Preston do not include variable, random practice. These researchers suggest therapy should proceed in a hierarchy of articulatory difficulty, as with traditional articulation therapy in the acquisition phase. They suggest that visualisation may be useful in the early stages of acquiring a speech sound, but is of less use in later stages when the focus is on transfer and retention of learning.

In terms of facilitating of generalisation and maintenance of target speech sounds using schemata theory, potential areas to explore include:

- Use of variable, random practice (e.g. Skelton, 2004; Skelton & Richard, 2016);
- Manipulation of feedback, e.g. reducing frequency of feedback, delay before giving feedback.

3.3.2.5 Limitations of Schema Theory

As it exists, Schema theory is very broad and it does not provide specific details on the structures and mechanisms for motor speech learning. Unanswered questions include, for example, what constitutes GMP and schemata? Do GMP involve words, parts of words, or individual phonemes? If GMP consist of words, are lexical stress patterns a feature of GMP, or are they controlled by schemata? How do GMP come about initially? Are these learnt, or are they in part innate? What are the causes of SSDs? Mass et al. (2008) suggests SSDs are the result of impairment or damage to GMP and schemata. If so, how did this impairment occur and what should be done to address this impairment/damage? It would appear more specific details relating to speech are needed, so that a more robust "causal" model can be developed to underpin intervention.

This theory also appears at odds with some of what we know about children developing typical phonology. Schmidt's schema theory suggests recall and recognition schemas are refined by a positive and negative feedback system whereby, in particular, errors are used to develop and improve schemas (Schmidt, 1975). This process assumes consistent, continuous improvement with development of speech sound production. However, this assumption is not met with typical children learning first phonology. Rather, typically developing children's early speech is characterised by intra-word variability (i.e. the same word is produced differently by individual children at the same point in time) and by the presence of regressive and progressive idioms (i.e. words are more or less advanced than what would be expected considering the child's overall speech sound system) (Ferguson & Farwell, 1975; Fletcher et al., 2004; Schwartz & Leonard, 1982; Vihman, 1996).

3.4 Section summary and conclusions

EPG is a type of visual feedback therapy used to assess and treat individuals with SSD involving misarticulation of lingual speech sounds. The patient is fitted with a custom made "artificial palate" which fits snuggly to the roof of the mouth. This plate contains sensors that detect tongue-palate contact. As the patient speaks, this dynamic contact is displayed in real time on a screen and this visual feedback is used to facilitate correct production of problem speech sounds. EPG is typically used with school-aged children with residual lingual speech errors which have not responded to standard speech therapy. EPG therapy has also been used with adults. Since individuals with the cleft condition often have long-standing difficulty with articulation of lingual speech sounds, EPG therapy is particularly suited to this patient group.

The current evidence base for this type of speech sound intervention with individuals with SSD secondary to cleft palate consists of within-participant case studies and two small group studies. A review of the literature in this chapter identified some methodological shortcomings, in particular, lack of reliability measures and absence of more holistic outcomes. Despite these shortcomings, studies indicate positive speech improvement for many patients receiving this type of instrumental treatment. However, not all patients respond to EPG therapy, and non-responders may have been under-reported in the past. In addition, fewer studies have examined and reported on transfer to and maintenance of the treatment effect to everyday speech. Clinical experience indicates that generalisation and maintenance can be more difficult for many patients.

Crucial to any therapy paradigm is the theoretical basis of the treatment technique. Teaching episodes and frequency of these teaching episodes stem directly from whatever theory underpins the treatment. Initially EPG therapy combined traditional articulation therapy with the visual feedback provided by EPG. However, traditional articulation therapy has a less clear theoretical basis. Consequently, identification of teaching episodes to facilitate generalisation and maintenance is more problematic. More recently, motor learning theory has been used as the theoretical rational for EPG therapy and application of these theories has refined the EPG therapeutic technique further. However, transfer and maintenance of new articulations remains problematic for some patients receiving more contemporary visual feedback therapy, including EPG. Schema theory suggests use of variable practice schedules may be important in the transfer and retention of new speech motor skills, and this may be an avenue for further development of biofeedback techniques. Manipulation of feedback schedules may be a further area for development. Some early work with manipulation of practice and feedback schedules has already been done (e.g. Hitchcock & McAllister Byun, 2015; Preston et al., 2017; Skelton, 2004; Skelton & Richard, 2016). Another avenue for development, which is undertaken in this research, is consideration of, and use of, alternative theory to underpin EPG therapy.

4 USAGE-BASED PHONOLOGY

As with most areas of speech and language therapy clinical practice, understanding of, and intervention for, SSD has been heavily influenced by rule-based, generative linguistic theory (Hatchard, 2015; McLeod & Baker, 2017). Generative linguistic theory first arose in the 1950s (Chomsky, 1957). According to this linguistic theory, speakers are perceived to have abstract linguistic knowledge that is, at least in part, innate. A series of rules operate to convert this abstract knowledge to surface forms, such as spoken speech sounds, words and sentences (Chomsky, 1957, 1968, 1972). Generative theory was originally developed to provide economical, elegant descriptions of speech and language in the typical adult (Chomsky, 1957, 1968), rather than to provide a causal explanation of speech and language development over time, or an explanation of atypical speech and language. Subsequently, the theory has been widely applied in understanding of and intervention for child and adult speech and language disorders (e.g. Ball & Kent, 1997; Williams et al., 2021), often supplemented by additional theories (e.g. Williams et al., 2021).

The influence of generative theory on SSD for children emerged in the 1970s (Ingram, 1976). Prior to this time intervention for SSD largely consisted on articulation therapy, as developed by Van Riper (1939 onwards), and as described in Chapter Three of this thesis. At this time (mid-19th century), all SSD were attributed to difficulty with the physical production of speech. However, the 1970s and 1980s saw a change in thinking. This change occurred with the application of linguistic theory to SSD intervention. In particular generative phonology (Chomsky, 1968), the related theory of natural phonology (Stampe, 1979), and distinctive feature theory (Jakobson, 1972) was influential in understanding of and treatment for SSD at this time (Ingram, 1976). Based on the ideas of generative theory, many academics and clinicians began to make a distinction between a). cognitive-linguistic storage and processing and b). the motor execution of speech. Cognitive-linguistic storage and processing is perceived as the storage and access of lexical representations of words, including phonological, semantic, syntactic, orthographic information (McLeod & Baker, 2017). It was argued that the source of SSD for many children is at this abstract cognitive-linguistic level. For example, if a 4 year old child is substituting [t] and [d] for /k/ and /g/ in words, but can produce [k] and [g] in isolation, the child's difficulty with /k/ and /g/ in words is attributed to difficulty at the cognitive-linguistic level, rather than a difficulty with the physical production of these speech sounds. This shift in thinking lead to the emergence of "phonological" intervention techniques (e.g. Blache et al., 1981; Hodson, 1978; Weiner, 1981; Weiner, 1984). Phonological therapy seeks to increase the child's awareness or knowledge of the structure of their ambient speech sound system, and is seen as appropriate for children whose speech difficulties arise at the cognitive-linguistic level. Depending on the linguistic model used by the particular phonological therapy, this may include increased awareness of "phonological rules" (Weiner, 1981, 1984), or natural sound classes/distinctive features (Blache et al., 1981). Baker (2006) surmises this shift in thinking and practice may have come about because of a change in speech and language clinical caseloads in the 1970s. In addition to the treatment of school-aged children with SSD, referral of pre-school children with complex SSD became increasingly commonplace. Baker (2006) suggests traditional articulation therapy was not an efficient therapy approach with this younger group, though she does not consider the reason/s for this in her paper. Aside from theoretical considerations (i.e. difficulty at the cognitive-linguistic level versus difficulty with the physical production of these speech sounds), reported inefficiency with the traditional approach may be linked to: a). younger children having more difficulty in attending to formal drill work, and/or; b). the need for a degree of metalinguistic awareness with this type of therapy that younger children may not possess.

Many authors view the emergence of "phonological" interventions as a significant paradigm shift (Baker, 2006; Ball, 2016; McLeod & Baker, 2017), and this motor execution/cognitive-linguistic distinction continues to the present day. Use of motor-based speech interventions, such as traditional articulation therapy, is seen as appropriate for SSDs arising from difficulty with the motor execution of speech, while difficulties with abstract phonological knowledge/ representations require phonological intervention. Part of the SLT's task in assessing a child with SSDs is to diagnose the basis of the child's SSD, and then to go onto select an appropriate treatment technique, i.e. articulation or phonological (McLeod & Baker, 2017). More recently psycholinguistic interventions have become prevalent in the treatment of SSD. Most psycholinguistic models used with SSD hold onto the idea of levels of processing, with a generative-type distinction between abstract cognitive-linguistic representations and the motor execution of speech. In terms of cleft palate speech, SSDs seen with this client group are typically attributed to difficulties with the motor execution of speech. However, as identified in Chapter One of this thesis, cleft palate speech is also associated with speech behaviour that can be better described using phonological patterns, such as backing of all anterior sounds to more posterior placement, and use of non-oral sounds to replace all sibilant sounds (Harding & Grunwell, 1996). As discussed previously, visual biofeedback intervention techniques are typically seen as appropriate for use with SSD due to difficulty with the physical production of speech, though recently ultrasound has also been used with children surmised to have developmental phonological difficulties (Cleland et al., 2019).

In recent times, generative, rule-based linguistic theory has been challenged in a number of areas, such as child phonology, child language and sociophonetics (Goldinger, 1998; Vihman & Croft, 2007; Vihman & Keren-Portnoy, 2013). Most of this criticism relates to the inability of generative theory to account for variability in the speech production of individual children, and in groups of people over time. Further, generative theory has more difficulty accounting for across-languages variances. Such disparities have led to development of alternative linguistic theory. Usage-based linguistic theory is one such alternative that has been growing in importance in recent times (Bybee, 2001, 2010; Goldinger, 1998; Menn et al., 2013; Pierrehumbert, 2001; Vihman & Croft, 2007; Vihman & Keren-Portnoy, 2013).

Section one of this chapter describes usage-based phonology, and will discuss key usage-based models proposed by leading academics in this area. This chapter will then go on to consider the implications of this theory to intervention with cleft palate speech. In particular, direction for generalisation and maintenance of speech learning is considered, since transfer and retention of learning has proven particularly challenging in the past. Further, since many individuals with persistent cleft palate speech are older, direction for intervention with school-aged children and adults is sought. The chapter concludes with the aim of the study and research questions.

4.1 Usage-based phonology

The term "usage-based" was first used by Langacker (1987) in his book "Foundations of Grammar" and is used to describe an emergent model of language where linguistic processing, acquisition and change comes about through language use, and the subsequent cognitive generalisations/associations made following these usage events. This emergent theory of linguistic development, also known as "cognitive phonology" (Bybee, 2010; Vogel Sosa & Bybee, 2008), subsequently has been applied to phonology, i.e. the sub-discipline of linguistics concerned with the sounds of a language (Bybee, 2001). Usage-based phonology differs to generative rule, and related constraint-based phonological theories, (Chomsky, 1972; Prince & Smolensky, 1997), in a number of ways:

- Generative theory assumes humans are born with a degree of innate, abstract, linguistic knowledge. In contrast, in usage-based theory does not view linguistic knowledge as innate or abstract – all knowledge and abilities arise from communicative experiences, and the cognitive associations made following these experiences.

- Generative theory conceptualises different levels of speech processing and makes a distinction between phonetics and phonology (where phonology is defined more narrowly as the systematic organisation of speech sounds in languages). In contrast, usage-based models are typically flat and de-emphasise the distinction between phonetics and phonology. Rather, the term "phonology" is used more broadly and encompasses all processing and structures relating to the production and perception of speech sounds.

- Generative theories represent top-down models of language processing – abstract cognitive-linguistic representations are subjected to rules (or constraints) to bring about surface forms. In contrast, usage-based models are typically bottom up models – experiences with speaking and listening result in speech learning and organisation.

Bybee (2001) was among the first to consider the development of the speech sounds of a language from a usage-based theoretical perspective. Bybee's seminal work, first detailed in her book "Phonology and Language Use" is largely motivated by explanation of the mechanisms for typical adult speech change over time. Bybee's work, and the work of others theorists adopting a usage-based stance (e.g. Goldinger, 1998; Johnson, 1997; Pierrehumbert, 2001), has become increasingly influential in the field of sociophonetics (Docherty & Khattab, 2008; Docherty & Foulkes, 2013; Docherty et al., 2013). More recently, other researchers and theorists have begun to apply this theory to typical child phonology (Edwards et al., 2004; Menn et al., 2013; Munson et al., 2005; Pierrehumbert, 2003; Vihman & Keren-Portnoy, 2013), with Vihman (Vihman & Croft, 2007; Vihman & Keren-Portnoy, 2013) and Menn et al. (2013) in particular producing significant work. To date, this theory has had very little application to the understanding and treatment of SSD. Likewise, it has had little application to other areas of communication impairment, though some pioneering work (Hatchard, 2015) has been done in the area of aphasia.

4.1.1 Bybee's usage-based phonology

The basic assumption of Bybee's usage based phonology theory (and wider usage-based theory) is that all human learning and behaviour, including language and speech, is governed by the same core principles and processes (Bybee, 2001, 2010). Key principles include our human anatomy, in particular our large memory capacity. Key processes include, for example, our ability to attend, to imitate, to carry out fine motor movements, and to categorise and make inferences. Central to most usage-based phonology models is exemplar theory. Arising from the field of general psychology, exemplar theory considers how humans categorise objects and ideas, and proposes that individuals make category judgements by comparing new experiences with experiences already stored in memory. Similar memory traces are grouped together and a memory trace is more central or more marginal depending on the number and nature of shared features. Frequency of use will impact on the core of a category. The more frequent a memory trace, the more central that trace will become. However, the core of a category can shift, depending on the experience of the individual.

Extrapolating from the above assumptions, the starting point of Bybee's (2001) usage-based phonology is a human being's advanced vocal tract with its capacity for sound and modification of sound. Bybee suggests that each instance of speech/vocal tract use leaves a trace in memory. Gradually, over time, utilising our large memory capacity and our ability to compare and infer, these memory traces are grouped together. For example, a 'word' will consist of all the motor/phonetic memories associated with the word; the core memory trace (i.e. the most frequently produced articulatory gesture) is the "exemplar". These phonetic memories will also be associated with "the meanings of the word and the contexts in which it has been used, which themselves form an exemplar cluster" (Bybee, 2010, p. 19). Therefore, according to Bybee's theory, it is an individual's speech use that generates their phonological system. Further, since speech use typically occurs in communicative settings, phonology is seen as being intrinsically linked to language use within communicative contexts. In addition, speech experiences can produce changes to an individual's phonological system over the lifespan.

An important tenet of general exemplar theory is frequency of use (Bybee, 2001). Bybee applies this principle in usage-based phonology: a neuromotor routine involving the vocal tract acquires strength the more it is used by the individual. Bybee (2001) writes about two types of frequency: token frequency and type frequency. Token frequency is how often a unit occurs. A unit is usually a word, but can be a part of a word or a stretch of words. Type frequency relates to how often a pattern occurs. For example, a young child developing first language may have a preferred pattern, such a consonant-vowel reduplicated pattern. Another example of a type frequency is use of a particular neuromotor routine in word initial position. However, it should be noted that type frequencies are not innate/universal rules – they have "no existence independent of the lexical units from which they emerge" (Bybee, 2001, p. 27).

Another important aspect of general exemplar theory is flexibility of categories (Bybee, 2001, 2010). As detailed above, similar memory traces are grouped together, and the core of a category can shift depending on the experience of an individual. Flexibility of categorisation provides for explanations for variability and atypical instances within categories. This flexibility is seen as a particular strength of exemplar theory. Bybee uses this flexibility of categories to explain how phonology can change gradually over time with instances of speech use. Data cited and reported by Bybee (2001) mostly relates to speech change in adults over time, and includes, for example, vowel shifts in San Francisco English, vowel reduction in Dutch, and t/d deletion in American English. Bybee labels this gradual change in speech as "gradience", (Bybee, 2010, p. 2).

As outlined, fundamental in usage-based theory is use of neuromotor routines involving the vocal tract (Bybee, 2001). Bybee suggests this neuromotor activity is subject to the tendencies that affect all human sequenced neuromotor activity, such as overlapping and reduction. Articulatory gestures will be influenced by surrounding gestures both within and across words. For example, in the word "stew" production of "s" will be accompanied by lip rounding in anticipation of the vowel "ew", and in the utterance "miss you", the gesture for /s/ will be influenced by the /j/, resulting in palatalisation of the /s/ with rapid speech. This omnipresent overlapping of articulatory movements is well described in the literature and is typically referred to as coarticulation (Hardcastle & Tjaden, 2008). Patterns such as /s/ to [ʃ] in the "miss you" example above, occur systematically across speakers and are known as connected speech patterns (Ellis & Hardcastle, 2002). Bybee suggests this overlapping and reduction is one mechanism to explain speech change in adults over time.

In Bybee's usage-based phonology speech production is presented as the key generator of phonological development and change. In her writings, Bybee has little to say in relation to the role of perception in phonological development, and this represents a significant gap in the model. However, Bybee suggests that any change in a neuromotor activity involving the vocal tract will produce an acoustic-perceptual change. Bybee further suggests that motor-articulatory representations and acoustic representations must be closely linked neurologically, however, she does not go on to theorise the nature of this link and its neural status.

4.1.2 Pierrehumbert's model

In comparison to Bybee's model, Pierrehumbert (2001) details a usage-based inspired model of phonology where speech perception is the primary generator of phonological learning/change. Pierrehumbert's model draws together exemplar learning theory with the mathematical constructs of computational linguistics. In this model, on-going acoustic experiences are laid down in the brain producing a "quantitative map". Pierrehumbert proposes that this phonetic space is categorised at a low level, and that this categorisation arises from frequency distributions over exemplars. Regarding production, Pierrehumbert writes:

> In production a label is first selected. A random sampling of the exemplar distribution is taken for that label. The neighbourhood of the selected exemplar is activated and the average properties of this neighbourhood constitute the production goal. The production goal is executed with noise corresponding to noise in the planning and execution of motor gestures (Pierrehumbert, 2003, p. 129).

In later writings (Pierrehumbert, 2006) Pierrehumbert acknowledges evidence pointing to dissociations between production and perception (Labov, 1994; Menn & Matthei, 1992; 2006). She therefore proposes separate perceptions and production systems, and suggests that these perception and production modules will carry their own frequency information. However, she does not detail how a separate production system might develop and function.

Bybee and Pierrehumbert's usage-based models have become increasingly influential in the field of sociophonetics. These models have been used by authors to account for findings of sociophonetic change (Docherty & Khattab, 2008; Rutter, 2014), and several journals have produced special issues on the contribution of usage-based linguistic theory to sociophonetics (Hinskens et al., 2014; Jannedy & Hay, 2006). However, Bybee and Pierrhumbert's models are somewhat deficient from a child perspective due to their limited attention to, and lack of empirical data relating to, child phonology. More recently, some researchers have begun to consider usage-based phonology with regard to typical children developing first phonology. Vihman, Menn and colleagues (Menn et al., 2013; Vihman & Croft, 2007; Vihman, M & Keren-Portnoy, 2013) in particular are at the fore-front of this work.

4.1.3 Vihman and Croft's "radical" template phonology

Vihman and Croft's radical template phonology focuses primarily on the first word period of a child's speech development (Vihman & Croft, 2007). The authors describe the development of early word forms/templates during this period which they argue emerge from infant's pre-word vocalisations, and which form the foundation of the child's early phonology. The term "templates" can be viewed as analogous to exemplars.

In the radical template model, a child's earliest word forms, or templates, are the result of the matching of the child's individual vocal patterns - produced in babble - to the child's input or perceptual representations of the word. As well as this matching/selection process, new templates develop when first words/templates are adapted in some way. Adapted templates are necessitated when the child's input representations of words are less similar to the child's existing output templates. In these circumstances, Vihman and Croft suggest the child is required to modify or adapt their output more radically to arrive at a template which better conforms (though not necessarily exactly) to the input pattern. As such, these authors view child phonology as emergent, rather than innate or universal, as it arises directly from the child's first word usage.

Vihman and Croft suggest templates are not lasting elements in a child's phonological system. Rather templates typically dominate around the middle to end point of the single word period, but subsequently fade as the child progressively masters more adult like speech sequences. However, these authors do not go on to describe how children's speech progresses past this first word stage.

4.1.4 Menn's linked-attractor model of phonological development

Menn et al.'s (2013) linked-attractor model of phonological development builds on from Vihman and Crofts (2007) radical template model and considers speech development beyond the first word stage. The linked attractor model contains three main elements: (1) production/output templates, (2) perceptual/input templates, and (3) mappings between input and output (whole word and part of word). Using Vihman and Croft's terminology, each of these three elements are termed "attractors", as they attract or direct the individual's production, perception and mapping between production and perception in some way. Menn et al. suggest that these three elements are built up progressively by experience with hearing, understanding and speaking. Attractor strength varies along a gradient from minimal to strongly entrenched. Menn and colleagues view speech as a redundant system, developed through millions of neural impulses/experiences relating to speech, and the associations between these experiences. Thus, these authors present a usage-based connectionist (as opposed to a box and arrow, model, e.g. Stackhouse and Wells, 1997) psycholinguistic model of phonology where the three elements of the model – output templates, input templates, mappings between input and output - exist in networks of interconnected brain cells.

As with Bybee's model, frequency of use is central in Menn et al.'s phonological model. Frequently activated items will become increasingly entrenched. However, while frequency is central, the authors suggest it is not the only factor contributing to template/exemplar strength. Other factors coming into play include, for example: the articulatory demands of a particular sound or sound sequence (e.g. some sounds/sound sequences are easier to produce than others); how easy a segment is to hear (e.g. unstressed syllables will be more difficult to perceive); and the emotional weight of a word/token (e.g. some words will have more emotional weight than others). Menn et al. suggest highly entrenched templates/ mappings will exert a strong "pull" and this will attract or direct the individual's production, perception and mapping between the two in some way. Thus, templates and mappings are conceptualised as "attractors". For example, in the first word stage, Menn and colleagues hypothesize that phonological idioms (i.e. words that are more or less advanced than what would be expected considering the child's overall phonological system) are infant words with high output frequency. As a result, they exert a strong pull and the child may continue to use these idioms even though they may not match the infant's input representations. Menn et al.'s model suggests the child will continue to use these idioms until the point where there is a greater pull from another attractor.

Menn et al. suggest an infant's first perceptual templates come about through their exposure to both the speech of others and the sound of their own babbled output. Neural links begin to form between clearly recurring situations and the words used in those situations, especially in situations arousing strong emotions. Menn et al. suggest during the late babble period, as the infant begins to produce more variegated, speech-like babble, some of the speech sounds the child produces will stimulate their memory of words stored in their auditory memory. This realisation will be reinforced if, when the child attempts production of a word, their parent or caregiver responds in a meaningful way. This marks the beginning of Vihman and Croft's template/first word period of development. Each attempt at producing a word will strengthen the association or mapping between the input and output templates, together with other usage information, such as referential meaning, emotional colouring, and so on.

How then, does a child move beyond this whole word stage, onto more systematic correspondences between child and adult forms? Menn et al.'s answer draws further on connectionist/network models of neural functioning. Connectionist/network models suggest that activation of any particular item spreads collaterally to items that are similar to it in some way (Sporns, 2011). Further, network models assume that simultaneously aroused items develop a coherent sub-network, that is, they develop representations of their own (Sporns, 2011). Menn et al. thus suggest that "each time a child says a word, words that have similar sounds and/or articulations are aroused to some extent. Because of this simultaneous arousal of similar forms, cross-representation will form between similar items" (p. 478). For example, each time a word containing a particular chunk of sounds is produced or perceived, a neural link will be made with templates of word/s containing similar chunks of sounds. Over time, with each instance of use, a stronger and stronger sub-network will develop, resulting in the gradual development of representations for this chunk of sounds. Other sub-network representations may include, for example, a particular intonation pattern, a certain syllable shape, specific articulatory gestures in word initial position, and so on. These representations are then available for the formation of "new" words/tokens. In this way, it can be seen the development of this sub-network is not necessarily at a conscious level. Menn et al.'s concept of a sub-network of representations is analogous to Bybee's "type frequencies".

Initially, parts of words/sounds will be tightly linked to whole words/tokens. However, as a child's vocabulary grows, Menn et al. suggest components become less tightly linked because each component recurs in so many words. As with input and output templates, Menn and colleagues suggest sub-network representations gradually build up attractor strength and become available/exert influence in the development of new words. Menn et al. suggest some sub-network representations may include representations that can be described as "phonological rules", such as input template containing /k/ being mapped to [t] in output templates, or /s/ being mapped to [t] in output. Menn et al. suggest that particular mappings, such as possible input of /k/ to output of [t] (i.e. fronting pattern) or /s/ to [t] (i.e. stopping pattern) can become attractors to new word forms. Menn et al. speculate the origin of some of these "rules" may lie with the poor motor control of unskilled speakers so that production frequently veers outside the appropriate articulation for that language. The authors go onto suggest these random shots may be pulled towards some other attractor off to the side.

Menn and colleagues suggest the linked attractor model complements and enriches some of the constructs included in generative phonology theory (Chomsky, 1972) and constraint-based frameworks. Constraint-based frameworks (Prince & Smolensky, 1997) are similar to generative theory in that these frameworks also (usually) assume innate, underlying phonological knowledge and surface forms. However, instead of "rules", surface forms are governed by "constraints", including faithfulness constraints, which stipulate speech output should adhere to adult forms as much as possible. Within Menn et al.'s usage-based model, "constraints" are seen to occur because of the limitations of a human being's vocal tract and cognition, together with the constraints of the ambient language to which an individual is exposed. Menn et al. argue the constraint-based concept of faithfulness - which the authors re-interpret as the "pull" to match the adult model - is an essential part of their developmental model, as it is this that drives phonological development and change. Generative-type rules are seen as comparable to the attractor model's mappings between representations. However, Menn and colleagues re-interpret these constraint and generative constructs within a usage-based model. Likewise, Pierrehumbert's model may be seen to bridge the gap between generative and usage-based models. Pierrehumbert's statistical categorisation of raw phonetic experience into a phonetic coding level, is analogous to the concept of levels of representation in generative models, and as a consequence, Pierrehumbert's model is described by some writers, as a "hybrid" model e.g. Docherty and Foulkes, (2013), Hinskens et al. (2014). Menn et al.'s model also enriches other usage-based accounts of phonology, such as Bybee's, Pierrehumbert's, Vihman and Croft's, not only by providing and extending a particular focus on child phonology, but also setting out a causal neurophysiological basis for phonological learning.

4.2 Clinical applications

This research uses usage-based theory to inform EPG intervention with individuals with SSD secondary to cleft palate +/- lip. This second section of this chapter will begin with discussion of previous work which has applied usage-based theory to the treatment of communication disorders. The section will then give a summary of the usage-based model used in the treatment study reported in this thesis. The section will go on to consider general implications for this theory on the management of cleft palate speech. This will be followed by specific therapeutic premises, and specific teaching episodes, for the usage-based EPG intervention used in this research.

4.2.1 Previous clinical application

Up to the present time, usage-based phonology theory has had limited direct clinical application. In a pioneering study, Hatchard (2015) applied usage-based language theory to aphasia. She examined the expressive language abilities of 12 individuals with aphasia according to word and contextual frequency effects, and to multiword retrieval. She concluded that participants' language difficulties were better explained within usage-based language theory, rather than generative theory.

Ball (2003) discusses broad clinical implications of Bybee's usage-based phonology on intervention for SSD. He suggests a usage-based "phonological" approach to intervention should involve drilling sets of words to "reinforce networks and allow specific units to emerge" (p. 66). He notes that this approach is in contrast to other phonological intervention, where the focus is on notions of contrast, such as in minimal pair therapy. Ball goes on to use usage-based theory to hypothesise the cause of speech difficulties seen in a 6-year old child with developmental SSD. This child's speech difficulties were attributed to entrenched articulatory gestural scores for problem sounds. Vogel Sosa and Bybee (2008), also discuss broad clinical implications of usage-based theory. They suggest speech change may be facilitated by manipulating frequency (both token and type). For example, they propose that low frequency words may be more disposed to phonological change in the therapy setting, while production of high frequency words may promote generalisation.

While not specifically labelled as "usage-based", some core concepts of usage-based theory also can be found in some clinical SSD work, perhaps suggesting some broader, pervasive, impact of this theory on clinical phonology. For example, Shriberg's speech attunement framework (Kwiatkowski & Shriberg, 1993, 1998; Shriberg et al., 2011) fits with usage-based theory. The speech attunement framework postulates that children need to "tune into" the articulation, voice and prosody of their ambient speech environment and to "tune up", so that their speech production matches those in their speech community (Shriberg et al., 2011). Another example is Scherer and Kaiser's Enhanced Milieu Teaching technique for treating young children with cleft palate +/- lip (Scherer & Kaiser, 2010). This therapy technique draws on a social interactional model. According to this model, language/speech is learned in the context of social interaction.

4.2.2 Usage-based phonology theory used in this thesis

The usage-based theory underpinning EPG intervention reported in this thesis derives from Bybee's (2001; 2010) and Menn et al.'s (2013) models, as described in section one of this chapter, and may be summarised as follows: At birth a baby's mind is like a blank slate. Individual experiences with vocalising and listening leads to first phonology and first words. Auditory experiences of speech lead to auditory memories, while vocalisations lead to motor memories (i.e. this is a two lexicon model, with separate input and output memories). Over time these input and output memories are linked together. Frequency of speaking and listening is a key component. Frequently produced vocalisations produce increasingly entrenched motor memories, while frequently heard speech produces increasingly entrenched auditory memories. When an individual goes to produce a word, they engage the most frequently used neuromotor memory used previously. For the infant developing typical speech, initially the basic phonological unit is the "word" or meaningful unit. Over time representations for part of words arise from sub-neural connections. Each time an individual hears a word or produces a word, words with similar speech sounds are aroused to an extent creating sub-network representations. However, it is use of words/tokens in meaningful units in communicative contexts that is the main driver for speech change. The mechanism by which generalisation occurs is the operation of this sub-neural network, i.e. change to an articulatory gesture in a treated word, for example /s/ in "see" will arouse other (untreated) words such as "seat", "so", leading to updating in these untreated words with time/practice. Phonological change may occur in an articulatory gradient way, with gradual phonetic adjustment to word exemplars over time. For example, a gradual adjustment of /t/ to /ʃ/ within words over time may look like: [t] → [s] →[s^j] → [ʃ]. As the child's motor skills improve with age and practice, overlapping of articulatory movements occurs, resulting in coarticulation and connected speech patterns. Use of typical connected speech patterns is important for speech which is understandable and does not draw attention to itself (Howard, 2013). Phonological learning occurs over the lifespan; the mechanisms for phonological change in the young child are the same as the mechanisms for change in the school-aged child/adult.

The importance of speech usage (i.e. speaking and listening events) in phonological development and change is one that will fit comfortably with most SLTs. SLTs manipulate speaking and listening events with the assumption that this manipulation and practice will lead to improvements with their client's speech sound production. However, usage-based theory's de-emphasis on the generative-type distinction between phonetics and phonology, and the concept that phonological learning is driven by phonetic and perceptual experiences rather than abstract phonological knowledge, is a very notable shift in thinking, and will be new to many SLTs working with SSDs.

4.2.3 General implications for the usage-based phonology model on intervention for cleft palate speech

In the management of the child with the cleft condition, the usage-based phonology model described above suggests primary palate repair to achieve a functioning speech mechanism, at or before speech-like babbling sounds emerge (i.e. at around 6-months of age for most children), is likely to be advantageous for the child's phonological development. Moreover, a particularly critical time for speech development is when the child begins to develop sub-network speech sound associations (i.e. at around 18-months for most children). Consequently, repair after this stage is predicted to be particularly disruptive to phonological development, as speech sound inaccuracies will "generalise" to other words/units. While cleft palate speech has not been considered from a usage-based perspective previously (to our knowledge), it is certainly the case that over the last 30-years or so the negative consequences of "late" cleft palate repair (i.e. beyond the first couple of years of life) on speech outcomes has become increasingly obvious to clinicians and researchers (Lohmander, 2011). Consequently, age of primary cleft repair has reduced over the years and today many cleft centres in the UK will have carried out primary cleft surgery by the time the infant is around 6-months of age. However, while the aim of primary cleft surgery is to fully repair the cleft and produce a functioning vocal tract, this is not always achieved, for reasons that are not fully understood (Lohmander, 2011). Common complications include oro-nasal fistula, velopharngeal insufficiency, impaired dentition and disrupted facial growth. Reported incidence of oro-nasal fistula ranges from 0% to 58% (Landheer et al., 2010), while the incidence of velopharyngeal insufficiency ranges from 5% to 40% (Enderby & Emerson, 1995). Individuals born with unilateral cleft lip and palate and bilateral cleft lip and palate universally will have impaired dentition, and many will have impaired dento-facial growth, the latter, which paradoxically, is usually a consequence of cleft repair at an early age (Lohmander, 2011; Mars & Houston, 1990). Thus, for many children born with the cleft condition, even after primary repair, they will continue to have an impaired vocal tract mechanism up until the time secondary deficits are corrected. In some of cases, even with modern surgical techniques, structural deficits cannot be completely corrected.

In addition to a trend for earlier surgical repair, over the past 10 to 15 years there has also been a growing movement for early speech and language therapy intervention with this client group (Golding-Kushner, 2001; Scherer & Kaiser, 2010). In the UK, the Royal College of Speech and Language Therapists (Enderby et al., 2009) recommend preventive advice and monitoring for babbling and early intervention is indicated in the child's second and third year of life. The American Cleft Palate-Craniofacial Associated (ACPA, 2009) also recommends close monitoring of early speech and language. In addition, this association recommends children born with cleft palate+/- lip are seen at least twice in the first 2-years of life, and that early intervention programmes should be administered if "the development of speech and language skills is not at an age-appropriate or developmentally appropriate level, or when early speech productions are deviant" (p. 26). Again, application of usage-based phonology points to the need for early speech and language therapy intervention.

Many individuals born with the cleft condition benefit from early surgical, audiology and speech and language therapy management, and go on to present with entirely normal speech beyond the first few years of life (Britton et al., 2014; Sell et al., 2015). However, as outlined in Chapter Two, around 20% of individuals born with the cleft condition present with persistent speech difficulties in their school years (Sell et al., 2015), and for some, problems can extend into adulthood (Whitehill et al., 1996). As discussed previously, currently, intervention for school aged children with cleft palate speech typically involves motor/articulation approaches (Golding-Kushner, 2001; Peterson-Falzone, 2006; Williams et al., 2021). In addition, phonological approaches to intervention have been used with this group of patients (Bessell et al., 2013; Williams et al., 2021). While many individuals benefit from this intervention, some individuals continue to experience difficulties with their speech. As stated previously, it is this group of patients which are the focus of this research.

4.2.4 EPG intervention: Application of usage-based phonology theory

Bybee (2001; 2010) and Menn et al.'s (2013) usage-based models are sufficiently detailed to permit identification of therapeutic premises, and from these premises, identification of specific teaching episodes for school aged children and adults with persistent cleft palate speech. Therapeutic premises and specific teaching episodes are listed below. In particular, the model summarised in 4.2.2 points to therapy premises and teaching episodes hypothesised to facilitate transfer and retention of new speech learning. Since the mechanism for phonological change is the same for children and adults, the same therapeutic technique can be used for both school-aged children and adults.

4.2.4.1 Therapeutic Premises

The following therapeutic premises are derived from the usage-based model summarised in 4.2.2:

- Since this is a two lexicon model (i.e. where separate production and auditory memory traces are assumed), speech production practice is essential to achieve speech production change.
- Since output memories consist of phonetic memories (i.e. memories of articulatory gestures) the focus needs to be on articulatory gestures and how to produce these in the vocal tract. EPG is well suited to providing visual feedback on lingual speech gestures to facilitate these gestures.
- Given the concept of frequency of use (i.e. neuromotor memories become increasingly entrenched with usage) a high dose of production trials is important. Further, manipulation of word frequency (in terms of how often that word is used in everyday talking, e.g. "the" is a high frequency word) is viewed as facilitative: Low

frequency words will have less entrenched memories and will be easier to produce in the early stages of therapy, while high frequency words will expedite generalisation in later stages of therapy, since high frequency words have stronger sub-network connections.

- Since change with speech production is often gradient, gradient change is desirable and can be facilitated through intervention.
- Since phonology is intrinsically liked to words/meaningful units, production of words in particular is important.
- Generalisation to everyday speech is possible through production of single words as change to word production will activate changes to sub-neural networks.
- Connected speech patterns can be taught and can facilitate speech understandability and speech acceptability.
- The mechanisms for phonological change is the same for children and adults.

4.2.4.2 EPG teaching episodes

Given the above premises, usage-based directed teaching episodes should involve:

- High volume speech production, with the focus on single word production.
- Production of low frequency words in the early stages of therapy, followed by production of high frequency words in the later stages.
- Specific feedback on articulatory gestures in words using EPG visual feedback, then subsequent fading of EPG to facilitate intrinsic speech monitoring systems.
- Facilitation of gradient speech change with articulatory gestures in words.
- Practice of connected speech patterns in connected speech.

Response to these teaching episodes can serve to strengthen or reject elements of the usage-based theory.

The usage-based EPG technique is described in detail in 5.3.

4.3 Study's aim and research questions

The aim of this study was to describe and evaluate cleft patients' response to a usage-based EPG therapeutic technique. It was hypothesised that the usage-based EPG intervention would improve accuracy of target phonemes post-intervention, and that improved accuracy of target phonemes would generalise to spontaneous connected speech outside the clinic environment. The research questions therefore were as follows:

1. Acquisition and generalisation: Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated words/pseudo- words?
2. Functional generalisation/ impact on quality of life: Does improved accuracy of target phonemes generalise to continuous, connected speech and, if so, does this result in improved ratings of speech understandability and speech acceptability and improved ratings of impact on quality of life, post-therapy?
3. Theoretical model: Do improvements with accuracy of targeted phonemes following therapy provide support for the usage-based theoretical model in general, and more specifically in terms of:
    a. Gradient phonetic speech change with target phonemes is shown over the course of therapy, with target production gradually moving from atypical production to production within normal parameters in the participant's dialect, e.g. [k] to [c] to [t̪] to [t].
    b. A word frequency effect is shown with low frequency words being easier for participants to learn compared to high frequency words, and high frequency words being the most resistant to change in everyday speech.

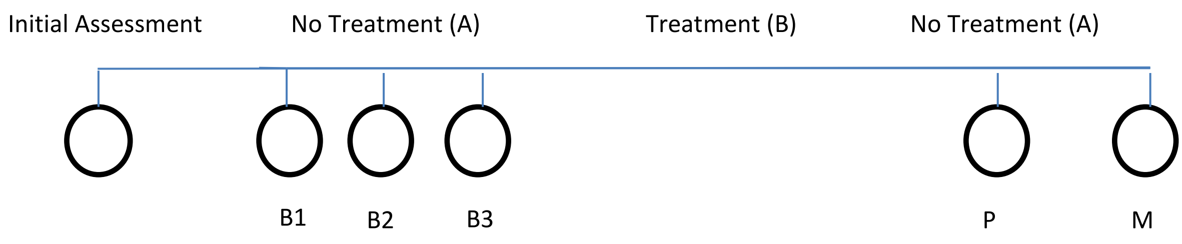
5 METHOD

Six patients with SSDs secondary to cleft palate were recruited and treated in a regional hospital setting. A multiple-baseline (ABA) within-participant design (Ebbels, 2017; Morley, 1994; Pring, 2005) was used for each patient treated within this study. As discussed in Chapter Three, within-participant designs are viewed as appropriate for examination of novel interventions, such as this usage-based EPG therapy technique, as they allow in-depth examination of participants' difficulties, together with detailed analysis of response to intervention (Ebbels, 2017; Pring, 2005).

Following recruitment, participants underwent an initial assessment to identify target speech sounds. Participants then completed three baseline assessments, at least 3-weeks apart (i.e. at least 6-weeks from baseline one to baseline three), to ensure speech sound errors were static (i.e. Phase A: No treatment). Participants were then seen for weekly usage-based EPG therapy (i.e. Phase B: Treatment). Individual speech sound errors were worked on in turn, i.e. speech target one was the focus of intervention, before moving onto speech target two, and so on. Words treated within the session were assessed for production at the end of each treatment session. At the end of each fifth treatment session, more detailed assessment occurred, including production of treated and untreated words and EPG assessment. Treatment continued until all speech sound errors were remediated and were being produced in the participant's everyday speech, or alternatively, the participant's progress had plateaued and no further gains were being seen. Detailed assessment was carried out at the end of therapy, then again 3-months after the end of treatment (i.e. Phase A: No Treatment). Speech assessment at the different testing points throughout the study was videoed and audio recorded and perceptual speech analysis came from these recordings. A schema of the study design is shown in Figure 5, and Table 2 details the treatment phase

Figure 5

Overall schema of ABA study design



Note: B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post-therapy; M = Maintenance assessment 3-months post-therapy.

Table 2

Schema of treatment (B) phase

Week No	1	2	3	4	5	6	7	8	9	10	11	….	End of intervention
Assessment	Tr1	Tr2	Tr3	Tr4	A5	Tr6	Tr7	Tr8	Tr9	A10	Tr11	….	P

Note: Tr1 = Assessment end of session 1; Tr2 = Assessment end of session 2, and so on; A5 = Assessment at end of five sessions; A10 = Assessment at the end of 10 sessions, as so on; P = Assessment immediately post-intervention.

This study received ethical approval from the North West – Greater Manchester East Research Ethics Committee (17/NW/0235). Written informed consent was obtained from adults and young people aged 16 – 17 years. Written informed proxy consent and written assent was obtained from the children 15-years and below.

5.1 Participants

Participants were recruited from the Royal Manchester Children's Hospital's cleft lip and palate service. Specialist SLTs working with the cleft team identified potential participants during the course of their everyday clinical practice using the following inclusion and exclusion criteria:

- Persistent (unchanged for six or more months) lingual speech sound difficulties (where the patient had been unable to produce, or had rarely produced, the speech sound, i.e. 0 – 10% accuracy with production), secondary to cleft palate +/- lip (including submucous cleft palate), adversely affecting speech understandability and/or speech acceptability. Lingual speech difficulties needed to show up on EPG;
- The patient had received at least two episodes or more widely available speech therapy interventions in the past which had failed to result in significant changes to speech (as determined from patient's clinical notes; this included gaps between episodes);
- Aged 7-years and over (i.e. children and young people aged 7-years and over, and adults), thereby ruling out any significant spontaneous developmental change that might be seen with younger children;
- No significant learning disabilities – schooled in mainstream school with no or limited support needed;
- Normal velopharyngeal function or mild signs of VPI;
- Able to attend for weekly speech and language therapy appointments at the Regional Cleft Unit, Royal Manchester Children's Hospital;
- Ability to practice speech targets on a daily basis, including speech practice using a portable EPG unit;

- Suitable dentition so that an EPG palate could be fitted and worn during speech practice for at least 4-months.

As seen with the above criteria, only patients who had long-standing lingual speech sound errors who had failed to progress with more widely available speech therapy interventions were selected to participate in this study.

If identified as a potential participant, the specialist SLT discussed the nature of the study with the patient, and in the case of children, the patient's parent. The patient/patient's family were told that participation was voluntary: it was made clear that they did not have to participate if they did not wish to do so. If the patient/patient's family wished to receive more information about the study, they were given a patient information sheet during this appointment. Separate patient information leaflets, as shown in 11.1, Appendix A, were available for:

- Adults and young people aged 16 – 17 years;
- Parents /guardians of children;
- Children aged 7 – 11 years;
- Young people aged 12 – 15 years.

The specialist SLT phoned the patient/patient's family 1 – 2 weeks after they were given written information on the study. If the patient/patient's family wished to participate, the researcher then made an appointment with the patient/patient's family at the hospital to discuss the patient's eligibility further, and to take written consent for participation.

Fifteen patients were given information sheets. All 15 patients were put forward to the researcher. Of these 15 patients, 5/15 were excluded (see Figure 6 for reason), leaving 10 who met the study criteria. All 10 patients consented. Of these 10 patients, three did not go to participate because they were unable to tolerate having a training plate in the mouth, leaving seven who received EPG therapy. One participant withdrew from the study because this participant was unable to attend hospital appointments because of the COVID-19 pandemic. Thus, the final study sample consisted of six participants. A flowchart of study recruitment is shown in Figure 6.

Figure 6

Flowchart for patients assessed, excluded, treated and followed-up

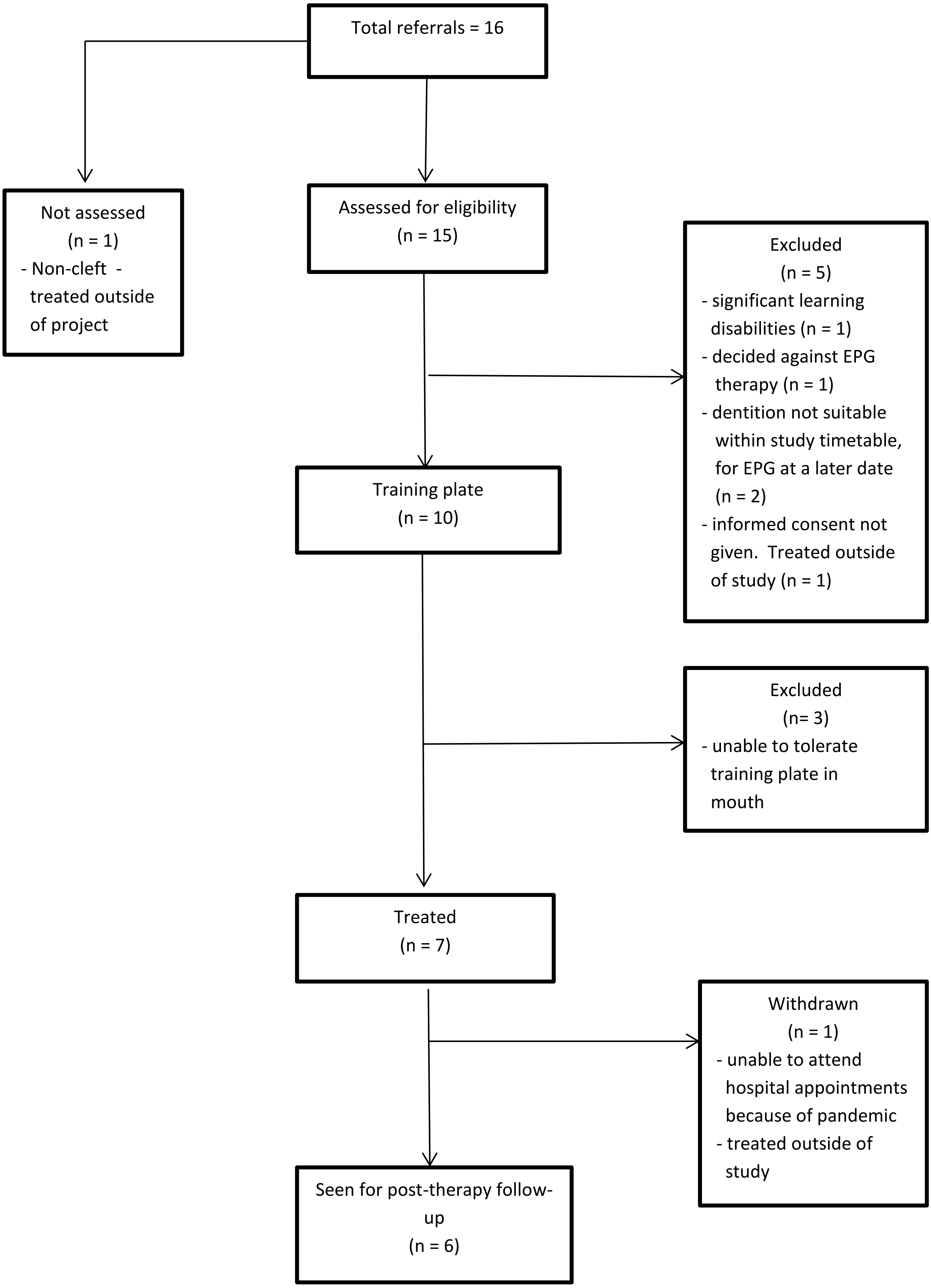


Table 3 summarises the demographic information of the final sample of six participants who participated in the intervention study. A pseudonym is used for each participant.

Table 3

Participants' demographic information

Participant	Sex	Age	Diagnosis	Co-occurring diagnosis	Speech sounds produced in error at word level
George	M	10	Cleft palate		z, ʃ, ʒ, tʃ, dʒ, ɹ, θ, ð
Olivia	F	20	Submucous cleft palate		s, z, tʃ, ɹ, θ, ð
Emma	F	11	Submucous cleft palate	22q1.1 deletion syndrome	t, d, n, l, s, z, ʃ, ʒ, tʃ, dʒ, ɹ, θ, ð
Victoria	F	27	Unilateral cleft lip and palate		s, z, ʃ, ʒ, tʃ, dʒ
Grace	F	7	Cleft palate		s, z
Poppy	F	17	Unilateral cleft lip and palate		n, l, θ, ð

5.2 Assessment and scoring

As discussed in 3.2, when evaluating the outcome of EPG therapy, both perceptual speech assessment and EPG assessment should be included. In addition, more holistic outcomes, such as speech understandability and speech acceptability and effect on daily life, require measurement. In this study, the following assessments/tasks were administered to identify phonemes to be targeted in therapy, and to measure response to intervention:

1. Perceptual speech assessment:
   - Great Ormond Street Speech Assessment (GOS.SP.ASS) (Sell et al., 1999);
   - Diagnostic Assessment of Articulation and Phonology (DEAP), phonology sub-test (Dodd et al., 2002);
   - Reading treated words task (task developed for this study);
   - Reading untreated words task (task developed for this study);

- Percentage of target sounds correct in connected speech task (task developed for this study).

2. EPG assessment:
    - Visual analysis of EPG palatograms;
    - EPG indices.
3. Speech Intelligibility/Quality of Life assessment:
    - Intelligibility in Context Scale (ICS) (McLeod et al., 2012): parent/spouse report of speech understandability;
    - Speech Understandability and Speech Acceptability Scales – ratings made from a two- minute connected speech sample (Henningsson et al., 2008);
    - Speech Participation and Activities of Children (SPAA-C) – ratings made through semi-structured interview with child (McLeod, 2004);
    - Quality of Life Instrument (QoL-Dys) – ratings made by adult (Piacentini et al., 2011)

Table 4 provides an overview of the administered assessments/tasks at each time point.

Table 4

Overview of administered tests/tasks at each assessment point

Testing point	Assessments used							
	GOS.SP.ASS	2-minute spontaneous connected speech sample	DEAP, phonology sub-test	ICS	SPAA-C or QoL_Dys	Reading treated words	Reading untreated words	EPG assessment
Initial assessment	X	X	X	X	X			
Baseline 1		X				X	X	
Baseline 2		X				X	X	
Baseline3 / pre-therapy	X	X		X		X	X	X
End of each treatment session						X		
End of each 5th treatment session		X				X	X	X
Immediately post-intervention	X	X	X	X	X	X	X	X
Maintenance 3-months post intervention	X	X	X	X	X	X	X	X

Note: GOS.SP.ASS = Great Ormond Street Speech Assessment; DEAP = Diagnostic Assessment of Articulation and Phonology; ICS = Intelligibility in Context Scale; SPAA-C = Speech Participation and Activities of Children; QoL-Dys = Quality of Life Instrument; Red crosses indicate that the assessment was videoed and audio recorded; Underlined crosses are the assessments blindly listened to and analysed by the two independent SLTs.

Each assessment used in this study is described in the sub-sections below:

5.2.1 The Great Ormond Street Assessment (GOS.SP.ASS)

GOS.SP.ASS (Sell et al., 1999) is an assessment developed specifically for the assessment of speech difficulties associated with cleft palate. GOS.SP.ASS is the standard assessment used by all cleft centres in the United Kingdom to assess cleft palate speech for treatment purposes. GOS.SP.ASS picture stimuli are used in the Cleft Audit Protocol for Speech - Augmented (John et al., 2006) which is used to report speech outcomes in the UK (Britton et al., 2014; Sell et al., 2001; Sell et al., 2015). This assessment was used in this study to identify lingual speech sounds in error and was used to develop treated and untreated word lists for each participant (see section 5.4.3).

This assessment was administered according to instructions, as follows: The SLT and the patient looked at the test's picture book (one picture per page). The SLT read a sentence relating to each of picture and the patient repeated the sentence (e.g. "Tim is putting a hat on"). Each sentence targets a speech sound (in word initial and word final position) that has been identified as particularly vulnerable to the effects of a cleft palate (Sell et al., 1994). A total of 22 sentences were repeated. The 22 sentences are listed in 11.2, Appendix B.

The SLT phonetically transcribed the target sound (in word initial and word final position) onto a table using narrow phonetic transcription. This table provided a visual account of the client's speech sound production.

5.2.2 Diagnostic Assessment of Articulation and Phonology (DEAP)

DEAP (Dodd et al., 2002) is a battery of assessments for children who present with SSDs. DEAP consist of five assessments, however, only the phonology picture naming assessment was used in this study. The phonological subtest calculates percentage of consonants correct (PCC) and identifies phonological patterns, such as backing of /t/ to [k]. This assessment was used to measure PCC and, alongside GOS.SP.ASS, was used to identify atypical speech sound production. Only PCC was calculated in this study; no phonological analysis was carried out.

The phonology picture naming test consists of 50 pictures. All English consonants in syllable-initial and syllable-final position are sampled in these 50 words. In this study, the SLT and the patient looked at the test picture book (one picture per page). The patient was asked the name the pictures. The SLT phonetically transcribed the participant's responses onto the DEAP phonology test form. This phonetic transcription was used to calculate PCC in the following manner: The total number of consonants correct was divided by the total number of consonants. Speech sounds in error were identified by examining the phonetic transcription and comparing this to the target.

5.2.3 Reading treated word list task

In a task developed specifically for this study, treated word lists were compiled for each participant, in accordance with their speech sound errors (see 5.4.4 for details regarding how treated word lists were developed and 11.4, Appendix D for word lists). This task was used to examine participant's response to intervention.

Treated words were written on a page, with one word per line, and participants were asked to read down the list. The SLT phonetically transcribed the participant's responses onto an assessment form developed specifically for this study. All phonemic errors and phonetic distortions were marked as incorrect, apart from the phonetic distortion of dental or very slight interdental production of alveolar consonants. This phonetic transcription was used to calculate percentage of target sounds correct (%TC) in the following manner: The total number of target sounds correct was tallied and was then divided by the total number of target sounds in the word list.

5.2.4 Reading untreated word list task

Like the previous task, this task was developed specifically for this study and individual untreated word lists were developed for each participant, in accordance with their speech sound errors (see 5.4.4 for details regarding how the untreated word lists were developed and 11.4, Appendix D for word lists). This task was used to examine generalisation of speech sound targets to untreated words.

As with the treated word task, untreated words were written on a page, and participants were asked to read down the list. The SLT phonetically transcribed the participant's responses, and percentage of target sounds correct (%TC) was calculated in the same manner as the treated word task.

5.2.5 Percentage of target sounds correct in connected speech task

In this assessment task, again developed specifically for this study, participants were asked to describe what they did the previous day. This event retell generated a sample of spontaneous continuous speech (i.e. connected sentences spontaneously generated by the participant) of around 2-minutes. This task was used to examine generalisation of speech sound targets to continuous connected speech.

Participants' were given the instruction: "Can you tell me everything you did yesterday from the time you woke up to the time you went to bed?" The researcher did not speak during this event re-tell. If the participant said very little, the researcher asked prompting questions (e.g. "Tell me about the TV programme you watched"), but such prompts were kept to a minimum.

The SLT phonetically transcribed the first 2-minutes of this speech sample. The total number of occurrences of a target sound was tallied. The number of correct productions of the target consonant was then tallied. As with percentage of target sounds correct in treated and untreated word lists, all phonemic errors and phonetic distortions were marked as incorrect, apart from the phonetic distortion of dental/slight interdental production of alveolar consonants. The number of correct instances of the target sound was divided by the total number of instances of the target sound to arrive at the percentage of target sounds correct (%TC) in connected speech. This task was used to examine generalisation of speech sound targets to continuous connected speech.

5.2.6 EPG assessment

This study used the WinEPG system, including Articulate Assistant software™ (Articulate Instruments Ltd, 2008), and Reading-style EPG palate. EPG analysis was used to confirm speech sound errors with placement and manner identified by phonetic transcription and to identify distortions and patterns not distinguishable by impressionistic perceptual speech assessment. In addition, EPG analysis was used to examine response to intervention.

EPG assessment involved the following: The participant wore their EPG palate and the plate was connected to the main EPG system. The participant read the speech stimuli listed in 11.3, Appendix C and EPG and acoustic recordings were taken. This stimulus consisted of:

- 20 repetitions of CV sequences (involving English phonemes), 10 repetitions involving closed vowels, 10 repetitions involving open vowels;
- Repetition of GOS.SP.ASS sentences.

This EPG assessment is similar to the EPG CLEFTNET speech word list (Gibbon & Wood, 2010) and allowed examination of tongue–palate placement for lingual phonemes at CV level and at sentence level. Each speaker's tongue–palate contact during speech production was recorded 100 frames per second in two-dimensional pictures (known as EPG frames or palatograms). Simultaneous, synchronised audio was also recorded using a Stageline EMA-20 headset microphone. The series of palatograms and acoustic information was saved.

Target sounds were later annotated by the researcher from EPG and acoustic information using the "analyse" function of Articulate Assistant™. Each target sound was annotated according to the articulatory and/or acoustic properties of the actual sound produced. For example, if the participant produced a [ʃ] for /s/, the [ʃ] was annotated. The system for annotating speech sounds is detailed in Table 5.

Table 5

System for annotating EPG data

Speech sound	Analysis (rationale)
Fricatives	Acoustic signal - onset to offset of friction (in order to capture the whole duration of the fricative).
Oral and nasal stops	First EPG frame of closure to release of closure using the acoustic signal (in order to capture the entire closure phase).
Affricatives	Analysed in two parts: 1. First EPG frame of closure to the release of closure using the acoustic signal (capturing the entire closure phase); 2. Acoustic signal - onset to offset of frication (capturing the entire duration of the fricative).
Glides	First EPG frame of lingual contact to release of contact (capturing the whole duration of contact).
Glottal stops and velar stops (not shown on EPG frames)	Acoustic signal – onset to offset (capturing the entire closure phase).

Figure 7 shows an example of annotations for /ʃ/, while Figure 8 gives an example of annotations for /t/.

Figure 7

*Example of /ʃ/ annotations (CVCV repetitions***)**

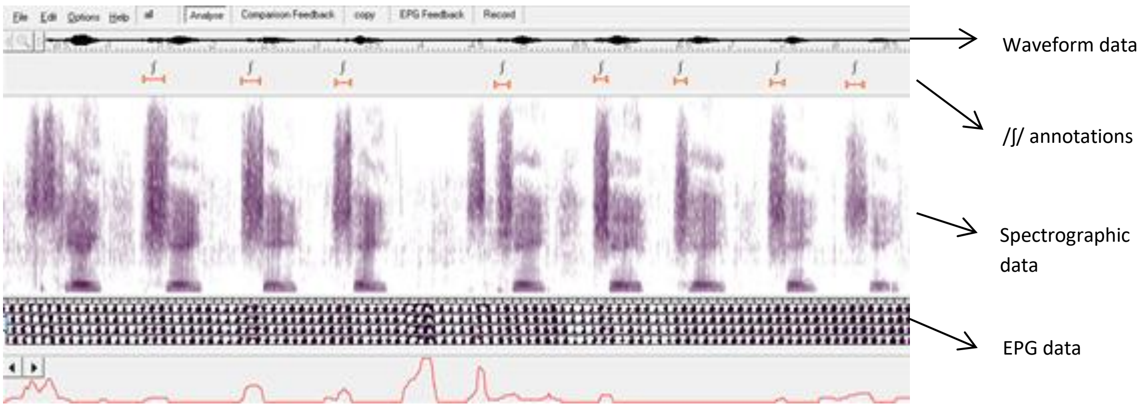
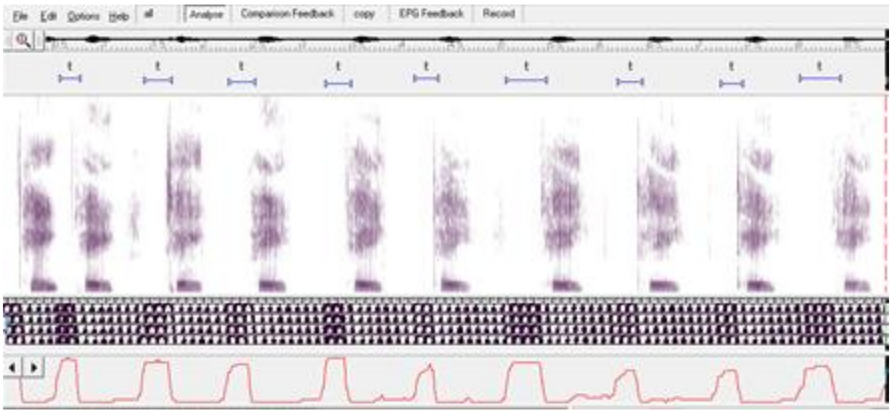


Figure 8

Example of /t/ annotations (CVCV repetitions)



On a few occasions the acoustic information did not appear to align with the EPG frames. Such occasional misalignment has been reported in other studies (Timmins, 2014). In these few instances, annotation was made from EPG patterns alone, if the EPG frames provided sufficient information. Each annotation included details of the time of recording (e.g. B3), target sound (e.g. /s/), repetition number (e.g. repetition 1), and type of vowel (e.g. high vowel).

Following this annotation of EPG images, further analysis was carried out: visual analysis of EPG palatograms and calculation of EPG indices.

5.2.6.1 Visual analysis of EPG palatograms

The EPG frames for target sounds were compared to standard EPG frames using reference frames as shown in McLeod and Singh (2009) and reference frames from the researcher's own speech. Cumulative frames of maximal contact was used to compare with reference frames, that is, each frame of maximum contact for each CV repetition (up to 20 in total) was superimposed to produce one cumulative picture. Participants' EPG frames were described using the classification system developed by Gibbon (2004), as summarised below and as described in more detail in 3.1.2:

- Increased contact;
- Retracted to palatal or velar placement;
- Fronted placement;
- Complete closure;
- Open pattern;
- Double articulation;
- Increased variability;
- Abnormal timing.

5.2.6.2 EPG indices

As discussed in 3.1.2, EPG indices reduce EPG data to single numerical values and selection of indices depends on the specific speech sound error. Table 6 outlines the speech sound errors evidenced by participants, and the EPG indices used, in this study. The frame of maximal contact was used to compute all EPG indices.

Table 6

Study EPG indices and speech sound errors

EPG indices	Speech sound errors	Explanation of EPG indices and rationale for use
AC	/s/ → [ɬ] /z/ → [ɮ] /ʃ/ → [ɬ] /tʃ/ → [ɬ] /dʒ/ → [ɮ] /z/ →[n]	AC relates to the connectivity between the two sides of the palate. The higher the AC score the more contact between the two sides of the palate. A decrease in scores across test points shows a change from lateral airflow to central tongue airflow.
AT and CoG	/s/ → [ʃ], [ç] /z/ → [ʒ], [ʝ] /t/ → [k] /d/ → [g] /n/ → [ŋ] /l/ → [j]	AT is the number of electrodes contacted in the first three rows of the EPG palate (possible total of 22) per EPG frame. An increase in scores across test points shows a change from posterior to anterior tongue - palate contact. CoG shows the position of the main concentration of electrodes across the palate per EPG frame. Small CoG values relate to posterior tongue-palate contact. CoG values rise with increases in anterior tongue-palate contact. AT and CoG therefore can show any changes from posterior to anterior tongue-palate contact
WT	/ʃ/ → [ħ]	WT represents the total number of electrodes contacted per EPG frame. WT was used to show a change from no tongue-palate to tongue palate contact, and also an increase in tongue-palate contact

Note. AC = Alveolar Closure; AT = Alveolar Total; CoG = Centre of Gravity; WT = Whole Total

5.2.7 Intelligibility in Context Scale (ICS)

The ICS is a parent, or alternatively spouse-reported, measure of speech understandability (McLeod et al., 2012). This assessment was administered to obtain a parent, or spouse-reported, measure of speech understandability, and was used to examine response to intervention.

The ICS consists of seven items. On the first item, the parent (or spouse) is asked to consider their child's (spouse's) speech over the past month and rate how well they have understood their child's speech using the following 5-point Likert scale:

- Never understand (1);
- Rarely understand (2);
- Sometimes understand (3);
- Usually understand (4);
- Always understand (5).

In the remaining six items, the parent (or spouse) is asked to consider how well six other communicative partners have understood their child over the past month, using the same five-point scale. The six other communicative partners include:

- Immediate family;
- Extended family;
- The child's friends;
- Acquaintances;
- Teachers;
- Strangers.

The scores for each of the seven items are added to result in a total score. In addition, the total score is divided by seven to give an average score.

5.2.8 Speech Understandability and Speech Acceptability Scales

The Speech understandability and Speech Acceptability Scales were developed by Henningsson and colleagues (Henningsson et al., 2008), and are part of a wider set of measures used to report speech outcomes in individuals born with cleft palate +/- lip. These scales were used to examine response to intervention, in terms of change to functional speech.

The two 4-point Likert rating scales rate speech from 0 – 3, with as score of zero being within normal limits and a score of three representing severe difficulties. The two scales are shown in Table 7. In this study, the rating scales were completed as follows: The SLT listened to the 2-minute sample of spontaneous speech and made a judgement of speech understandability by selecting the rating that best matched their perception of the participant's speech. This rating was marked on a form developed for this study. If speech understandability was not normal, the SLT wrote down why understandability was perceived to be different using free text. The SLT then listened to the participant's production of treated and untreated words. The rating of speech acceptability was made from both the spontaneous speech sample and production of words. Again, if speech acceptability was perceived as different, a free text note was made to outline the perceived cause of this difference.

Table 7

Speech understandability and speech acceptability scales (Henningsson et al., 2008)

Speech understandability
• Speech is always easy to understand (0 – within normal limits)
• Speech is occasionally hard to understand (1 – mild)
• Speech is often hard to understand (2 – moderate)
• Speech is hard to understand most or all of the time (3 – severe)
Speech acceptability
• Speech is normal (0 – within normal limits)
• Speech deviates from normal to a mild degree (1 – mild)
• Speech deviates from normal to a moderate degree (2 – moderate)
• Speech deviates from normal to a severe degree (3 – severe)

5.2.9 Speech Participation and Activities of Children (SPAA-C)

SPAA-C (McLeod, 2004) is a questionnaire which examines the impact of a child's speech difficulties on their everyday life. This questionnaire was used to examine response to intervention in terms of changes to quality of life. This questionnaire was used with the three children participating in this study.

The SPAA-C contains five sections:

1. Questions for the child.
2. Questions for friends.
3. Questions for parents.
4. Questions for teachers.
5. Questions for others.

The SLT asks the questions in the questionnaire using a semi-structured interview technique. Not all sections need to be completed: The SLT selects the sections relevant to the child and the interview context (e.g. hospital, clinic, school). Section 1 includes 10 questions on the child's feelings about talking in different communicative contexts (e.g. talking to parents, talking to teachers), and the child's responses are recorded using a 5-

point Likert scale: 1 = happy, 2 = in the middle, 3 = sad, 4 = another feeling, and 5 = don't know. In this study sections one and three were administered as this was considered relevant to the child and interview context (i.e. hospital). Responses to questions were documented using the SPAA-C form.

5.2.10 Quality of Life Instrument (QoL-Dys)

The QoL-Dys (Piacentini et al., 2011) is a patient-reported quality of life measure that was developed for adults with dysarthria. The QoL-Dys, was used to measure quality of life with the three participants, aged 17-years and over. QoL-Dys was selected in the absence of a quality of life measure specifically developed for young people aged 16- to 17-years and adults born with the cleft condition at the time this study was submitted for ethical approval. Since this time, the Cleft Q (Klassen et al., 2018) is now available to measure quality of life in individuals born with the cleft condition. Although developed with people with dysarthria, items contained in the QoL-Dys instrument are relevant to adults with speech sound disorders secondary to cleft palate +/- lip.

The QoL-Dys consists of 40 statements relating to the impact of a patient's speech difficulties on their everyday life. Statements include, for example, "My speech sound unnatural", "When I talk people pretend to understand me". The patient completes this instrument on their own and responds to each statement using a 5-point Likert scale: O = never, 1 = almost never, 2 = sometimes, 3 = almost always, 4 = always. The patient selects the response which best matches his/her experience. Scores are added to obtain a total score which ranges from 0 – 160.

5.2.11 Scoring and agreement procedures

Two independent SLT blindly phonetically transcribed participant's production of treated and untreated words (see 5.2.3 and 5.2.4), and made a blind judgement of speech understandability and speech acceptability (see 5.2.8) at the three baseline assessments and the two assessments post-therapy. When transcribing production of treated and untreated words, the independent SLTs made a tick beside words perceived as accurate and words perceived as inaccurate were transcribed using narrow phonetic transcription. All other assessment measures (i.e. initial assessment, all assessment carried out during intervention) were completed and scored by the main researcher. The two independent SLTs work as specialist cleft SLTs the Royal Manchester Children's Hospital; neither SLT had any involvement in participants' EPG treatment. The two independent SLTs did not transcribe and rate all assessment measures in this study due to time and cost restrictions.

5.2.11.1 Independent SLTs' inter-/intra-rater agreement

Inter-rater agreement for the two independent SLTs' phonetic transcription of treated and untreated words was computed on a point-by-point basis in the following way: The accuracy of each target sound for each SLT was compared and tallied. If both SLTs marked speech target one as accurate, this counted for an "A" tally in an agreement column. If speech target two was marked as accurate by one SLT and inaccurate by the second SLT, this counted for a “D” tally in the agreement column. The percentage of agreement (speech accuracy) reported was the total number of “A” tallies in the agreement column divided by the sum of the “A” and “D”. Inter-rater agreement for ratings of speech understandability and speech acceptability were calculated in the same way: The same rating score represented an agreement, while different ratings represent a disagreement.

Inter-rater agreement for the two independent SLTs’ phonetic transcription of treated and untreated words was 91.09%. Cohen’s Kappa (Cohen, 2016) was also run to examine agreement between the two independent raters’ transcription of words. This statistic takes change agreement into account. The Kappa value is interpreted as follows (Cohen, 2016):

- values equal to or less than zero = no agreement;
- 0.01 – 0.20 = none to slight agreement;
- 0.21 – 0.41 = fair agreement;
- 0.41 – 0.60 = moderate agreement;
- 0.61 – 0.80 = substantial agreement,
- 0.81 – 1.00 = almost perfect agreement.

Inter-rater agreement for the two independent SLTs’ phonetic transcription of treated and untreated words showed substantial agreement ($\kappa = .786$, $p < .000$).

Inter-rater agreement for the two independent SLT’s ratings of speech understandability was 60%, and agreement for speech acceptability was also 60%. Cohan’s kappa showed moderate agreement for understandability ratings ($\kappa = .417$, $p < .000$), while fair agreement was shown for acceptability ratings ($\kappa = .393$, $p < .001$). Thus, lower inter-rater agreement was achieved for these rating scales. However, agreement within two points (i.e. score of “0” and score of “1” rated as agreed, score of “1” and “2” rated as agreed, score of “0” and “2” rated as disagreed, score of “1” and “3” rated as disagreed, and so on) gave an agreement score of 93.33% and 100% for speech understandability and speech acceptability respectively. Agreement with ratings of speech understandability and acceptability is discussed further in Chapter Eight.

Both independent SLTs transcribed and rated a random 20% of speech data at least one month following original transcription to calculate intra-rater agreement. Agreement for transcription for the same data, assessed at two time-points (listen number one and listen number two) was computed on a point by point basis in the following way: The accuracy of each target sound for each time-point was compared and tallied. If the SLT marked speech target 1 as accurate on the first listen and speech target 1 as accurate on the second listen, this counted for an ”A” tally in an agreement column. If speech target 2 was marked as accurate on the first listen and inaccurate on the second listening, this counted for a “D” tally in the agreement column. The percentage of agreement (speech accuracy) reported was the total number of “A” tallies in the agreement column divided by the sum of the “A” and “D”. Intra-rater agreement for ratings of speech understandability and speech acceptability were calculated in the same way: The same rating score represented an agreement, while different ratings represent a disagreement.

Intra-rater agreement for phonetic transcription of treated and untreated words was 92.08% for the first independent SLT. Cohen’s kappa showed substantial agreement (κ = .729, p < .000). Intra-rater agreement for the second independent SLT’s transcription of treated and untreated words was 96.96%. Cohen’s kappa showed almost perfect agreement (κ = .923, p < .000). Intra-rater agreement with ratings of speech understandability and speech acceptability was 80% and 80% respectively for the first independent rater. Cohen’s kappa showed substantial agreement of ratings of understanding and acceptability (κ = .688, p <. 022, and κ = .615, p < .136 respectively). Intra-rater agreement with ratings of speech understandability and speech acceptability was 50% and 100% respectively for the second independent rater. Cohen’s kappa showed fair agreement for ratings of understandability (κ = .250, p < .257), and perfect agreement for acceptability ratings (κ = 1.00, p < .046). Thus, lower intra-rater agreement was shown for the second independent SLT on the speech understandability scale. However, intra-rater agreement with speech understandability and speech acceptability within 2 points gave agreements of 100% for both independent raters.

5.2.11.2 Researcher inter-/intra- rater agreement

Twenty percent of the researcher’s transcriptions and ratings were transcribed and rated by the two independent SLTs to produce inter-rater agreement for the researcher, using the method above. Percentage of agreement with transcription of treated and untreated words with the first independent rater was 89.75%. Cohen’s kappa showed substantial agreement (κ = .788, p < .000). Percentage of agreement on transcription of treated and untreated words with the second independent rater was 91.39%. Cohen’s kappa showed almost perfect agreement (κ = .813, p < .000).

Percentage of agreement with ratings of speech understandability and speech acceptability with the first independent rater was 53.33 and 80% respectively. Cohen’s kappa showed fair agreement for understandability ratings (κ = .272, p < .020) and moderate agreement for acceptability ratings (κ = .513, p<.000). However, agreement with speech understandability and speech acceptability within two points gave percentages of 93.33% and 100% respectively. Percentage of agreement with ratings of speech understandability and speech acceptability with the second independent rater was 60% and 47% respectively. Cohen’s kappa showed fair agreement for understandability and acceptability ratings (κ = .247, p < .051) and (κ = .270, p < .029), respectively. However, agreement with speech understandability and speech acceptability within 2 points gave agreements of 100%.

The researcher also re-rated 20% of the speech data at least one month following original transcription and ratings and intra-rater agreement was calculated. Intra-rater agreement with phonetic transcription of treated and untreated words was 97.07%. Cohen's kappa showed almost perfect agreement ($\kappa = .940$, $p < .000$). Intra-rater agreement with ratings of speech understandability and speech acceptability was 75% and 100% respectively. Cohen's kappa showed substantial agreement for speech understandability ($\kappa = .619$, $p < .009$). Agreement for speech acceptability showed almost perfect agreement ($\kappa = 1.00$, $p < .000$). Intra-rater agreement with speech understandability and speech acceptability within two points gave agreements of 100%.

Where disagreement amongst the raters occurred, this was not resolved by consensus. Percentage of targets correct for treated and untreated words at baseline one, baseline two, baseline three, assessment immediately post-intervention, and maintenance assessment were derived from an average of the two independent raters (see Figure 9 – 14). Ratings for participants' speech understandability and speech acceptability are given for both independent raters (see Table 17 and Table 18). All other assessment measure (i.e. initial assessment, all assessment carried out during intervention) come from the researcher's ratings.

5.3 Intervention

As discussed in 3.1.4, standard EPG therapy targets production of speech sounds successively in a hierarchy of motor speech difficulty including: production in isolation; syllable level; word level; and then finally sentence level (Gibbon & Wood, 2010). Patients are typically required to achieve 80 – 90 % accuracy of production before moving onto the next level in the hierarchy. Standard EPG therapy does not consider connected speech patterns. In this study, the EPG therapeutic technique was modified from the novel application of usage-based phonology theory, as discussed in 4.2. The resultant technique differs from standard EPG therapy in two main ways: Focus on speech production at word level, focus on production of connected speech patterns.

Focus on speech production at word level: In this usage-based phonology approach, production at sound and syllable level was not part of the therapeutic approach. Rather the focus of therapy was on production at word level, and on achieving accuracy at this level before moving onto sentence level. This immediate focus on words, rather than sounds or syllables, follows the usage-based phonology theory that speech change occurs at the level of the word (i.e. meaningful token) rather than single sounds (Bybee, 2001, 2010). The mechanism for system-wide generalisation is the operation of sub-neural networks, i.e. change to an articulatory gesture in a word will arouse other words containing the same or similar articulatory gestures, and these other words will be updated. Patients first practiced problem speech sounds in words occurring with low frequency, before going on to practice target speech sounds in high frequency words. Word frequency is a key concept of usage-based theory: the more frequent a word/string of words, the more entrenched the motor/perceptual memory becomes for that word/string of words (Bybee, 2001; Menn et al., 2013). Consequently, it was hypothesised that low frequency words may be easier for the patient to produce in the early stages of therapy, as they are less entrenched in memory, while in the later stages, high frequency words should expedite generalisation of target speech sounds through collateral neural activation. A further core concept of usage-based phonology therapy is the notion of gradience (Bybee, 2001, 2010). Phonological change is conceived to occur in a gradual way, with gradient phonetic adjustment occurring in words/production tokens over time. Thus, with this technique gradient articulatory change was promoted and expected.

Focus on production of connected speech patterns: This modified EPG technique taught and practiced use of connected speech patterns in sentences. Since use of atypical connected speech patterns can have a negative effect on speech understandability and speech acceptability (Howard, 2013), it was predicted that teaching connected speech patterns would have a positive effect on speech understandability and acceptability.

The usage-based therapeutic technique, as used in this study, is described in detail below.

5.3.1 Initial stages of therapy

In the first session, the researcher explained and demonstrated how the EPG system (WinEPG, Articulate Instruments Ltd, 2008) works, i.e. different tongue-palate contact corresponds to different patterns on the visual display unit.

EPG visual feedback was then used to facilitate production of problem speech sounds. The researcher went through each speech sound produced atypically and attempted to elicit the sound. This elicitation was attempted through working through the following steps. The first three elicitation techniques are described in previous literature (Gibbon & Wood, 2010; Hardcastle et al., 1991; Lee et al., 2007), the final one is novel to this approach:

- Production in isolation (i.e. single sound), possibly with an initial intermediate step of getting silent posturing, e.g. getting and holding a horse-shoe shaped EPG pattern with tongue contact with the upper dental arch as an intermediate step for production of /t/ and /d/;
- Adaption of existing speech sounds at single sound level, e.g. producing a /ʃ/, then bringing tongue forward to achieve a /s/;
- Production of target speech sounds at syllable level, using facilitating vowel contexts, e.g. production of /k/ with a facilitative high vowel /i/ in /ki/;
- Adaption of existing speech sounds at word level, e.g. replacing /s/ with /t/ in “soap”.

The researcher selected the lingual speech sound that was the mostly easily elicited (using the above techniques), and this sound was the first target for therapy.

In the first session, the patient was asked to focus on the visual representation accompanying the speech sound production together with the associated proprioceptive/sensory feedback, rather than auditory perception (e.g. “Try to match my display of lights. Can you feel where your tongue is when those sensors light up?”). In addition, the researcher’s spoken models were kept to a minimum at this stage, so that the patient did not receive any auditory input. This focus on output was because therapy is aiming to achieve new motor memory traces. Usage-based phonology suggests old, inaccurate motor memory traces will be closely linked to auditory memory traces (Menn et al., 2013). Therefore, directing attention away from hearing may assist with new motor traces.

Within the space of one session practice of a target speech sound occurred at word level, even if production of the target speech sound did not exactly match the adult realisation, in terms of placement, voicing and manner. Initially low frequency words were practiced. Low frequency words are words occurring with low frequency in the ambient language (see 5.4.4 for further description and 11.4, Appendix D for low frequency English words used in this study). In addition, therapy specifically focused on those low frequency words whose phonetic contexts were most facilitative to the patient. Facilitative contexts were identified from initial assessment. Some variability of production of target words, in terms of placement, voicing and manner, was allowed and expected. However, therapy worked towards more accurate production of target speech sounds as therapy progressed. For example, initially a [t̪] for a /s/ target within a word might be accepted, then a heavily aspirated [t̪^{h}] accepted, before moving onto a [s̪], then finally a [s]. The patients/patient’s carers were counselled that initial productions were intermediate steps towards a match with the adult realisation. This progressive approximation of words is unique to this treatment approach and is based on the usage-based phonology principle of gradience, as discussed above. If the patient struggled to produce successively closer productions of the target sound at word level within one session, then the researcher moved to another target lingual sound, and went back to the previous speech sound target at a later date.

Once the patient was able to produce the low frequency target words with 90% accuracy, with adult realisation of the target speech sound (i.e. use of allophone productions typically of the participant’s dialect) using EPG feedback, these words were then practiced without the EPG palate. When the participant was able to produce the low frequency target words with 90% accuracy without EPG feedback, high frequency words were practiced with EPG feedback. High frequency words are words occurring many times in the ambient language (see 5.2.3. for further description and 11.4, Appendix D, for high frequency English words used in this study). Once the participant was able to produce the high frequency target words with 90% accuracy with EPG feedback, these words were practiced without the EPG palate.

5.3.2 Next stages of therapy

Once the patient was able to produce the first target speech sound at word level, the researcher then targeted another speech sound produced in error, and therapy followed the stages listed in the previous section.

Once all target speech sounds were produced at word level practice moved onto production of the target sounds at connected word level. The patient practiced the target speech sound in connected sentence tasks, such as reading and event retell. Normally, the patient did not wear their EPG palate during these tasks. The researcher monitored the patient's speech production during sessions through listening to the patient's connected speech and gave feedback. If speech sound errors were made, feedback was given on these. Initially this feedback was specific, e.g. "You used your old "s" sound at the end of that sentence", before providing indirect feedback, e.g. "Pardon? Say that again". If atypical connected speech patterns were evident, these were worked on directly in the following way. The researcher explained to the patient that speech sounds in words can change when they are produced in sentences. The researcher gave examples of open connected speech patterns (i.e. patterns that serve to make a clear separation between two words), such as pausing between words, and then examples of closed connected speech patterns, i.e. patterns which serve to bind sounds/words together, such as elision and assimilation) (Wells, 1994). If the patient over-used open connected speech patterns, closed patterns were taught. Likewise, if closed juncture patterns were over-used, open patterns are taught. Initially the patient produced these patterns at sentence level given a direct model from the researcher (e.g. relating to a /s/ target, "He was sad" [hiwɑsæd], where /z/ is assimilated to /s/), then the sentence was practiced with a delayed model, then without any modelling. Use of appropriate connected speech patterns were reinforced verbally, e.g. "Good you combined those words together smoothly". In addition, the patient was expected to monitoring their production through auditory and proprioceptive feedback.

In each session, at least 200 production trials were expected. In this study, an average around 250 production trials occurred for all participants over the course of their treatment (as calculated through tallying of production trials during each session).

Throughout intervention, more typical productions were encouraged, while unwanted responses were discouraged, in the manner of operant learning (Scherer & Kaiser, 2010). Initially this feedback was very specific; over time this feedback became less specific and less regular.

5.3.3 Home practice

Participants were expected to practice targets worked on in therapy sessions at home at least 5 times per week for at least 10 – 15 minutes per home practice episode. This practice was carried out using a portable EPG machine until the participant could produce all speech sounds produced in error accurately in high frequency words. Once high frequency words could be produced, home practice continued without a portable EPG machine.

5.4 Procedure

This section outlines the study's procedure including: speech recording and blinding of independent SLTs; initial assessment; target selection following initial assessment; baseline assessment; intervention; home practice; and post-treatment assessment.

5.4.1 Speech recording

Participants' speech was video- and audio- recorded on initial assessment and at all test points throughout the ABA phases of the Study (see Table 4). All speech results reported in this thesis derive from ratings taken from speech recordings. The study's recording procedure followed recommendations for recording cleft palate speech, as described by the Cleft Palate International Speech Issue (CLISPI) website.

Speech was recorded using the following sound and visual equipment:

- JVC-GY-HM100E Solid state video recorder and Rode NT4 microphone. Speech was videoed using MP4 format.
- Zoom H4next audio recorder and Rode NT4 microphone. This audio recording was in uncompressed WAV format.

The camera was positioned to point directly at the participant's face. The two external microphones (Rode NTE microphone) were placed at a distance of 40 cm from the participant. A blue backdrop was used for the video recording in order that a clear image of the participants face was recorded with no visual distraction. The participant's head and shoulders were within the frame of the recording so articulatory movements of consonants were be visible and could be assessed. Participants were instructed to look directly at the camera while speaking.

Phonetic symbols and diacritics used by the extended International Phonetic Alphabet (Full IPA Chart, 2015), together with phonetic symbols and diacritics described and developed for transcribing cleft palate speech (Sell et al., 2009) were used for all phonetic transcription in this study.

5.4.2 Blinding of independent SLTs

The two independent SLTs blindly listened to speech recordings at the three baseline assessments and the two post-therapy assessments. The independent SLTs listened to one randomly selected assessment session, before listening to another, i.e. time points were randomised, rather than individual items. Randomisation was carried out through the following process: all time-points were written onto separate cards and put into a bag. A person unrelated to the study blindly pulled out each card to produce an order for listening and rating.

The procedure for listening to untreated and untreated words, and rating speech understandability and speech acceptability was as follows: Wearing headphones the SLT first watched the event retell and then made a rating of speech understandability. The SLT then watched the videos of the participant reading the treated and untreated words list. Words perceived to be accurate were given a tick, atypical productions were phonetically transcribed. Finally, the SLT made judgement on speech acceptability. The SLT was able to listen to connected speech samples and single word reading as many times as they wanted.

5.4.3 Initial assessment

Once written consent was obtained, each participant attended an initial assessment appointment with the researcher at the hospital. The initial assessment battery included: GOS.SP.ASS, DEAP, phonology sub-section, 2-minute spontaneous connected speech sample, ICS, and SPAA-C or Dys-QoL, the later depending on the participants age (see Table 4)

5.4.4 Target selection following initial assessment

Initial assessment on GOS.ASS.ASS and DEAP identified which speech sounds the participants produced with 0 – 10 % accuracy, and which speech sounds could potentially be assisted with EPG (i.e. lingual speech sounds that were produced incorrectly in terms of placement and manner).

Following this identification, a list of 16 treated and 16 untreated words for each speech sound produced in error was developed for each participant. Treated and untreated words were matched on the following criteria: Word position; word frequency; word length, and vowel.

5.4.4.1 Word position

The target lingual speech sound was in word initial position in eight of the 16 words, while the remaining eight words the target sound was in word final position. This mix provides a representation of speech sounds in everyday talking. Word medial position was not included to keep the word list to an acceptable length in testing.

5.4.4.2 Word frequency

Eight words were high frequency (four word initial, four word final), and eight words were low frequency (four word initial, four word final). Words were selected from a spoken and written word frequency corpus published by Leech et al. (2001). Words with a frequency of less than 100 (rounded frequency per million word tokens) were classified as low frequency words, while words with a frequency of more than 100 were classed as high frequency words. Control of word frequency in this study was included since usage-based theory suggests low frequency words are easier to learn, while high frequency words may be important for generalisation at the later stages of therapy (see sections 4.2.2 for further discussion).

5.4.4.3 Word length

Treated and untreated words were one to two syllables in length and consisted of CV (consonant, vowel), CVC (consonant, vowel, consonant), CVCV (consonant, vowel, consonant, vowel), CVCVC (consonant, vowel, consonant, vowel, consonant) combinations. Word lists were matched for word length, and syllable shape as these two factors may impact on production of speech sounds.

5.4.4.4 Vowel

Words were matched for vowels occurring close together in terms of the English vowel chart, i.e. matched in terms of mouth opening and position of tongue in mouth. Treated and untreated word lists were matched for vowels, as vowel position may impact on production of speech sounds.

Treated words were used in therapy, while untreated words were never used in therapy and were used to measure lexical generalisation and maintenance. /z/ word lists consisted of 12 untreated and 12 treated words (rather than 16) due to no high frequency word initial words being available. /ʃ/ word lists consisted of 15 untreated and 15 treated words due to only three high frequency word final words being available. Word lists for each lingual speech sound are shown in 11.4, Appendix D.

5.4.5 Baseline assessment

Following initial assessment, participants then attended three subsequent appointments to fit the dental plates (training plate and EPG palate) required for EPG therapy. These appointments were 3 – 8 weeks apart. During these appointments, baseline testing occurred. Baseline testing involved the following: reading the treated word list; reading the untreated (probe) word list; and a 2-minute spontaneous connected speech sample (see Table 4). Speech was recorded using the procedure described in 5.4.1. On the final baseline, baseline 3, testing also included EPG assessment, GOS.SP.ASS and ICS. Throughout the entire study, the EPG palate was only ever worn during EPG assessment, i.e. for all other speech assessments the EPG plate was not worn.

5.4.6 Intervention

Therapy commenced immediately following baseline 3 assessments. Participants were offered therapy once a week at the hospital in a cleft SLT treatment room and were asked to carry out speech practice at home for 10 – 15 minutes five times per week. The researcher delivered the therapy and sessions lasted between 50 – 60 minutes. The intervention delivered is described in section 5.3 of this chapter. Each participant was asked to wear their training plate for 1-2 hours prior to each therapy session so that they were accommodated to having a plate in the mouth.

At the end of every treatment session, the participant was asked to read aloud words from the treated word list which related to the speech sound targeted in therapy during that session. For example, if the participant worked on production of /s/, the /s/ therapy words were named. The participant's speech was recorded in the manner described in 5.4.1 and was later transcribed by the researcher in the way described in 5.4.1.

At the end of every fifth treatment session, the participant underwent the same assessments as carried out in the baseline phase (see Table 1):

- Single word reading of treated words;
- Single word reading of untreated words;
- 2-minutes of spontaneous, connected speech.

This speech was recorded and was transcribed at a later time by the researcher in the way described in 5.4.1. In addition, EPG assessment data was collected at the end of every fifth session of therapy, as described in section 5.2.6 (see Table 4).

Therapy continued until such time as:

- All lingual (tongue) speech sounds were being used correctly in connected speech (as assessed by the researcher on listening to the 2-minute sample of spontaneous speech);
- Despite initial improvements, progress had stopped with no subsequent improvement over eight sessions (as assessed by the researcher on assessment tasks at the end of each therapy session);
- No improvement had occurred over eight sessions (as assessed by the researcher on assessment tasks at the end of each therapy session).

5.4.7 Home practice

Speech practice done at home was recorded by the participant/participant's parent or caregiver on a written sheet and was collected by the researcher to assist with monitoring adherence to home practice.

5.4.8 Post-treatment assessment

Immediately following completion of therapy, and then 3-months following cessation of therapy, the participants were reassessed on (see Table 4): GOS.SP.ASS; single word picture naming from the phonology sub-test of the DEAP; 2-minutes of spontaneous, continuous speech where the participant is asked to recount what they did the day before; ICS; SPAA-C questionnaire (for children) or QoL-Dys (for young people and adults); reading treated words; reading untreated words.

5.5 Data analysis

This section outlines how data was analysed to address each research question.

Research question 1: Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated words/pseudo- words.

To answer this question, percentage of targets correct (%TC) in treated words at the three baseline assessment and two post-therapy assessment time-points is displayed in a line graph for each participant in the results chapter (Figure 9 – 16). Visual analysis of this line graph allows for identification of improvements with %TC. Likewise, %TC in untreated words at the same test points is displayed in a line graph for each participant in results (Figure 9 – 16) and visual analysis is carried out in the same way. In addition, an adaption of the effect size (ES) index of standard mean difference with correction for continuity, *d*, (Gierut et al., 2015) is calculated for untreated words, %TC, and is shown in Figure 9 - 16. The effect size (ES) index of standard mean difference with correction for continuity, *d*, statistic is used with within-participant studies where there is a lack of independence between samples (e.g. speech samples from the same individual at different time-points). Since it is unit-free, this index permits comparison across different studies. This statistic is computed as follows: Mean accuracy on pre-intervention baselines is calculated, together with mean accuracy on assessment during and post-intervention assessment. The difference between these means is then divided by a pooled standard deviation for pre-intervention baselines. Benchmarks for interpretation of this statistic for SSD outcome studies are shown in Table 8 (Gierut et al., 2015).

Table 8

Interpretation of effect size index of standard mean difference with correction for continuity for SSD outcome studies

Score	Effect size for learning
1.4	Small
3.6	Medium
10.1	Large

This study used an adaption of this ES statistic, since independent SLT assessment was not completed mid-therapy. In this study ES statistic was computed as follows: Mean accuracy on pre-intervention baselines was calculated, together with mean accuracy on post-intervention assessment. The difference between post-intervention and pre-intervention means was then divided by a pooled standard deviation for pre-intervention baselines. The pooled standard deviation is derived from the mean baselines of the 5/6 participants of this study. The sixth participant (Poppy) was excluded from the pooled standard deviation as the independent SLTs transcribed this participant's production of /n/ and /l/ with high levels of accuracy. However, EPG analysis showed consistent velar tongue-palate contact for these target sounds in pseudo-words. This removal is in line with Gierut et al. (2015)'s study, where outliers who posed a risk to the index's internal and external validity were removed. Since this ES statistic has been adapted for this study, caution is needed in using the benchmarks listed in Table 8. This point is discussed further in Chapter Eight.

Research question number one is also addressed through examination and calculation of EPG data/indices. Cumulative EPG palatograms are shown on %TC line graphs across the intervention period and permit visual analysis of changes to tongue-palate contact (Figure 15 - 22). In addition, EPG indices over test points are shown in Figure 28 in the results, and the Wilcoxon signed-rank test is used to determine statistically significant differences (Table 10). The Wilcoxon signed-rank test was selected as EPG indices were not normally distributed, therefore a non-parametric test was used.

Research question 2: Does improved accuracy of target phonemes generalise to continuous, connected speech and, if so, does this result in improved ratings of speech intelligibility and improved ratings of impact on quality of life, post-therapy?

To answer this question, %TC was computed for the continuous connected speech sample at all test points (B1, B2, B3, A5, A10…P, M) and this is presented in a line graph and is shown in the results (Figure 21). Visual analysis of this line graph allows for identification of any improvements with targets correct in connected speech. To measure any changes to speech understandability and acceptability, ICS scores at each test point (IA, B3, P, M) and speech understandability and speech acceptability scores at each test point (B1, B2, B3, P, M) are presented in a graph and are shown in results (Table 15 - 18). Likewise, scores on the quality of life measures at each test point (IA, P, M) are presented in a table (Table 19) and are described in the text in Chapter Six.

Research question 3: Do improvements with accuracy of targeted phonemes following therapy provide support for the usage-based theoretical model in general and more specifically in terms of:

> **a). Gradient phonetic speech change with target phonemes is shown over the course of therapy, with target production gradually moving from error production to production within normal parameters in the participants' dialect;**
> **b). A word frequency effect is shown with low frequency words being easier for participants to learn compared to high frequency words?**

Support for the theoretical model in general comes from answers to research questions one and two, as described above (i.e. did the usage-based phonology technique produce change in speech sound production, and did this occur in a manner predicted by the theoretical model?). To answer part a). of this question relating to gradient phonetic speech change, cumulative EPG frame pictures are examined over each test point (B3, A5, A10...P and M), in addition to examination of narrow phonetic transcription at each test point (Figure 22 – 29).

To answer part b). the word frequency effect is examined by comparing the %TC for low frequency and high frequency words at test points (A5 and A10) and the percentages are presented in a graph and are shown in results (Table 13). A Wilcoxon signed-rank test is used to determine statistical significance. In addition, the word frequency effect is examined in continuous connected speech samples by comparing the %TC for low frequency and high frequency words at test points (Table 14). Again, a Wilcoxon signed-rank test is used to determine statistical significance.

6 RESULTS – IMPRESSIONISTIC PERCEPTUAL SPEECH AND QUALITY OF LIFE OUTCOMES

The results of this study are divided into two parts. Chapter Six presents impressionistic perceptual speech outcomes together with quality of life outcomes. Chapter Seven presents EPG outcomes. Chapter Six begins with a description of the six participants' speech on initial assessment. Section two of this chapter presents the impressionistic perceptual speech findings prior to, during and post-therapy. These perceptual results address the study's first research question relating to acquisition and generalisation. Section three of this chapter provides information on speech understandability, speech acceptability, and quality of life measures pre- and post-therapy. These findings address the study's second research question concerning functional generalisation and impact on quality of life. Chapter Seven presents EPG findings prior to, during and post-therapy. EPG results contribute further to the study's first research question regarding acquisition and generalisation. The study's third research question, concerning support for the theoretical model, is addressed in both results chapters.

6.1 Initial assessment

Participants' speech on initial assessment (PCC on phonology subtest of the DEAP and findings from GOS.SP.ASS) is shown in Table 9 and is described below. Table 9 also includes findings from repeat assessment at the end of therapy and at maintenance assessment, 3-months post-therapy; these are discussed in 6.2.5. All initial assessment speech analysis was completed by the researcher.

6.1.1 Resonance and nasal airflow

As shown in Table 9, speech resonance was assessed as normal for Emma and Poppy on initial assessment. George, Olivia and Grace showed hyernasality on closed vowels, i.e. mild hyponasality and Victoria showed hypernasality on open and closed vowels, i.e. moderate hypernasality. In terms of nasal airflow, two participants, George and Grace, showed slight nasal turbulence. It was more difficult to assess Victoria's nasal airflow as she produced all oral pressure consonants weakly. The three remaining participants did not present with any errors with nasal airflow on initial assessment.

6.1.2 Consonant production

Participants presented with a range of cleft speech characteristics on initial assessment. See Table 10 for a summary of participants' main cleft speech characteristics and Table 9 for detailed transcriptions.

Table 9

Participants' resonance, nasal airflow and consonant production on GOS.SP.ASS and PCC on the phonology subtest of the DEAP on initial assessment, end of therapy assessment and maintenance assessment

a). George

Partic.	Assessment	Resonance	Nasal Airflow	Consonant Production																					PCC on DEAP
George	Initial assessment	Grade 1 hyponasality (nasal consonants slightly denasal)	No nasal emission Grade 1 nasal turbulence (slight nasal turbulence)		Labial					Alveolar						Post-alveolar			Velar			Glottal			72.7 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h		θ/ð	
				SIWI										s̪͆ / s̪͆ˈ	ɮ	ɬ	ɬ	ɮ						f/v	
				SFWF										s̪͆	ɮ	ɬ	ɬ	ɮ						f/v	
	End of Therapy assessment	Grade 1 hypernasal-ity (hypernasal-ity heard on closed vowels)	No nasal emission Grade 2 nasal turbulence (marked nasal turbulence)		Labial					Alveolar						Post-alveolar			Velar			Glottal			95.8 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h		θ/ð	
				SIWI										s̪	z̪		tʃ /tj	dʒ /dj						f/v	
				SFWF										s̪	z̪		tʃ /tj	dʒ /dj						f/v	
	Mainten. assessment	Grade 1 hypernasal-ity (hypernasal-ity heard on closed vowels)	No nasal emission Grade 2 nasal turbulence (marked nasal turbulence)		Labial					Alveolar						Post-alveolar			Velar			Glottal			94.4 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h		θ/ð	
				SIWI										s̪	z̪			dʒ /dj						f/v	
				SFWF										s̪	z̪			dʒ /dj						f/v	

b). Olivia

Partic.	Assessment	Resonance	Nasal Airflow	Consonant Production																				PCC on DEAP
Olivia	Initial assessment	Grade 1 hyponasality	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		79 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI										ʃ	ʒ		ʃ						f/v	
				SFWF										ʃ	ʒ		tʃ						f/v	
	End of therapy assessment	Grade 1 hyponasality	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		94.4 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI																				
				SFWF																				
	Mainten. assessment	Grade 1 hyponasality	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		94.4 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI																				
				SFWF																				

c). Emma

Partic.	**Assessment**	**Resonance**	**Nasal Airflow**	**Consonant Production**																				**PCC on DEAP**
Emma	Initial assessment	Normal	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		52%
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI						ŋ	ɰ	k	g	ɬ	ɬ̬	ɬ	kɬ	gɬ̬					d/v	
				SFWF						ŋ	ɰ	k	g	ɬ	ɬ̬	ɬ	kɬ	gɬ̬					d/v	
	End of therapy assessment	Grade 1 hypernasal-ity (hypernasal-ity heard on closed vowels)	No nasal emission Grade 1 nasal turbulence		Labial					Alveolar						G			Velar			Glottal		92.3 %
					m	p	B	F	v		l		d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI																			θ /v	
				SFWF																			θ /v	
	Mainten. assessment	Normal	No nasal emission Grade 1 nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glotta		93.7 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI													tʃ /tj						θ/v	
				SFWF													tʃ						θ/v	

d). Victoria

Partic.	Assessment	Resonance	Nasal Airflow	Consonant Production																				PCC on DEAP
Victoria	Initial assessment	Grade 1 hyper-nasality (heard on vowels)	No nasal emission No nasal turbulence (but weak production of oral pressure consonants)		Labial					Alveolar						Post-alveolar			Velar			Glottal		70%
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI										s̪͡ħ	n	ħ	ħ	nj						
				SFWF										s̪͡ħ	n	ħ	ħ	ʔ						
	End of therapy assessment	Grade 2 hyper-nasality (heard on vowels and voiced oral pressure consonants)	Grade 1 nasal emission No nasal turbulence (but weak production of oral pressure consonants)		Labial					Alveolar						Post-alvolar			Velar			Glottal		97.2 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI											s			dʒ / tʃ						
				SFWF											s			dʒ / tʃ						
	Mainten. assessment	Grade 2 hyper-nasality (heard on vowels and voiced oral pressure consonants)	Grade 1 nasal emission (slight) No nasal turbulence (but weak production of oral pressure consonants)		Labial					Alveolar						Post-alveolar			Velar			Glottal		96.5 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI											s			dʒ						
				SFWF											z			dz						

e). Grace

Partic.	Assessment	Resonance	Nasal Airflow	Consonant Production																				PCC on DEAP
Grace	Initial assessment	Grade 1 hyponasality (nasal consonants slightly denasal	No nasal emission Grade 1 nasal turbulence (slight and inconsistent		Labial					Alveolar						Post-alveolar			Velar			Glottal		87%
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI										ç	ʝ									
				SFWF							j			ç	ʝ									
	End of therapy assessment	Normal	No nasal emission Grade 1 nasal turbulence (slight)		Labial					Alveolar						Post-alveolar			Velar			Glottal		99.3 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	k	g	h	θ/ð	
				SIWI										s̪	z̪									
				SFWF										s̪	z̪									
	Mainten. assessment	Grade 1 hypernasal-ity (hypernasal-ity perceived on closed vowels)	No nasal emission Grade 1 nasal turbulence (slight)		Labial					Alveolar						Post-alveolar			Velar			Gltta		100%
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	K	g	h	θ/ð	
				SIWI										s̪	z̪									
				SFWF										s̪	z̪									

f). Poppy

Partic.	**Assessment**	**Resonance**	**Nasal Airflow**	**Consonant Production**																				**PCC on DEAP**
Poppy	Initial assessment	Normal	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		79%
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	K	g	h	θð	
				SIWI						ŋ	ɰ	t̪	d̪	s̪	z̪								f/l	
				SFWF						ŋ	ɰ	t̪	d̪	s̪	z̪								f/l	
	End of therapy assessment	Normal	No nasal emission No nasal turbulence		Labial					Alveolar						Post-alveolar			Velar			Glottal		97.2 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	K	g	h	θð	
				SIWI						n	l, l̪	t̪	d̪											
				SFWF						n	l	t̪	d̪											
	Mainten. asseessment	Normal	No nasal emission No nasal turbulence		Labal					lveolar						Post-alveolar			Velar			Glottal		96.5 %
					m	p	b	f	v	n	l	t	d	s	z	ʃ	tʃ	dʒ	ŋ	K	g	h	θð	
				SIWI						n	l, l̪	t̪	d̪											
				SFWF						n	l	t̪	d̪											

Note: Green shading indicates consonants present in participants' inventory with sentence repetition on GOS.SP.ASS; GOS.SP.ASS = Great Ormond Street Speech Assessment; DEAP = Diagnostic Evaluation of Articulation and Phonology; SIWI = Syllable initial, word initial; SFWF = Syllable final, word final; Manten. = maintenance; Partic. = Participant; PCC = Percentage of consonants correct.

Table 10

Summary of participants' main cleft speech characteristics at word level

Participant	Main cleft speech characteristics at word level
George	Lateral production of lingual sibilants
Olivia	Post-alveolar production of /s/ and /z/
Emma	Backing and lateral production of lingual speech sounds
Victoria	Non-oral production of lingual sibilants
Grace	Palatal production of /s/ and/z/
Poppy	Backing of /n/ and /l/

George's main cleft speech characteristics on initial assessment included lateral release of alveolar and post-alveolar sibilants, i.e. /z/ → [ɮ], /ʃ/ → [ɬ], /tʃ/ → [ɬ] and /dʒ/ → [ɮ] (single word naming, sentence repetition on GOS.SP.ASS and connected speech).

Olivia's speech was characterised by patterns of alveolar to post-alveolar placement, deaffrication, i.e. /s/ → [ʃ], /z/ → [ʒ], /tʃ/ → [ʃ] (single word naming and sentence repetition on GOS.SP.ASS). In connected speech, Olivia's cleft speech characteristics were more extensive and /s/, /z/, and /ʃ/ sounds were realised as [ħ], /tʃ/ as [ʃ] respectively and at times /t/ and /d/ were produced as [ħ].

Emma showed the largest number of cleft speech characteristics on initial assessment. Her production was characterised by a backing pattern of alveolar to velar placement and lateral release of alveolar and post-alveolar sibilants, i.e. /n/ → [ŋ], /l/ → [ɰ], /t/ → [k], /d/ → [g], /s/ → [𝼄] (voiceless velar lateral), /z/ → [𝼄̬] (voiced velar lateral), /ʃ/ → [𝼄], /tʃ/ → [k 𝼄], and /dʒ/ → [g𝼄̬] (single word naming, sentence repetition on GOS.SP.ASS and connected speech).

Victoria's cleft speech was characterised by a pattern where lingual sibilants were typically replaced by non-oral sounds, i.e. /s/ → [s̡͡ħ], /z/ → [n], /ʃ/ → [ħ], /tʃ/ → [ħ], /dʒ/ → [nj]. Victoria also presented with weakened production of oral pressure consonants, i.e. without complete closure (single word naming, sentence repetition on GOS.SP.ASS and connected speech).

Grace showed palatal production of /s/ and /z/, i.e. /s/ → [ç] and /z/ → [ʝ] (single word naming, sentence repetition on GOS.SP.ASS and connected speech).

Poppy's main cleft speech characteristics included backing, with /n/ → [ŋ], and /l/ → [ɰ]. In addition, she evidenced dental production of /t/ and /d/ and interdental production of /s/

and /z/ (single word naming and sentence repetition). In connected speech, Poppy's speech sound errors were more extensive and in addition to a backing pattern, lingual sibilants were often produced with non-oral productions, i.e., /s/, /z/, /ʃ/, /ʒ/, /tʃ/ and /dʒ/ → [ħ].

Initial assessment of consonant production identified which speech sounds could potentially be assisted with EPG (i.e. lingual speech sounds that were produced incorrectly in terms of placement and manner). Table 11 shows speech sounds targeted in therapy for each participant.

Table 11

Lingual speech sound targets for each participant

Participant	Speech targets
George	z, ʃ, tʃ, dʒ
Olivia	tʃ, s, z
Emma	n, l, t, d, s, z, ʃ, tʃ, dʒ
Victoria	s, z, ʃ, tʃ, dʒ
Grace	s, z
Poppy	n, l

6.1.3 Percentage of consonants correct on DEAP

Participants' PCC on the DEAP assessment ranged from 52% to 87% (see Table 9).

6.2 Impressionistic perceptual speech outcomes

Change in percentage of targeted sounds correct prior to, during and post-treatment was examined through impressionistic phonetic transcription (treated words, untreated words and connected speech) and EPG analysis (pseudo-words). Impressionistic perceptual speech outcomes are discussed here under four sub-headings: Percentage of targets correct with single word production (treated and untreated words) at the three baseline pre-therapy (B1, B2, B3) and the two assessments post-therapy (P, M); Percentage of targets correct for treated words during the treatment phase; Percentage of targets correct in continuous connected speech at the three baseline assessments, assessment at the end of every fifth treatment session (A5, A10, A15 and so on) and the two assessments post-therapy; and PCC on phonology subtest of the DEAP and findings from GOS.SP.ASS on the two assessment post-therapy. The results outlined in this section contribute to answering research questions number one and number two: **Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated words/pseudo-words? Does improved accuracy of target phonemes generalise to continuous connected speech?** In addition, results in this section contribute to answering research question number three: **Do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in terms of a word frequency effect?**

6.2.1 Percentage of targets correct at the three baselines, at the end of therapy and 3-months post-therapy

Percentage of targets correct in treated words at the three baseline assessments and the two assessments post-therapy are shown for each participant in Figures 9a, 10a, 11a, 12a, 13a, 14a. Percentage of targets correct is derived from independent SLT analysis (average of scores from the two independent listeners). Likewise, percentage of targets correct in untreated words over the same assessment time-points are shown for each participant in Figures 9b, 10b, 11b, 12b, 13b, 14b. Again, percentage of targets correct derives from independent SLT analysis. Figures showing percentage of targets correct in untreated words also include calculation of effect size. Percentage of targets correct increased for all target sounds, for all participants, in both treated and untreated words, as discussed further below.

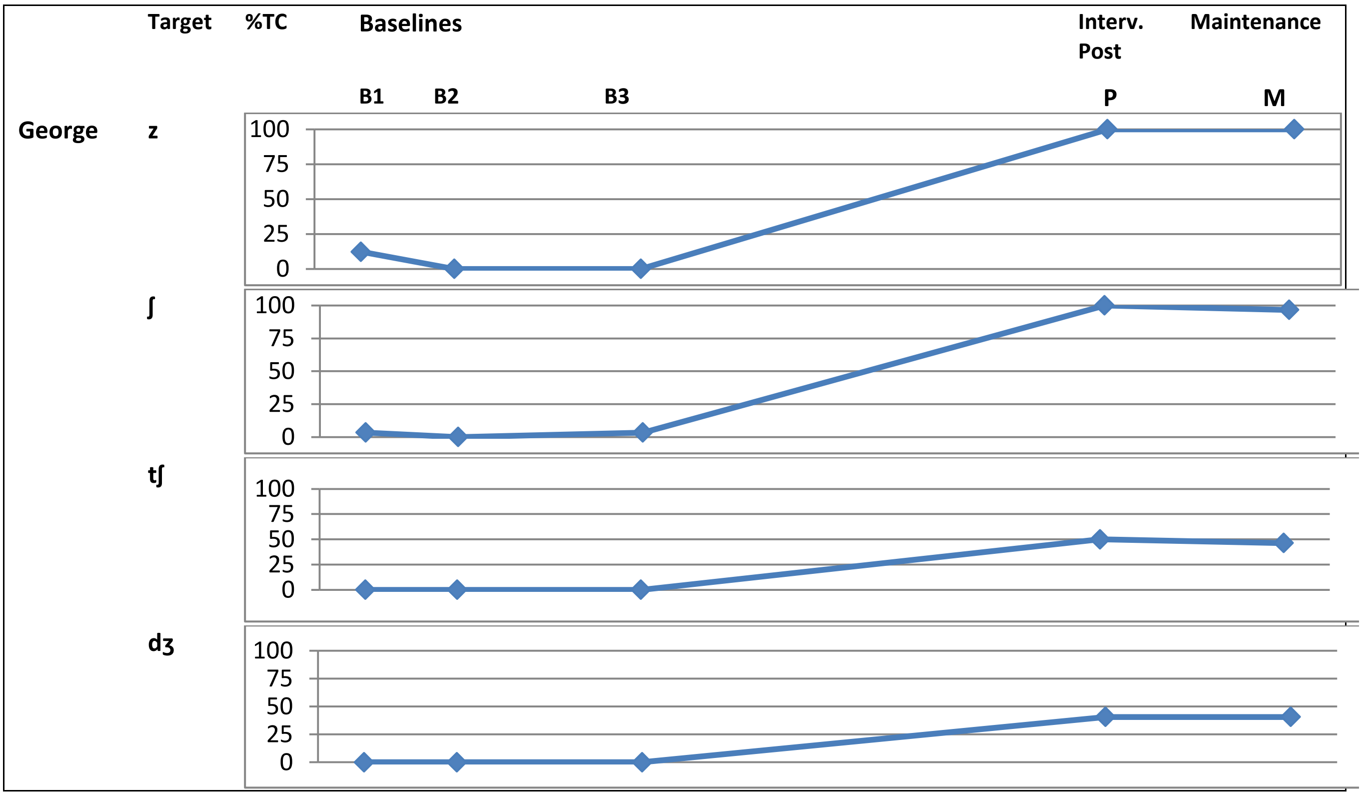
At the three baseline assessments George scored near 0% accuracy for production of /z/, /ʃ/, /tʃ/, and /dʒ/ in treated and untreated words (see Figure 9 a and b). Independent SLT transcription matched that of the research SLT; these sounds were transcribed as alveolar lateral sounds (/ʃ/, /tʃ/ → [ɬ]; /dʒ/ → [ɮ]). Immediately following therapy (P) he scored 100% or near 100% accuracy for /z/ and /ʃ/ in both treated and untreated words. This high level of accuracy was sustained on maintenance assessment (M). He performed less well with /tʃ/ and /dʒ/. These speech sounds were assessed as correct around 50% of the time at the end of treatment and on maintenance assessment for both treated and untreated word production. However, /tʃ/ and /dʒ/ words assessed as incorrect by the independent listeners showed improvement from the three baseline measures: these sounds were transcribed as [tj] and [dj] respectively, i.e. they were no longer produced with lateral release and were impressionistically similar to typical speech.

Visual analysis of the line graphs in Figure 9 a and b shows George's production of untreated words was much the same as production of treated words on post-therapy assessments (P and M). This visual analysis was confirmed statistically. A Wilcoxon signed-rank test showed no significant difference in percentage of targets correct for treated and untreated words for /z/ ($Z = -1.41$, $p = .157$), /ʃ/ ($Z = -1.41$, $p = .157$), /tʃ/ ($Z = -.45$, $p = .655$) and /dʒ/ ($Z = -1.34$, $p = .180$) (see Table 12). Lack of difference indicates improvement with %TC with treated words was matched by a similar level of improvement with untreated words. Thus transfer of learning from treated to untreated words is inferred. Large treatment effect sizes were shown for /z/ and /ʃ/, and medium to large effect sizes were seen for/tʃ/ and /dʒ/, using Gierut et al. (2015) benchmarks (see Table 8).

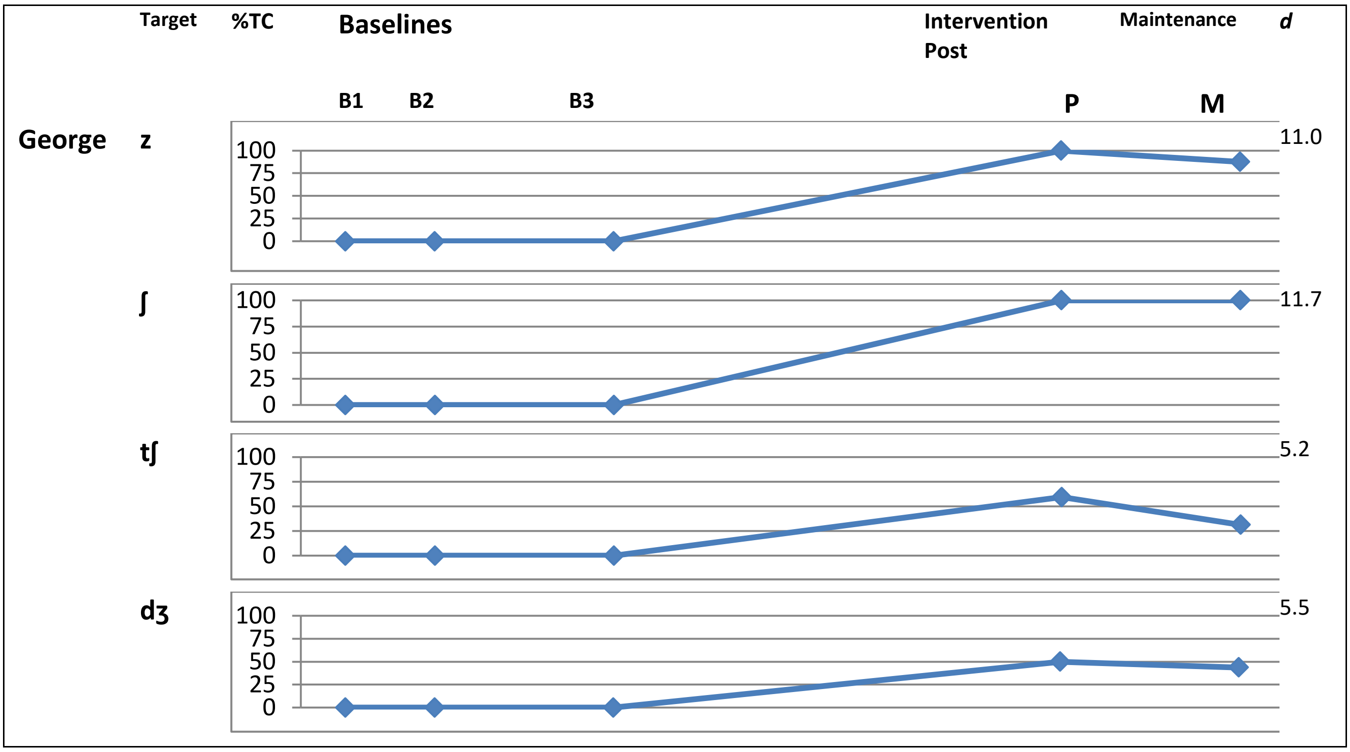
Figure 9

George's percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners' scoring (average score)

a) Treated words



b). Untreated words



Note: %TC = Percentage of targets correct; B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

Table 12

Differences in percentage of targets correct for treated and treated words (Wilcoxon sign-ranks test) to show if lexical generalisation from treated to untreated words occurred

Participant	Speech sound target	Percentage Targets Correct Mean			
George		Treated	Untreated	Z score	p value
	z	42.50	37.50	-1.41	.157
	tʃ	19.28	18.12	-.45	.655
	dʒ	16.25	18.75	-1.34	.180
	ʃ	41.33	40.00	-1.41	.157
Olivia	s	47.50	42.95	-.82	.414
	z	37.50	39.16	-.27	.786
	tʃ	37.5	43.75	-1.63	.102
Emma	n	60.64	48.75	-.730	.465
	t	50.62	48.12	-1.07	.285
	d	46.87	48.12	.00	1.000
	l	57.49	55.62	-.53	.593
	s	39.37	39.37	.00	1.000
	z	34.16	35.00	-.45	.655
	ʃ	40.00	39.33	-.27	.785
	tʃ	40.00	36.87	-1.34	.180
	dʒ	38.75	38.75	-1.60	.109
Victoria	s	41.20	38.80	-1.41	.157
	ʃ	43.75	46.65	-.53	.593
	z	30.83	34.16	-.82	.414
	tʃ	41.26	39.37	-.27	.785
	dʒ	33.00	30.64	.00	1.000
Grace	s	40.00	40.00	.00	1.000
	z	40.00	40.00	.00	1.000
Poppy	n	69.25	65.22	-.40	.686
	l	87.60	88.2	-1.73	.083

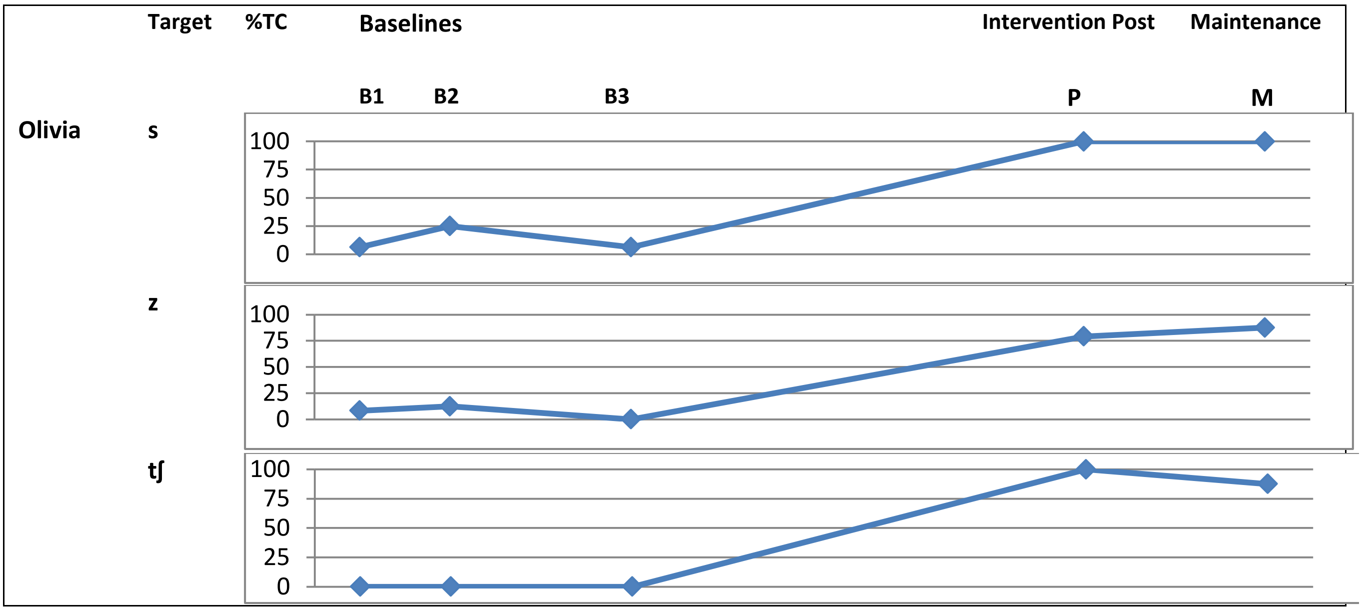
Note: Percentage of targets correct come from independent speech and language therapists' analysis at baseline 1, baseline 2, baseline 3, post-intervention and maintenance assessment.

At the three baseline assessments Olivia scored between 0 – 25% accuracy on production of treated and untreated /s/, /z/, and /tʃ/ (see Figure 10 a and b). The independent SLT transcribed /s/ as [ʃ], [ç] or [s], /z/ as [ʒ], [ʝ] or [z] and /tʃ/ as [ʃ]. On EPG assessment, /s/ and /z/ were never produced with typical tongue-palate contact in pseudo-words (see Figure 16), highlighting a discrepancy between independent SLT impressionistic and instrumental analysis. Following therapy, these speech sounds were assessed as correct in treated and untreated words 79 – 100% of the time on assessment at the end of therapy and on maintenance assessment. Again, transfer of learning occurred from treated and untreated words (see Table 12), with no statistical difference between %TC for treated and untreated words for /s/ (Z = -.82, p = .414), /z/ (Z = -.27, p = .414) and /tʃ/ (Z = -1.63, p = .102). Large treatment effects sizes were shown for all target speech sounds (see Figure 10 b).

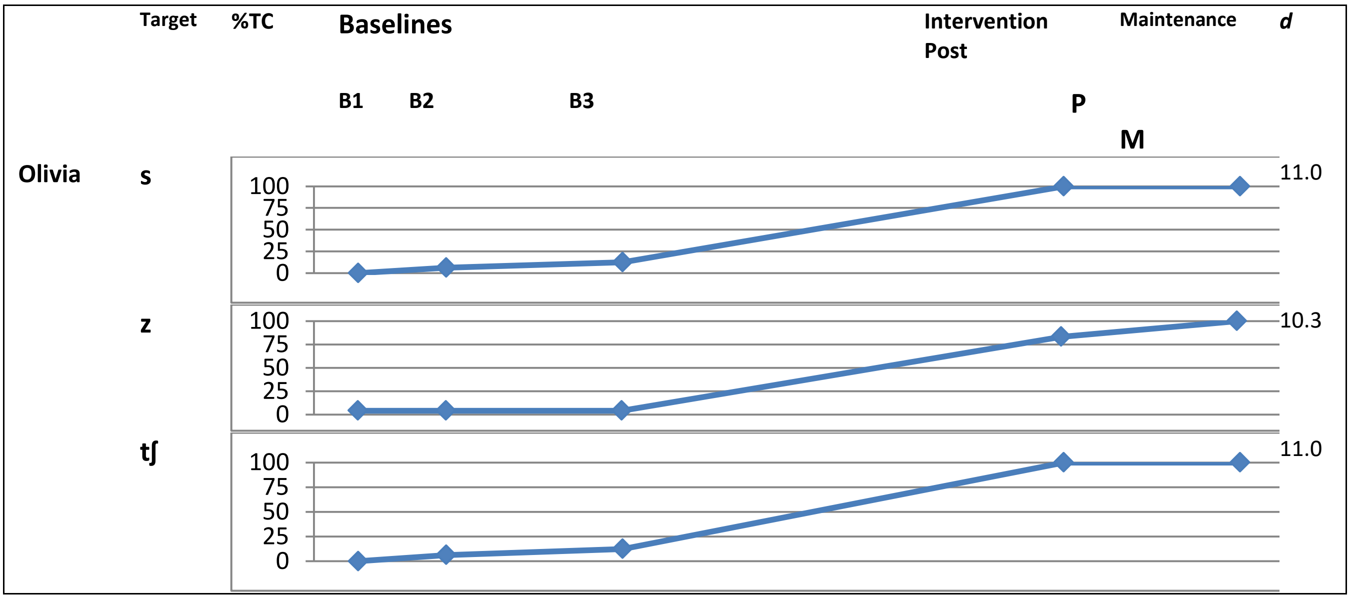
Figure 10

Olivia's percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners scoring (average score)

a) Treated words



b) Untreated words



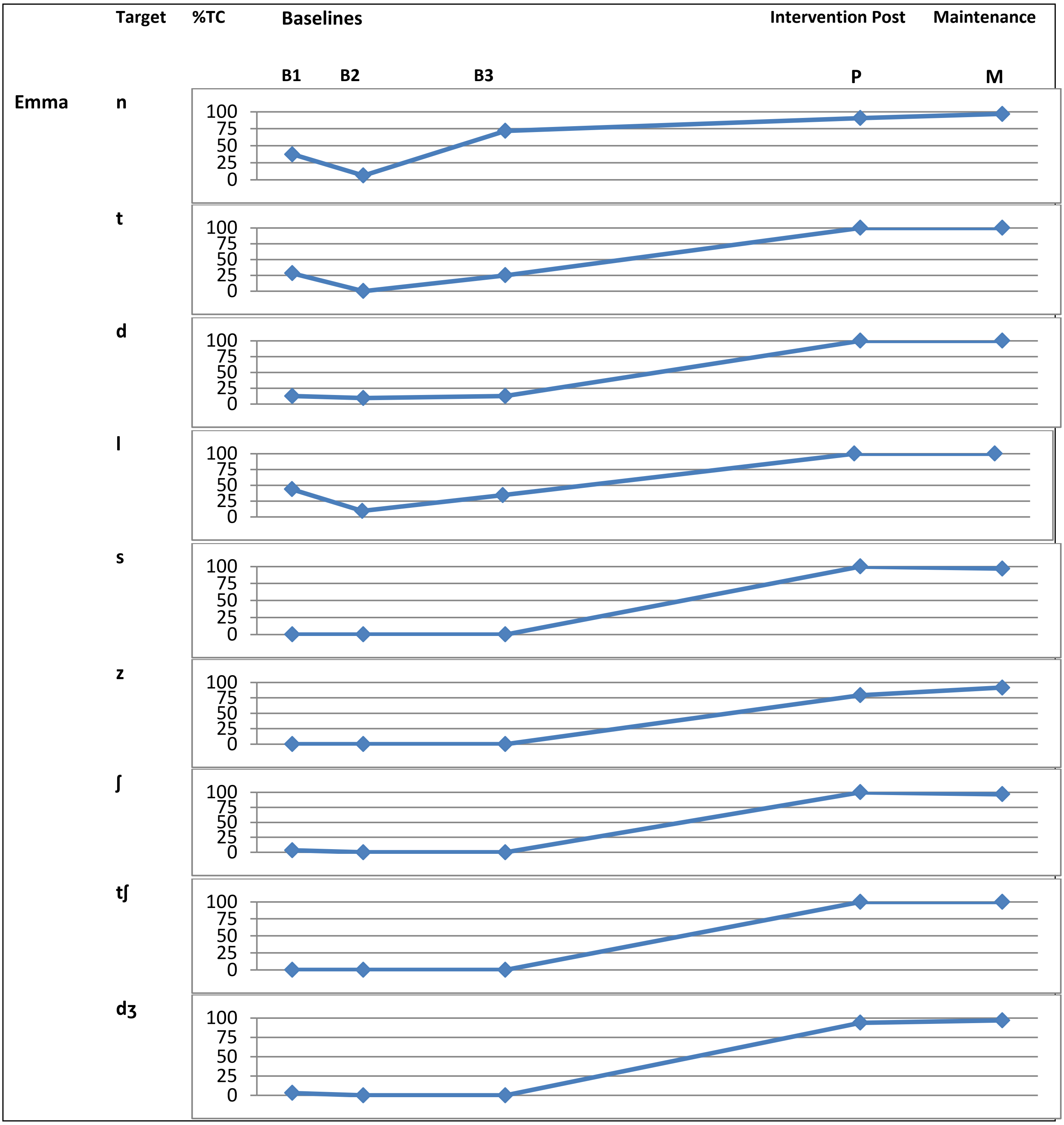
Note: % TC= Percentage of targets correct; B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

At the three baseline assessments, Emma scored near 0% accuracy for production of /s/, /z/, /ʃ/, /tʃ/ and /dʒ/ in treated and untreated words (see Figure 11 a and b). However, some inconsistency with transcription occurred. One independent SLT transcribed these speech sounds as palatal fricatives ([ç],[ʝ]), while the other SLT transcribed them as alveolar laterals ([ɬ], [ɮ]). EPG analysis showed these speech sounds were retracted and produced without a central groove. Lateral airflow was determined given EPG patterns of complete closure and accompanied fricative noise heard and seen on the spectrogram (i.e. in agreement with the research SLT phonetic transcription on initial assessment). Independent SLTs' accuracy levels for production of /n/, /t/, /d/, /l/ were higher and more variable, ranging from 0 - 43.75% correct. Accuracy of these sounds at the three baseline assessments did not match EPG analysis at baseline 3. On EPG analysis these sounds were always backed to velar (see Figure 17). This large discrepancy is likely due to Emma's double articulation of these sounds, making them more difficult to transcribe perceptually. Immediately following therapy, Emma showed 100% or near 100% accuracy for all speech sounds targeted in therapy in both treated and untreated words. Like George and Olivia, no differences were shown with Emma's production of treated and untreated words (see Table 12) for /n/ ($Z = -.730$, $p = .465$), /t/ ($Z = -1.07$, $p = .285$), /d/ ($Z = .00$, $p = 1.000$), /l/ ($Z = -.53$, $p = .593$), /s/ ($Z = .00$, $p = 1.000$), /z/ ($Z = -.45$, $p = .655$), /ʃ/ ($Z = -.27$, $p = .785$), /tʃ/ ($Z = -1.34$, $p = .180$), and /dʒ/ ($Z = -1.60$, $p = .109$). Large treatment effect sizes were shown for all target sounds (see Figure 11 b).

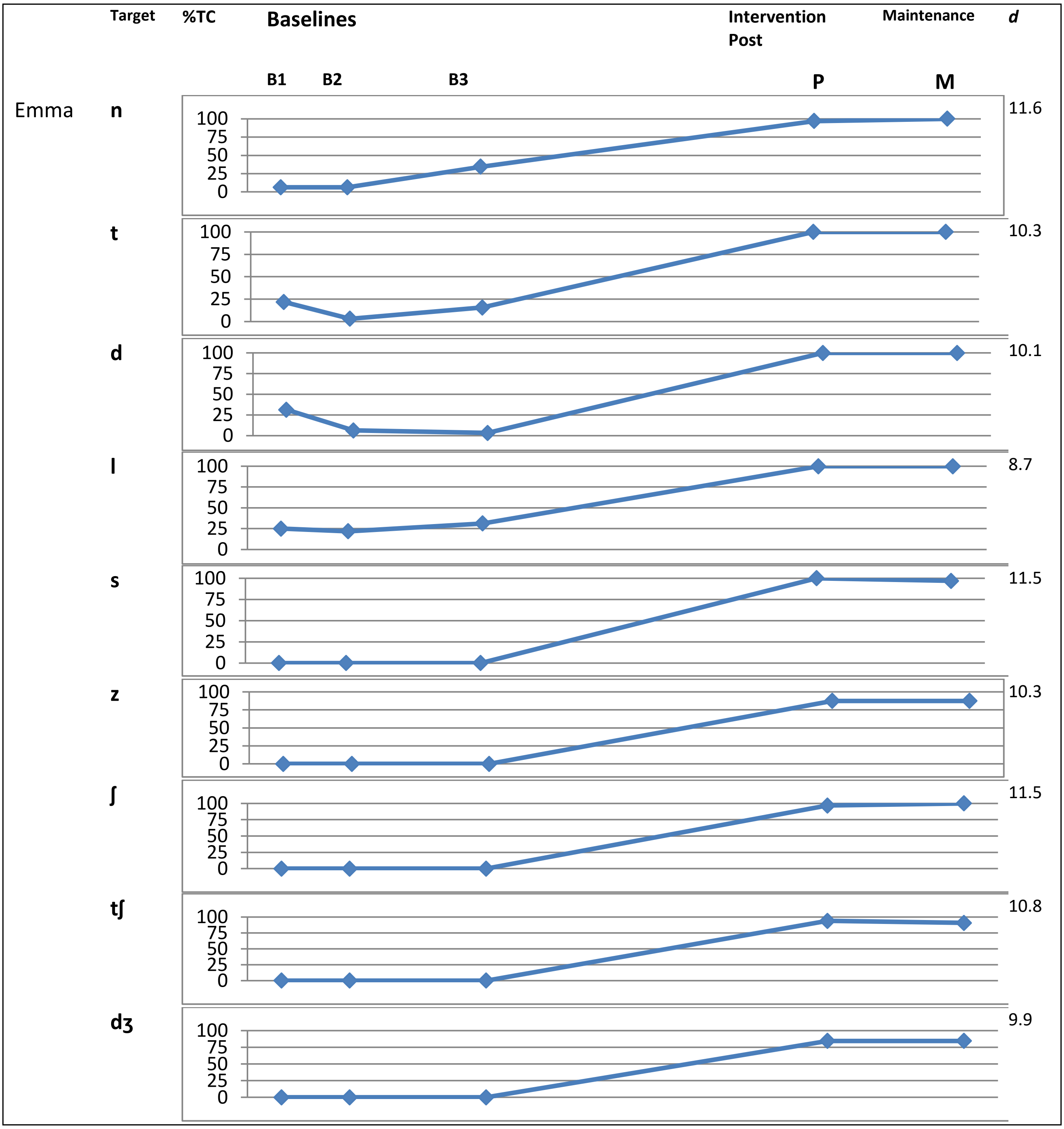
Figure 11

Emma*'s percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners scoring (average score)*

a) Treated words



b) Untreated words



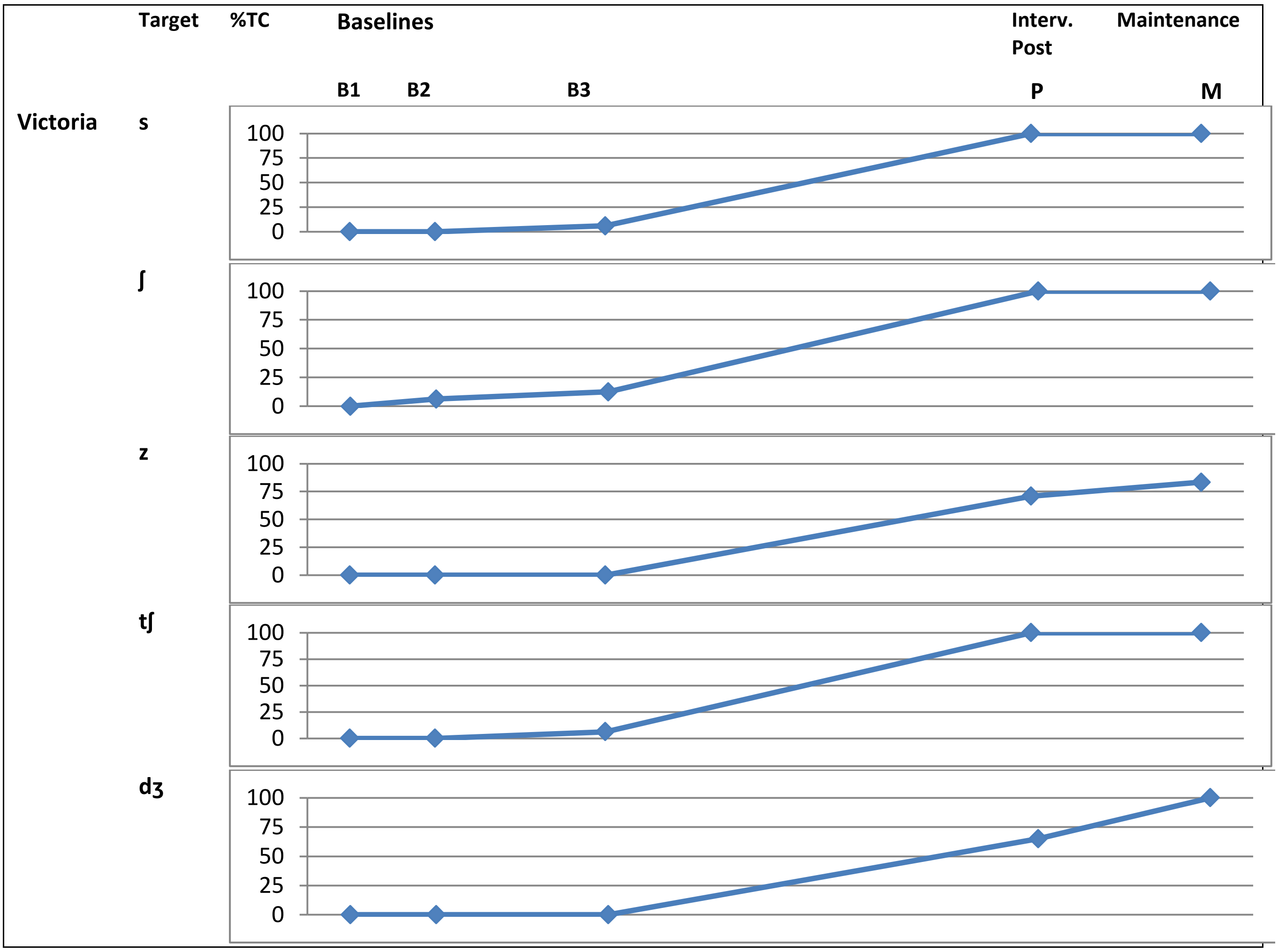
Note: %TC = Percentage of targets correct; B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

At the three baseline assessments Victoria scored low levels of accuracy for production of /s/, /ʃ/, /z/, /tʃ/ and /dʒ/ in treated and untreated words (see Figure 12 a and b). Independent SLT transcription was variable for /s/, /z/. One SLT transcribed /s/ as [ħ], while the other transcribed this speech sound as [ŋ̥̊] or [ħ]. One SLT transcribed /z/ as [n], while the other SLT transcribed this as [n̥̊]. EPG analysis (see Figure 25) for /s/ showed horseshoe shaped tongue-palate contact, with a midline groove, consistent with the researcher's transcription of a weak /s/, double articulated with /ħ/. EPG analysis for /z/ showed complete horseshoe shaped contact, consistent with /n/ or /n̥̊/. Following therapy, /s/, /ʃ/, and /tʃ/ were assessed as correct in treated and untreated words 94 – 100% of the time on post-therapy assessments. Lower levels of accuracy were shown for /z/ and /dʒ/. Post-therapy, these speech sounds were produced accurately 65 – 100% of the time. Errors on production of /z/ and /dʒ/ were /s/ and /tʃ/, respectively. This represents an improvement from pre-therapy productions of [n] and [h] respectively. Transfer of learning occurred from treated to untreated words for/s/ ($Z = -1.41$, $p = .157$), /ʃ/ ($Z = .53$, $p = .593$), /z/ ($Z = -.82$, $p = .414$), /tʃ/ ($Z = -.27$, $p = .785$), and /dʒ/ ($Z = .00$, $p = 1.000$), (see Table 12). Large treatment effect sizes were shown for all target sounds (see Figure 12 b).

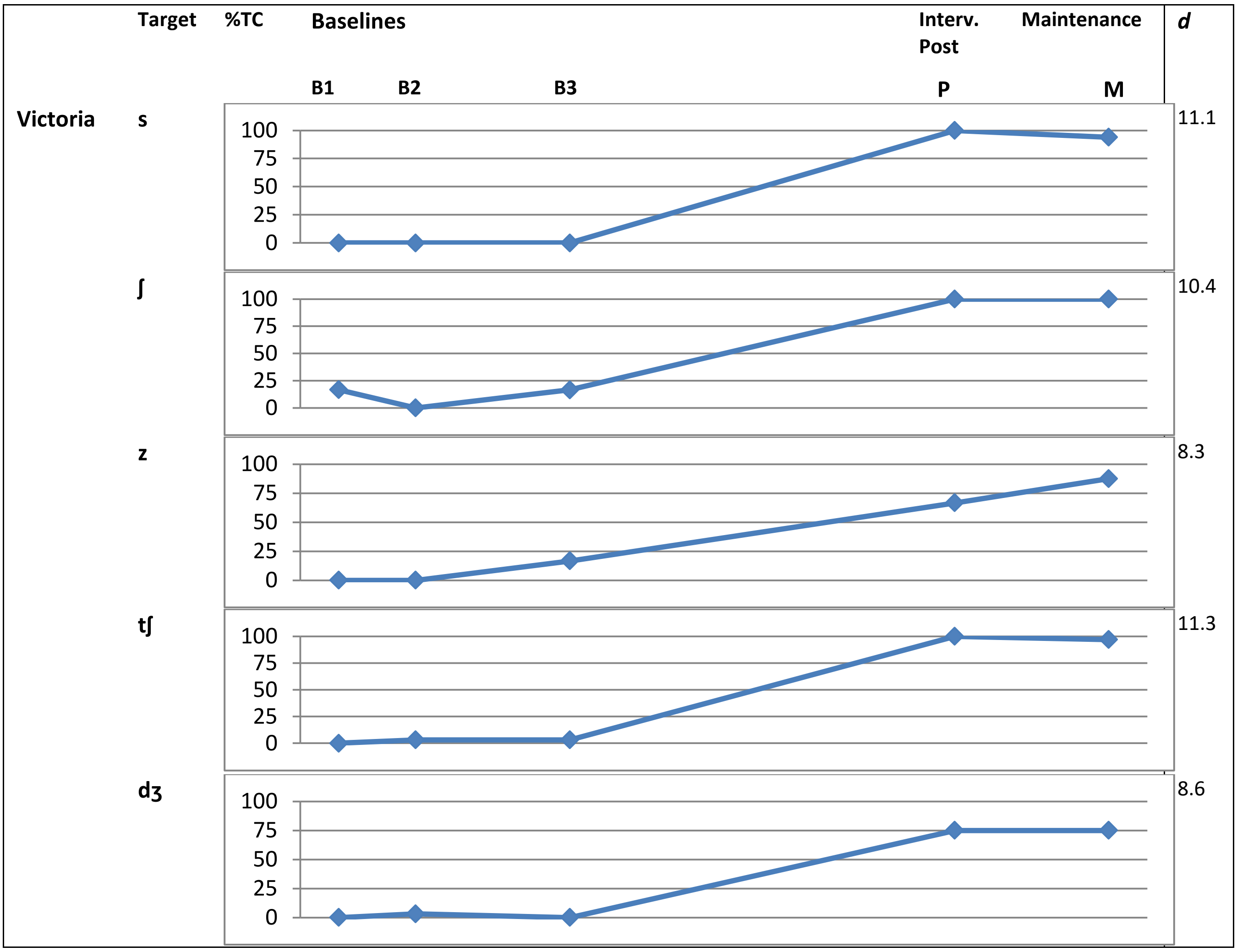
Figure 12

Victoria's percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners (average score)

a) Treated words



Untreated words



Note: %TC = Percentage of targets correct B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

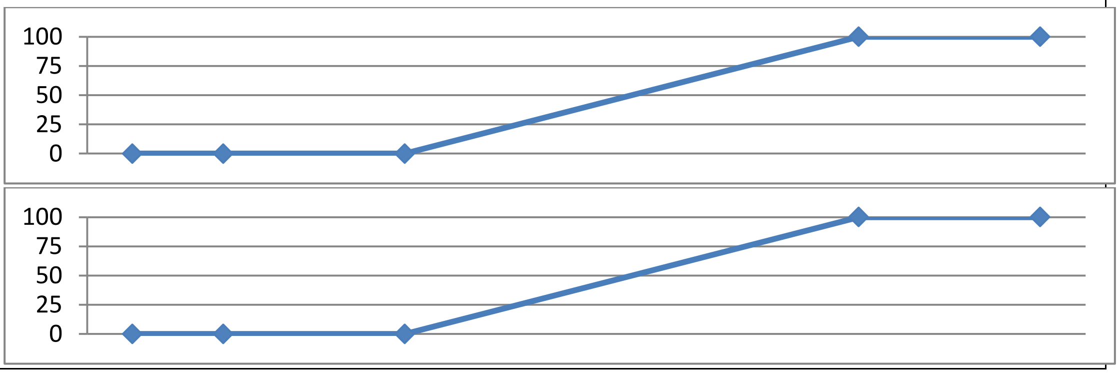
On the three baseline assessments, Grace scored 0% accuracy on production of treated and untreated /s/ and /z/ (see Figure 13 a and b). The independent SLTs transcribed these speech sounds as [ç] and [ʝ] respectively. Following therapy, these speech sounds were assessed as 100% correct in treated and untreated words on the two post-therapy assessments. Transfer of learning occurred from treated to untreated words for /s/ (Z = .00, p = 1.000) and /z/ (Z = .00, p = 1.000) (see Table 12). Large treatment effect sizes were shown (see Figure 13 b).

Figure 13

Grace's percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners (average score)

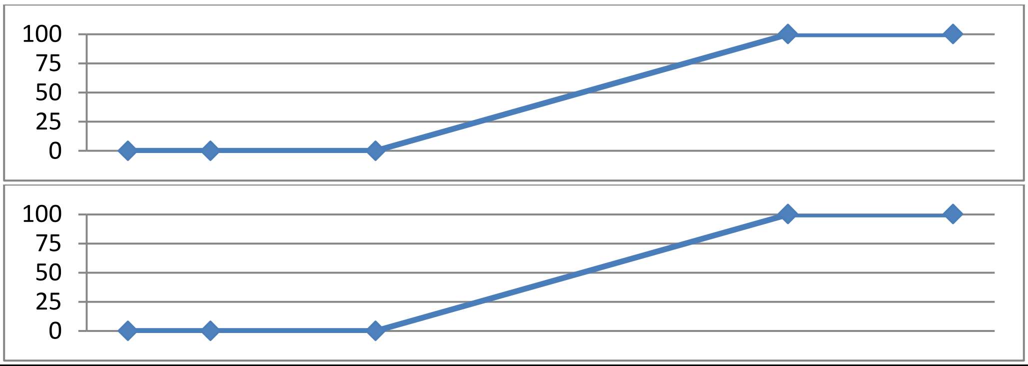
a) Treated words

	Target	%TC	Baselines	Intervention Post	Maintenance
			B1 B2 B3	P	M
Grace	s				
	z				



b) Untreated words

	Target	%TC	Baselines	Intervention Post	Mainten.	*d*
			B1 B2 B3	P	M	
Grace	s					11.7
	z					11.7



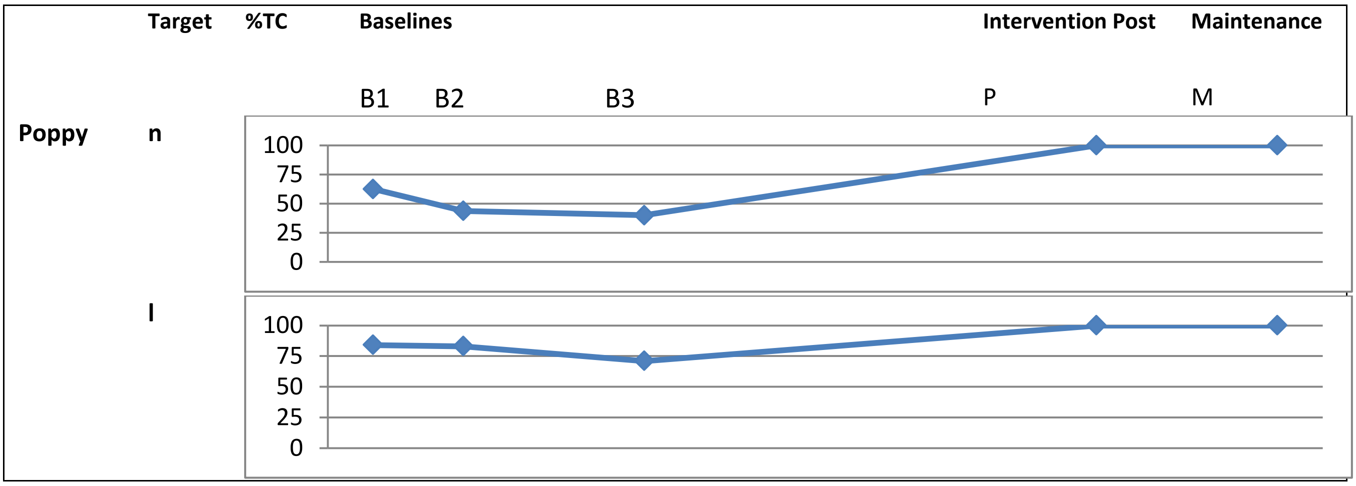
Note: %TC = Percentage of targets correct B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

At the three baseline assessments, the independent SLTs scored Poppy between 71 – 85% accuracy on production of /l/ in treated and untreated words and 40 – 62% accuracy on production of /n/ in treated and untreated words (see Figure 14 a and b). In comparison, the research SLT the research SLT transcribed these speech sounds with 0% accuracy on the three baseline assessments. EPG analysis in the third baseline showed that both speech sounds were consistently produced with velar tongue-palate contact in word initial position in pseudo-words (see Figure 20). Some double articulation (/n/ →[n͡ŋ]) was shown on EPG, and this may explain why the independent SLTs were at times perceiving this sound as accurate. In addition, it is speculated that Poppy's production of /l/ in postvocalic position was very close to a typical dark /l/ (Narayanan et al., 1997) and thus was perceived as typical by the independent SLTs. This point is discussed further in Chapter Eight. Following therapy, /n/ was assessed as 90.62 – 100 % correct by the independent SLT, and /l/ with 100% accuracy. Transfer of learning occurred from treated to untreated words for /n/ (Z = -.40, p = .686) and /l/ (Z = -1.73, p = .083) (see Table 12). A medium to large treatment effect size was shown for /n/ and a small to medium treatment effect size was shown for /l/.

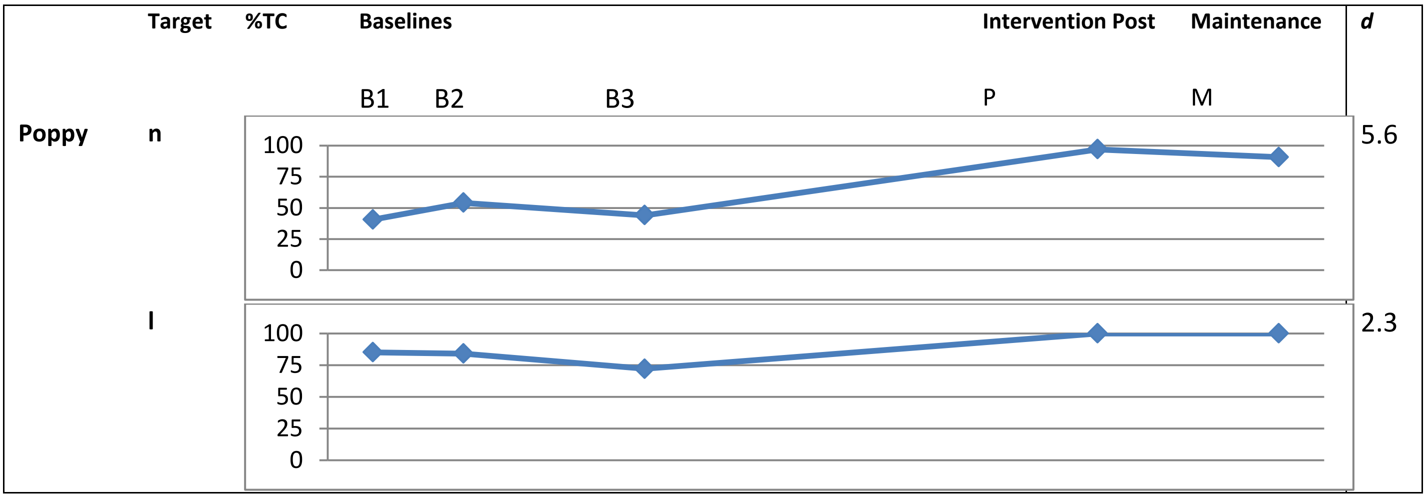
Figure 14

Poppy's percentage of targets correct in treated and untreated words at B1, B2, B3, P, M assessments – from independent listeners (average score)

a). Treated



b). Untreated



Note: %TC = Percentage of targets correct B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention; *d* = Effect size.

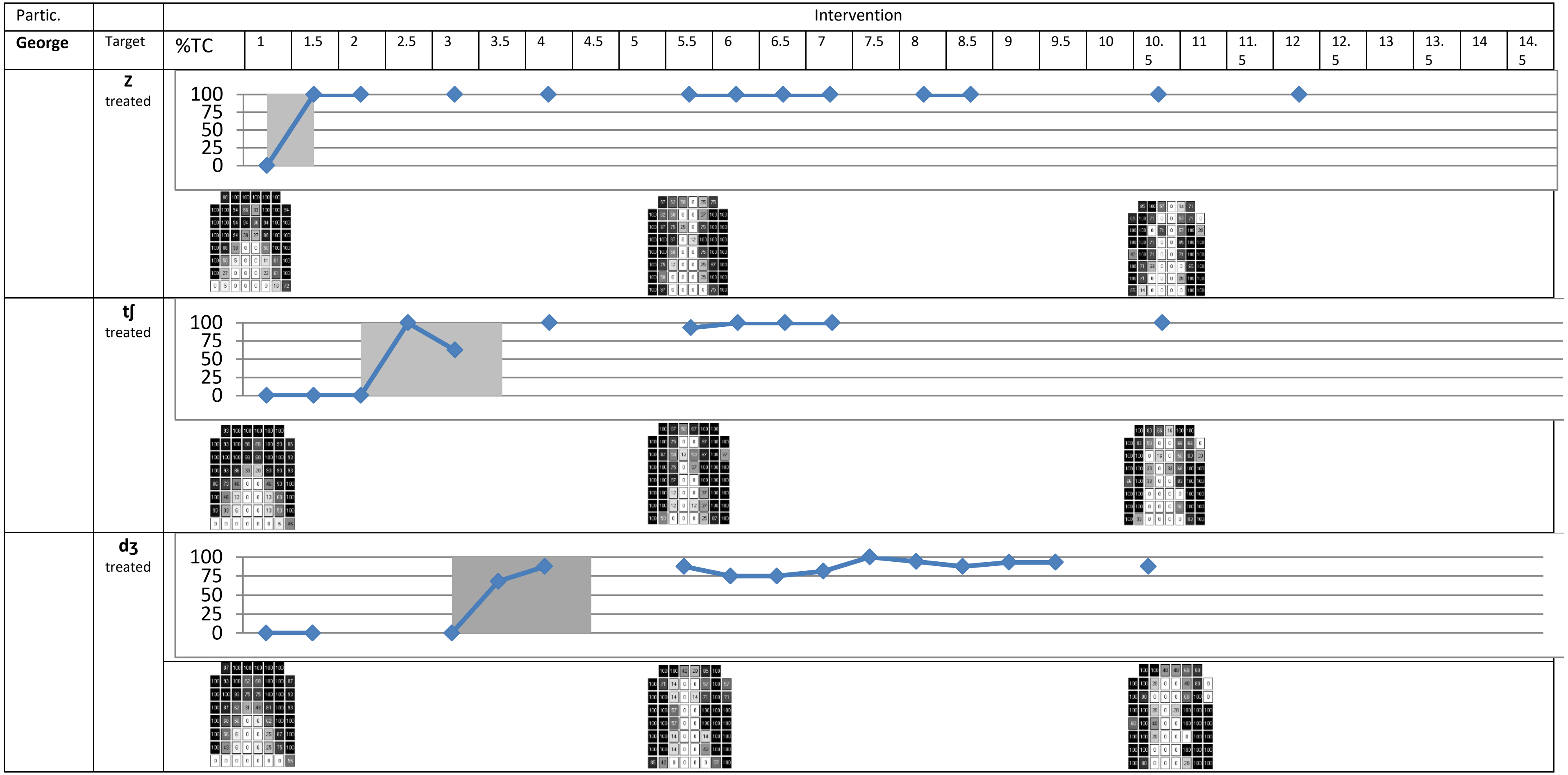
6.2.2 Percentage of targets correct in treated words over the treatment phase

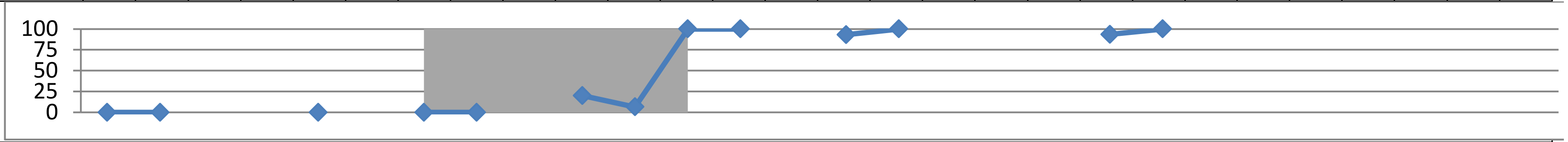
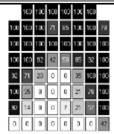
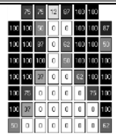
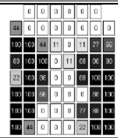
Percentage of targets correct in treated words at the beginning and/or end of separate treatment sessions is shown in Figures 15 – 20. Percentage of targets correct in these figures is derived from the researcher's analysis. The shaded areas on the figures represent when a speech sound was targeted for acquisition (to word level) in therapy. Each figure shows total number of sessions within the treatment phase, i.e. both acquisition and generalisation phases. The acquisition phase involved accurate or near-accurate production at treated word level. The generalisation phase involved production of target sounds in untreated words and spontaneous, connected speech, and is discussed in 5.3.2. Figures 15 - 20 also contain composite EPG pictures for pseudo-words taken at five session intervals throughout intervention.

Figure 15 shows that George's percentage of targets correct only increased when each sound became the focus of therapy. George responded rapidly to intervention. Lateral production was no longer a feature of single word production by the end of the sixth session. It can be noted that percentage of targets correct for /tʃ/ and /dʒ/ was higher in the treatment phase, compared to percentage of targets correct for /tʃ/ and /dʒ/ at the end of therapy and at the 3-month maintenance assessment in Figure 9. This reflects a discrepancy in the research SLT and independent SLT ratings; the research SLT was more likely to rate /tʃ/ and /dʒ/ as correct, compared to the independent SLTs, the later who rated George as either [tʃ] or [tj] or [dʒ] or [dj].

Figure 15

George's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo-words) at EPG assessment points during intervention



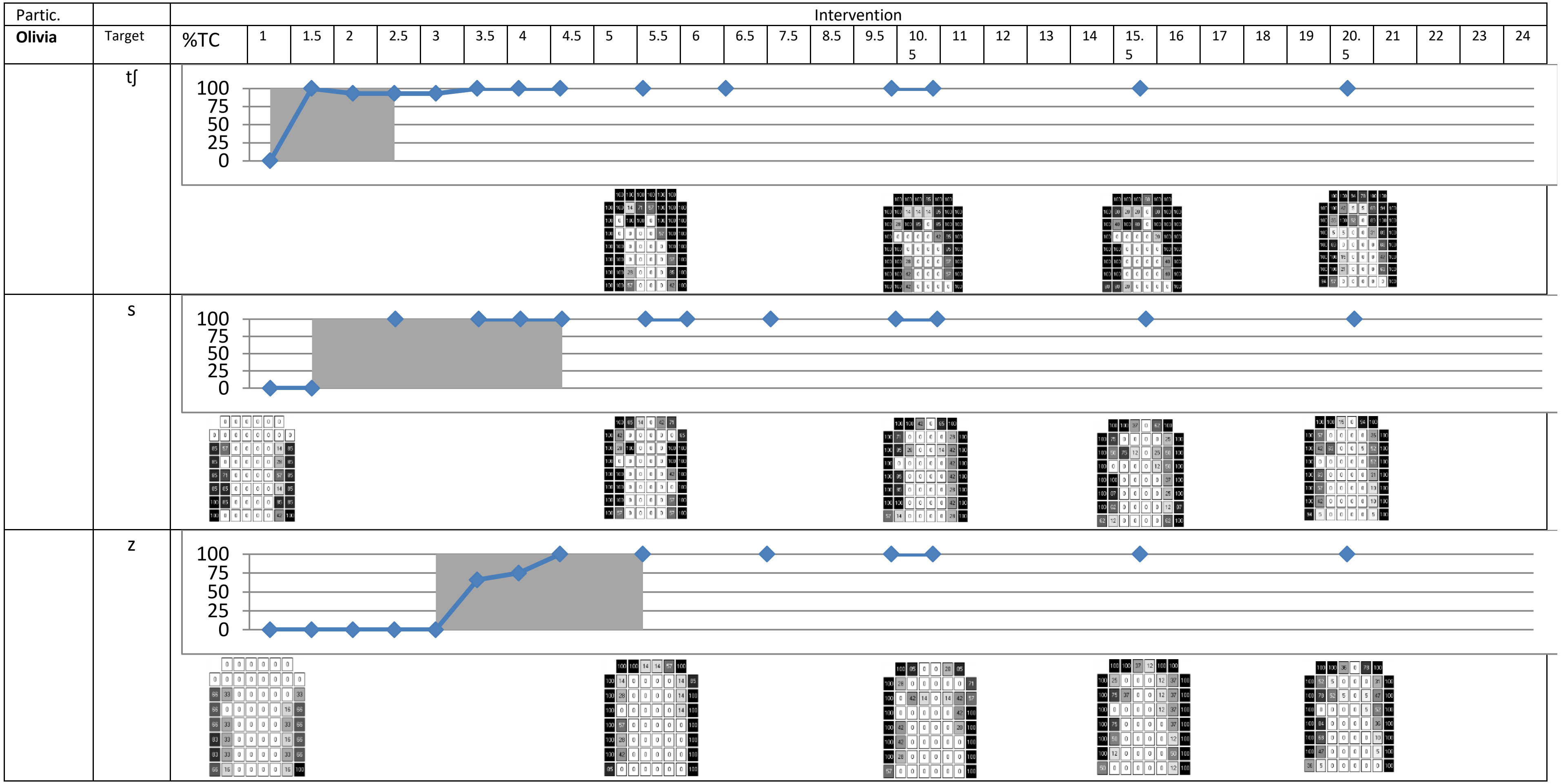
George	Target	%TC	1	1.5	2	2.5	3	3.5	4	4.5	5	5.5	6	6.5	7	7.5	8	8.5	9	9.5	10	10.5	11	11.5	12	12.5	13	13.5	14	14.5
	ʃ treated																													
		  																												

Note: % TC = Percentage targets correct; 1, 2, 3 etc. = Beginning of session 1, 2, 3 etc.; 1.5, 2.5, 3.5 etc. = End of session 1, 2, 3 etc.; Shaded area indicates intervention period for target sound, word level. Composite EPG pictures for/ tʃ/ and /dʒ/ are frames of maximum contact for the fricative element of the speech sound. Partic. = Participant.

Figure 16 shows a clear treatment effect for all three speech sounds targeted in Olivia's therapy. All three speech sounds achieved 100% accuracy at word level by five sessions of therapy. A similar treatment effect is shown for Emma in Figure 17. Percentage of targets correct increased as each lingual speech sound became the focus of therapy. However, Emma also showed generalisation of some aspects of target speech sound production prior to each speech sound becoming the focus of therapy. The pattern was for an anterior placement and less lateralised production, as can be seen on EPG composite frames for pseudo-words (e.g. see frames for /l/, /tʃ/ and /dʒ/). Emma achieved 100% or near 100% accuracy at word level for all target sounds by session 19.

Figure 16

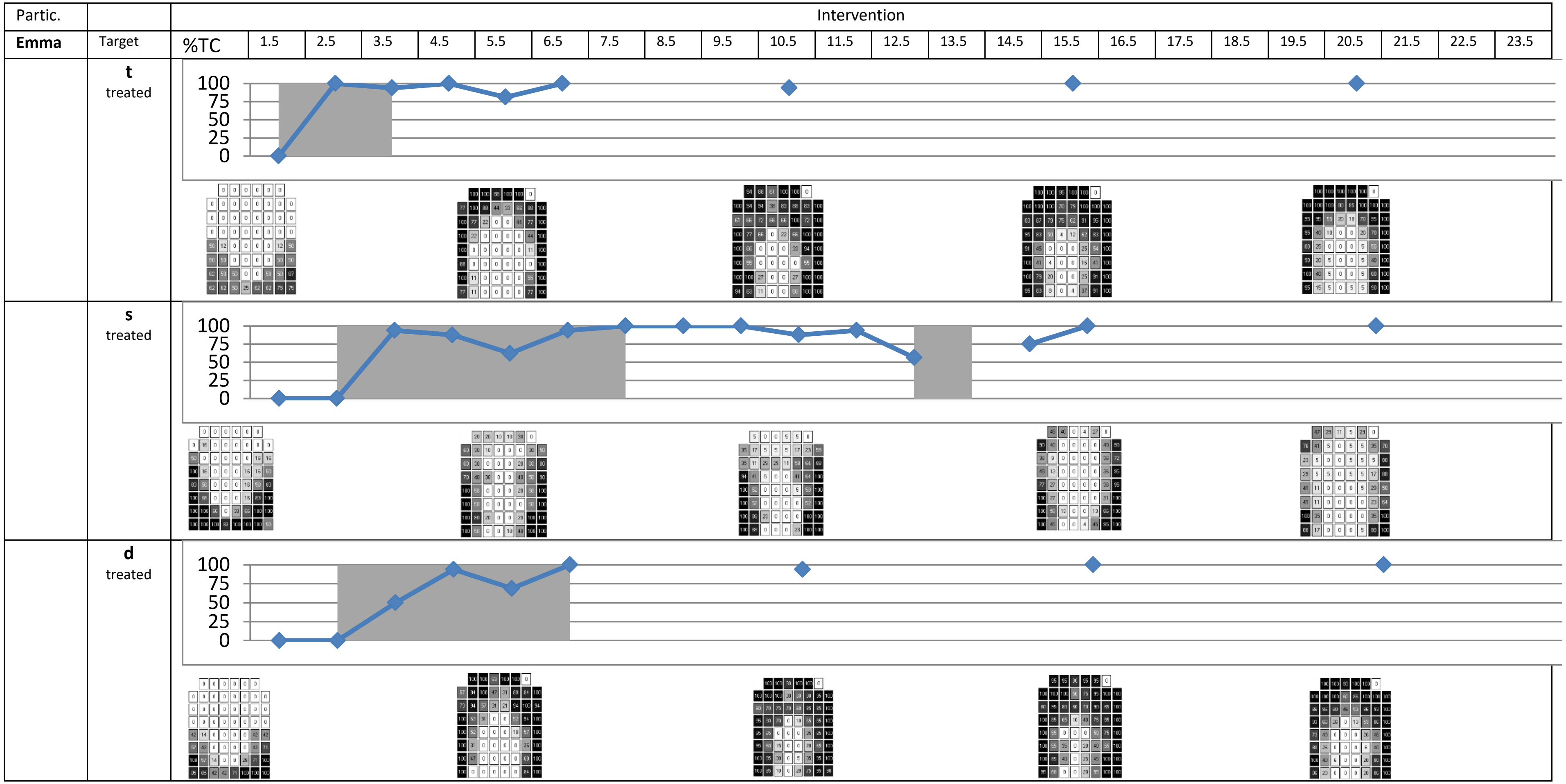
Olivia's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo-words) at EPG assessment points during intervention

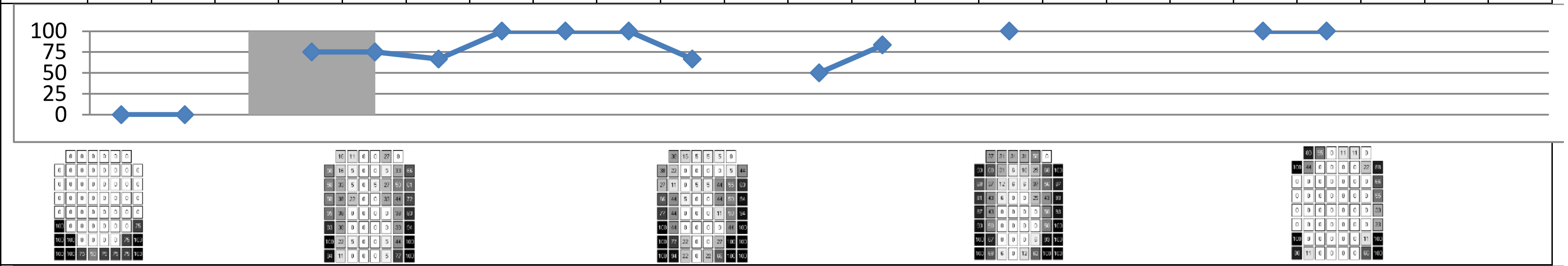
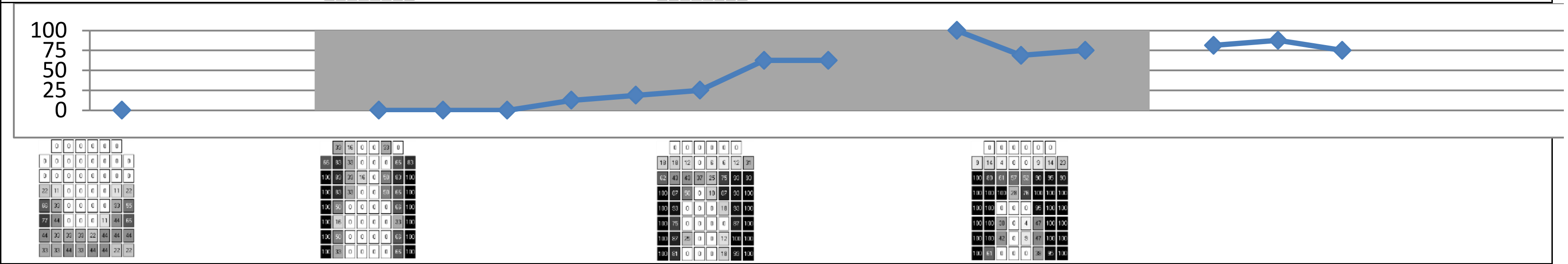
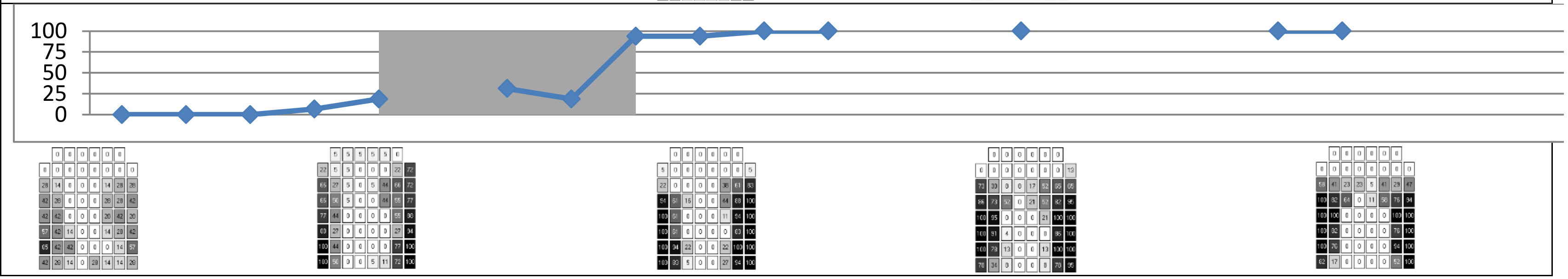


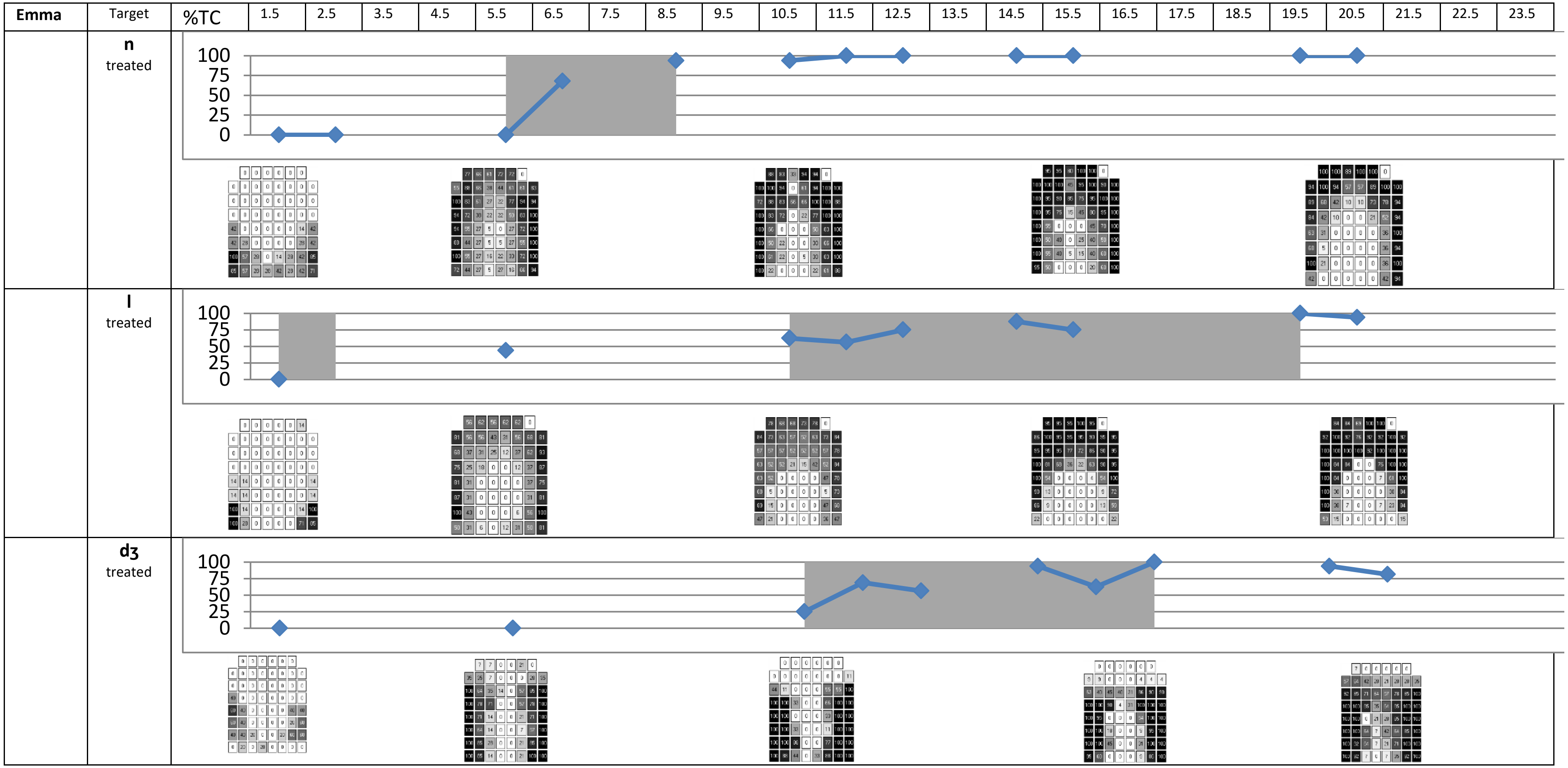
Note: % TC = Percentage targets correct; 1, 2, 3 etc. = Beginning of session 1, 2, 3 etc.; 1.5, 2.5, 3.5 etc. = End of session 1, 2, 3 etc.; Shaded area indicates intervention period for target sound, word level; Composite EPG pictures for /tʃ/ are frames of maximum contact for the stop element of the speech sound. At start of treatment /tʃ/ did not contain a stop element, therefore no EPG picture is shown; Partic. = Participant.

Figure 17

Emma's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo words) at EPG assessment points during intervention



Emma	Target	%TC	1.5	2.5	3.5	4.5	5.5	6.5	7.5	8.5	9.5	10.5	11.5	12.5	13.5	14.5	15.5	16.5	17.5	18.5	19.5	20.5	21.5	22.5	23.5
	z treated																								
	tʃ treated																								
	ʃ treated																								

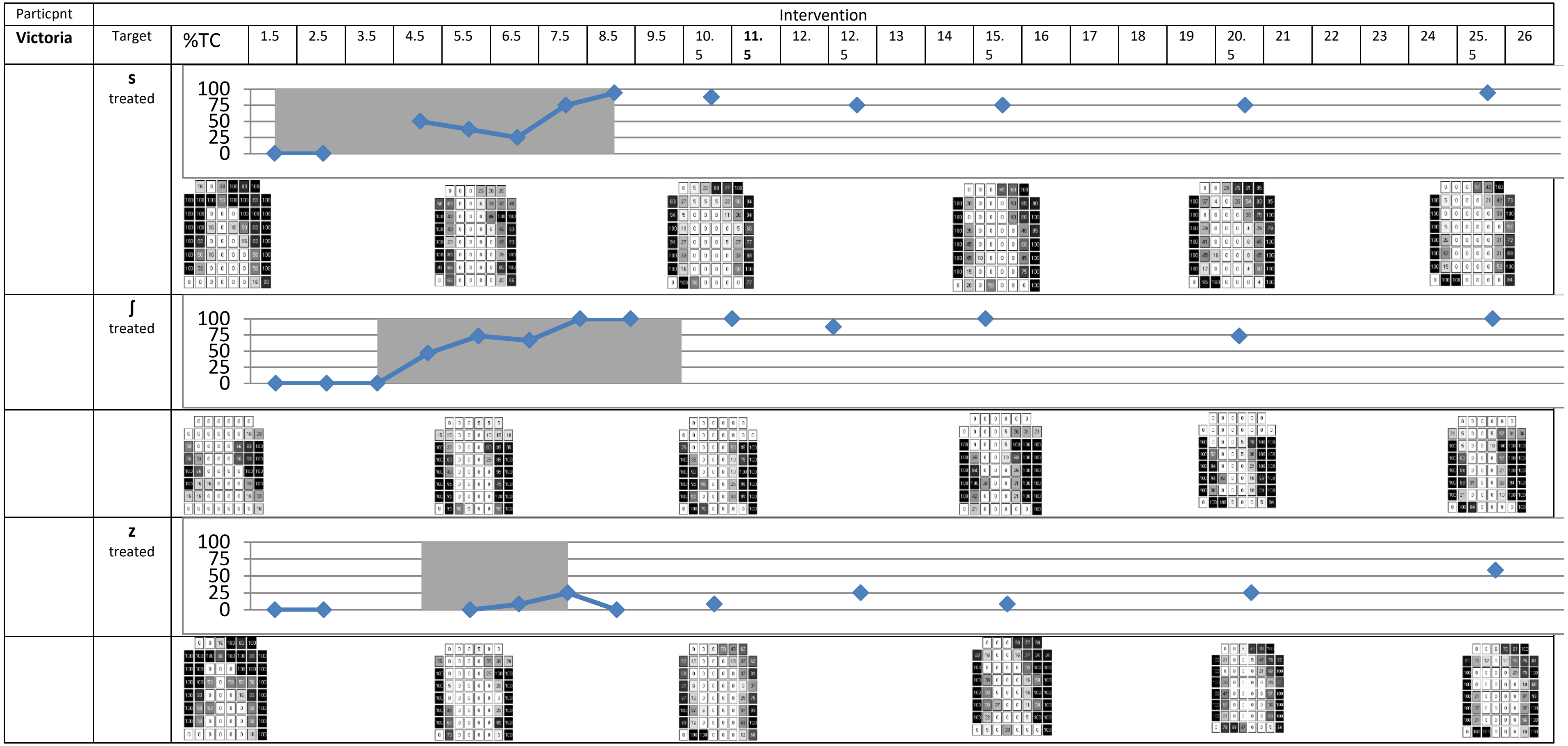


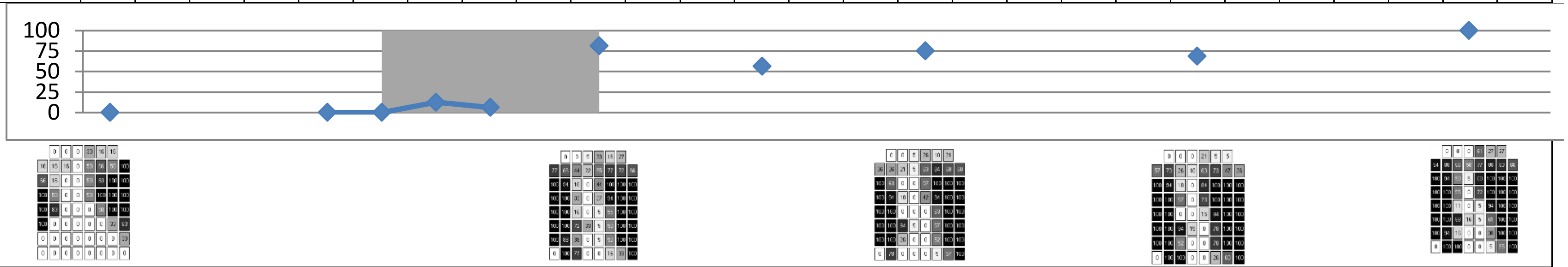
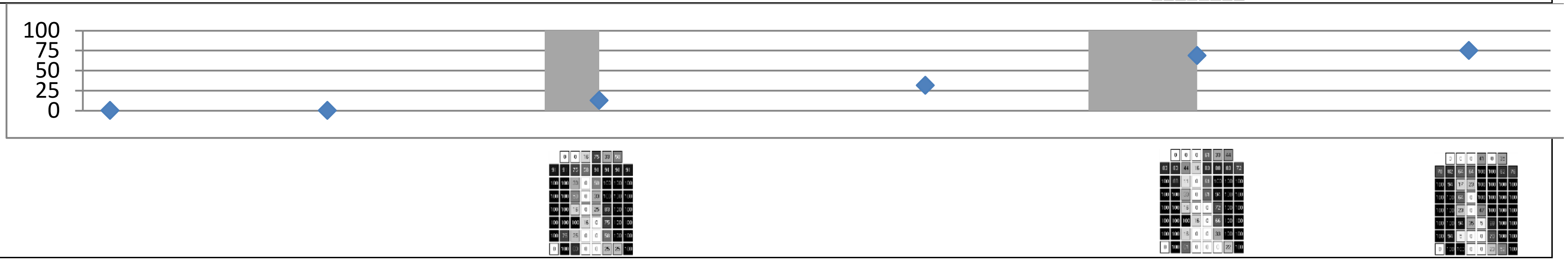
Note: % TC = Percentage targets correct.; 1.5, 2.5, 3.5 etc. = End of session 1, 2, 3 etc.; Shaded area indicates intervention period for target sound, word level; Composite EPG pictures for /tʃ/ and /dʒ/ are frames of maximum contact for the fricative element of the speech sound, with missing data at 20.5 for /tʃ/; Partic. = Participant.

As shown in Figure 18, Victoria responded less rapidly to therapy and had more difficulty acquiring the sibilant speech sounds targeted in therapy. Victoria was fully engaged in therapy, but as therapy progressed, it became apparent that VPI was a major obstacle for production of target sounds. Victoria had difficulty building up oral pressure to produce these speech sounds. It was observed that, when putting more effort into her speech, voiced pressure consonants became nasalised (grade 2 hypernasality on GOS.SP.ASS). Since Victoria had undergone multiple speech surgeries in the past, further surgery to the velo-pharyngeal sphincter was not an option. Despite apparent VPI, with therapy, percentage of targets correct increased from 0% to 58 – 100% at word level by 25 sessions.

Figure 18

Victoria's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo-words) at EPG assessment points during intervention



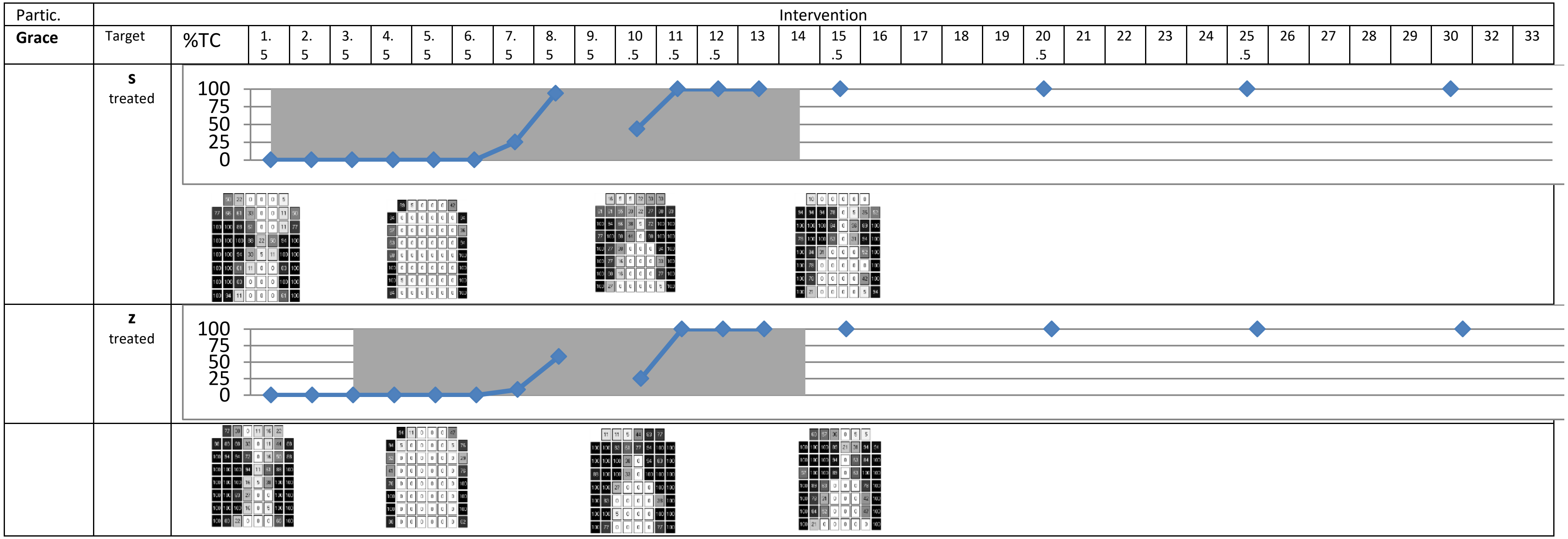
Victoria	Target	%TC	1.5	2.5	3.5	4.5	5.5	6.5	7.5	8.5	9.5	10.5	**11.5**	12.	12.5	13	14	15.5	16	17	18	19	20.5	21	22	23	24	25.5	26
	tʃ treated																												
	dʒ treated																												

Note: % TC = Percentage targets correct; 1, 2, 3 etc. = Beginning of session 1, 2, 3 etc.; 1.5, 2.5, 3.5 etc. = End of session 1, 2, 3 etc.; Shaded area indicates intervention period for target sound, word level; Composite EPG pictures for /tʃ/ and /dʒ/ are frames of maximum contact for the fricative element of the speech sound, with missing data at 5.5 for /tʃ/ since production did not include a fricative element, likewise for /dʒ/ at 1.5, 5.5 and 15.5.

Grace also responded less rapidly to therapy, compared to George, Olivia and Emma (see Figure 19). In the initial stages of therapy, she produced an interdental /s/ and /z/, but these reverted to a palatal sound when she was given the instruction to draw her tongue back in her mouth. However, by session 11 she was consistently producing a perceptually accurate /s/ and /z/ at treated word level. The researcher observed that changes to /s/ production immediately transferred to production of /z/, without instruction from the researcher.

Figure 19

Grace's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo words) at EPG assessment points during intervention

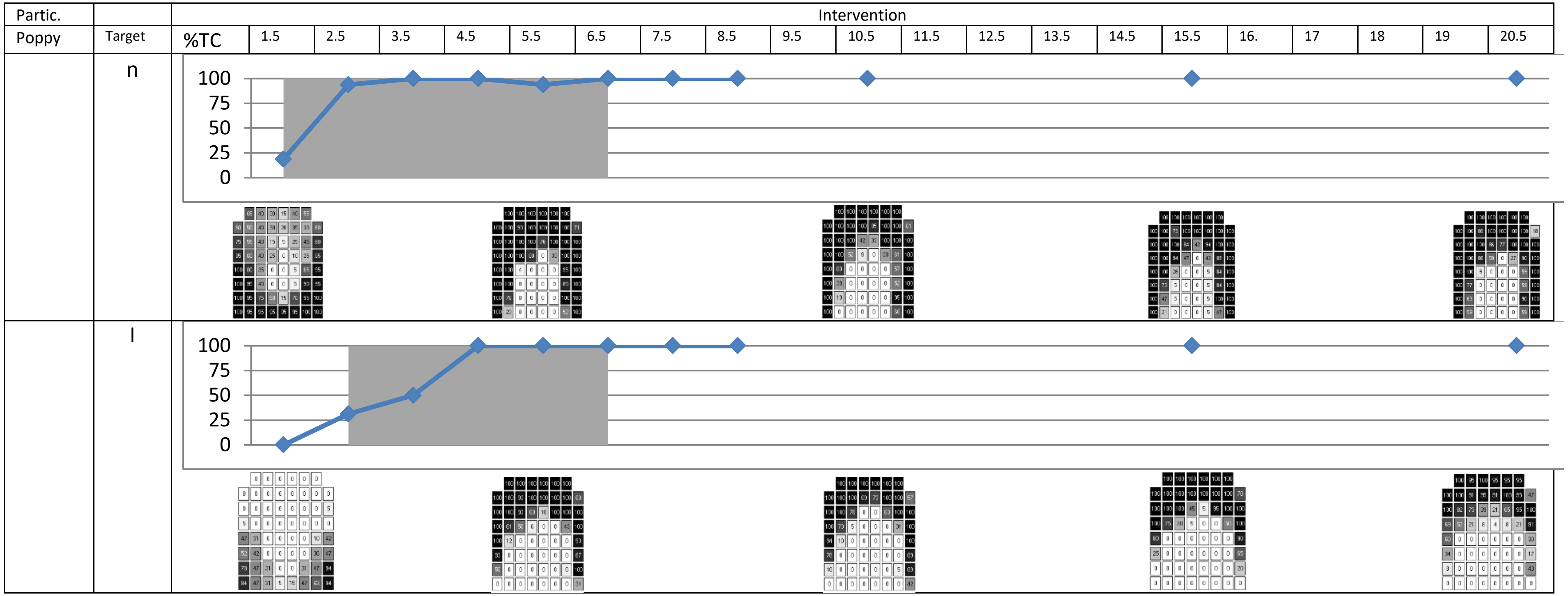


Note: % TC = Percentage targets correct; 1.5, 2.5, 3.5 etc. = End of session 1, 2, 3 etc.; Partic. = Participant; Shaded area indicates intervention period for target sound, word level.

As shown in Figure 20, Poppy responded rapidly to therapy. She produced a perceptually accurate /n/ by the end of session 2 and a perceptually accurate /l/ by the end of session 4.

Figure 20

Poppy's percentage of targets correct (in treated words) across the intervention period and composite EPG frames (pseudo-words) at EPG assessment points during intervention



Note: % TC = Percentage targets correct 1.5, 2.5, 3.5 etc = end of session 1, 2, 3 etc.; Partic. = Participant; Shaded area indicates intervention period for target sound, word level.

6.2.3 Percentage of targets correct in connected speech over baseline, treatment and post treatment phases

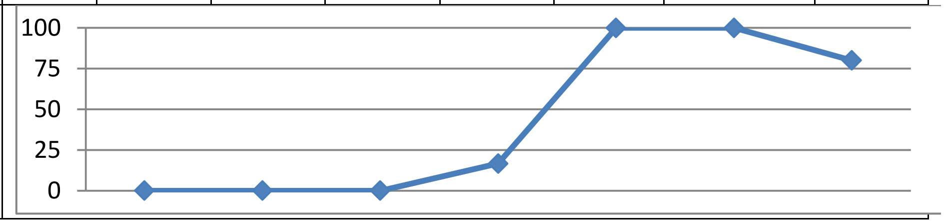
Participants' percentage of target's correct in connected speech over the baseline, therapy and post-therapy phases is shown in Figure 21. The results outlined in this section contribute to answering research question number two: **Does improved accuracy of target phonemes generalise to continuous, connected speech, and, if so, does this result in improved ratings of speech intelligibility and improved ratings of impact on quality of life, post-intervention?** Percentage of targets correct comes from the researcher's transcription of the 2-minute spontaneous connected speech samples. Percentage of targets correct is combined in this figure, e.g. percentage of targets correct in connected speech for George includes his accuracy for /z/, /ʃ/, /tʃ/ and /dʒ/ combined. As can be seen from this figure, with therapy, target consonants generalised to connected speech for all participants, but to varying degrees.

Figure 21

Percentage of targets correct in continuous speech (2-minute connected speech sample) at baseline, intervention and post-therapy assessments

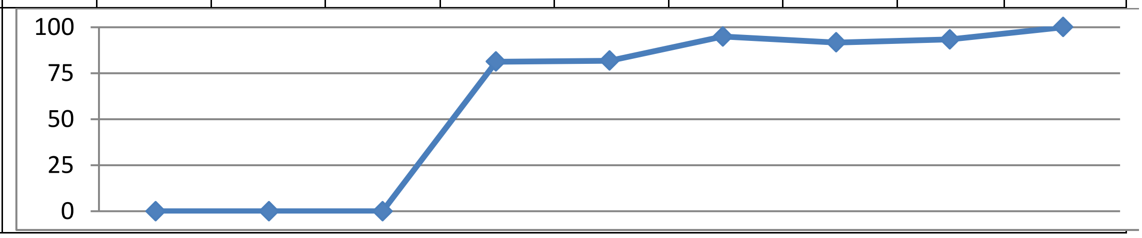
a) George

Partic.		Baselines			Intervention		Intervention post	Mainten.
George	%TC	B1	B2	B3	A5	A10	P	M



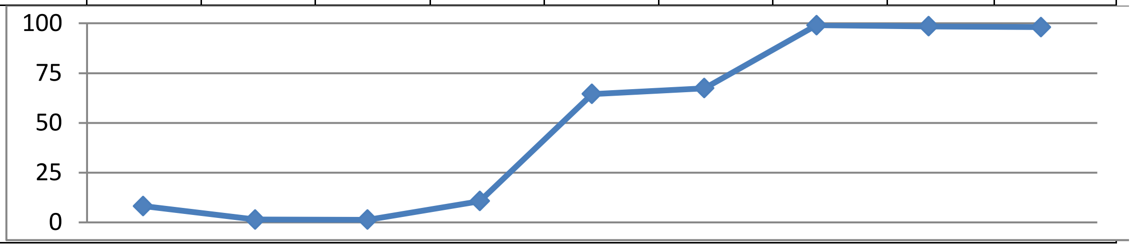
b) Olivia

Partic.		Baselines			Intervention				Interven post	Mainten.
Olivia	%TC	B1	B2	B3	A5	A10	A15	A20	P	M



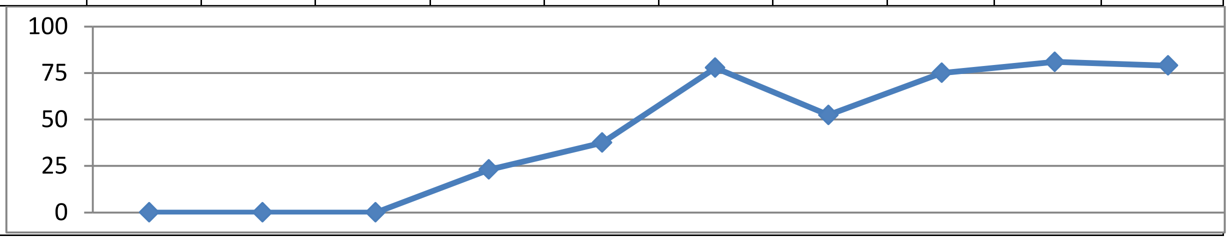
c) Emma

Partic.		Baselines			Intervention				Interven post	Mainten.
Emma	%TC	B1	B2	B3	A5	A10	A15	A20	P	M



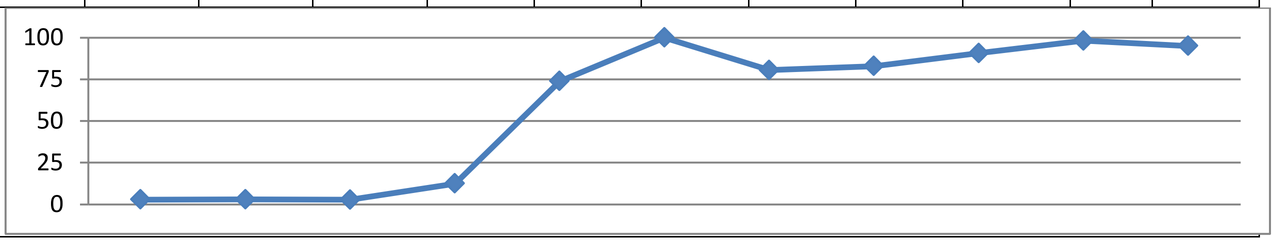
d) Victoria

Partic.		Baselines			Intervention					Interven post	Mainten.
Victoria	%TC	B1	B2	B3	A5	A10	A15	A20	A25	P	M



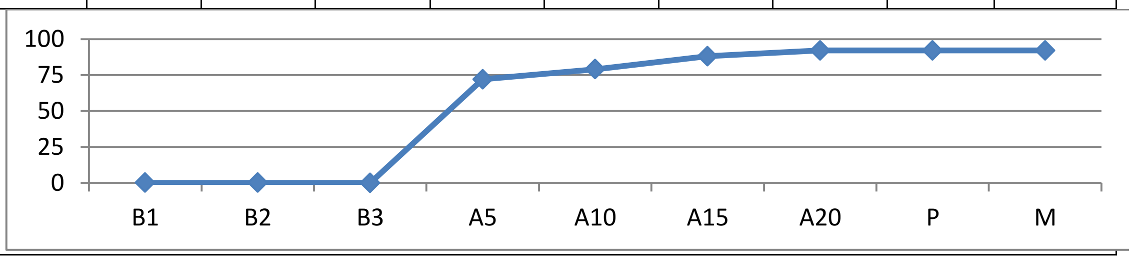
e) Grace

Partic.		Baselines			Intervention						Inter post	Mainten
Grace	%TC	B1	B2	B3	A5	A10	A15	A20	A25	A30	P	M



f). Poppy

Partic.		Baselines			Intervention				Interven post	Mainten.
Poppy	%TC	B1	B2	B3	A5	A10	A15	A20	P	M



Note: B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; A5 = Assessment end 5 sessions; A10 = Assessment end of 10 session; A15 = Assessment end of 15 sessions, and so on; P = Assessment immediately post intervention; M = Maintenance assessment 3-months post-intervention. Partic. = Participant.

George's accuracy with target sounds at word level quickly generalised to spontaneous speech, and he achieved high levels of accuracy in continuous connected speech by assessment at the end of ten sessions (see Figure 21 a). On maintenance assessment, percentage of targets correct was 80% in the event retell task. This mirrored his performance with connected speech outside of the therapy setting according to his parents. Targets produced in error involved /tʃ/ and /dʒ/ which were transcribed as [tʃ] or [tj] and [dʒ] or [dj] respectively by the research SLT. As discussed in 6.2.1, this represents an improvement from [ɬ] and [ɮ] production pre-therapy (though see 8.1.1 for further discussion). However, George may have benefited from further therapy to generalise /tʃ/ and /dʒ/.

Olivia acquired 100% accuracy of target sounds to word level relatively quickly (four sessions – see Figure 21 b). However, she required a further 15 sessions before she was consistently using her target sounds correctly in the retell task, and then a further five sessions before accurately producing speech targets in all speech settings. Olivia's accuracy in spontaneous speech was maintained on maintenance assessment.

Emma showed steady generalisation of target sounds into her everyday speech. This generalisation was aided by regular home practice. By 20 sessions she was using targets sounds accurately in the event retell task (see Figure 21 c). She was seen for another three sessions to achieve functional generalisation (i.e. accurate production in all speech settings). Emma's progress was maintained on maintenance assessment.

Victoria had more difficulty with use of target speech sounds in connected speech. As with use of target sounds in treated and untreated words, difficulty with generalisation appeared due to VPI and production of target sounds in connected speech was particularly effortful. However, despite apparent on-going structural difficulties, Victoria achieved around 80% accuracy of target sounds correct in connected speech after 26 sessions of therapy and this level was unchanged on maintenance assessment (see Figure 21 d).

Although arguably having the mildest SSD out of all the participants, Grace had the most difficulty integrating her remediated /s/ and /z/ into her everyday talking. Having achieved a perceptually accurate /s/ and /z/ in words after 11 sessions of therapy, Grace received a further 22 sessions (see Figure 21 e). Grace's practice of speech outside of therapy was less regular and she was at times sensitive to adults' attempts to correct her speech. However, by the end of therapy her /s/ and /z/ were produced correctly 98.18% of the time in the retell task. On maintenance assessment accuracy was 95%. Errors consisted of mild palatisation of /s/ and /z/. Grace also showed increased signs of VPI over the course of therapy (grade 1 hypernasality on GOS.SP.ASS with hypernasality perceived on closed vowels and more noticeable nasal turbulence) which may possibly have made production of /s/ and /z/ more difficult.

Like Olivia, Poppy acquired 100% accuracy of target sounds to word level relatively quickly (four sessions – see Figure 21 f). However, she required a further 15 sessions before these target sounds were being used 92% of the time in the retell task. Errors with production at end of therapy assessment and maintenance assessment mostly occurred with /l/ production, with error production consisting of [ɰ].

6.2.4 Percentage of targets correct in low and high frequency words

Differences in percentage of targets correct in low frequency words and high frequency words was examined at word level (treated and untreated words) and connected speech level (2-minute connected speech sample). A Wilcoxon signed-rank test showed no difference in percentage of targets correct in production of low and high frequency words after five sessions (A5) ($Z = -.53$, $p = .593$) after ten sessions (A10) ($Z = .00$, $p = 1.000$) for participants, as shown in Table 13 ($p > .05$; researcher's phonetic transcription). However, a difference was shown in percentage of targets correct in low and high frequency words in connected speech. A Wilcoxon signed- rank test showed significant difference between percentage of targets correct in low and high frequency words ($Z = -2.20$, $p = .028$) in continuous connected speech (see Table 14). Percentage of targets correct in connected speech samples is derived from George at A10, Olivia at A5, Emma at A10, Victoria at A25, Grace at A20, Poppy at A5 and comes from the researcher's transcription. These varying assessment times were selected for participants as they represented assessment points where most target sounds had been worked on in therapy, but errors were still seen in spontaneous speech (see Figure 21). This significant difference suggests participants had more difficulty producing target sounds in words occurring with high frequency (e.g. "at", "and") in connected speech, compared to words occurring with low frequency (e.g. "lesson", "chicken") during the acquisition phase of treatment.

Table 13

Differences in percentage targets correct for low frequency and high frequency words in treated and untreated words at A5 and A10 (Wilcoxon signed-rank test) for all participants (combined)

	Percentage of Targets Correct Mean			
Assessment	Low frequency words	High frequency words	Z score	p value
A5	57.64	58.77	-.53	.593
A10	88.00	88.63	.00	1.000

Note: A5 = Assessment end of five sessions; A10 = Assessment end of 10 sessions; Percentage of targets correct from all treatment targets for all six participants (research SLT transcription)

Table 14

Differences in percentage targets correct for low frequency and high frequency words in continuous connected speech sample for all participants (Wilcoxon signed-rank test)

Percentage of Targets Correct Mean			
Low frequency words	High frequency words	Z score	p value
84.48	39.53	-2.20	.028

Note: Percentage of targets correct in connected speech taken from George at A10, Olivia A5, Emma A10, Victoria A25, Grace A20, Poppy A5 (see text for further information)

6.2.5 Percentage consonant correct on phonology subtest of the DEAP and findings from GOS.SPP.ASS on post-therapy assessment

Participant's PCC on phonology subtest of the DEAP and findings from GOS.SPP.ASS at the end of therapy (P) and 3-months after completion of therapy (M) is shown in Table 9.

6.2.5.1 Resonance and nasal airflow

Compared to initial assessment, George showed increased signs of VPI on the two post-therapy assessments. His resonance went from grade 1 hyponasal (slight denasalisation of nasal sounds) to grade 1 hypernasality (hypernasality perceived on closed vowels) on assessment immediately following therapy and on maintenance assessment. His grade 1 nasal turbulence (slight turbulence) increased to grade 2 (marked) on post-therapy assessment. Likewise, Emma, Victoria and Grace all showed evidence of VPI, or worsening signs of VPI over the duration of therapy. Emma went from normal resonance and no nasal emission or turbulence on initial assessment, to grade 1 hypernasality (hypernasality perceived on closed vowels) and grade 1 nasal turbulence on post-therapy assessment. Victoria was assessed as having grade 1 hypernasality (hypernasality perceived on open and closed vowels) on initial assessment. On post-therapy assessment, hypernasality was rated as grade 2 (perceived on vowels and voiced pressure consonants) and grade 1 audible nasal emission (slight emission). Grace showed grade 1 hyponasality and grade 1 nasal turbulence on initial assessment. On maintenance assessment she was assessed as having grade 1 hypernasality (perceived on closed vowels) and slight, but more consistent nasal turbulence. Thus, for 4/6 participants the trend was for increased signs of VPI following treatment.

6.2.5.2 Consonant production on GOS.SP.ASS

All participants showed improved consonant production on the GOS.SP.ASS on post-treatment assessment, and this improvement mirrors gains described in 6.2.1 (i.e. independent SLT assessment of treated and untreated words). George's residual errors included, at times /tʃ/ → [tj], /dʒ/ → [dj], /θ/ → [f], and /ð/ →[v]. Olivia's speech production was typical on GOS.SP.ASS. As well as lingual EPG targets, Olivia's therapy has also included teaching /θ/ and /ð/ and these sounds were assessed as typical on GOS.SP.ASS on post-treatment assessment. Emma's consonant production on GOS.SP.ASS was also typical, apart from /ð/ production. Emma was given limited instruction with /θ/ and /ð/ towards the end of her intervention. Victoria's consonant production was accurate, apart from at times /z/ was produced as [s] and /dʒ/ as [tʃ] or [dz] on post-therapy assessment. Grace's speech production was typical on GOS.SP.ASS on post-intervention assessment. Likewise, Poppy's speech production was typical on GOS.SP.ASS on post-therapy assessment. It should be noted that in this study dentalisation/minimal interdental production of alveolar targets was deemed "typical".

6.2.5.3 Percentage of consonants correct on DEAP

Percentage of consonants correct on DEAP ranged from 92.3% - 100% on post-therapy assessment. Prior to intervention, PCC on DEAP ranged from 52% - 87%.

6.2.6 Summary of impressionistic perceptual speech outcomes

The results outlined in this section contribute to answering research questions number one and number two: **Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated words/pseudo-words? Does improved accuracy of target phonemes generalise to continuous connected speech?**

All participants showed improved accuracy of targeted phonemes in treated words during intervention and on post-intervention assessment. Most target phoneme production went from near zero levels of accuracy on production of treated words on baseline assessments to near 100 per-cent levels of accuracy for most target phonemes on post-therapy assessment. This improved accuracy generalised to untreated words for all participants and large treatment effect sizes were shown for most target speech sounds. For most target sounds, accuracy only increased when these were worked on in therapy. Improved accuracy with production of target phonemes in words also showed generalisation to connected, continuous speech for all participants on maintenance assessment, though to varying degrees. George showed 80% generalisation to connected speech, Olivia 100%, Emma 98.07%, Victoria 78.94%, Grace 95% and finally Poppy achieved 92% generalisation to continuous speech.

The results in this section also contribute to answering research question number three: **Do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in terms of a word frequency effect?**

This study predicted target phonemes contained in high frequency words would be more resistant to improved production, compared with improvement of target phonemes in low frequency words. This frequency effect was not shown at word level, i.e. no difference in production was seen with low and high frequency word production on assessment at the end of five sessions and the end of ten sessions. However, target phonemes in high frequency words were more likely to be produced in error in continuous connected speech, than target phonemes in low frequency words in the acquisition and early generalisation phase of therapy, and this difference was statistically significant ($p < .05$).

6.3 Speech understandability, speech acceptability and quality of life measurement

This section describes findings from speech understandability, speech acceptability and quality of life measurement. The results outlined in this section contribute to answering research question number two: **Does improved accuracy of target phonemes generalise to continuous, connected speech, and, if so, does this result in improved ratings of speech understandability and speech acceptability and improved ratings of impact on quality of life, post-intervention?**

6.3.1 Speech Understandability

Table 15 shows scores on the Intelligibility in Context Scale. Table 16 shows further breakdown of scores into the seven speaking situations.

Table 15

Intelligibility in context scale scores at assessment points (average score across seven different speaking conditions)

Participant	ICS score			
	Initial Assessment	Baseline 3	End of therapy	Maintenance
George	3.85	3.71	4.00	3.85
Olivia	3.42	3.50	4.00	4.33
Emma	3.50	3.14	4.28	5.00
Victoria	3.80	3.80	4.80	4.40
Grace	4.57	4.57	4.71	4.71
Poppy	4.28	3.80	5.00	5.00

Note: ICS = Intelligibility in context; score of 3 = sometimes understood; score of 4 = usually understood; score of 5 = always understood.

Table 16

Intelligibility in Context Scale responses for the seven listener situations for each participant

Participant	Listeners	Initial assessment	Baseline 3	Post-therapy	Maintenance
George	Parents				
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers				
	Strangers				
Olivia	Parents				
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers	N/A	N/A	N/A	N/A
	Strangers				
Emma	Parents				
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers				
	Strangers				
Victoria	Parents	N/A	N/A	N/A	N/A
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers	N/A	N/A	N/A	N/A
	Strangers				
Grace	Parents				
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers				
	Strangers				
Poppy	Parents				
	Immediate members of family				
	Extended members of family				
	Child's friends				
	Acquaintances				
	Child's teachers				
	Strangers				

Note: N/A = Not applicable; Green shading = Always understands; Light green shading = Usually understands; Orange shading = Sometimes understands; Red shading = Rarely understands.

Tables 15 and 16 show all participants' speech was rated as 3 – 4 pre-therapy (i.e. speech sometimes to usually understandable) in some listener situations. All participants showed improvements with ICS scores post therapy, apart from George, whose score on maintenance was unchanged from initial assessment (3.85). George's parent's score of 3.85 on maintenance was attributed to a fast rate of speech and low speech volume, rather than distorted speech sound production or nasal turbulence, the later which had increased over the course of therapy. None of the participants achieved a full score of 5 (i.e. speech is always understood) post-therapy, apart from Emma on maintenance and Poppy on post-therapy and maintenance assessments. Emma's mother reported her score of 4.28 immediately following therapy was because she felt Emma's speaking was affected by low confidence. On maintenance assessment, Emma's mother reported that Emma's general confidence with speaking had greatly improved, and consequently scored Emma 5 across all speaking situations. Poppy's mother felt that Poppy's speech was always understood post-therapy.

Table 17 shows the independent SLTs' ratings of speech understandability at the three baseline assessments and the two assessments post-therapy. These ratings were made after listening to the 2-minute sample of spontaneous speech. Table 18 shows the independent SLTs' ratings of speech acceptability at the same assessment points. These ratings were made after listening to the 2-minute sample of spontaneous speech together with participants' single word production of treated and untreated words.

Table 17

Independent SLTs' ratings of participants' speech understandability (ratings from 2-minute connected speech sample) at B1, B2, B3, P and M

Participant	Independent SLT rating	Assessment				
		B1	B2	B3	P	M
George	SLT no. 1	Orange	Light green	Light green	Light green	Orange
	SLT no. 2	Orange	Light green	Light green	Light green	Orange
Olivia	SLT no. 1	Orange	Orange	Orange	Green	Green
	SLT no. 2	Orange	Light green	Light green	Green	Green
Emma	SLT no. 1	Orange	Orange	Orange	Light green	Light green
	SLT no. 2	Light green	Orange	Orange	Light green	Green
Victoria	SLT no. 1	Red	Red	Red	Light green	Light green
	SLT no. 2	Light green	Orange	Light green	Light green	Light green
Grace	SLT no. 1	Green	Green	Light green	Light green	Light green
	SLT no. 2	Green	Green	Light green	Green	Green
Poppy	SLT no. 1	Green	Light green	Light green	Green	Green
	SLT no. 2	Green	Light green	Green	Green	Green

Note: SLT = Speech and language therapist; B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 Immediately pre-therapy; P = Assessment immediately post-intervention; M = Maintenance assessment 3-months post-intervention; Green shading = Always easy to understand; Light green shading = Occasionally hard to understand; Orange shading = Often hard to understand; Red shading = Hard to understand most of the time.

Table 18

Independent SLT's ratings of participants' speech acceptability (ratings from 2-minute connected speech sample and single word production) at B1, B2, B3, P and M

Participant	Independent SLT	Assessment				
		B1	B2	B3	P	M
George	SLT no. 1	Orange	Orange	Orange	Light green	Orange
	SLT no. 2	Orange	Orange	Orange	Light green	Orange
Olivia	SLT no. 1	Orange	Orange	Orange	Light green	Light green
	SLT no. 2	Orange	Orange	Light green	Light green	Green
Emma	SLT no. 1	Red	Red	Red	Light green	Light green
	SLT no. 2	Orange	Orange	Orange	Light green	Light green
Victoria	SLT no. 1	Red	Red	Red	Orange	Orange
	SLT no. 2	Orange	Red	Orange	Light green	Light green
Grace	SLT no. 1	Light green	Light green	Orange	Light green	Light green
	SLT no. 2	Light green	Light green	Light green	Light green	Light green
Poppy	SLT no. 1	Light green	Orange	Light green	Green	Green
	SLT no. 2	Light green	Light green	Light green	Light green	Green

Note: SLT = Speech and language therapist; B1 = Baseline 1; B2 = Baseline 2; B3 = Baseline 3 immediately pre-therapy; P = Assessment immediately post-intervention; M = Maintenance assessment 3-months post-intervention; Green shading = Normal acceptability; Light green shading = Mild deviation; Orange shading = Moderate deviation; Red shading = Severe deviation.

Olivia, Emma, Victoria and Poppy showed improved speech understandability following therapy. George's speech understandability on maintenance assessment was unchanged from baseline 1 assessment (often hard to understand). The independent SLTs attributed reduced scores with speech understandability on maintenance assessment to noticeable nasal turbulence, rather than distorted articulation, as had been the case on the three baseline assessments. As discussed previously, George's nasal turbulence increased in severity over the course of therapy. Grace's speech understandability was not particularly affected by her SSD on baseline assessment. Post-therapy, understandability was rated down by one SLT due to the presence of nasal turbulence, which had increased over the course of therapy. Only 2/6 participants were rated as dark green (i.e. always easy to understand) on maintenance assessment (Olivia and Poppy). Emma was received as score of light green due to mild hypernasality and nasal turbulence, along with a distorted /ɹ/. Victoria's understandability was rated down due to hypernasality and weak oral pressure consonant production. As discussed above, George and Grace's reduced scores with understandability on pre-therapy assessment were due to nasal turbulence.

Olivia, Emma, Victoria, Grace and Poppy showed improved speech acceptability post-therapy (see Table 18). George's acceptability was unchanged on maintenance assessment, despite having improved his speech sound production. Reduced speech acceptability was attributed to nasal turbulence, which had increased over the course of therapy. Only Poppy was rated as having normal speech acceptability by both independent SLTs on maintenance assessment. Olivia's reduced score on maintenance assessment by one SLT was attributed to a stilted speech quality, mild hyponasality and a higher pitch quality. Emma's reduced scores on maintenance were attributed to mild hypernasality, occasional nasal turbulence and errors with /ɹ/ and /l/. Victoria's reduced speech acceptability post-therapy was ascribed to hypernasal resonance, audible nasal emission and somewhat stilted speech production. Graces' lower score on maintenance assessment was due to nasal turbulence and dentalisation of /s/ and /z/.

6.3.2 Quality of life

The SPAA-C was used to examine the impact of the SSD on everyday life in child participants, pre- and post-therapy, while the QoL-Dys questionnaire was used to assess quality of life pre- and post-therapy in adult participants.

6.3.2.1 SPAA-C

The SPAA-C was administered to the three child participants, George, Emma and Grace. Pre-therapy, George felt happy about his speech in all speaking situations, apart from when he spoke to the whole class and when people did not understand what he said; in these situations, he felt "in the middle" between happy and sad. Post-therapy his ratings were unchanged. Pre-intervention, Emma felt her talking was different to other people. She said she did not like talking to strangers, for example in restaurants. She reported she was happy when talking to her mum and dad, her best friend and her teachers. She did not know how she felt when teachers asked her a question, when she played on her own or when people did not understand what she said. On post-treatment assessment, all ratings improved and she reported feeling happy with her speech in all speaking situations. She said everyone could understand her and she felt happy talking at home, at school and outside of home and school. Emma's parents reported Emma's general confidence with talking was greatly increased post-therapy. Prior to therapy, Grace's speech difficulties had a limited impact on her quality of life. She felt happy about her speech in all speaking situations, apart from when she spoke to the whole class and when people did not understand what she said. In these situations, she felt "in the middle" between happy and sad. Post-therapy, she reported that at times it was difficult to "speak well". She felt happy talking to her family. Her recent schooling had been online due to the coronavirus pandemic and she reported she felt nervous speaking to her teachers and peers online; it was not clear if this was speech-related or due to general nervousness with using a new medium of communication.

6.3.2.2 QoL-Dys

Table 19 shows Olivia, Victoria and Poppy's self-rating scores on the QoL-Dys quality of life questionnaire on initial assessment, assessment immediately following therapy and assessment 3-months following intervention. The maximum score for this questionnaire is 160. A score of 0 occurs when an individual's speech difficulties are having no impact on their everyday functioning. A score of 160 represents severe impact in every aspect of life. All three adult participants felt their SSD was impacting on some aspects of their everyday functioning pre-therapy, though for Poppy this impact was minimal. Olivia scored 62 on initial assessment. Her ratings on this self-assessment showed:

- Strangers almost always had difficulty understanding her;
- She felt her speech almost always sounded unnatural;
- She almost always experienced difficulty making herself understood on the phone.

Post-therapy, she scored 41 on post-therapy assessment and 39 on maintenance assessment. She rated herself as:

- Almost never having difficulty talking to strangers;
- Almost never having difficulty talking on the phone;
- Sometimes felt her speech sounded unnatural.

Prior to intervention Olivia had been unemployed and had not engaged in any activities out of the house that involved contact with less familiar people. On maintenance assessment, Olivia had secured a part-time job and was increasingly engaging in activities out of the house independently. Oliver's scores of 41 and 39 post-therapy related to her on-going hearing difficulties.

Table 19

Adult participants' scores on the QoL-Dys measure

Participant	Scores on QoL- Dys		
	Initial assessment	Assessment immediately following therapy	Maintenance assessment
Olivia	62	41	39
Victoria	66	22	35
Poppy	18	26	21

Note: QoL-Dys = Quality of Life, Dysarthria instrument*;* 0 = Speech never impacts on everyday life; 160 = Maximum score where speech always impacting on life

Victoria scored 66 on initial assessment. Her ratings showed:

- Strangers sometimes had difficulty understanding her speech;
- She almost always having difficulty making herself understood on the phone;
- She almost always felt her speech sounded unnatural.

Victoria reported use of a range of compensatory strategies, including getting people's attention before she spoke, positioning herself so she could be seen and using different wording if she hadn't been able to make herself understood. Because of her speech, she felt:

- Sometimes people treated her as if she wasn't very bright;
- Sometimes people got irritated with her;
- Sometimes people spoke louder to her or ignored her because of her speech difficulties.

Post intervention, Victoria scored 22 on post-therapy assessment and 35 on maintenance assessment. She reported strangers still sometimes had difficulty understanding her and she still needed to use compensatory strategies, such as facing people, when she talked. However, she reported:

- She no longer had difficulty talking on the phone;
- People no longer treated her as if she wasn't very bright;
- People no longer ignored her;
- People no longer got irritated or spoke louder.

Over the course of therapy Victoria changed jobs to progress her career and post-therapy was very successfully working in a job requiring spoken communication.

Poppy scored herself 18 on initial assessment, i.e. speech difficulties were only having a mild impact on her quality of life. Her ratings showed:

- Strangers at times had difficulty understanding her;

- She felt her speech was unnatural sounding;
- She didn't like speaking in class;
- She had difficulty communicating when she was upset.

Post-therapy her scores on the QoL-Dys increased slightly to 26 and 21. Poppy rated herself as no longer thinking her speech was difficult to understand or was unnatural sounding. However, she was reporting more difficulty talking up in class and speaking on the phone. The reason for these increases in scores post-therapy was not clear, but may have been related to the on-going COVID-19 pandemic which had isolated Poppy to an extent.

6.3.3 Summary of speech understandability, speech acceptability and quality of life measurement

The results in this section contribute to answering research question number three: **Does improved accuracy of target phonemes generalise to continuous, connected speech, and, if so, does this result in improved ratings of speech understandability and speech acceptability and improved ratings of impact on quality of life, post-intervention?**

Most of the participants could make themselves understood to familiar listeners pre-therapy, so scores on the ICS for familiar listeners ranged between 3 - 4 (speech sometimes to usually understandable). All participants showed improvement with speech understandability post-therapy (on ICS and independent SLT assessment), reflecting improvements with speech sound production following therapy. The exception to this was George. Prior to therapy, George's reduced speech understandability was attributed to his lateral speech productions. Post-therapy, reduced speech understandability was attributed to nasal turbulence, which had increased over the course of therapy, a fast rate of speech and low volume of voice, rather that lateral production. Although all participants' speech sound production improved following therapy, only Olivia and Poppy's speech was rated as "normal" by the independent SLTs post-therapy. According to independent SLT ratings, the remaining participants' decreased scores were attributed to presence of hypernasality, audible nasal emission and nasal turbulence.

Likewise, all participants showed improved speech acceptability post-therapy, apart from George. Only Poppy's speech was rated as "normal" by the independent SLTs. Independent SLTs' reduced speech acceptability ratings were attributed to hypernasality, audible nasal emission, nasal turbulence and mild developmental speech distortions.

Quality of life for 2/3 child participants was not particularly affected by their SSD, as measured by the SPAA-C, and their quality of life was largely unchanged post-therapy. Quality of life for the other child participant was markedly affected by her SSD pre-therapy. This child had the greatest number of speech sounds produced in error pre-therapy and was also older. Post-therapy, this child's quality of life was markedly improved. For 2/3 adults, quality of life was noticeably affected by their SSDs pre-therapy, as measured by the QoL-Dys. Post therapy, quality of life for these two adult participants was noticeably improved.

For the other adult, quality of life was not noticeably impacted by her SSD. Post-therapy this adult's quality of life was slightly reduced, though this reduction may have been due to the COVID-19 pandemic.

7 RESULTS: EPG OUTCOMES

This chapter presents findings from EPG analysis prior to, during, and post therapy. EPG analysis is discussed under two sub-headings: Qualitative analysis and quantitative analysis. Qualitative analysis consists of visual and descriptive analysis of EPG frames. Quantitative analysis comprises numerical indices relating to EPG frames. All EPG annotation and analysis was completed by the researcher. EPG results contribute towards answering research question number one: **Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated/pseudo- words?** In addition, qualitative analysis of EPG frames contributes towards research question number three: **Do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in terms of demonstration of gradient phonetic change with target phonemes.**

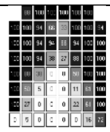
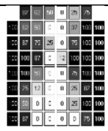
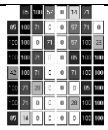
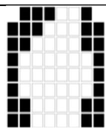
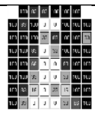
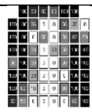
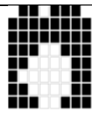
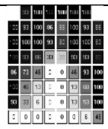
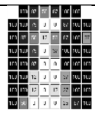
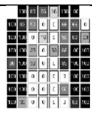
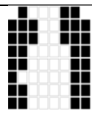
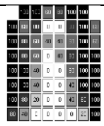
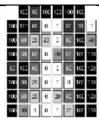
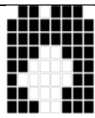
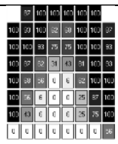
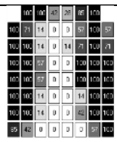
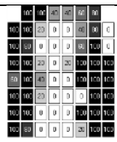
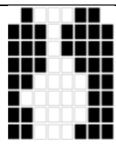
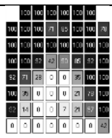
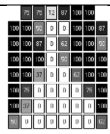
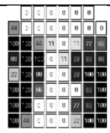
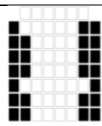
7.1 Qualitative EPG analysis (visual and descriptive)

Figures 22 - 27 show cumulative EPG frames for target speech sounds at each EPG assessment point, for each participant. EPG cumulative frames show frames of maximum contact and come from repetitive production of pseudo-words. George, Emma and Grace outgrew their EPG plates during the course of therapy. Consequently, EPG assessment was only possible for these participants while their EPG palates still fitted. For George this was up to and including assessment at the end of 10 sessions, for Emma up to and including assessment at the end of 20 sessions, and for Grace up to and including assessment at the end of 15 sessions.

On perceptual speech analysis, George presented with a pattern of lateral production with his production of /z/, /ʃ/, /tʃ/, and /dʒ/ (these sounds were transcribed as [ɮ], [ɬ], [ɬ], and [ɮ] respectively by the three transcribing SLTs). This perceptual analysis of lateral production with anterior tongue-palate placement was confirmed with EPG qualitative analysis, as shown in Figure 22. Cumulative EPG frames taken on baseline 3 show complete anterior closure for production of /z/ and /ʃ/, and complete anterior closure for the fricative elements of /tʃ/, and /dʒ/. In all frames, lateral release around the lateral margins occurred on the right. On EPG analysis /tʃ/, and /dʒ/ did not include a stop element and there was no observable difference between /z/ and /dʒ/ and /ʃ/ and /tʃ/, confirming the loss of phonemic contrast between these speech sounds, as suggested by perceptual speech analysis. By assessment at the end of 5 sessions, George's lateral pattern was no longer evident in pseudo-word production. Instead, a central groove was shown, where air was being directed over the centre of the tongue. In addition, by assessment at the end of five sessions George was also evidencing use of the stop element in tongue-palate contact for his affricatives. However, the central groove for the fricative element of these affricative sounds was more anterior compared to reference frames. In addition, the central groove for production of /ʃ/ was more anterior (and was transcribed perceptually as [s]). By assessment at the end of 10 sessions, the central groove for /ʃ/ had retracted to post-alveolar placement and was similar to the reference frame, and was transcribed perceptually as [ʃ]. However, the central groove for the fricative element of /tʃ/, and /dʒ/ remained more anterior, and likewise, the stop element for these affricatives was more anterior than reference frames. George's production of /tʃ/, and /dʒ/ was transcribed perceptually in words as [tj] or [tʃ] and [dj] or [/dʒ/] post-therapy and on maintenance assessment.

Figure 22

Cumulative EPG frames (pseudo-words) of maximal contact for George's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and researcher's transcriptions at each time point

Participant	Speech sound / speech element	B3	A5	A10	Reference frame
George	/z/	 Complete closure			 Typical
	Phonetic transcription	[ɮ]	[z]	[z]	[z]
	/tʃ/ - stop element	No stop element			 Typical
	Phonetic transcription		[t]	[t]	[t]
	/tʃ/ - fricative element	 Complete closure			 Typical
	Phonetic transcription	[ɬ]	[s]	[s]	[ʃ]
	/dʒ/ - stop element	No stop element			 Typical
	Phonetic transcription		[d]	[d]	[d]
	/dʒ/ - fricative element	 Complete closure			 Typical
	Phonetic transcription	[ɮ]	[z]	[z]	[ʒ]
	/ʃ/	 Complete closure			 Typical
	Phonetic transcription	[ɬ]	[s]	[ʃ]	[ʃ]

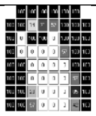
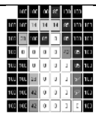
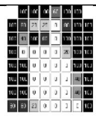
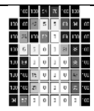
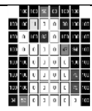
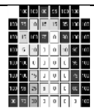
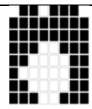
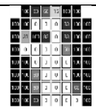
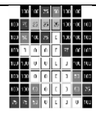
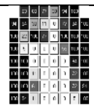
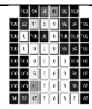
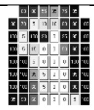
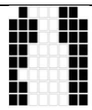
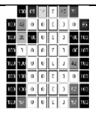
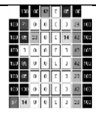
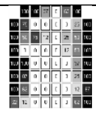
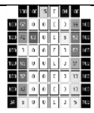
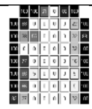
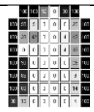
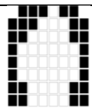
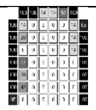
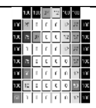
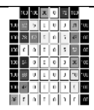
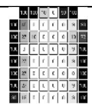
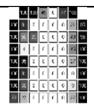
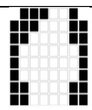
Note: B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; Reference frames are researcher's productions of target sounds (frame of maximum contact).

On perceptual speech analysis, Olivia showed a pattern of post-alveolar or palatal production, with /s/ and /z/ transcribed as [ʃ] or [ç] and [ʒ] or [ʝ] by the independent SLTs.

In addition, /tʃ/ was transcribed as [ʃ]. This perceptual analysis of retracted placement for /s/ and /z/, and deaffrication of /tʃ/ was confirmed on EPG visual analysis (see Figure 23). By assessment at the end of 5 sessions Olivia showed stop tongue-palate contact for /tʃ/. By assessment at the end of 5 sessions tongue-palate contact for /s/ and /z/ had moved to alveolar placement, and these sounds were transcribed perceptually as [s] and [z]. However, as can be seen on Figure 23, width of groove for these sounds became smaller on subsequent assessment points, suggesting further phonetic refinement of these sounds as the functional maintenance stage of therapy progressed.

Figure 23

Cumulative EPG frames (pseudo-words) of maximal contact for Olivia's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and researcher's transcriptions at each time point

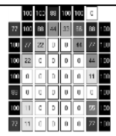
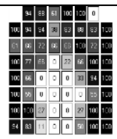
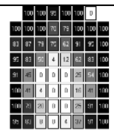
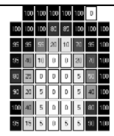
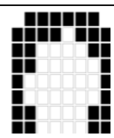
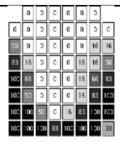
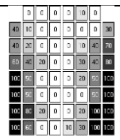
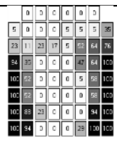
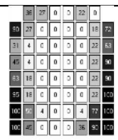
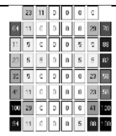
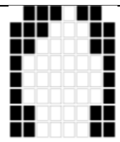
<table>
<tr><th>Partic.</th><th>Speech sound / speech element</th><th>B3</th><th>A5</th><th>A10</th><th>A15</th><th>A20</th><th>P</th><th>M</th><th>Reference frame</th></tr>
<tr><td rowspan="8">Olivia</td><td>/tʃ/- stop element</td><td>No stop element</td><td></td><td></td><td></td><td></td><td></td><td></td><td>
Typical</td></tr>
<tr><td>Phonetic transcription</td><td></td><td>[t]</td><td>[t]</td><td>[t]</td><td>[t]</td><td>[t]</td><td>[t]</td><td>[t]</td></tr>
<tr><td>/tʃ/ - fricative element</td><td></td><td></td><td></td><td></td><td></td><td></td><td></td><td>
Typical</td></tr>
<tr><td>Phonetic transcription</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td><td>[ʃ]</td></tr>
<tr><td>/s/</td><td>
Retracted</td><td></td><td></td><td></td><td></td><td></td><td></td><td>
Typical</td></tr>
<tr><td>Phonetic transcription</td><td>[ç]</td><td>[s]</td><td>[s]</td><td>[s]</td><td>[s]</td><td>[s]</td><td>[s]</td><td>[s]</td></tr>
<tr><td>/z/</td><td>
Retracted</td><td></td><td></td><td></td><td></td><td></td><td>
l</td><td>
Typical</td></tr>
<tr><td>Phonetic transcription</td><td>[j̹]</td><td>[z]</td><td>[z]</td><td>[z]</td><td>[z]</td><td>[z]</td><td>[z]</td><td>[z]</td></tr>
</table>

Note: B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; A15 = Assessment end of 15 sessions; A20 = Assessment end of 20 sessions; P = end of therapy assessment; M = Maintenance assessment. Reference frames are researcher's productions of target sounds (frame of maximum contact); Partic.= Participant.

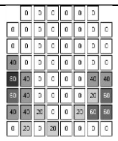
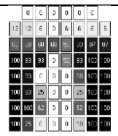
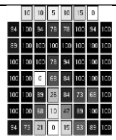
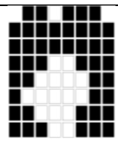
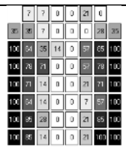
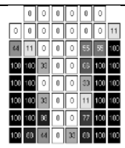
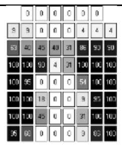
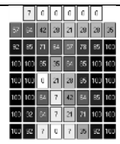
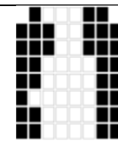
Emma's speech patterns on perceptual speech assessment included backing and lateral production. The independent SLTs showed some variation in transcription of target sounds. Although always transcribed as being produced in error, the sounds /s/, /z/, /ʃ/, /tʃ/ and /dʒ/ were at times transcribed as [ç] (in the case of /s/, /ʃ/, and /tʃ/) and [ʝ] (in the case of /z/ and /dʒ/) (i.e. palatal), and at other times transcribed as [ɬ] or [ɮ] (i.e. lateral fricative). The researcher transcribed these sounds as velars with lateral release (i.e. ([𝼄] and [𝼄̬]). EPG assessment at baseline 3 (see Figure 24) showed that /s/ and /z/ were always retracted to velar and complete closure was shown, confirming the researcher's perceptual transcription of velar lateral sounds, ([𝼄] and [𝼄̬]). Likewise, /tʃ/ showed a pattern of posterior placement with lateral release. The /tʃ/ sound had a stop element (perceptual spectrogram and waveform analysis), and the signs were that this stop element was posterior, likely in the posterior velar/uvular region (i.e. not completely visible by EPG). No clear central groove was seen for the fricative element of this speech sound, indicating lateral release. On production of pseudo-words /dʒ/ had an acoustic and perceptual stop element and again the signs were that this stop element was posterior, likely in the posterior velar/uvular region (i.e. not completely visible by EPG). This speech sound did not have a fricative element. The /ʃ/ sound was transcribed perceptually as [ç] or [ʃʲ] (independent SLTs) or ([𝼄] (researcher). EPG analysis suggested tongue-palate contact was again more posterior. Although transcribed perceptually as the same as /s/, on EPG analysis /ʃ/ was distinct from /s/ (i.e. with tongue-palate contact further back for/ʃ/).

Figure 24

Cumulative EPG frames (pseudo-words) of maximal contact for Emma's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and researcher's transcriptions at each time point

Participant	**Speech sound / speech element**	**B3**	**A5**	**A10**	**A15**	**A20**	**Reference frame**
Emma	**/t/**	 Retracted to posterior placement					 Typical
	Phonetic transcription	[k]	[t̪]	[t]	[t]	[t]	[t]
	/s/	 Retracted to velar; complete closure					 Typical
	Phonetic transcription	[𝼄]	[s̠]	[s̠]	[s]	[s]	[s]

	/d/	Retracted to posterior placement					Typical
	Phonetic transcription	[g]	[d̪]	[d]	[d]	[d]	[d]
	/z/	Retracted to velar; complete closure					Typical
	Phonetic transcription	[ɫ̰]	[z̪̄]	[z̪̄]	[z]	[z]	[z]
	/tʃ/- stop element	Retracted to posterior					Typical
	Phonetic transcription	[k]	[t] or [s]	[t]	[t]	[t]	[t]
	/tʃ/- fricative element					No fricative element	Typical
	Phonetic transcription	[ɫ]	Prolonged aspiration from /t/	Prolonged aspiration from /t/	Prolonged aspiration from /t/		[ʃ]
	/ʃ/	Retracted to posterior					Typical
	Phonetic transcription	[ɫ]	[s̪] or [t̪]	[ʃ]	[ʃ]	[ʃ]	[ʃ]
	/n/	Retracted to posterior					Typical
	Phonetic transcription	[ŋ]	[n]	[n]	[n]	[n]	[n]
	/l/	Retracted to posterior					Typical
	Phonetic transcription	[ɰ]	[n] or [l]	[l]	[l]	[l]	[l]

	/dʒ/ - stop element	 Retracted to posterior	No stop element	No stop element			 Typical
	Phonetic transcription	[g]			[d]	[d]	
	/dʒ/ - fricative element	No fricative element					 Typical
	Phonetic transcription		[s]	[ʃ]	[ʒ]	[ʒ]	[ʒ]

Note: B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; A15 = Assessment end of 15 sessions; A20 = Assessment end of 20 sessions; reference frames are researcher's productions of target sounds (frame of maximum contact).

By assessment at the end of five sessions, Emma's lateral pattern was no longer in evidence, with a clear midline groove shown with production of /s/, /z/, /ʃ/, /tʃ/ and /dʒ/. Emma's /s/ and /z/ were typically transcribed as [s̪] and [z̪] at word level, /ʃ/ was transcribed as [s̪], /tʃ/ as [ts̪] or [tj] and /dʒ/ as [d], [dj] or [dz]. As therapy progressed EPG shows changes with tongue-palate contact for /s/ and /z/, with anterior movement of tongue-palate contact. By assessment at the end of 10 sessions, Emma's /ʃ/ was perceptually accurate, and this change is shown on EPG frames with increased lateral contact. Again, slight changes are shown with subsequent EPG assessment for /ʃ/, with a more defined palatal groove formation as time progressed. Emma's production of /tʃ/ and /dʒ/ was somewhat variable. Although Emma's EPG images were looking closer to reference frames, these speech sounds were not consistently produced accurately by assessment at the end of 20 session (i.e. the last EPG assessment point before Emma outgrew her EPG palate). At this point, /tʃ/ was transcribed as [tʃ] or [tj] and /dʒ/ as [dʒ] or [dj]. Emma's production of /tʃ/ was targeted for therapy in session number four. Emma's /dʒ/ production was not targeted until session number 10. However, changes to this later sound were seen at assessment at the end of five sessions (transcribed perceptually as a weak [zʲ], indicating a degree of cross-phonemic generalisation).

The independent SLTs transcription of Emma's production of /n/, /t/, /d/, /l/ was variable; at times these were transcribed as accurate, at other times they were transcribed as backed to velar or with double articulation (alveolar-velar in case of stops or /j/ for /l/). In comparison, the researcher transcribed these are being consistently produced in error with backing to velar (in case of /n/, /t/, /d/ and /l/). The researcher also at times transcribed /n/, /t/, and /d/ with double articulation (dental alveolar and velar). EPG showed consistent retraction to velar placement for all these speech sounds and these speech sounds were never produced accurately. No double articulation was evidenced on EPG. This may have been because the tongue tip was elevated, but not making contact with the palate. Alternatively, tongue tip contact may have been dental or interdental and not shown on

EPG. By assessment at the end of five sessions, /n/, /t/, and /d/ showed anterior placement. Over time, EPG contact patterns for these sounds showed slight change with less tongue-palate contact, suggesting phonetic refinement in production of these speech sounds as therapy progressed. Production of /l/ had also altered by assessment at the end of five sessions, despite this speech sound not yet being worked on in therapy. This speech sound was transcribed perceptually as [n] or [l] on production of pseudo-words at assessment at the end of 5 sessions. This suggests probable cross-phonemic generalisation. By assessment at the end of 10 sessions, this sound had increased in accuracy and was looking more similar to the reference /l/ frame. By assessment at the end of 20 sessions, this speech sound was consistently produced accurately, though visual EPG analysis shows increased contact compared with the reference frame.

On perceptual assessment, Victoria's main cleft speech characteristic was pharyngeal articulation of a number of lingual sibilants. On initial assessment the researcher transcribed her production of /s/ as [s̪͡ħ], /z/ was [n], /ʃ/ was transcribed as [ħ], /tʃ/ as [ħ], and /dʒ/ as [nj]. Independent SLT transcription on baseline assessment was similar to the researcher's transcription, though one independent SLT's transcribed /s/ as [ñ̥] or [ħ]. EPG at baseline 3 showed tongue-palate placement for /s/, but little evidence of a mid-line groove and an incomplete horseshoe shape on the left suggesting reduced oral pressure, and consistent with [s̪͡ħ] (see Figure 25). Tongue-palate contact for /z/ showed complete anterior closure consistent with /n/. Some tongue-palate contact was seen laterally for production of /ʃ/, suggesting double articulation of this sound, though perceptually this was transcribed as [ħ]. Since incomplete lateral bracing was evident, it would appear this tongue-palate contact involved silent posturing. Victoria's /tʃ/ EPG contact pattern was identical to her /ʃ/ EPG pattern. Her /dʒ/ EPG pattern was one of complete alveolar contact, consistent with the perceptual transcription of [nj] and no fricative element was seen on EPG. Thus, Victoria showed lack of phonemic contrast between /z/ and /n/, and /ʃ/ and /tʃ/. By assessment at the end of 5 sessions, Victoria showed anterior groove formation for /s/ and /z/ and production was not accompanied by pharyngeal friction. However, this groove formation was wide and slightly retracted. Perceptually /s/ and /z/ were transcribed as [sʲ] and [zʲ]. By assessment at the end of 10 sessions, placement for /s/ and /z/ had moved forward and Victoria's production of these sounds was perceptually accurate. EPG cumulative frames at assessment at the end of 10 sessions show more anterior tongue-palate placement, though groove formation was wide. This groove did reduce in size over time. Nevertheless, it remained wider than that seen in the reference frames. This appeared due to Victoria's difficulty in building up pressure in her mouth to make these sibilant sounds. This difficulty in achieving oral pressure was likely due to VPI. In addition, although Victoria's /z/ EPG frames matched reference frames, perceptually /z/ was often produced as [s]. Voiced production of this sound did increase over time, but by the end of therapy /z/ was still at times produced as [s]. By assessment at the end of five sessions, Victoria was producing a perceptually accurate /ʃ/ and her EPG frames show lateral bracing and are similar to the reference EPG frame. By assessment at the end of 10 sessions, Victoria's EPG frames for /tʃ/ were similar to reference frames for this speech sound. Victoria struggled more with production of /dʒ/. This speech sound was similar to reference EPG frames by assessment at the end of 20 sessions, though perceptually this sound was at times produced as [tʃ].

Figure 25

Cumulative EPG frames (pseudo-words) of maximal contact for Victoria's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and researcher's transcriptions at each time point

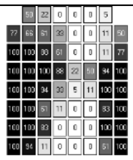
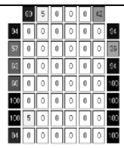
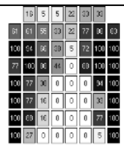
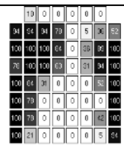
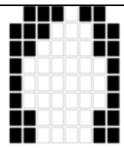
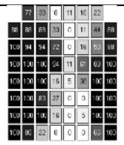
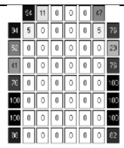
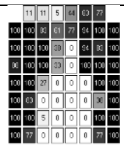
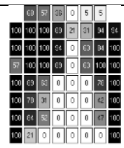
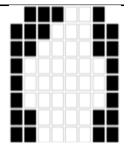
Part.	Speech sound/ speech element	B1	A5	A10	A15	A20	A25	P	M	Ref. frame
Vict.	/s/	Complete contact								Typical
	Phonetic transcrip.	[s̪͡ħ]	[ʃ̪̃]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s]
	/ʃ/	Double artic.								Typical
		[ħ]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ]
	/z/	Complete contact								Typical
	Phonetic transcrip.	[n]	[ʃ̪̃]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]	[s̪̃̇]/[z̪̃̇]	[s̪̃̇]/ [z̪̃̇]	[z]
	/tʃ/ - stop elem.	No stop element								Typical
	Phonetic transcrip.		[t̪]	[t̪]	[t̪]	[t̪]	[t̪]	[t̪]	[t̪]	[t]
	/tʃ/ - fric. elem.		No fric. element							Typical
	Phonetic transcrip.	[ħ]		[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]	[ʃ]
	/dʒ/ - stop elem.									Typical
	Phonetic transcrip.	[nj]	[t̪]	[t̪]	[t̪]	[t̪]	[t̪]	[t̪]/ [d̪]	[t̪]/ [d̪]	[d]
	/dʒ/ - fric. elem.	No fric element	No fric element		No fric element					Typical
	Phonetic transcrip.			[ʃ̪̃]		[ʃ̪̃]	[ʃ̪̃]	[ʃ̪̃]/[ʒ̪̃̇]	[ʃ̪̃]/[ʒ̪̃̇]	[ʒ]

Note: Part. = Participant; Vict. = Victoria; B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; A15 = Assessment end of 15 sessions; A20 = Assessment end of 20 sessions; A25 = Assessment end of 20 session; P = Assessment post-therapy; M = Maintenance assessment; Ref. = Reference frame; Reference frames are researcher's productions of target sounds (frame of maximum contact); Transcrip. – Transcription.

On perceptual analysis, Grace's cleft speech characteristic was palatal production of /s/ and /z/. All three SLTs transcribed /s/ as [ç] and /z/ as [ʝ]. This perceptual characteristic was confirmed on EPG on baseline three, with retracted palatal tongue-palate placement with a central groove (see Figure 68). By assessment at the end of five sessions, tongue-palate contact had moved forward for both /s/ and /z/, and these sounds were transcribed perceptually as [s̪͆] and [z̪͆[. Interdental production had been taught in therapy. By assessment at the end of 10 sessions, Grace was no longer producing an interdental /s/, rather her production of these speech sounds was typically transcribed as [ts] or [dz]. By assessment at the end of 15 sessions, Grace was producing a perceptually accurate /s/ and /z/. EPG frames on this assessment indicate slightly retracted placement, compared to the reference frame. However, production was perceptually accurate. Visual differences do exist between composite EPG frames at baseline 3 and assessment at the end of 15 sessions. At baseline 3 maximum grooving for these sibilants is occurring on the fourth row from the front, while on assessment at the end of 15 sessions tongue-palate contact is slightly more anterior with grooving apparently occurring on the third row. It is possible this tongue-palate contact may have shown further changes as therapy progressed, however, Grace had out-grown her EPG palate by assessment at the end of 20 sessions.

Figure 26

Cumulative EPG frames (pseudo-words) of maximal contact for Grace's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and researcher's transcriptions at each time point

Participant	Speech sound / speech sound element	B3	A5	A10	A15	Reference frame
Grace	**s**	 Retracted to palatal				 Typical
	Phonetic transcription	[ç]	[s̪͆]	[s̪]	[s̪]	[s]
	z	 Retracted to palatal				 Typical
	Phonetic transcription	[ʝ]	[z̪͆]	[z̪]	[z̪]	[z]

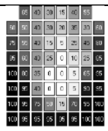
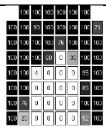
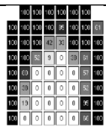
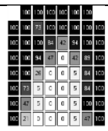
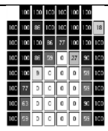
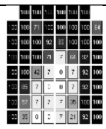
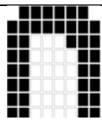
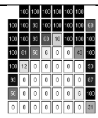
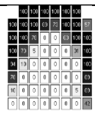
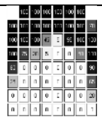
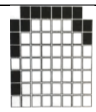
Note: B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; A15 = Assessment end of 15 sessions; Reference frames are researcher's productions of target sounds (frame of maximum contact).

On perceptual speech assessment, Poppy presented with a backing pattern for /n/ and /l/ production. The researcher consistently transcribed /n/ and /l/ as [ŋ] and [ɰ] respectively. However, the independent SLTs transcribed /n/ with 40 - 62% accuracy and /l/ with 71 - 85%

accuracy in single words on baseline assessment. The independent SLT transcribed errors were as /n/ → [ŋ], /l/ → [j]. Thus major discrepancies were evident between the researcher and the independent SLTs acoustic perceptual phonetic transcription. EPG assessment at baseline 3, showed that tongue-palate contact for /n/ and /l/ was always retracted to velar position for /n/ and /l/ (see Figure 27) in word initial position in pseudo-words. At times double articulation was seen for /n/, i.e. /n/ → [n͡ŋ] (see 8.1.2 for further discussion). By assessment at the end of session five, Poppy was producing all sounds accurately, perceptually, in pseudo-words. Tongue-palate placement had moved forward for /n/ and /l/ and production better matched reference frames.

Figure 27

Cumulative EPG frames (pseudo-words) of maximal contact for Poppy's target speech sounds at EPG assessment points (plus reference EPG frame of typical adult speaker for comparison) and research SLT's transcriptions at each time point.

Participant	**Speech sound / speech sound element**	**B3**	**A5**	**A10**	**A15**	**P**	**M**	**Reference frame**
Poppy	**n**	 Retracted to velar						 Typical
	Phonetic transcription	[ŋ]	[n]	[n]	[n]	[n]	[n]	[n]
	l	 Retracted to velar						 Typical
	Phonetic transcription	[ɰ]	[l]	[l]	[l]	[l]	[l]	[l]

Note: SLT = Speech and language therapist B3 = Baseline 3; A5 = Assessment end of 5 treatment sessions; A10 = Assessment end of 10 treatment sessions; A15 = Assessment end of 15 sessions; A20 = Assessment end of 20 sessions; P = end of therapy assessment; M = Maintenance assessment. Reference frames are research SLT's productions of target sounds (frame of maximum contact).

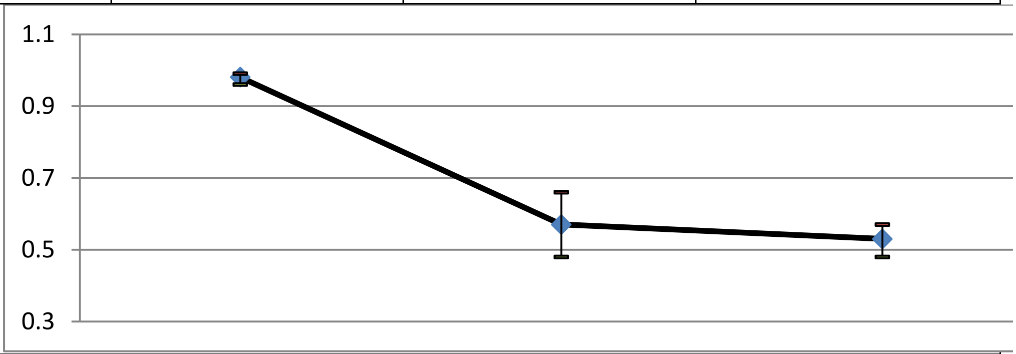
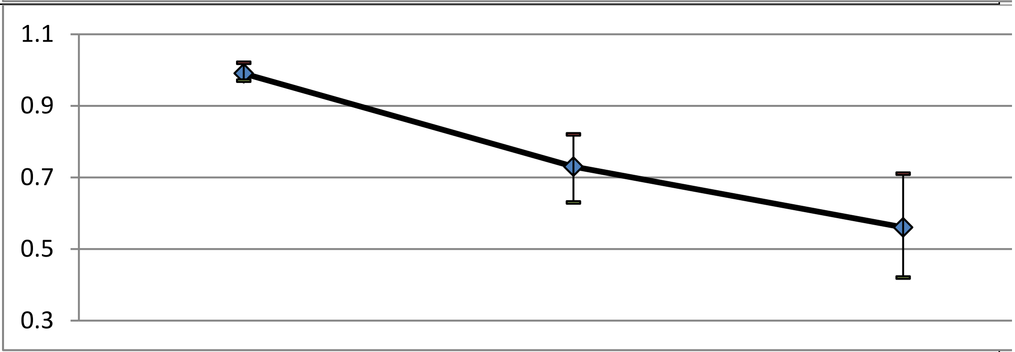
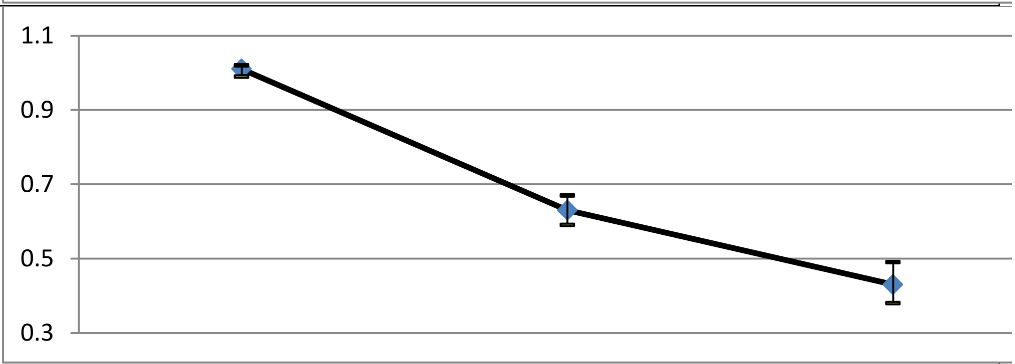
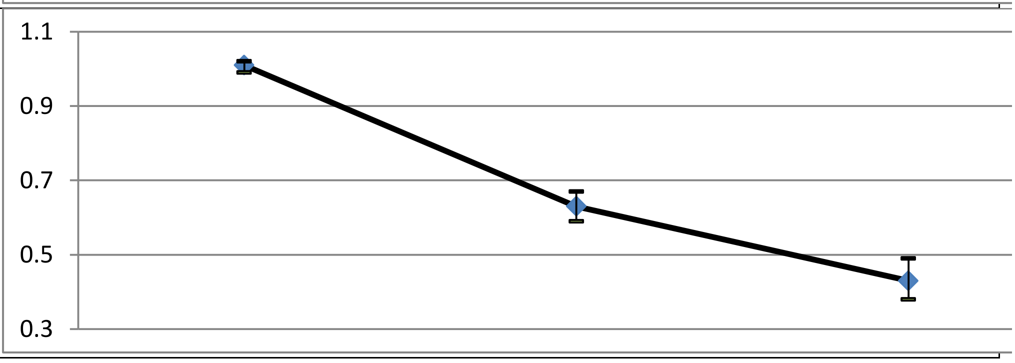
7.2 Quantitative EPG analysis (EPG indices)

EPG indices for participant's target sound at each EPG assessment point are shown in Figure 28 a - f by way of line graphs. Line graphs show mean index scores from repetitive production of pseudo-words at each assessment point and include error bars showing 95% confidence intervals. Selection of EPG indices and rationale for this selection is described in 5.2.6. Visual analysis of these graphs show change in indices pre-therapy, during and following therapy in the expected direction of improvement, and this is the case for all target sounds, for all participants. Increases in alveolar total (AT) and centre of gravity (CoG) scores, represents improvement with tongue-palate contact moving from posterior to more anterior. Decrease in alveolar closure (AC) scores represents improvement with a change from no central oral airflow to central tongue flow. Increase in whole total (WT) represents improvement with a change from little tongue-palate contact, to increased tongue-palate contact.

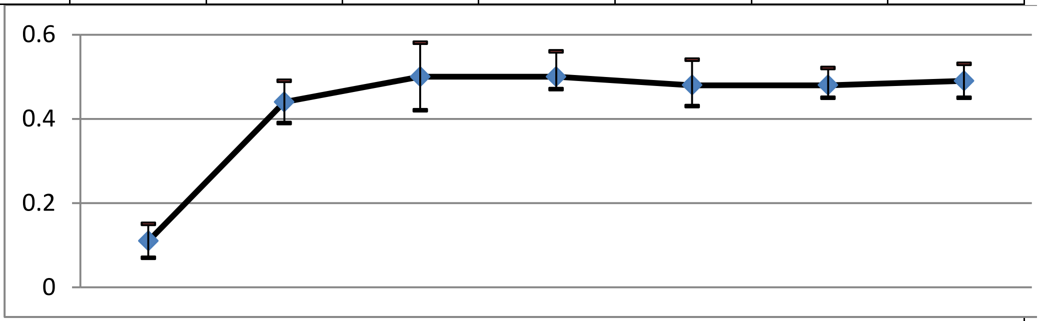
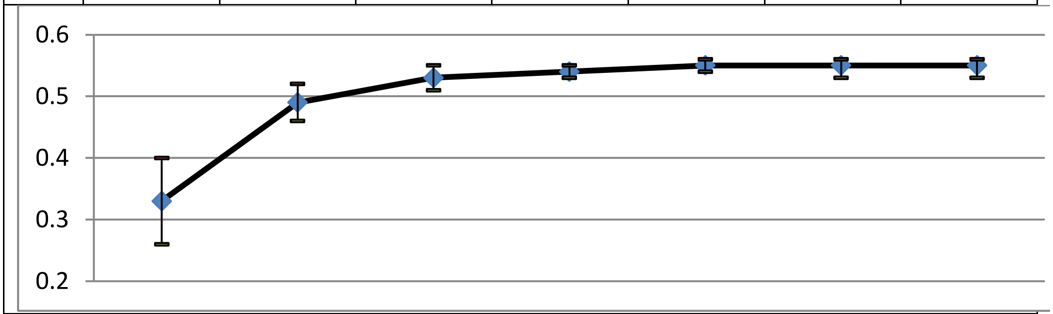
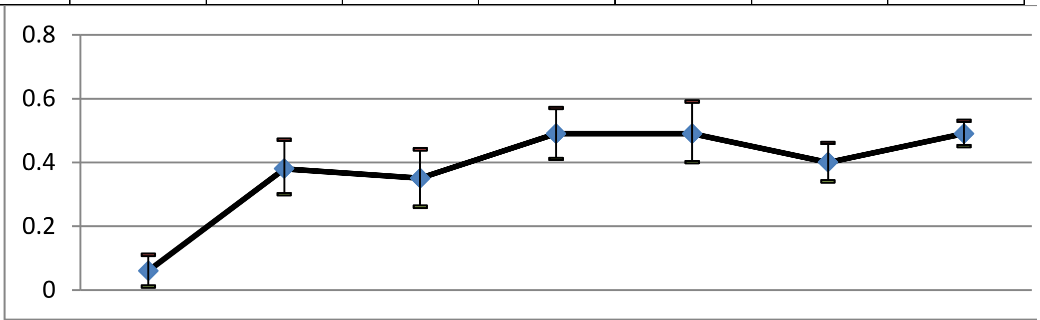
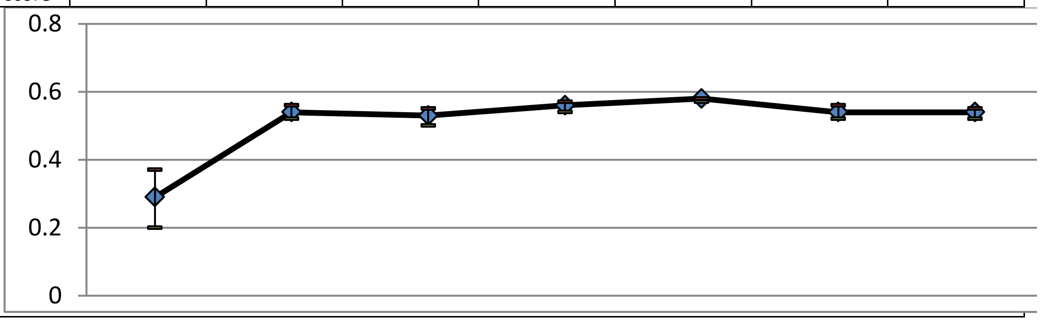
Figure 28

Average EPG indices scores for target sounds in pseudo-words at EPG assessment points with 95% confidence intervals

a). George

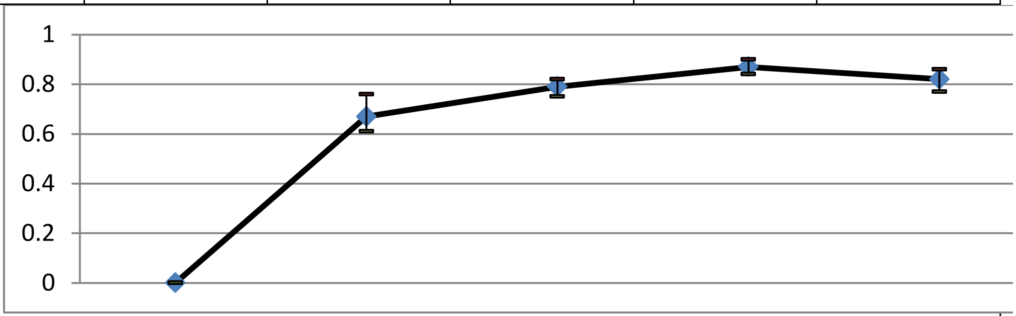
Partic.	Speech target	AC score	B3	A5	A10
George	**/z/**				
	/tʃ/ fric. elem.				
	/dʒ/ fric. elem.				
	/ʃ/				

b). Olivia

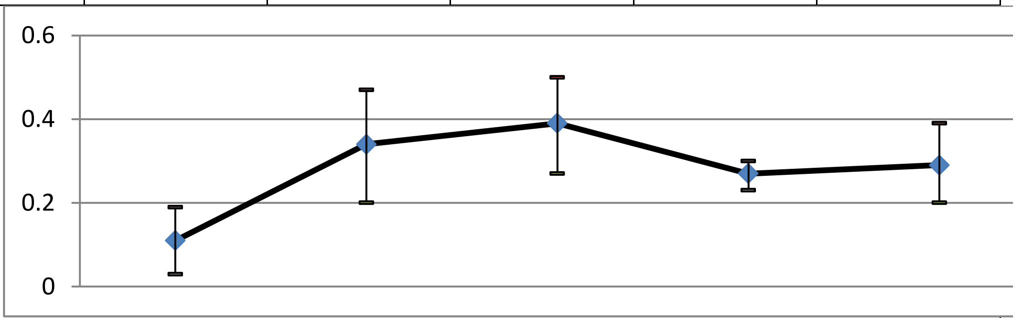
<table>
<tr><td>Partic.</td><td>Speech sound target</td><td>AT score</td><td>B3</td><td>A5</td><td>A10</td><td>A15</td><td>A20</td><td>P</td><td>M</td></tr>
<tr><td rowspan="8">Olivia</td><td rowspan="5">/s/</td><td colspan="8">
</td></tr>
<tr><td>CoG score</td><td>B3</td><td>A5</td><td>A10</td><td>A15</td><td>A20</td><td>P</td><td>M</td></tr>
<tr><td colspan="8">
</td></tr>
<tr><td>AT score</td><td>B3</td><td>A5</td><td>A10</td><td>A15</td><td>A20</td><td>P</td><td>M</td></tr>
<tr><td colspan="8"></td></tr>
<tr><td rowspan="3">/z/</td><td colspan="8">
</td></tr>
<tr><td>CoG score</td><td>B3</td><td>A5</td><td>A10</td><td>A15</td><td>A20</td><td>P</td><td>M</td></tr>
<tr><td colspan="8">
</td></tr>
</table>

c). Emma

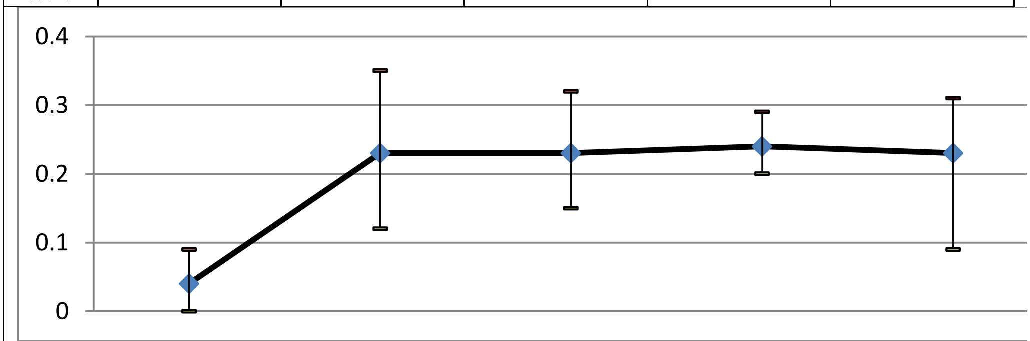
Participant	Speech sound target	AT score	B3	A5	A10	A15	A20
Emma	**/t/**						



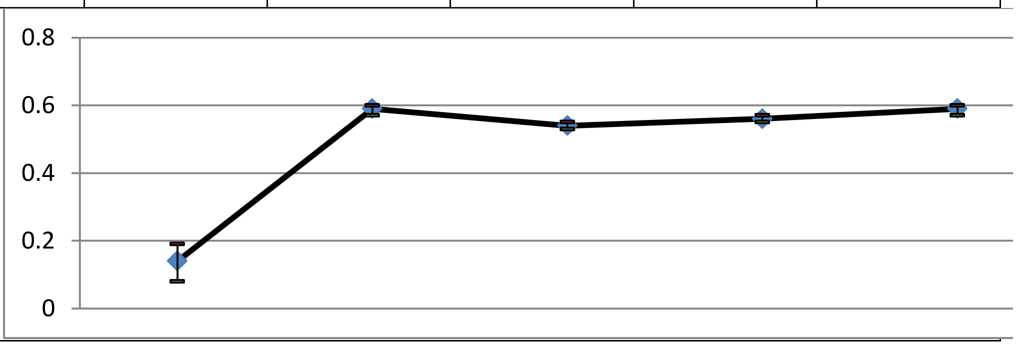
	Speech sound target	AC score	B3	A5	A10	A15	A20
	/s/						

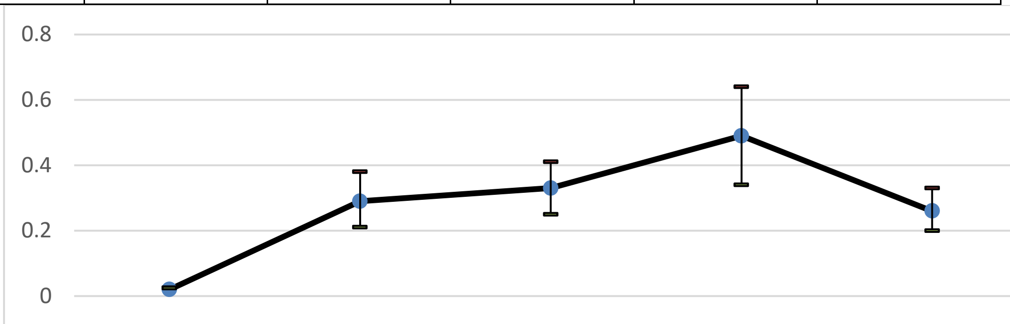
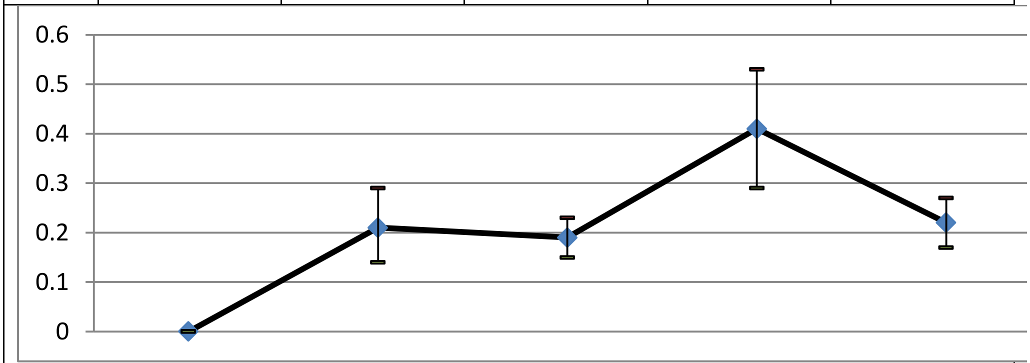
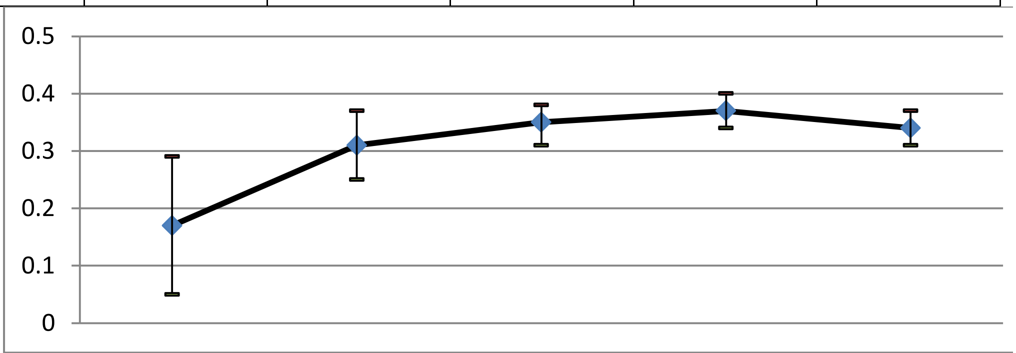
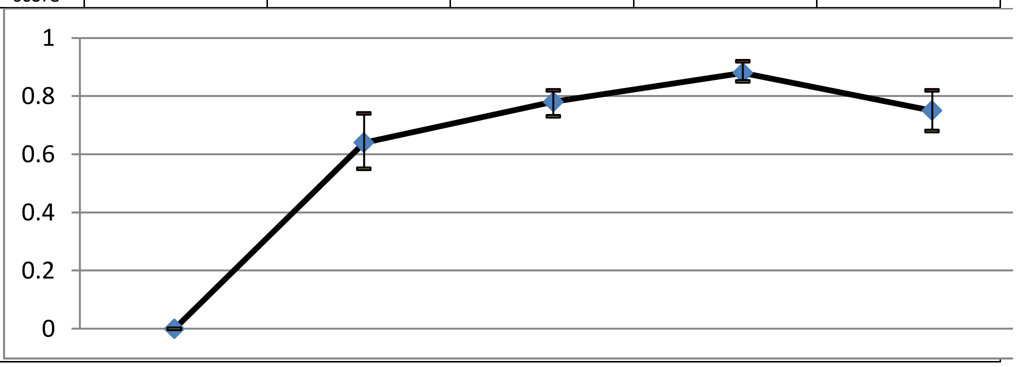


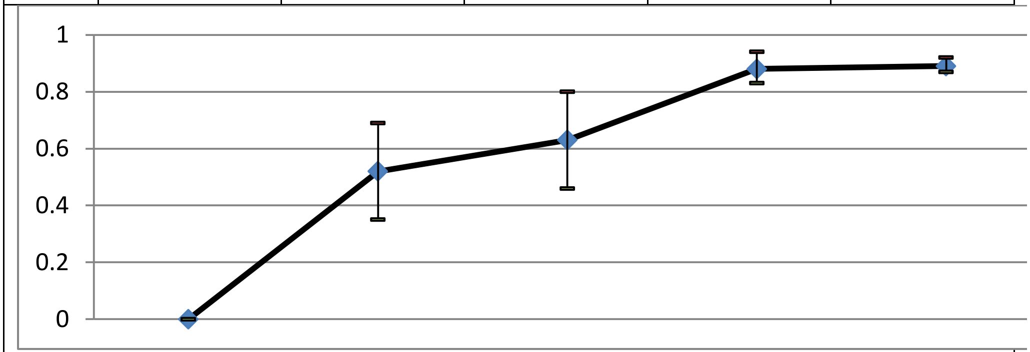
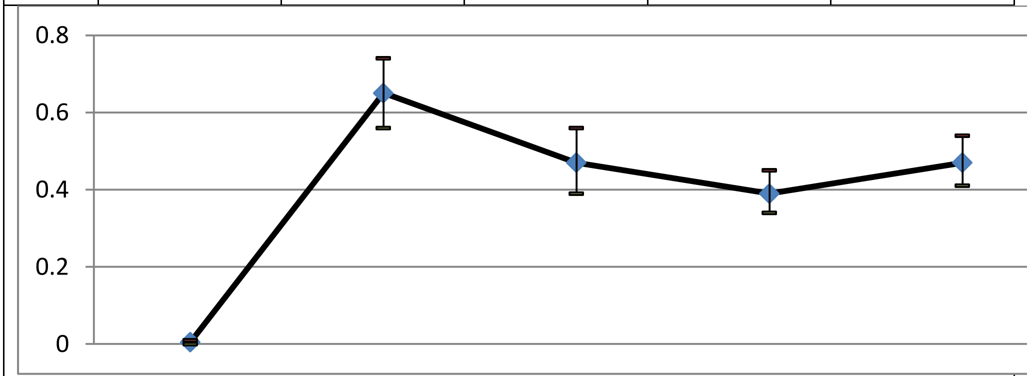
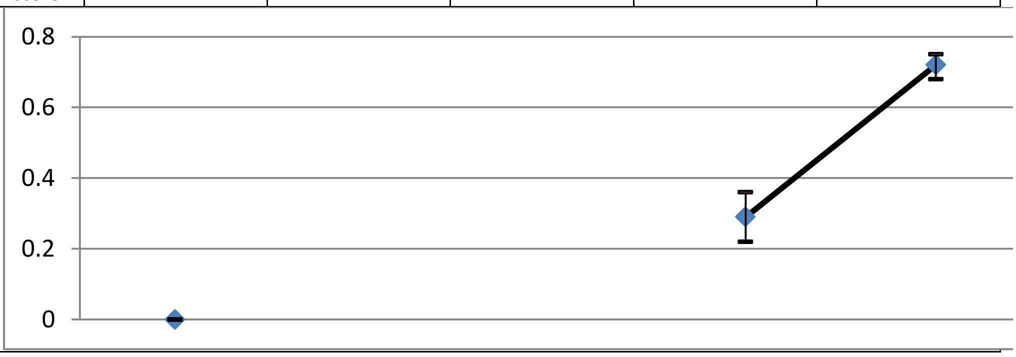
AT score	B3	A5	A10	A15	A20



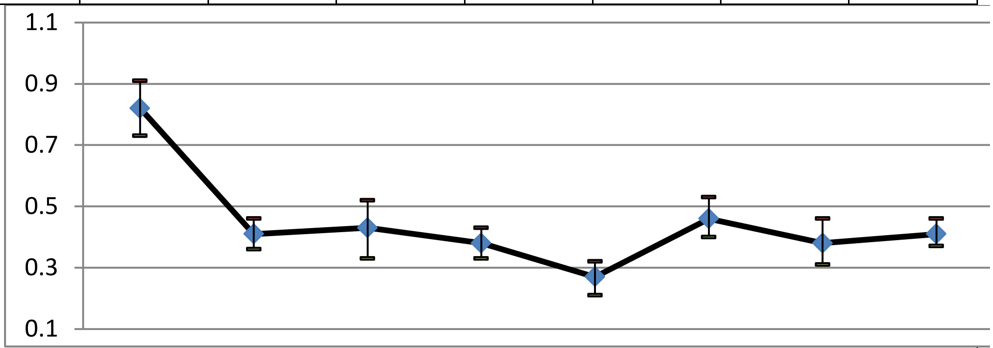
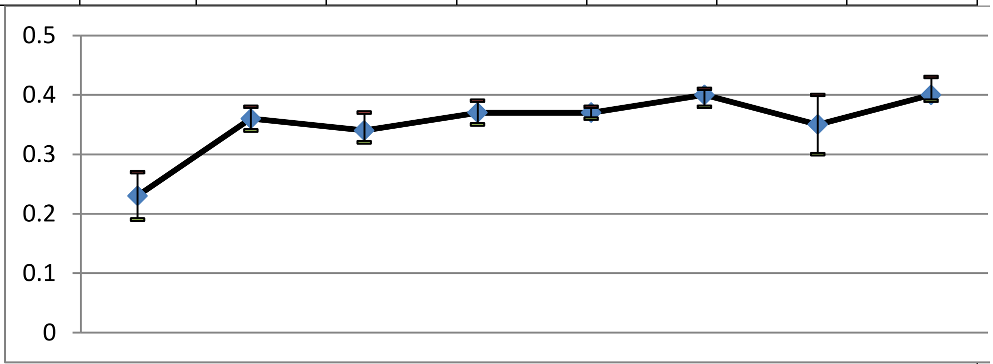
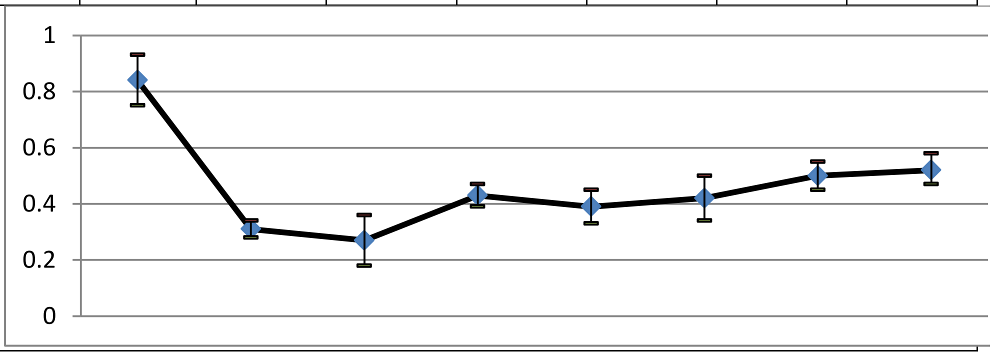
	Speech sound target	AT score	B3	A5	A10	A15	A20
	/d/						



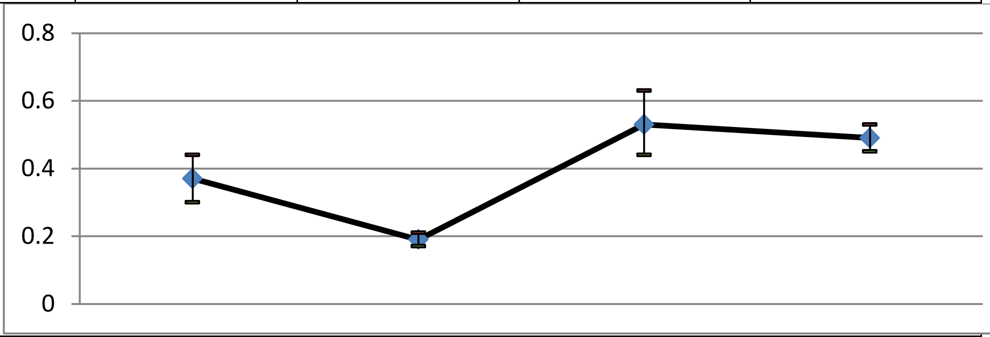
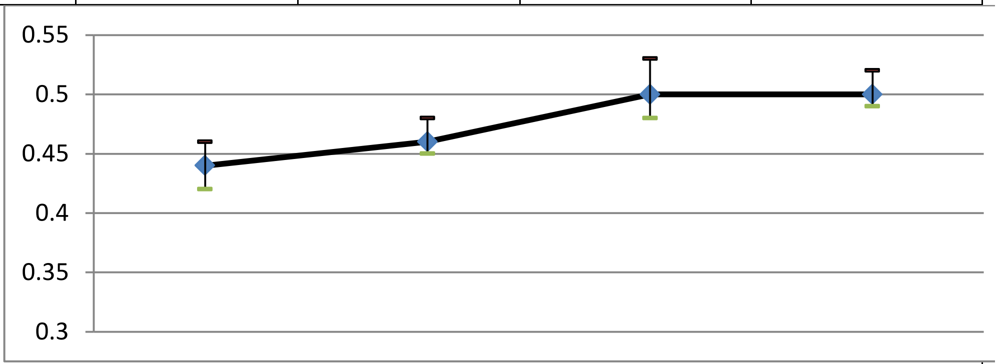
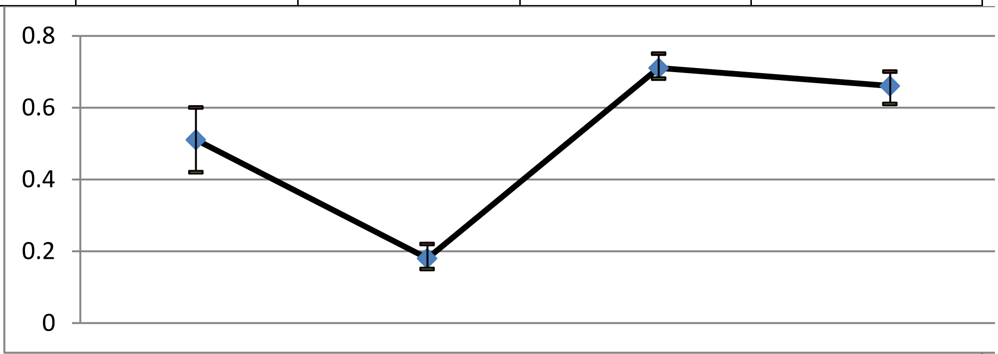
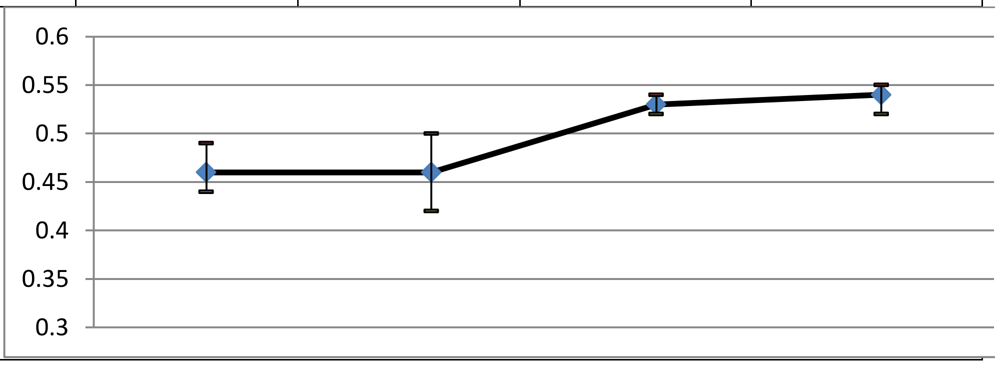
Participant	Speech sound target	AC score	B3	A5	A10	A15	A20
Emma	**/z/**						
		AT score	B3	A5	A10	A15	A20
							
	Speech sound target	WT score	B3	A5	A10	A15	A20
	/ʃ/						
		AT score	B3	A5	A10	A15	A20
	/n/						

Emma	**/l/**	AT score	B3	A5	A10	A15	A20
							
	/tʃ/ stop elem.	AT score	B3	A5	A10	A15	A20
							
	/dʒ/ stop elem	AT score	B3	A5	A10	A15	A20
							

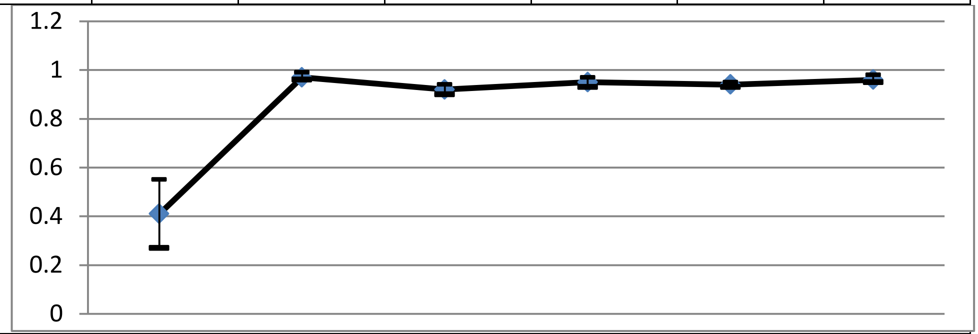
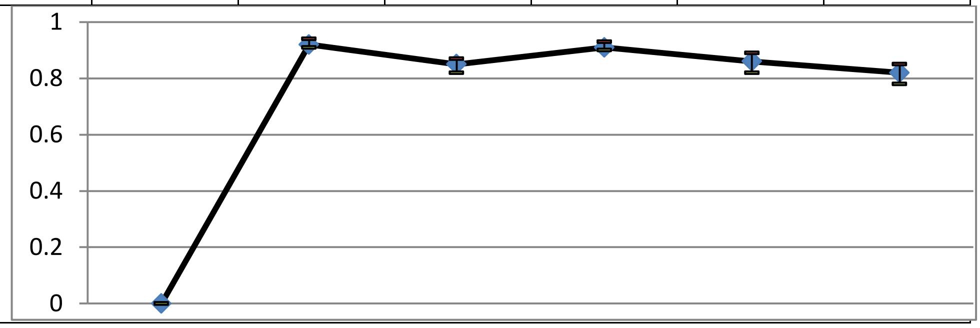
d). Victoria

Participant	Speech sound target	AC score	B3	A5	A10	A15	A20	P	M
Victoria	**/s/**								
	Speech sound target	WT score	B3	A5	A10	A15	A20	P	M
	/ʃ/								
	Speech sound target	AC score	B3	A5	A10	A15	A20	P	M
	/z/								

e). Grace

Participant	Speech sound target	AT score	B3	A5	A10	A15
Grace	**/s/**					
	Speech sound target	COG score	B3	A5	A10	A15
	/s/					
	Speech sound target	AT score	B3	A5	A10	A15
	/z/					
	Speech sound target	CoG score	B3	A5	A10	A15
	/z/					

f). Poppy

<table>
<tr><th>Participant</th><th>Speech sound target</th><th>AT score</th><th>B1</th><th>A5</th><th>A10</th><th>A15</th><th>P</th><th>M</th></tr>
<tr><td rowspan="3">Poppy</td><td rowspan="2">/n/</td><td colspan="7">
</td></tr>
<tr><td>AT score</td><td>B1</td><td>A5</td><td>A10</td><td>A15</td><td>P</td><td>M</td></tr>
<tr><td>/l/</td><td colspan="7">
</td></tr>
</table>

Note: AC = Alveolar closure; AT = Alveolar Total; CoG = Centre of gravity; WT= Whole total; B3 = Baseline 3; A5= Assessment session five; A10 = Assessment session 10; A15 = Assessment session 15; A20 = Assessment session20; P = Assessment following completion of therapy; M = Maintenance assessment 3-months post therapy; partic. = Participant; fric. = Fricative; elem = Element. Increases in AT and CoG represents improvement with tongue-palate contact moving from posterior to more anterior. Decrease in AC score represents improvement with a change from no central oral airflow to central tongue flow. Increase in WT represents improvement with a change from little tongue-palate contact, to increased tongue-palate contact.

Mean index scores pre-therapy (i.e. baseline 3) and on final EPG assessment were further examined for statistical significance using the Wilcoxon signed-rank test, and findings are presented in Table 20 a - f. As shown in this table, differences in mean index scores pre-therapy and following treatment were statistically significant ($p < .05$) for all target sounds, for all participants, apart from AT calculation of Emma's production of /z/ at baseline 3 and assessment at the end of 20 sessions. Despite this lack of significance, visual analysis clearly shows a change in tongue-palate placement for this speech sound (see Figure 24).

Table 20

Differences in EPG indices at Baseline 3 and final EPG assessment in pseudo-words (Wilcoxon signed-ranks Test)

a). George

Participant	Speech sound target	Alveolar Closure Mean			
George		B3	A10	Z score	p value
	z	0.98	0.53	-2.37	.018
	tʃ - fricative element	0.99	0.56	-2.20	.028
	dʒ - fricative element	0.99	.54	-2.02	.043
	ʃ	1.01	0.43	-2.66	.008

Note: B3 = Baseline 3; A10 = Assessment end of 10 sessions. George outgrew his EPG plate after 10 sessions, so this represents the last EPG data collection point.

b) Olivia

Participant	Speech sound target	Alveolar Total Mean			
Olivia		B3	M	Z score	p value
	s	0.11	0.5	-2.37	.018
	z	0.06	0.49	-2.21	.027
	Speech sound target	**Centre of Gravity Mean**			
		Before (B3)	Following (M)	Z value	p value
	s	0.32	0.54	-2.37	.018
	z	0.29	0.54	-2.20	.028

Note: B3 = Baseline 3; M = Maintenance assessment.

c). Emma

Participant	Speech sound target	Alveolar Total Mean			
Emma		B3	A20	Z score	p value
	t	0	0.8	-2.21	.011
	s	0.04	0.23	-2.21	.027
	d	0	0.85	-2.39	.017
	z	0	0.22	-1.84	.066
	n	0	0.75	-2.37	.018
	l	0	0.9	-2.38	.017
	tʃ - stop element	0	0.47	-2.67	.007
	dʒ - stop element	0	0.72	-2.06	.039
	Speech sound target	**Alveolar Closure Mean**			
		Before (B3)	Following (A20)	Z score	p value
	s	0.11	0.29	-1.99	.046
	z	0.25	0.26	-1.83	.068

Note: B3 = Baseline 3; A20 = Assessment end of 20 sessions. Emma outgrew her EPG plate after 20 sessions, so this represents the last EPG data collection point.

c). Victoria

Participant	Speech sound target	Alveolar Closure Mean			
Victoria		B3	M	Z score	p value
	s	0.82	0.42	-2.20	.028
	z	0.84	0.52	-2.21	.027
	Speech sound target	**Whole Total Mean**			
		Before (B3)	Following (M)	Z score	p value
	ʃ	0.23	0.41	-2.20	.028

Note: B3 = Baseline 3; M = Maintenance assessment.

d). Grace

Participant	Speech sound target	Alveolar Total Mean			
Grace		Before B3	A15	Z score	p value
	s	0.37	0.49	-2.30	.021
	z	0.51	0.66	-1.97	.049
	Speech sound target	**Centre of Gravity Mean**			
		Before (B3)	Following (A15)	Z score	p value
	s	0.44	0.51	-3.38	.001
	z	0.46	0.53	-3.72	.000

Note: B3 = Baseline 3; A15 = Assessment end of 15 sessions.

f). Poppy

Participant	Speech sound target	Alveolar Total Mean			
Poppy		Before (B3)	Following (M)	Z score	p value
	n	0.41	0.96	-3.30	.001
	l	0.00	0.82	-3.85	.000

Note: B3 = Baseline 3; M = Maintenance assessment.

7.3 Summary of EPG results

EPG results contribute towards answering research question number one: **Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy/post-therapy and, if so, does this improved accuracy generalise to untreated/pseudo- words?** In addition, qualitative analysis of EPG frames contributes towards research question number three: **Do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in terms of demonstration of gradient phonetic change with target phonemes.**

Quantitative and quantitative EPG analysis showed tongue-palate contact moved in the direction of improvement, and better matched reference frames for all target sounds in pseudo, for all participants. Further, the difference between EPG indices pre-therapy and with therapy, was statistically significant (Wilcoxon signed-rank test, $p < .05$), apart from Emma's production of /z/ in pseudo-words. However, visual analysis of EPG frames pre- and post-therapy clearly shows change in tongue-palate placement for the speech sound (see Figure 24). EPG frames and indices showed gradient articulatory change on assessment over time. This gradient change occurred during both the acquisition and generalisation phases of therapy.

On assessment pre-therapy, EPG analysis differed from the independent SLTs' acoustic perceptual transcription for 3/6 participants (Emma, Victoria and Poppy) in terms of place and manner. In addition, independent SLTs' phonetic transcription was variable for these three participants for transcription of /s/, /z/, /ʃ/, /tʃ/, /dʒ/, /n/ and /l/. The objective data provided by EPG thus provides important additional information to supplement subjective acoustic phonetic transcriptions.

8 DISCUSSION

The aim of this study was to describe and evaluate patients' response to a usage-based EPG therapeutic technique. A total of six, consecutively treated, patients with SSDs secondary to cleft palate +/- lip participated in this study and response to therapy was examined through a within-participant ABA multiple baseline design. Usage-based phonology was the theoretical underpining for the intervention (Bybee, 2001, 2010; Menn et al., 2013). As discussed in the literature review, usage-based phonology is an alternative theory to generative, rule-based linguistic theory (Chomsky, 1957, 1968; Stampe, 1979), the later theory having underpinned most SSD intervention since the 1970s (Bowen, 2015; Ingram, 1976; McLeod & Baker, 2017). The motivation for application of this newer phonological theory was the development of a treatment technique which might better facilitate functional generalisation of target speech sounds. Previous EPG intervention research indicates functional generalisation can be challenging for this client group (Cleland & Preston, 2021; Gibbon & Paterson, 2006; Lee et al., 2009). In addition, this research aspired to develop a technique that would be effective for both children and adults with persistent cleft palate speech. The main features of the usage-based therapeutic technique included: high number of production trials; a focus on word production instead of sounds; teaching low frequency words before high frequency words; facilitation of phonetic gradience; and teaching connected speech patterns (Bybee, 2001, 2010; Menn et al., 2013).

Three of the participants recruited to this study were school-aged, the remaining three were adults. All study participants had shown persistent, unchanging cleft speech characteristics for two or more years prior to the study, despite having received standard intervention for their SSDs. Participants' cleft speech characteristics included: backing of anterior lingual sounds to more posterior placement; post-alveolar production of alveolar sibilants; lateral production of lingual fricatives; and non-oral production of lingual sibilants. As such, participants' cleft speech features were representative of the speech characteristics typically seen in individuals with persistent cleft palate speech (CRANE database, see 2.2.4.3). Participants were treated until target speech sounds were produced typically, on auditory perception, or until no further progress had occurred over eight sessions.

This study had three research questions relating to participants' response to a usage-based EPG therapeutic technique. The first research question related to acquisition and lexical generalisation and asked: **Does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy and post-therapy, and if so, does this improved accuracy generalise to untreated/pseudo- words?** All six participants showed acquisition of typical/more typical productions of target phonemes in treated words with therapy (see Figure 9 – 14). Likewise, all six participants showed generalisation of typical/more typical productions of target phonemes to untreated words/pseudo-words following therapy (see Figure 9 – 14). The second research question related to functional generalisation and impact of quality of life and asked: **Does improved accuracy of target**

phonemes generalise to continuous, connected speech and, if so, does this result in improved ratings of speech understandability and speech acceptability and improved ratings of impact on quality of life, post-therapy? All participants showed generalisation of target speech sounds to continuous connected speech, though to varying degrees. Individual's use of target sounds in continuous connected speech ranged from 78.94 – 100% (mean = 90.66%). Parent and SLTs' ratings of speech understandability and speech acceptability improved for 5/6 participants. Self-reported improvements in quality of life occurred for 3/6 participants. The third and final research question related to the theoretical model and asked: **Do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in general, and more specifically in terms of: gradient speech change is shown; and a word frequency effect is shown.** Participants' positive response to the usage-based EPG therapeutic technique (in terms of improved speech change) provides some general support for the therapeutic model. Gradient articulatory speech change with target phonemes was shown over the course of therapy, as shown with EPG analysis. A word frequency effect was shown with high frequency words proving more resistant to articulatory change in everyday speech (see 6.2.6, 6.3.3 and 7.3 for a more comprehensive summary of study findings).

The first three sections of this chapter discuss the study's three main areas of inquiry, as reflected in the study's research questions: 1). acquisition and lexical generalisation; 2). functional generalisation and impact on quality of life; and 3). support for the theoretical model. Each section concludes with an answer to the research question relating to the section, based on the proceeding discussion. The chapter then goes on to discuss the limitations of EPG as a biofeedback therapy, followed by a discussion of patient selection. The chapter then discusses the strengths and limitations of the study and directions for future research. The chapter concludes with clinical and theoretical implications.

8.1 Acquisition and lexical generalisation

All participants enrolled in this study acquired typical/more typical lingual speech sound productions during therapy. Prior to therapy, all participants in this study had received many sessions of standard speech and language therapy and had been unable to improve their speech sound production. Further, for 5/6 participants in this study, this acquisition of target lingual sounds was rapid, with more typical speech production occurring in treated words within one to two sessions of therapy. In addition, lexical generalisation was rapid and occurred within five sessions. For the remaining participant, accurate production occurred after 11 sessions. This finding of improved speech sound production with EPG visual feedback is not unique. Rapid acquisition of lingual speech sounds for some patients has been reported in EPG intervention studies involving individuals with cleft palate speech (Gibbon et al., 2001; Stokes et al., 1995; Whitehill et al., 1996), and with EPG intervention studies involving other client groups (Gibbon et al., 2003; Martin et al., 2007). As discussed in Chapter Two, since lingual articulation is hidden away in the mouth, cannot be seen and is fleeting, EPG provides information that is not normally available to the patient or SLT. This study supports other EPG intervention research (Fujiwara, 2007; Lohmander et al., 2010; Martin et al., 2007; Whitehill et al., 1996) suggesting that the visual feedback provided by EPG assists the patient's acquisition of more typical lingual speech sounds. In addition, utilising EPG information, the SLT is better placed to provide the patient with detailed and exact phonetic cues for lingual production as therapy progresses (Gibbon & Wood, 2010; Lee, 2021).

While rapid speech sound acquisition for some patients is a finding of this, and other EPG intervention studies (Martin et al., 2007; Stokes et al., 1995; Whitehill et al., 1996), previous research also shows that a proportion of individuals put forward for this type of therapy exhibit no response (Gibbon et al., 2001; Gibbon & Paterson, 2006). As discussed in Chapter Two, Cleland and Preston (2021) suggest biofeedback intervention studies carried out in the 1980s and 1990s, including EPG studies, may have been subject to publication bias, in that non-responders (i.e. participants not responding to biofeedback intervention in any way) were not reported. These authors point out that more recent biofeedback interventions show a greater number of non-responders, compared with earlier studies. In order to gain an accurate record of non-responders, studies need to: report consecutively treated patients; clearly outline inclusion and exclusion criteria; and give information on the progress of all participants through the study. Such information is shown in many contemporary SSD intervention studies and is typically given by way of a flow diagram (e.g. Cleland et al., 2019; Sweeney et al., 2020). None of the previous Cleft EPG intervention studies involving more than one participant have included such information (see Table 1 for a list of previous studies), making it more difficult to judge if these studies are biased in terms of patients reported. In the seven cleft EPG intervention studies reviewed in Chapter Three, three out of a total of 28 participants were non-responders, and these three came from an intervention study carried out in 2001 (Gibbon et al., 2001). These non-responders received a total of four sessions of EPG therapy. In the present study, one participant required 11 sessions before acquisition of a target sound occurred. It is possible that change may have occurred for the three non-responders in Gibbon et al.'s cleft EPG intervention study, had these participants received a greater number of EPG therapy sessions. The present study involved a consecutively treated series of patients, and flow of patients through the study is shown in Figure 6. Although all participants of this series responded to intervention, it is possible that non-responders may have occurred, had greater numbers of participants been recruited. This study also excluded individuals with severe learning disabilities and this resulted in one person being excluded from this study. The Gibbon et al. (2001) cleft EPG study involving three non-responders does not give details on whether or not individuals with learning disabilities were included or excluded; it is possible such individuals were included.

In this study, speech sound acquisition was measured by percentage of target consonants correct with production of treated words (SLTs' phonetic transcription). Lexical generalisation was measured through a number of measures: percentage of targets correct in untreated words from (SLTs' phonetic transcription); and EPG analysis (researcher's analysis). The following two sub-sections discuss these methods for measuring acquisition and lexical generalisation.

8.1.1 Percentage of targets correct as an outcome measure for speech acquisition and lexical generalisation

SSD intervention research involving cleft palate speech has utilised a range of measures to report speech outcomes (Sell & Pereira, 2015). Percentage of consonants correct, and the related measure of percentage of targets correct, are amongst the most frequently used tools to measure speech outcomes with this client group (Sell & Sweeney, 2020). Percentage of targets correct, as used in this study, is derived from acoustic phonetic transcription. In this study, when listening to speech, SLTs gave a tick for words perceived as accurate, while atypical productions were transcribed narrowly using phonetic transcription. Percentage of targets correct was the total number of target sounds perceived as correct, divided by the total number of instances of that target sound. The disadvantage of acoustic phonetic transcription is that such transcription is subjective and therefore susceptible to variability and bias, even for highly trained SLTs and phoneticians (Lohmander & Olsson, 2004; Sell, 2005; Sell & Pereira, 2016). For example, Buckingham and Yule (1987) describe the phenomenon of phonetic false evaluation, where categorical perception leads the listener to transcribe speech productions within their own phonological system, despite the fact that sub-phonetic contrasts or subtle phonetic errors may be occurring. Likewise, SLTs not blinded to intervention may be unconsciously biased to knowledge of intervention and may, for example, report improved outcomes following therapy when none or little change has occurred. As discussed in Chapter Three, given this variability and bias, researchers and clinicians reporting impressionistic perceptual speech ratings need to adopt procedures to reduce these threats, such as use of standard speech samples, ensuring listeners are blind to test time point, more than one SLT being involved with transcription, and using instrumental methods, such as EPG (Cleland & Preston, 2021; Dalston et al., 1988; Lee, 2021; Lohmander & Olsson, 2004; Sell, 2005; Sell & Pereira, 2016) (see 3.2.2 for further discussion). Such procedures to counter variability and bias were lacking in earlier cleft EPG intervention studies, but were used in this research. All procedures used in this research were viewed as important, and future research should continue to adopt such procedures, as described in 3.2.2.

When using subjective measures, such as phonetic transcription, researchers and clinicians are also obliged to show reproducibility of ratings, through agreement or reliability scores, so that readers can judge results (Chapman et al., 2016). Both inter-rater (agreement between different raters, independently rating the same material) and intra-rater (agreement by a single rater, rating the same material on two separate occasions) agreement/reliability should be reported (Dalston et al., 1988; Lohmander & Olsson, 2004; Sell, 2005), as was done in this study. High inter-rater agreement/reliability indicates different listeners are applying the same criterion when rating the material. High intra-rater agreement/reliability indicates the rating is reproducible under similar conditions (Chapman et al., 2016). Levels of agreement/reliability are important for confidence in results. High agreement may produce high confidence in results, while low agreement may indicate caution is needed when interpreting results. Previous literature considering agreement for phonetic transcription of SSD, as a whole, suggests agreement levels of 90 - 95% for broad transcription and 80% for narrow transcription are required for satisfactory agreement (Shriberg et al., 1997; Shriberg & Lof, 1991). Although these studies are now dated, these benchmarks remain the gold-standard (Stemberger & Bernhardt, 2020). However, it is recognised that such levels of transcription agreement may not be achievable with children with more complex and severe speech difficulties (Cleland et al., 2019; Gooch et al., 2001). In particular, transcription of anterior cleft characteristics, including lateralisation/lateral production, palatalisation/palatal production, and double articulation have shown lower inter- and intra-rater reliability (Chapman et al., 2016; Sell et al., 2009) (see 2.2.4 for description of cleft speech characteristics). For example, Chapman et al. (2016) and Sell (2009) reported poor to moderate reliability with SLT's judgement of anterior oral cleft speech characteristics using kappa and interclass correlation coefficient (ICC) statistics.

As discussed in Chapter Two, deficiencies with reliability reporting are a significant shortcoming with previous EPG intervention studies with this client group (Fujiwara, 2007; Gibbon et al., 2001; Michi et al., 1986; Michi et al., 1993; Whitehill et al., 1996). Consequently, it is more difficult to be confident of, and/or judge, the outcomes reported in these studies. In this study, both inter- and intra-agreement/reliability was reported for perceptual speech assessments. Percentage of agreement for the two independent SLT's phonetic transcription was 91.09%. Intra-rater agreement for the two independent SLTs was 92.08% and 96.96% respectively (re-rating of 20% of speech data). Percentage of agreement with the researcher and each independent SLT was 89.75% and 91.39% (20% of speech data), respectively. The researcher's intra-rater agreement was 97% (re-rating of 20% of speech data). Thus, acceptable levels of agreement were achieved in this study and these levels may provide a degree of confidence with interpretation of findings. In this study SLTs' disagreements in phonetic transcription largely related to /l/ and /n/, and to a lesser extent, /t/ and /d/. Disagreement often occurred in the presence of double articulation. Double articulation involving anterior consonants is classified as an anterior cleft speech characteristic. Thus, rater disagreement shown in this study aligns with Chapman et al. (2016) and Sell et al. (2009) findings of lower agreement with SLT's judgement of anterior oral cleft characteristics, as discussed in the previous paragraph. This point is discussed further in 8.1.2 below.

In this study, for each of the six participants, percentage of targets correct at the different assessment time-points was presented on line graphs. Visual inspection is the most common method for analysing speech outcomes using within-participant designs, in general, and with intervention for cleft palate speech (Lohmander et al., 2010; Martin et al., 2007; Pring, 2005). In this study, visual inspection of the line graphs clearly shows increases in percentage of targets correct in treated and untreated words with therapy (see 6.2.1 and 6.2.3). However, the limitation of visual inspection in single-subject research is that findings cannot be generalised to other patients (Howard et al., 2015; Pring, 2005; Vallino-Napoli, 2011). More recently, statistical measures have also been used with single-subject research designs (Gierut et al., 2015). Effect size is one such statistical measure. Effect size indices examine magnitude of change following intervention. Effect size allows for lack of independence between samples (e.g. speech samples from the same individual at different time-points), as occurs with within-participant studies. As such, effect size indices permit comparison across a range of within-participant studies and allow for a degree of meta-analysis. Meta-analysis can establish trends for a body of related intervention research, thereby contributing to evidence-based practice. Standard mean difference, with correction for continuity, is an effect size index used with SSD interventions involving within-participant designs (Cleland et al., 2019; Gierut et al., 2015). Gierut et al. (2015) present benchmarking for this statistic. Numbers corresponding to large, medium and small treatment effects are provided for children with functional SSDs (see Table 8). This study used an adaption of this effect size index (see 5.5), and large treatment effects were shown (mean effect size = 9.47). Although derived from children with functional SSD, use of these benchmarks appeared a reasonable description of the treatment gains achieved by the participants in this study. Thus, use of these benchmarks with future cleft palate speech intervention studies is suggested.

A limitation of percentage of targets correct as an outcome measure is that gradient changes to speech sound production may not be shown (Cleland et al., 2019). However, small changes with speech sound production may be clinically significant. For example, Cleland et al. (2019) report on change in the articulation of /s/ for two participants with developmental SSD. Pre-therapy, /s/ was transcribed as a pharyngeal sound. Post therapy, this sound was produced as ([ʃ] or [ɬ]). These authors suggest this change represents a significant improvement in speech acceptability for both participants. Consequently Cleland et al. (2019) write "this speaks to a need for future studies to move away from right/wrong judgements and to adopt a more gradient approach to correctness" (p. 240).

In this study, in addition to examination of narrow phonetic transcription of speech, gradient speech change was shown through visual analysis of EPG palatograms, and through changes to EPG indices. Visual analysis of EPG palatograms and EPG indices is straight-forward and has the advantage over perceptual speech transcription of being objective. In this study, examination of EPG frames and indices over time showed gradient change in tongue-palate placement for speech sounds and, along with sound and spectrographic information, showed changes with manner. However, a limitation of this type of assessment is that change with sounds not involving tongue-palate contact, such as /p/, /b/, /m/, /f/ and /v/, will not be shown, though it should be noted that if a speaker did show tongue palate contact with production of a non-lingual sound (e.g. /p/ produced as [p͡k]), this would represent an error and elimination of such lingual contact would be a goal of EPG treatment (Lee, 2021). Also, change with voicing and resonance, and possibly other aspects of manner, will not be identified with EPG images alone (Gibbon & Wood, 2010). In addition, significant time is needed to extract EPG frames and calculate EPG indices. Potentially, EPG systems could be developed so this information is automatically compiled by software programmes, therefore making this information more readily accessible.

In this study, George's use of /tʃ/ and /dʒ/ went from 0% correct on baseline assessments to around 50% correct on post intervention and maintenance assessment. George's 50% error production post-therapy consisted of /tʃ/ → [tj] and /dʒ/ → [dj], which, arguably, represents improvement from his lateral production ([ɬ] and [ɮ] respectively of these target sounds pre-therapy. This gradient change was captured on EPG and perceptual speech assessment. Victoria's atypical productions of /z/ and /dʒ/ post-intervention included /z/ →[s] and /dʒ/ → [tʃ]. This could also be viewed as an improvement from /z/ → [n] and /dʒ/ → [nj] on pre-intervention assessment. This gradient change was not identified by EPG assessment, as EPG analysis does not pick up changes with voicing, but was shown with perceptual assessment. Although the SLTs involved with this study felt George and Victoria's gradient speech change was clinically important, naïve listeners may disagree. In addition to perceptual and EPG analysis, future studies could have naïve listeners make judgements on speech correctness, through having multiple listeners rate which productions are closer to the target (Lohmander et al., 2010; Roxburgh, 2018), or through listeners' use of use of visual analogue scales (Baylis et al., 2011; Pereira, 2012).

8.1.2 EPG data as an outcome measure for speech acquisition and lexical generalisation

In this study, EPG data was examined quantitatively and qualitatively to measure speech sound acquisition and generalisation to pseudo-words. Quantitative analysis comprised numerical indices relating to EPG frames. Qualitative analysis consisted of visual and descriptive analysis of EPG frames. For all target speech sounds, tongue-palate contact moved in the direction of improvement, and better matched reference frames. In this study the centre of gravity (CoG), alveolar total (AT) and whole total (WT) indexes were effective in identifying change with anterior-posterior tongue-palate placement, as evidenced with Olivia, Emma, Grace and Poppy (see 5.2.6 for description of these indexes). The Alveolar Closure (AC) index, as used with George and Emma, measured change with production from complete closure (i.e. lateral production with lingual fricatives) to central oral airflow. In addition AC was used with Victoria's production of /s/ and /z/ where again central airflow was absent prior to intervention. Victoria's change with production of /tʃ/ and /dʒ/ was not analysed quantitatively using EPG indices, as no index clearly showed change with production, so this represented a shortcoming with existing EPG indices. However qualitative data showed change with production of these two speech sounds (see Figure 25).

EPG palatograms were examined qualitatively using the classification system developed by Gibbon et al. (2001), including: increased contact, retracted to palatal or velar placement, fronted placement, complete closure, open patterns, double articulation, increased variability, abnormal timing (see section 3.1.2 for further details). This classification system has also been used to describe ultrasound images for children with cleft and functional SSDs (Cleland et al., 2020; Sugden & Cleland, 2021). In this study, participants' EPG contact patterns for sounds produced atypically fitted into this classification system. Since Gibbon's system was developed for cleft palate speech data, this finding is unsurprising. George and Emma showed patterns of complete closure. Olivia and Grace showed retracted to palatal placement patterns. Emma and Poppy showed patterns of retracted to velar placement, though for some of Emma's speech sounds, retraction occurred further back, to probably uvular placement. Gibbon labels such uvular productions as "open", though arguably uvular productions are very different to other speech sounds labelled as "open", such as glottal substitutions of lingual sounds. Victoria's palatograms were less usual, with lingual placement accompanying pharyngeal production. However, her palatograms fit into Gibbon et al.'s classification system under "double articulation". The atypical EPG patterns shown by participants in this study are similar to those reported in existing cleft palate EPG intervention research (Fujiwara, 2007; Lohmander et al., 2010; Michi et al., 1993; Stokes et al., 1995). Therefore, the participants in this study appear representative of the population of individuals with persistent cleft palate speech in terms of atypical tongue-palate contact and manner for production of lingual speech sounds.

As well as providing another measure of speech outcome (i.e. in addition to percentage of targets correct), Lee (2021) and Cleland and Preston (2021) suggest a key strength of instrumental assessment is its capacity to supplement perceptual phonetic transcription. Findings from this study support this contention. In this study differences with EPG assessment and phonetic transcription existed. Differences related to a). agreement with accurate and inaccurate production, and b). agreement with inaccurate production. Each of these is discussed below.

Agreement with accurate and inaccurate production: A total of 25 sounds were targeted in this intervention study. Of these targets, 19/25 scored above 0%TC on baseline assessments with independent SLT transcription, i.e. at times these target sounds were heard as correct by the independent SLTs in single word production. However, contrary to impressionistic phonetic transcription, EPG analysis prior to intervention showed that no target sound was ever produced with typical placement and/or manner in word initial position in pseudo-words (see section 6.1), compared with typical speakers. Similar discrepancies with blind impressionistic phonetic transcription and instrumental assessment were shown by Cleland et al. (2019). As discussed in 3.1.2, it is important to highlight that caution is needed when comparing tongue-palate contact in individuals with the cleft condition with that shown by speakers without the cleft condition. This is because the structure of the palate will be affected in individuals with cleft lip and palate. At present, little normative EPG data exists for individuals born with the cleft condition, though some preliminary work has been done by Yamamoto (2020). Yamamoto found tongue-palate contact patterns for Japanese speakers with unilateral cleft lip and palate matched those of typical speakers, and this matching occurred for a range of speech sounds. However, all 15 participants in this study had normal occlusion following dental treatment. While caution is needed in making comparisons between typical speakers and speakers with cleft lip and palate, in this study, differences in tongue-palate contact for participants were very marked (see sections 6.2.1 and 6.2.2). For example, Emma's production of /n/ shows consistent velar tongue-palate contact on assessment prior to therapy, despite the independent listeners perceiving this sound with 53.17 % accuracy in single word production at the same assessment time-point. Likewise, Poppy's production of /n/ shows consistent velar tongue-palate contact on assessment prior to therapy, despite the independent listeners perceiving this with 42% accuracy in single word production at the same time point. Thus, one can be more confident that EPG palatograms were identifying meaningful differences in tongue-palate placement, compared to typical speakers and individuals with cleft with typical speech.

In this study, the most striking disagreement between independent SLT transcription and EPG Analysis occurred for Poppy. The independent SLTs scored Poppy with high levels of accuracy for /l/ and /n/ on baseline assessments (range: 40% - 84%). However EPG analysis pre-therapy showed all productions of /l/ and /n/ in pseudo words were retracted to velar placement. Emma's production of /l/ and /n/ also received higher scores of accuracy (range 6.25 – 53.17%). This suggests these two phonemes were more difficult to transcribe accurately with phonetic transcription, in terms of placement, given the listening conditions of this study. This contention is further supported by the finding of lower inter- and intra-agreement for the independent SLTs' phonetic transcription of these two phonemes, as discussed above.

For Poppy, double articulation of /n/ was evident (/n/ → [n͡ŋ]) at times (simultaneous double articulation), and this may have made acoustic phonetic transcription more difficult in terms of identifying phonetic placement. With regard to /l/, phonetic transcription may be more difficult due to reported variability in typical production of this phoneme. Imaging research shows production of /l/ can vary dependent on word position (McLeod & Singh, 2009; Narayanan et al., 1997) and on the speaker. Narayanan and colleagues (1997) used MRI and EPG to image /l/ production in four typical adult speakers. Prevocalic /l/ production was characterised by complete tongue-palate contact in the alveolar region. In contrast, postvocalic contact showed variable tongue-palate contact and variable positioning of the body of the tongue amongst the four speakers, including no alveolar contact and approximation of the dorsum of the tongue with the posterior part of the palate. Therefore, it may be that Poppy and Emma's "backed" production of /l/ in word final production prior to treatment was within the boundaries of typical production, thus explaining why the independent SLTs often assessed these as accurate.

Agreement with inaccurate production: Anomalies between perceptual phonetic transcription and EPG assessment with inaccurate production were seen with Emma and

Victoria’s speech. One independent SLTs transcribed Emma’s /s/, /z/, /ʃ/, /tʃ/ and /dʒ/ as palatal ([ç], [ɟ̟]), while the second independent SLT transcribed these speech sounds as lateral fricatives ([ɬ], [ɮ]) on baseline assessment. Similar listener differences in phonetic transcription of these lingual fricatives are suggested by Sell et al. (2009) and Chapman et al. (2016). Emma’s EPG assessment at baseline 3 provided an answer to this listener disagreement. EPG assessment at baseline 3 showed a consistent retracted pattern to velar with lateral airflow. Lateral airflow was determined given EPG patterns of complete closure and accompanying fricative noise heard and seen on the spectrogram. This pattern of backing and lateral airflow was transcribed perceptually by the researcher on initial assessment, despite no access to EPG images. It may be that the researcher’s “unblind” knowledge of Emma’s speech facilitated phonetic transcription. In addition, the researcher’s knowledge of lateral fricatives through her experience with EPG may have facilitated transcription. This knowledge is in comparison to the independent SLTs who do not have the same experience with EPG.

Likewise, independent SLT phonetic transcription of Victoria’s speech was variable. One independent SLT transcribed /s/ as [ħ], while the other transcribed this speech sound as [ŋ̥̃̇] or [ħ]. EPG analysis showed some horse-shoe shaped contact for /s/, consistent with a weak /s/, double articulated with [ħ]. The sounds /ʃ/, /tʃ/ were transcribed as [ħ], but EPG assessment showed some silent tongue-palate contact for these phonemes. Such anomalies between phonetic transcription and EPG analysis are also reported elsewhere. Dagenais et al. (1994) describes the treatment of two children with a lateral dental fricative. While a dental lateral fricative was heard perceptually, EPG analysis (and accompanying sound) showed tongue-palate placement and manner was different for the two children.

The above differences in phonetic transcription and instrument assessment, as found in this study, and other studies (Cleland et al., 2019; Dagenais et al., 1994) reinforce the benefits of EPG in supplementing phonetic transcription. Such objective EPG assessment can serve to explain the perceptual speech data. This objective data can: a). assist with treatment planning; b). provide objective speech outcomes, and c). increase confidence in study findings. A further supplementary benefit of EPG is its ability to identify covert contrasts (Gibbon, 1999). Covert contrasts is the term used to describe instances where instrumental differences are seen with atypical productions of phonemes by a single individual, but where these differences are not identified perceptually (Hewlett, 1988). For example, where /ʃ/ and /tʃ/ are produced with different tongue placement on EPG, but where both are heard as /ɬ/ perceptually (Hewlett, 1988). In this research, covert contrasts were evidenced with Emma. Although Emma’s /s/ and /ʃ/ were transcribed as a voiceless lateral velar fricative by the researcher on baseline assessments, EPG assessment showed different tongue-palate contact for these two speech sounds. For /s/, tongue-palate contact was in the velar region, while for /ʃ/ tongue-palate contact appeared to be further back (i.e. not completely visible by EPG). The same was the case for /t/ and /k/, /d/ and / g/, and /n/ and /ŋ/. The sounds /t/ and /k/ were transcribed as [k], /d/ and /g/ as [g], and /n/ and /ŋ/ as [ŋ]. However, differences were seen on EPG, with tongue-palate contact for /t/, /d/ and /ŋ/ seen in the velar region, while contact for /k/, /g/, and /ŋ/ not visible on EPG (i.e. presumably further back).

The findings of this study supports the recommendations of Lee et al. (2009) in that cleft EPG intervention studies should report both perceptual and EPG outcomes. Perceptual speech assessment best represents the reality of human's perception of speech, and remains the "gold standard" for reporting cleft speech outcomes (Sell & Pereira, 2016). However, EPG assessment can supplement these perceptual outcomes in important ways, as discussed above.

8.1.3 Acquisition and lexical generalisation: Summary and conclusions

All six participants showed acquisition of typical/more typical productions of target phonemes in treated words with therapy. Likewise, all six participants showed generalisation of typical/more typical production of target phonemes to untreated words/pseudo-words following therapy. Further, calculation of effect sizes showed large treatment effects for production of untreated words. Acquisition and lexical generalisation were measured by percentage of targets correct. Percentage of targets correct was derived from phonetic transcription carried out by two independent SLTs, blind to test point. Inter- and intra-rater reliability for the independent SLTs' phonetic transcription was high. As such, the study's positive results with acquisition and lexical generalisation come with a degree of confidence. Acquisition and lexical generalisation were further measured by EPG assessment. EPG assessment showed change with target speech production, in that tongue-palate contact for target sounds moved in the direction of improvement, and better matched reference frames post-therapy. Thus, the answer to the first research question – **does the usage-based EPG technique improve accuracy of targeted phonemes in treated words during therapy and post-therapy, and if so, does this improved accuracy generalise to untreated/pseudo- words?** – is **yes**. However, whether this would have remained the case had more patients been recruited is unknown. It may be that non-responders would have occurred with larger number of participants.

8.2 Functional generalisation and impact of quality of life

This section discusses functional generalisation before considering impact of therapy on quality of life.

8.2.1 Functional generalisation

The ultimate goal of any intervention for SSD is functional generalisation of target sounds, i.e. use of target sound/s in everyday, continuous speech, in all speech settings. As discussed previously, functional generalisation has proven problematic for individuals undergoing visual biofeedback therapy, including EPG, in the past (Cleland & Preston, 2021; Gibbon & Paterson, 2006; Lee et al., 2009). Acquisition of target speech sounds occurs for many, but transfer of learning to everyday speech, and maintenance of that learning, has proven challenging. Since patients put forward for EPG and other biofeedback therapies are typically individuals with very entrenched SSDs, difficulty with functional generalisation is not unexpected. Consequently, any findings for this group of hard-to-treat patients need to be considered in this context. Nevertheless, as argued in Chapter Three (see 3.2), if visual biofeedback therapies are to be used, functional improvement with speech is needed to justify their wider use in clinical settings.

A limitation of intervention studies for SSD, in general, is that experimental treatment does not typically include therapy to discharge (Sugden et al., 2018), i.e. seemingly, intervention studies do not include the treatment phase involving functional generalisation. This is presumably because many studies are time-limited due to finite research funding or the length of post-graduate courses. A strength of cleft EPG studies to date is that many report on therapy to discharge (Fujiwara, 2007; Stokes et al., 1995; Whitehill et al., 1996). Despite this, reports of complete functional generalisation are low (see Table 1 for a review of cleft EPG intervention studies). However, the number of previous cleft EPG studies is very small (i.e. a total of seven experimental studies) and most studies are within participant designs or small group studies (see Table 1). As such, the current evidence base is low.

To date, the most comprehensive information on generalisation following EPG therapy with children with SSDs comes from Gibbon and Patterson's (2006) clinical survey. These authors surveyed SLTs' views on EPG therapy outcomes for children with SSD in Scotland over a 10-year period. SLTs returned questionnaires for 95% of the patients who had EPG palates made for them in Scotland over this time period (n = 71). Close to half of these patients had SSD related to cleft palate. Of those undergoing EPG treatment (n = 60), 12.5% of patients achieved total success, 41.1% showed "moderate success where some or all sounds targeted in therapy showed improvement and were used in some speaking contexts" (p 283), 19% showed "slight improvement in some speaking contexts" (p 283), while 12.5% showed no change in any speaking contexts.

Using Gibbon and Patterson's taxonomy for success, in the study presented in this thesis, 33% of participants showed total success (Olivia and Emma), 50% showed near total success (George, Grace and Poppy) and 16% showed moderate success (Victoria). These levels of generalisation are encouraging. However, such results may not have occurred had this study involved a greater number of participants. With larger numbers of participants, it is possible some subsequent participants may have shown lower levels of, or no, generalisation. In addition, maintenance assessment for this study was taken 3-months following the end of therapy. It is possible that the maintenance assessment time point (i.e. 3-months post-therapy) was too short. Since these participants will be seen for follow-up, as part of their clinical care, it will be important to see if improvements with functional generalisation are maintained longer term, for example 12-months post-therapy. In the present study, a 12-month post-therapy maintenance assessment would have been difficult to accommodate, given the time-frame of the researcher's programme of study.

It is also important to note that functional generalisation did not occur rapidly for participants in this study. Although production of target phonemes was achieved at untreated word level for 5/6 participants within 5 sessions, functional generalisation into everyday continuous speech took many more sessions. Participants required an average of 11 additional sessions to achieve functional generalisation. As identified above, in past accounts of intervention for SSD, this phase of therapy (i.e. functional generalisation) has received much less attention (McLeod, 2017; Williams et al., 2010). For the participants of this study, the conclusion is that this phase of therapy was long.

In this study, functional generalisation was measured through percentage of targets correct in a 2-minute sample of connected speech, together with parent- and spouse-reported ratings of speech understandability and SLT's ratings of speech understandability and speech acceptability. Percentage of targets correct as an outcome measure was discussed in the previous section. What follows is discussion of parent-reported ratings of speech understandability and SLTs' ratings of speech understandability and speech acceptability

8.2.2 Parent report of speech understandability

As previously defined, speech understandability relates to how well a listener can understand another person's speech in a particular communicative context (Hustad et al., 2015; Yorkson et al., 1996). In this study, parent report of speech understandability was measured using the Intelligibility in Context Scale (ICS) (McLeod et al., 2012), as described in 5.2.7. According to parent and spouse report, all the participants in this study could make themselves understood to people who were familiar to the participant (e.g. family), even those participants with a large number of affected consonants. This finding is in line with other research showing higher speech understandability scores for people familiar with the speaker (Flipsen, 1995; McLeod et al., 2012). However, previous research does not speculate what it is, specifically, that affords familiar listeners this advantage, apart from sheer contact with the child. In this study, high levels of speech understandability may have been related to participants' consistency with production. Little variability with cleft speech characteristics was shown, i.e. production of speech sounds was typically consistent. Consequently, a familiar listener could easily "tune into" a participant's cleft speech characteristics. In this study, Emma presented with the largest number of affected consonants. Acoustic phonetic transcription identified a large number of phoneme collapses (e.g. /t/, /k/→ [k], /d/, /g/ → [n]), /n/, /ŋ/→ [n]), which, by definition, is associated with reduced speech understandability. However, EPG analysis showed covert contrasts for these phonemes (i.e. differences in tongue-palate contact for phoneme collapses – see 8.1.2), which perhaps listeners actually perceived at some level, especially those very familiar with Emma's speech. Perhaps this also explains her higher scores with familiar listeners.

Despite scores in the mid- to high- range for familiar listeners, improvements in ICS scores were shown on re-assessment post-intervention for all participants apart from George, that is, the scale was sensitive to improvements with speech sound production for 5/6 participants. George's score on maintenance assessment was the same as his score on initial assessment. This occurred despite the fact that George's speech sound production had markedly improved. Emma was scored as 4.28 on assessment at the end of therapy. Her score on maintenance, 3-months later, was five, despite the fact that her speech sound production was unchanged from her previous assessment, immediately following therapy. These findings point to multiple factors contributing to speech understandability, i.e. not just accuracy of speech sound production. These findings are in line with other researchers who highlight the complexity of this entity, and its measurement (Hustad et al., 2015; McLeod et al., 2012; Miller, 2013; Whitehill et al., 2011). As the ICS currently exists, no opportunity is given for the parent/carer to describe what it is about their child's speech that makes them less understandable. Miller (2013) argues strongly for greater diagnostic assessment of speech understandability, that is, what it is about an individual's speech that makes it less easy to understand. He argues this information is important for intervention and to assist our understanding of this aspect of speech. Thus, having parents provide information on exactly what it is that makes their child less understandable would be helpful addition to the ICS.

8.2.3 SLT rating of speech understandability and speech acceptability

As defined previously, speech acceptability relates to how close an individual's speech is to their peers, or the degree to which a person's speech draws attention to itself, outside of the content of the spoken message (Henningsson et al., 2008). As identified by Miller (2013), even when speech understandability is not particularly affected, identifiable differences with sound production may be associated with marked psychosocial consequences for the speaker. In this study, the SLTs made judgements of speech understandability and speech acceptability using the scales developed by Henningsson et al. (2008), as described in section 5.2.8. These scales are part of a wider set of measures used to report speech outcomes in individuals born with cleft palate (Henningsson et al., 2008).

All participants showed improved ratings of speech understandability and speech acceptability post-therapy, apart from George. On baseline assessment, the SLTs attributed George's reduced understandability and acceptability to his atypical speech sound productions. Post-therapy reduced ratings were attributed to nasal turbulence which had increased over the course of therapy. It should be noted that lower levels of inter-rater and intra-rater agreement were shown with these rating scales. Lower levels of agreement were, in particular, shown for ratings of speech understandability. Inter-rater agreement for the two independent SLTs was 60% for both the speech understandability and speech acceptability scales. Intra-rater agreement was 80% for both scales for one independent SLT, and 50% (understandability) and 100% (acceptability) for the other independent SLT. The researcher's agreement with each independent SLT was 53% (understandability) and 80% (acceptability). Intra-rater agreement for the researcher was 75% (understandability) and 100% (acceptability). However, agreement within two points (i.e. a score of "0" and "1" rated as agreed etc.) produced high agreement scores for all agreement calculations relating to these measures (see 5.2.11). This suggests that the SLTs were applying slightly different criteria when using these scales. To date, we are not aware of any other research that has used these ordinal scales. Lower agreement means that caution is needed in interpreting the speech understandably and speech acceptability ratings in this study. These scales may benefit from further development. For example, better quantification of the descriptors used in this scale, for example, "deviates to a mild degree", "deviates to a moderate degree" etc., may increase agreement between raters. Use of visual analogue scales and word recognition tests are alternatives to the measurement of speech understandability and acceptability (Miller, 2013). However, these tools are also associated with lower inter- and intra-rater agreement (Miller, 2013). Lower agreement with different measures again highlights the complexity of speech understandability and acceptability in terms of defining and measuring these holistic outcomes. Development of tools to measure these aspects of speech remains a challenge for researchers (Hustad et al., 2015; Miller, 2013; Whitehill et al., 2011; Yorkson et al., 1996).

In addition to SLTs' assessment of speech acceptability, as suggested by Lohmander (2010) and Sell and Pereira (2016), having naïve listeners make a judgement of speech acceptability would have been useful. As discussed above, in this study, George's change with /tʃ/ to [tj] and /dʒ/ to [dj] following therapy was seen as improvement by the researcher. Likewise, Victoria's change with /z/ to [s] and /dʒ/ to [tʃ] was viewed as an improvement in production. However, naïve listeners' judgement may have differed from the SLTs' opinion. In addition, having the participants' opinion on their own speech acceptability would have been useful. Such assessment was not completed in this study, as the main focus for this exploratory research was with changes to speech sound production, as shown by phonetic transcription and EPG analysis.

8.2.4 Quality of life

As far back as the early 1990s, Enderby (1992) identified the potential negative impact of communication difficulties on daily life and satisfaction with communication as important concerns for speech and language therapy intervention. However, measurements of change to daily activities and general satisfaction with speech following therapy have been slow to appear in intervention studies involving cleft palate speech (Howard & Lohmander, 2011). This may be because, traditionally, management of the cleft condition has been firmly embedded within a medical model (Stackhouse & Wells, 1997). Nevertheless, in recent times there has been a growing awareness of the importance of measurement of these wider outcomes with this client group (Howard & Lohmander, 2011; van Eeden et al., 2019). As identified previously, to date, no EPG intervention study has measured the impact of intervention on quality of life. In this study, quality of life was measured through Speech Participation and Activities of Children (SPAA-C) tool (McLeod, 2004), in the case of children, and the Quality of Life (QoL-Dys) instrument (Piacentini et al., 2011), in the case of the adult participants.

In this research, 2/3 children's reported quality of life was not particularly affected by their SSD and reported quality of life was largely unchanged post-therapy. For one child (Grace) this may have been because her SSD was relatively mild. Alternatively, since the SPAA-C was not developed specifically for cleft palate, this tool may lack sensitivity for this group of children (i.e. tool not measuring important aspects of quality of life for children with cleft palate speech). In addition, because of their age, these two children may have lacked ability to report their own feelings. If so, a proxy measure involving their parents may have been useful. For the third child, reported quality of life on the SPAA-C was notably affected by her SSD and reported quality of life improved following intervention. This child had the most extensive SSD of all three children and was also the oldest.

For the three adult participants, 2/3 (Olivia and Victoria) felt that their SSDs notably affected their quality of life and both reported improved quality of life following intervention. This improvement was most marked for Victoria, who, for this case series, had the poorest outcome in terms of percentage of targets correct in connected speech. Thus, ratings of quality of life did not necessarily correlate with amount of improvement with speech sound production. Pre-therapy, self-ratings of quality of life for the remaining adult were not overly affected by her SSD. However, discussion with her family indicated this participant's SSD had very notably impacted on her quality of life in the past and this continued to be the case. Thus, it appeared that the QoL-Dys was not a sensitive measure for this participant. As discussed in the method chapter, the QoL-Dys was developed for adults with dysarthria, and was chosen in the absence of a quality of life measure for individuals with cleft at the time of this study's application for ethical approval. Since this time, a self-reported measure of quality of life has been developed for individuals born with the cleft condition, the CLEFT-Q (Klassen et al., 2018; Wong Riff et al., 2019). The CLEFT-Q was rigorously developed from data from nearly two and a half thousand children and adults born with cleft palate +/- lip in 12 different countries. The CLEFT-Q may have identified quality of life issues for this third adult and is likely to have been a more sensitive and comprehensive measure for the other participants in this series. The Therapy Outcome Measure (Enderby, 2006) has also been used as a quality of life outcome measure for individuals born with the cleft condition (Rees et al., 2016), and this may be a useful measure for future studies. It is possible that the adult not identifying significant quality of life issues, may have had difficulty reporting her own feelings and experiences. If so, a proxy measure involving her family may have been useful.

8.2.5 Functional generalisation and impact of quality of life: Summary and conclusions

All participants showed generalisation of target speech sounds to continuous connected speech, though to varying degrees. Individual's use of target sounds in continuous connected speech ranged from 78.94% - 100 (mean = 90.66%). Parent and SLTs' ratings of speech understandability and speech acceptability improved for 5/6 participants. Self-reported improvements in quality of life occurred for 3/6 participants. Speech understandability and speech acceptability was rated by SLTs using Henningson et al.'s rating scales (Henningsson et al., 2008). Poorer inter and intra-rater reliability was shown with these scales, therefore caution is needed in evaluating findings related to these functional outcomes. Likewise, the quality of life tools used in this study were not developed specifically for individuals with cleft palate speech, and therefore caution is needed with consideration of outcomes. Thus, the answer to research question number two - **does improved accuracy of target phonemes generalise to continuous, connected speech, and, if so, does this result in improved ratings of speech understandability and speech acceptability and improved ratings of impact on quality of life, post-therapy?** – is:

- **yes**, improved accuracy of target phonemes generalised to continuous, connected speech of all participants, to high levels;
- **partially,** 5/6 participants showed improved speech understandability and acceptability while the remaining participant showed no change. However, this was due to increase in nasal turbulence over the course of therapy, rather than unchanged speech sound production (but see below);
- **partially**, 3/6 reported improved quality of life while the remaining three participants reported no improvement with quality of life post therapy. However, these three participants did not report any current quality of life issues pre-therapy (but see below).

However, given the shortcomings of the tools used to measure speech understandability, speech acceptability and quality of life, as discussed above, caution is needed with bullet points two and three above.

8.3 Support for the theoretical model

This study's third and final research question related to support for the theoretical model. The therapeutic technique used in this research was informed by usage-based phonology theory (Bybee, 2001, 2010; Menn et al., 2013). This theory, as summarised in 4.2.2, informed therapeutic premises, and specific teaching events came from these therapeutic premises. Therapeutic premises and teaching episodes are detailed in 4.2.4. The resultant usage-based EPG therapy delivered in this study involved: a high number of production trials; use of EPG to provide visual feedback on tongue-palate contact; a focus on word production rather than single sounds; teaching low frequency words before high frequency words; facilitation of phonetic gradience; and teaching connected speech patterns. The therapy delivered in this study produced improvement with everyday speech for all participants (as determined by the multiple baseline ABA design), thereby provided some support for the theoretical model. What follows is discussion of the therapy's key therapeutic premises/teaching episodes in relation to study findings and previous research: focus on single words/frequency of use; gradient speech change; generalisation through sub-neural connections; connected speech patterns; continuity of speech learning. This is followed by comparison of the usage-based therapeutic model with therapy based on schema theory. As identified in Chapter Two, schema theory is the theoretical basis for contemporary articulation and visual feedback therapies.

8.3.1 Focus on single words/frequency of use

According to the usage-based model, phonology is intrinsically linked to meaningful units such as the word (Bybee, 2001). Further, the process of phonological learning and change occurs with use of words/meaningful units in communicative settings (Bybee, 2001). Thus, the theoretical model predicts greater and more rapid change to speech will occur when the focus of therapy is with meaningful units, such as the word. In the therapy delivered in this study, practice of target sounds at word level occurred from session one. In this study, participants were able to alter their production of target sounds at word level relatively easily with EPG visual feedback, despite never or rarely being able to produce target speech sounds accurately (in either isolation or in words) prior to EPG intervention. It helped that exact production of target sounds in words was not an immediate requirement, using the concept of gradience (see 8.3.2 below). Rather intermittent, step-wise articulatory change in words was permitted. This immediate focus on words is novel and is in contrast to traditional articulation therapy, where practice of target sounds occurs in isolation and syllables before moving to words. In many past studies involving articulation therapy, a large number of sessions have been given over to production of target sounds in isolation and syllable level (Dobbelsteyn et al., 2014; Preston et al., 2016; Sjolie et al., 2016). In the present study, this single sound and syllable practice did not occur. However, whether or not the study's immediate focus on words expedited therapy, from start to finish, cannot be determined. To do this, a design which compared this approach with one practicing target sounds in isolation, syllable etc. would be needed.

According to the usage-based theoretical model, words are associated with articulatory memories. These memories become increasingly entrenched with production practice (Bybee, 2001; Menn et al., 2013). Thus, the theoretical model predicts that any change with articulatory gestures will require speech production practice. Further, in the treatment of SSDs, the theoretical model predicts that, in general, greater amounts of production practice will be needed for older children and adults, compared with younger children. This is because older children and adults will have more entrenched articulatory gestures than younger children. This study involved a high dose of production trials within sessions. On average, 250 production trials occurred per treatment session. In this study, George received 15 sessions of therapy over a five-month period. Olivia received 25 sessions over an eight-month period. Emma had 24 sessions over 10-months. Victoria was seen for a total of 26 sessions over a 16-month period. Grace received 33 sessions over a 15-month period. Finally, Poppy received 20 sessions over a seven-month period. Cumulative intervention (production dosage x number of sessions) for participants treated within this research was therefore: 3,750 (George); 6,250 (Olivia); 6,000 (Emma); 6,500 (Victoria); 8,250 (Grace); and 5,000 (Poppy). These figures do not include practice done at home. Although weekly sessions were offered, sessions were less frequent due to illness, holiday, absence due to school exams/events and other family commitments. Thus, as predicted by the theoretical model, participants required a large amount of production practice to achieve full/near generalisation of target sounds into continuous connected speech.

At present, total cumulative intervention needed to cure a SSD in a child, whatever the aetiology, is largely unknown (Baker & McLeod, 2011; Sugden et al., 2018). The reason for this is two-fold. Firstly, SSD intervention studies have rarely provided comprehensive details on dosage (Sugden et al., 2018). Secondly, as discussed earlier, few studies report of the entire duration of treatment, that is, from initial assessment to discharge (Baker & McLeod, 2011; Sugden et al., 2018). In this study, dose frequency (i.e. how often a child receives therapy, and for how long, see 3.2.1) was similar to four of the seven cleft EPG intervention studies reviewed in 3.2. A shortcoming of previous cleft studies is that dose (i.e. how many times a teaching episode is delivered per session, see 3.2.1) has not been reported. Consequently, cumulative dosage cannot be calculated for previous cleft intervention studies. Thus, the cumulative intervention figures calculated in this study have no direct comparators.

Hitchcock and colleagues (2019) examined treatment intensity in published reports of visual biofeedback intervention for SSD. Intervention included EPG, ultrasound and visual acoustic biofeedback. Participants with a structural anomaly, e.g. cleft palate +/- lip were excluded from this review. Cumulative intervention could be calculated for 18 of the 29 studies and this ranged from 840 – 5,117. However, whether or not this represented total duration of therapy (i.e. therapy to discharge) was not stated in this analysis. These authors found a weak, but statistically significant, correlation between cumulative intervention and treatment outcomes, with higher cumulative intervention being correlated with better speech outcomes. Future cleft intervention studies, and SSD intervention studies in general, need to provide details of cumulative intervention, ideally for total duration of therapy (i.e. to discharge). In doing so, comparisons amongst intervention studies will be possible.

As well as a focus on words/meaningful units, the usage-based model adopted in this study predicted speech targets in low frequency words (i.e. words occurring with low frequency in the English language) would be easier to learn compared with high frequency words (i.e. words occurring with high frequency in the English language). According to the theoretical model, low frequency words have less entrenched neuromotor memories, while high frequency words have highly entrenched neuromotor memories (Bybee, 2001, 2010; Menn et al., 2013; Vogel Sosa & Bybee, 2008). Consequently, in this intervention research, low frequency words were taught first, followed by high frequency words.

To date few SSD intervention studies have considered word frequency. In a series of three within-participant intervention studies, (Gierut & Morrisette, 2012; Gierut, Morrisette, & Hust Champion, 1999; Morrisette & Gierut, 2002) word frequency was manipulated in the treatment of pre-school children with developmental SSDs. Children were taught either low frequency words or high frequency words (Gierut, Morrisette, & Champion, 1999;

Morrisette & Gierut, 2002) or individual participants were taught low and high frequency words alternatively (Gierut & Morrisette, 2012). Findings showed that teaching high frequency words promoted better system-wide speech change. The authors suggested that teaching high frequency words may be advantageous due to the easing of language processing demands. In contrast to this research, these authors approached speech production from a levels-of-processing viewpoint. They proposed high frequency words may be retrieved from the lexicon more rapidly than low frequency words. They suggested this easing of processing allows the child to focus on successful learning of the treated sound. In addition, these researchers suggested extra processing capacity can be given to generalisation.

In this study, as a group, no difference in percentage of targets correct was seen for high versus low frequency words on assessment at the end of five sessions and assessment at the end of ten sessions (Wilcoxon signed-rank test, $p > .05$). However, a marked difference was seen in percentage of targets correct in low versus high frequency words in connected speech samples for all participants during the acquisition and lexical generalisation phases. Participants were much more likely to produce high frequency words in error in connected speech, and this difference was statistically significant (Wilcoxon signed-rank test, $p < .05$). This would suggest that a strong word frequency effect was operating in connected speech. Applying usage-based theory, this finding is interpreted in the following way: high frequency words, with their highly entrenched motor memories, were exerting a strong pull to old exemplars, and were more resistant to updating, compared with words with less entrenched motor memories (Menn et al., 2013).

The finding of a word frequency effect provides some support for usage-based models. According to usage-based models, “words” consist of a). motor/phonetic memories, b). memories of meanings of the words, and c). memories of the contexts in which these motor memories have been used (Bybee, 2001, 2010). At birth, an infant’s mind is perceived to be like a blank slate. Linguistic memories develop with exposure to language. Accordingly, these memories become more entrenched with usage. When a child goes to produce a word, they execute the most entrenched motor memory associated with that word (Bybee, 2001; Menn, 1983) (see 4.1). Thus, a word frequency effect is predicted with this model. This view is in contrast to generative theory. Generative theory views a child’s underlying lexical representation as innate and as approximating the target. Rules operate to produce surface forms (Chomsky, 1991). For example, a child who produces [ku] for “two” is assumed to have some kind of representation for [tu] in their mental lexicon, despite their misarticulation of the word. According to the generative stance, misarticulation occurs with the linking of the lexical representation and the depiction of the target sounds in the phonological system (Storkel, 2018). Evidence for this perspective comes from experience showing children often perceive the difference between a word and their own production substitute in listening tasks, for example, [tu] versus [ku], as in the example above (McGregor & Schwartz, 1992; Tyler et al., 1990). However, other researchers cite evidence related to children's difficulty with the same perception tasks (Macken, 1980; Vihman, 1982). Storkel (2018) presents a hybrid approach. She holds onto the generative idea of lexical representations and separate phonological systems (in contrast to usage-based models). However, she suggests lexical representations in the young child may be underspecified or lacking in detail. If so, some kind of frequency effect is likely, as speech usage will impact on underspecified lexical representations. However, this frequency effect is likely to be less than that predicted by usage-based theory.

In this study, it was also predicted that high volume production of high frequency words in the later stages of therapy would advance generalisation. According to the usage-based model, words occurring with high frequency will have strong sub-neural connections with other word tokens (Menn et al., 2013). In this phonological theory, it is this sub-neural network that produces system-wide change, and consequently generalisation to non-treated tokens (see 4.1.4). However, whether or not generalisation was, in particular, facilitated by a focus on high frequency words at the later stage of therapy, as predicted by the theoretical model, can't be determined with the current study's research design. In order to determine this, a design which compared this approach with another approach without a high frequency word focus, would be needed, as was done in the Gierut et al. (1999, 2002, 2012) studies discussed above. In addition, according to the usage-based theoretical model, any production practice will arouse similar speech tokens, thereby developing and updating the sub-network of representations. As discussed above, large volumes of production trials were executed by study participants per session. It may be that sheer volume of production trials was the key factor in achieving generalisation and maintenance of speech change following therapy, rather than the focus on high frequency words.

8.3.2 Gradient speech change

A core concept of usage-based phonological theory is the notion of gradience (Bybee, 2001). Phonological change is conceived to occur in a gradual way, with gradient phonetic adjustment occurring in words/production tokens over time. For example, a gradual adjustment of /t/ to /ʃ/ within words over time may look like: [t] → [s] →[s^j] → [ʃ]. The usage-based basis for this is exemplar theory: The core neuromotor trace (i.e exemplar) for a spoken token shifts given the motor control and speech experience of the individual (Bybee, 2001; Menn et al., 2013). Gradient phonetic change is a well-known phenomenon in young, typically developing children acquiring their first language. Studies of the speech of typically developing, children over time shows that speech sound production develops in a broadly step-wise manner towards mature phonology (McLeod, 2007). Studies of sociolinguistic speech change in typical adult speakers also demonstrates gradient speech change (Bybee, 2001, 2010). In contrast, to date, the possibility of gradient phonetic change in children/adults with SSD has received little attention. In an innovative study by Cleland and Scobbie (2021), gradient speech change was shown in five children with SSD.

Using ultrasound imaging, these researchers demonstrated that five children with persistent velar fronting, aged 6;1 – 13;2 years, acquired /k/ in an articulatory gradient manner.

In this study, gradient change of target sounds within words was seen for all participants. In the early stages of therapy this gradient change was directly due to the treatment: The researcher specifically gave instructions for intermediate phonetic step-change towards more accurate production. In this study, early in therapy, George's production of [ts] for /tʃ/, [dz] for /dʒ/, and [s] for /ʃ/ was taught in single word production. The participant was told that this production was an intermediate step towards typical production. Likewise, Emma's production of [ʃ] for /s/ and [ts] for /tʃ/ was initially taught in words. Olivia's production of [s] for /z/ was taught in the initial stages of intervention. Victoria's production of [sʲ] for /s/, [ʃ] for [tʃ/ and [s] for /z/ was initially taught. Initially a marked interdental production of /s/ was taught for Emma. Finally, in Poppy's case, an interdental production of /n/ and /l/ in words was facilitated. Shaping of single sounds (e.g. where /s/ is elicited from /ʃ/ with a cue to draw the tongue back) is a familiar technique in articulation therapy (Cleland & Preston, 2021; Cleland et al., 2015; Gibbon & Wood, 2010; Preston & Leece, 2021). In this study, such shaping occurred at a word level, rather than at single sound level.

Gradient change in tongue-palate contact was also seen in later stages of therapy for some participants without instruction from the researcher. This change occurred after the time in which the participant had achieved what the researcher thought was a perceptually acceptable speech sound. For example, Olivia had achieved a perceptually acceptable /s/ and /z/ by assessment at the end of five sessions. However, production of these sounds showed further refinement on EPG over the generalisation phases of therapy, with examination of successive EPG palatograms in pseudo-word showing increasingly narrow groove formation (see Figure 23). Likewise, progressive phonetic change and refinement is shown for Emma's production of /t/, /d/, and /n/ in pseudo-words. This change occurred after these sounds had been acquired and assessed as perceptually accurate. These speech sounds show a pattern of reduced tongue-palate contact on EPG, so that they better match reference frames (see Figure 24). This gradience presumably occurred through a mechanism whereby participants' auditory memories of speech tokens (participant's own speech and the speech of others) were used to compare and contrast to further refine subsequent output speech tokens, in a similar manner to the speech attunement framework (Kwiatkowski & Shriberg, 1993, 1998; Shriberg et al., 2011) (see 4.2.1 for discussion of this framework). Such gradient speech change would also be expected with intervention based on motor learning theory, where speech movements are refined with speech practice (Maas et al., 2008; Preston & Leece, 2021) (see 8.3.6 for further discussion regarding motor learning theory and usage-based theory).

8.3.3 Generalisation through sub-neural connections

According to usage-based phonology theory, the mechanism for system-wide generalisation is the operation of sub-neural networks. Change to an articulatory gesture in a treated word, for example /t/ in "to" will arouse neuromotor memories of untreated words with /t/, such as "toe" and "tea". Over time, repeated practice of treated words will update articulatory gestures in untreated words (Menn et al., 2013). Further, according to usage-based theory, articulatory memories associated with a word are linked with the meaning of the word and memories of the contexts in which they have been used (Bybee, 2001). In this way, generalisation occurs across words (i.e. from treated words to untreated words), and across situations (i.e. from the clinical setting to school, home, work etc.). In this study, across word generalisation occurred, as discussed in 8.1. In addition, across situation generalisation occurred, according to parent report on the Intelligibility in Context Scale.

Another type of generalisation often seen in SSD intervention is across-sound and across-feature generalisation (Bankson et al., 2017). This is when generalisation occurs within sound classes or with sounds that at phonetically similar (e.g. /s/ to /z/; /ʃ/ to /tʃ/). This type of generalisation is expected with phonological treatments which are have generative theory as their underpinning, such as minimal pair therapy (Baker, 2010) and multiple oppositions therapy (Williams, 2010). These phonological therapies work by seeking to increase the patient's awareness or knowledge of the rule-based structure of their ambient speech sound system. For example, if a child is exhibiting a pattern of "fronting", where velar sounds /k/, / g/, /ŋ/ are produced as [t], [d], and [n] respectively, remediation of /k/ may generalise to other sounds within the same class (i.e. sound with posterior placement), so that /g/ and /ŋ/ are produced accurately, without any specific work on these related speech sounds.

Across feature generalisation was shown in Whitehill et al.'s (1996) EPG intervention study with an Cantonese speaking adult with cleft palate speech. In this study EPG therapy for /s/ generalised to the Cantonese affricatives /ts/ and /tsʰ/ following EPG feedback and production work for /s/, despite no specific work on these affricative sounds. However, in this paper, the authors did not propose a reason for this across-feature generalisation. Across-sound and across-feature effects were seen in the present study. Changes to Graces' production of /s/ (i.e. more forward placement) generalised to her production of /z/, before specific work on this voiced sound commenced. However, the most obvious example of across-feature generalisation occurred for Emma and can be seen on EPG frames (see Figure 17). Prior to EPG treatment, Emma presented with patterns of backing and lateral airflow release for lingual sibilant sounds. Treatment of /t/, /d/, /s/, /z/ and /tʃ/ in the early stages of therapy had an influence on production of untreated /ʃ/, /dʒ/, /n/ and /l/. All showed some evidence of more anterior placement and removal of lateral airflow at pseudo-word level on EPG assessment after 5 sessions of therapy, as shown in Figure 17, despite these speech sounds not being treated until **after** session five. Across-sound and across-feature generalisation with production practice can be explained using usage-based phonology theory. Change to an articulatory gesture in a word will evoke motor and auditory memories of words with the same, but also similar, articulatory gestures, for example, production of /s/ will evoke memories for all lingual sibilants to an extent (Menn et al., 2013). In this way, over time, production practice of, for example, a particular word, will update words containing similar articulatory gestures.

8.3.4 Connected speech

Connected speech relates to the systematic way words abut together in continuous, connected speech (Ellis & Hardcastle, 2002). For example, in rapid connected speech, for most speakers, the phrase "red car" becomes [ɹɛg ka] (i.e. /d/ is backed to [g] in anticipation of /k/) and "miss you" becomes [mɪ ʃu] (i.e. /s/ is realised as [ʃ] in expectancy of /j/ in "you"). Wells (1994) makes the distinction between open and closed connected speech patterns at word and syllable boundaries. Closed patterns act to bind words together and include, for example, assimilation patterns as in the "red car" example above, and coalescence patterns, as in the "miss you" example (Howard et al., 2008). In comparison, open connected speech patterns seek to make a clear separation between two words, and include such behaviours and the insertion of a glottal stop, or a slight pause between words. Howard et al. (2008) suggest that as humans "we display our mastery of the phonology of the language as much by the ways in which we connect words up – our realisation of word junctures – as we do by our pronunciation of individual words" (p. 583). These authors consequently argue an essential part of an individual's phonology includes learning the phonology of connected speech and they contend this is an area in need of greater study and understanding. Howard (2013) suggests atypical use of connected speech patterns can affect both speech understandability and speech acceptability. Howard (2013) presents the case of a 9-year-old child with cleft palate speech who demonstrated an over-use of open connected speech patterns. Although this child's speech was understandable, his over-use of open connected speech patterns resulted in reduced speech acceptability.

To date few phonological theories have considered connected speech. Although not specifically considered previously (to our knowledge), usage-based theory suggests the phonology of connected speech may have several dimensions. Overlapping and reduction in connected speech will be a direct consequence of rapid sequenced movement (Bybee, 2001). It is also likely that this production will result in exemplars for groups of words and segments within words (exemplar = neuromotor memory). For example, an utterance including the words "have to", with usage, may result in an exemplar where /v/ becomes [f] whenever this meaning unit is used within a particular context. The same mechanism is likely to be the case for phonetic devices applied to preserve word and syllable boundaries in connected speech, such as the use of a glottal stop. In addition, an individual's connected speech is likely to be influenced by their input auditory exemplars of connected speech, and they are likely to match their output to speakers within their social and/or geographical group. Ellis and Hardcastle (2002) report EPG data which provides some support for the novel usage-based account of connected speech suggested here. These researchers examined alveolar to velar assimilation in 10 adults using EPG. The speakers repeated a number of test sentences which included potential alveolar to velar assimilation. The speakers showed individual differences in assimilation strategy. Two speakers never assimilated. Two speakers showed gradual assimilation, suggesting the assimilation was a consequence of coarticulation. Four speakers showed complete assimilation, perhaps suggesting application of a connected speech exemplar. The remaining two produced full alveolars or complete assimilations, perhaps suggesting variable application of a connected speech exemplar. Further tentative support for the ideas presented here comes from the study by Rutter (2014). In this study, the production of /str/ in words varied across adult speakers, and that the speech of some children appeared to match the usage patterns of their mothers.

To date, we are not aware of any therapeutic technique that has included teaching connected speech patterns. In this study both closed and open connected speech patterns were taught in the later stages of therapy, with the assumption that what was being taught were connected speech exemplars. This teaching was straight-forward and can be summarised as follows: The researcher explained that as mature speakers, at times we do things to blend words together (i.e. closed connected speech patterns), while at other times we do things to make words more distinct (i.e. open connected speech patterns). The researcher then modelled different patterns at sentence, and connected sentence level. The participants were asked to copy given a direct model. Gradually this modelling was reduced. All participants quickly responded to this teaching and were able to copy and subsequently incorporate connected speech patterns into their continuous connected speech.

Given her clinical experience, the researcher had predicted most participants would present with hyper-articulation following the acquisition and word generalisation stages of therapy, thereby necessitating the need for teaching closed connected speech patterns. This was the case for Emma and Poppy. Following the acquisition and word generalisation stages, Emma and Poppy's speech was characterised by hyper-articulation, with an over-emphasis on target sounds and over-use of open speech patterns (researcher's subjective impression). Both responded to being taught closed connected speech patterns and this instruction resulted in more natural sounding speech (subjective impression). However, for all other participants, hyper-articulation was not a feature of their speech (subjective impression). Although target sounds were modelled and taught in closed connected speech patterns, the researcher also had to encourage use of open connected speech patterns, as generally the trend was for hyperelision (i.e. over-use of closed connected speech patterns) among the other four participants. In three of the four (George, Victoria and Emma) this appeared, at least in part, related to very fast rate of speech.

In this study it was predicted that teaching connected speech patterns would accelerate therapy and improve participants' speech acceptability. Following treatment, speech acceptability improved in 4/6 participants. Use of connected speech patterns may have been a factor in improved speech acceptability. However, it may be appropriate use of connected speech patterns simply came with speech practice at word and sentence level.

Studies of typical speakers suggest, with practice, speakers start to incorporate aspects such as coarticulation naturally (Bybee, 2001, 2010). Further research is needed to examine if teaching connected speech patterns accelerates speech acceptability. This could be done by comparing a treatment technique which included teaching connected speech patterns with one that did not contain such teaching.

8.3.5 Continuity of speech learning

Usage-based phonology theory assumes continuity between child and adult phonology. The mechanisms of phonological change in children is the same as for adults, i.e. speech usage events and the cognitive associations made following these usage events to develop and reinforce neural networks (Bybee, 2001, 2010). Thus, according to this theory, no pivotal age exists, after which time speech change is not possible, though, one might expect that speech change may be slower in adults given the concept of increasing entrenchment of neuromotor memories with speech usage over time (see discussion above). This assumption is in contrast to the critical age hypothesis (Lenneberg, 1967), where acquisition of speech and language occurs in a restricted window of time, usually the first few years of life, possibly up to puberty, after which time little or greatly reduced learning ensues. This later hypothesis is widely assumed in the management of developmental speech and language disorders (Baker & McLeod, 2011; Bishop & Adams, 1990; McWilliams et al., 1990). The critical age hypothesis has, in particular, been of interest in the field of second language learning. Some researchers argue strongly for a critical period for learning, after which time native-like competence of a second language is unachievable, and provide evidence to support this claim (Johnson, 1992; Johnson & Newport, 1991). In comparison, others present evidence not in support of this hypothesis (Birdsong & Molis, 2001; Hakuta et al., 2003). These latter writers point to instances of older learners who achieve native-like competence in second languages, together with a lack of marked change in learning immediately following the end of the critical period.

This study involved treatment of three school aged children and three adults. Studies reporting intervention for adults with cleft palate speech are scant. Generally, the prevailing view is that adults do not benefit from therapy (McWilliams, 1984; Noordhoff et al., 1987). Whitehill et al. (1996) report on the EPG treatment of an 18 year old female with cleft palate speech. This participant showed improved articulation at word level and some generalisation to connected speech following 23 one-hour sessions of EPG therapy.

In this study, the adults were treated in the same way as the child participants. All three adults showed increased accuracy of target phonemes at word level and this improvement generalised to continuous connected speech, to varying degrees. On maintenance assessment, Olivia showed 100% of targets correct in connected speech while Poppy showed 92% of targets correct. For the third adult, Victoria, less functional generalisation occurred, with 78% accuracy of target sounds in connected speech on maintenance assessment, though persistent VPI may have made functional generalisation more difficult for this adult (see 8.5 below for further discussion). The findings of this study support the usage-based assumption of the possibility for speech change in adults and that this change occurs using the same mechanisms children, i.e. speech use and the sub-neural links made following this speech use. As such, findings go against the critical age-hypothesis (Lenneberg, 1967). However, this change with the adults in this study did not come quickly. Adult participants required a high dosage of between 20 - 26 therapy sessions. This aligns with the usage-based assumption that speech change may be slow in adults given the concept of neuromotor entrenchment with speech usage over time.

8.3.6 Usage-based phonology and motor learning theory

As identified in Chapter Two, recently, motor learning theory has been used as the theoretical rationale for articulation therapy and visual feedback therapy (Cleland & Preston, 2021; Gibbon & Wood, 2010; Preston & Leece, 2021). In particular, schema theory (Schmidt, 1975) is used as the theoretical basis for articulation and visual feedback therapies (Cleland & Preston, 2021; Maas et al., 2008; Preston & Leece, 2021). Motor learning intervention based on schema theory has parallels with the usage-based therapy described in this thesis. Both focus on the motor execution of speech and the need for a high number of production trials during therapy sessions. Both use operant learning. However, usage-based phonology theory differs fundamentally from schema theory in a number of ways. According to usage-based phonology theory, phonological units and speech learning emerges in a bottom-up way, from raw speech and hearing events (Bybee, 2001, 2010; Menn et al., 2013). In contrast, schema theory is a top-down model: The origin of motor movement is seen to lie with generalised motor programmes (GMP). GMPs are abstract, innate, frameworks stored in memory that capture the invariant aspects of a particular set of movements. GMPs are adapted to produce specific movements through schema. These schemas may be conceived as “rules” which operate to provide detailed instructions to the musculature (Schmidt, 2003).

Table 21 compares schemata versus usage-based phonology therapy for SSDs. In this table, blue shading represents no difference between the two treatment approaches, while red shading identifies differences. As can be seen from Table 21, the main differences between the two therapies lie with practice schedules: Schemata theory suggests practice of simple targets (e.g. single sounds/words) is needed in the acquisition phase, while practice of more complex targets should occur in the motor learning/generalisation phases (Preston & Leece, 2021). Schema theory also suggests variable and random schedules of production trials are required for motor learning/generalisation (Schmidt, 1975). In addition, some production errors are seen as important for both acquisition and motor learning (Hitchcock & McAllister Byun, 2015; Schmidt, 1975). Variable practice and errors in production lead to more detailed schemas (see 3.3.2.3). In comparison, usage-based phonology theory (Bybee, 2001, 2010) suggests practice of word/meaningful single token targets should occur in both the acquisition and generalisation phases. In addition, usage-based theory suggests all practice should be blocked and constant, including in the generalisation phase. Further, this approach has no particular requirement for production errors.

Table 21

Comparison of schemata versus usage-based therapy for SSD

	Schemata Theory		**Usage-based Phonology Theory**	
	Acquisition	**Motor learning/ Generalisation**	**Acquisition**	**Generalisation**
Feedback	Frequent	Infrequent	Frequent	Infrequent
	Immediate	Delayed	Immediate	Delayed
	Knowledge of performance	Knowledge of results	Knowledge of performance	Knowledge of results
	Shaping of speech sound production at phoneme level		Facilitation of gradient articulatory change at word level	
Practice	Blocked schedule of production trials	Random schedule of production trials	Blocked schedule of production trials	Blocked schedule of production trials
	Constant practice	Variable practice	Constant practice	Constant practice
	Simple targets	Complex targets	Word / meaningful single token targets	Word / meaningful single token targets
			Low frequency then high frequency word/token targets	Low frequency then high frequency word/token targets
			Connected speech tokens	Connected speech tokens
	Practice in therapy setting	Practice in a range of communicative settings	Practice in therapy setting	Practice in a range of communicative settings (but not essential for generalisation)
	Some production errors important	Some production errors important		
Target populations	SSDs associated with articulatory distortions		All SSDs (theoretically)	

Note: SSD = Speech sound disorder; Blue shading = No differences between two therapy approaches; Red shading = Differences between two therapy approaches

(adapted from Preston & Leece, 2021)

Therapy based on Schema theory is seen as appropriate for SSDs associated with articulatory distortions (Cleland & Preston, 2021; Preston & Leece, 2021). In comparison, usage-based therapy is theoretically suitable for all SSDs. Although the usage-based therapy approach focuses exclusively on production, the basis of this therapy approach is a single, comprehensive, causal, phonological theory. Usage-based phonology surmises that phonological learning and change comes about from phonetic and auditory experiences (Bybee, 2001, 2010; Menn et al., 2013). As such, usage-based therapy can be viewed as a type of "phonological therapy", and theoretically could be used with all SSDs, including those children traditionally viewed as having "phonological" difficulties, i.e. children who are able to articulate phonemes in certain circumstances, such as production in isolation given instruction, but whose speech difficulties may be described in terms of phonological patterns (McLeod & Baker, 2017). Applying usage-based theory, the phonological patterns shown with these children are due to the operation of sub-network neural representations where, words containing a particular sound are mapped to other articulatory gestures. For example, in the case of a "fronting" pattern, /k/ and /g/ are mapped to [t] and [d] respectively in word production (Menn et al., 2013). The rationale for use of the usage-based therapy technique with this group of patients is as follows: production practice of words containing target sounds produces neuromotor memories for these words. Each time the individual produces a word, other words with motor memories containing similar sounds will be evoked to an extent, thereby producing neural links. With time and speech output, speech sounds in untreated words will be updated, as new, therapy-induced articulatory gestures create a pull away from gestures previously produced atypically.

While the findings of this study appear to provide some support for usage-based phonology theory, study findings can be re-interpreted using schema theory. Schema theory predicts a high volume of production trials will result in improved speech sound production. This is especially predicted given the "knowledge of performance" provided by EPG visual feedback (see 3.3.2). Further, gradient speech change may be due to progressive refinement of recall and recognition schema (see 3.3.2.1 for details on recall and recognition schema). However, as discussed in 3.3.2.5, schema theory has some limitations. As it exists, Schema theory is very broad and it does not provide specific details on the structures and mechanisms for motor speech learning. Unanswered questions include, for example, what constitutes GMP and schemata? Do GMP involve words, parts of words, or individual phonemes? If GMP consist of words, are lexical stress patterns a feature of GMP, or are they controlled by schemata? How do GMP come about initially? Are these learnt, or are they in part innate? What are the causes of SSDs? It would appear more specific details relating to speech are needed.

8.3.7 Support for the theoretical model (Research question number three): Summary and conclusions

Participants' positive response to the usage-based EPG therapy technique (in terms of positive speech sound change) provides some general support for the theoretical model. The usage-based model predicts a large number of production trials are needed to produce generalisation of target sounds to everyday connected speech. In this study, participants received a high dose of production trials per session (mean = 250) and a high cumulative dosage (mean = 5,958). The model further predicts generalisation to untreated words and continuous connected speech occurs through sub-neural networks: production of target sounds in treated words arouses target sounds in untreated words and, with time and practice, generalisation with untreated words/units occurs. In this study, production of target sounds in treated words resulted in generalisation to untreated words, and to target sounds in continuous connected speech. The usage-based model predicts a word frequency effect, with target speech sounds in high frequency words being more resistant to updating, than those in low frequency words. In this study, a frequency effect was shown in connected speech during the acquisition and lexical generalisation phases of therapy. The model anticipates gradient speech change will occur over time. In this study, articulatory gradient change in tongue-palate contact was shown on EPG analysis. The model further predicts speech change is possible in adults, using the same mechanism for phonological change as children. In this study, clinically meaningful improvement with speech occurred for all three adult participants. Thus, the answer to the research question number three - **do improvements with accuracy of targeted phonemes following intervention provide support for the usage-based theoretical model in general, and more specifically in terms of: gradient speech change is shown; and a word frequency effect is shown?** - is **yes**. However, more research is needed to investigate if a focus on high frequency words in the later stages of therapy expedites generalisation. Further, the findings of this study can be re-interpreted in terms of schema theory. Schema therapy predicts improvement with target sounds given a large number of production trials. Further, gradient speech change could be attributed to the successive refinement of schemata as motor learning occurs.

8.4 EPG: Limitations of this biofeedback tool

The EPG biofeedback used in this study mostly occurred in the early stages of therapy, and as discussed in 8.1, change with lingual speech targets occurred after only a few sessions for 5/6 participants. Previously study participants had received much standard therapy, with no change to lingual speech targets. As such, EPG biofeedback appeared crucial in the acquisition phase of therapy. However, this visual biofeedback tool does have some limitations requiring discussion.

Not all patients put forward for the study were able to participate in, and potentially benefit from, this type of treatment. As shown in the flowchart for patients assessed, excluded, treated and followed-up (see Figure 6), 3/15 patients assessed for eligibility (i.e. 20%) were excluded because they could not tolerate having a training plate in the mouth. To date, no study has reported on percentage of patients unable to tolerate a plate in the mouth and this large percentage of 20% is note-worthy. In addition, two patients (13%) were excluded as their dentition was not favourable for having a plate within the time-frame of the study (one patient's dentition was unstable and the other was due for orthodontic treatment). These individuals were offered EPG at a later time. However, for these patients, this meant delay in receiving any potential benefits from this type of treatment, highlighting the reliance on a custom-made palate for this type of treatment.

EPG therapy has some other factors that make it less accessible. EPG computer systems are expensive, though this is a one-off cost. EPG palates need to be made by specialist dental technicians, usually off site. These are time-consuming to make and are consequently expensive to buy (£450 - £530 in 2021). Since EPG is a low incidence therapy, historically only a few dental laboratories have made EPG palates and this continues to be the case. The technique is therefore vulnerable to laboratories ceasing to do the work because of low demand or being unable to produce palates at an affordable price. Further, if patient's teeth move, even slightly, the EPG palate will no longer fit and another will need to be made if treatment is not complete. Fortunately, as discussed above, speech sound acquisition with EPG visual feedback is often rapid, so EPG palates may only be needed for a short time.

Ultrasound is another type of instrumental technique and has been presented as an alternative to EPG (Cleland et al., 2020; Cleland & Preston, 2021; Lloyd et al., 1918). Ultrasound provides information on the location and movement of the tongue. To do this an ultrasound probe is placed under the chin. This probe emits and pulses very high frequency sound waves. When a pulse reaches a tissue boundary, a proportion of the wave energy is reflected back to the probe. In this way a real time image is produced which is displayed on a computer screen (Cleland & Preston, 2021). Ultrasound has a number of advantages over EPG (Cleland, 2021). It is not dependent on dentition. It does not require an expensive custom-made plate. Individuals who cannot tolerate a plate in the mouth are not excluded. Ultrasound assessment can be carried out immediately. Ultrasound provides better images of the back of the tongue and gives information on what part of the tongue is making contact for lingual speech sounds (e.g. tongue tip, tongue blade etc.). However, EPG does have some advantages over ultrasound. At present ultrasound systems include few quantitative assessment methods (Cleland & Preston, 2021). Current ultrasound systems also provide less easily interpretable information on tongue-palate contact in the coronal plane (Cleland, 2021). Consequently, ultrasound may be less useful for assessing and treating lateral and palatal production of lingual sibilants (i.e. /s/, /z/, /ʃ/, /ʒ/, /tʃ/, /dʒ/). As identified in Chapter Two, lateral and palatal cleft characteristics are the speech features mostly commonly seen with persistent SSD secondary to cleft (CRANE database).

8.5 EPG therapy: Patient selection

As identified in Chapter Two, previous research shows that some patients benefit more than others from EPG therapy (Gibbon et al., 2001; Gibbon & Paterson, 2006). In addition, some patients show no response to this instrumental therapy tool (Gibbon et al., 2001). Consequently, identification of which patients do best with EPG, and other visual biofeedback techniques in general, has been identified as an important question for research (Cleland & Preston, 2021; Lee et al., 2009). A conundrum of patient selection for EPG therapy is that it is not until the patient has received their expensive, custom-made EPG palate that the SLT can ascertain if the patient is stimulable for problem articulatory gestures, and if the individual can benefit from the feedback provided by palatograms as therapy proceeds. This is in contrast to standard therapy, where stimulability testing can occur before therapy commences.

Over the years, different clinicians and writers (Carter & Edwards, 2004; Gibbon et al., 1998; Gibbon & Wood, 2010; Hardcastle et al., 1991) have attempted to ascertain critical factors for success. As discussed in Chapter Two, Hardcastle et al. (1991) identify patient-related factors which, from the authors' clinical experience, appear to influence success / progress with EPG therapy. These include:

- Age and cognitive ability of the patient, with increasing age and more advanced cognitive ability resulting in better outcomes;
- Severity of the patient's SSD, with more severe SSD affecting a large number of sound classes requiring longer treatment;
- Awareness and motivation, with patients with higher levels of awareness and motivation resulting in more rapid progress;
- Concomitant difficulties, with patients with additional problems, such as profound hearing loss, significant VPI, and significantly impaired dentition progressing more slowly and perhaps being unable to achieve "normal" speech.

Given the small number of participants in this study, findings regarding patient selection cannot be generalised. However, since patient selection has been identified as a key area for research, the experience of this study may add to the experience of previous studies, and is worthy of some cautious discussion.

In this study, patients with severe cognitive disabilities were excluded. In this consecutive series of patients, this resulted in one patient being excluded. Two of the participants, Olivia and Emma, had additional difficulties requiring extra support at school. These additional difficulties may have slowed progress for Olivia, though the amount of therapy she received was similar to other participants. Additional difficulties did not appear to slow Emma's progress and she achieved amongst the best outcomes for this cohort of patients.

Emma presented with the most severe SSD, with difficulties involving a large number of sound classes. However, she did not require notably longer treatment than the other participants. Grace, with the mildest SSD, in terms of number of speech sounds produced in error, took the second longest amount of time to complete therapy.

As therapy progressed, it became apparent that Victoria had significant residual VPI. Prior to EPG therapy, Victoria's speech was characterised by use of non-oral speech sounds to replace sibilant sounds. When instructed to produce these sibilant sounds orally, Victoria struggled to build up oral pressure and significant audible nasal emission accompanied on-target production of these oral sibilants. This significant VPI did appear to limit her progress with functional generalisation. In addition, 3/5 of the remaining participants (George, Emma, Grace) showed increased signs of VPI (increased hypernasality and nasal turbulence) over the duration of therapy, though to a much lesser extent. It should be noted that EPG therapy is not expected to improve resonance or nasal airflow errors. However, this tendency for increased signs of VPI over the course of EPG therapy has also been observed in the researcher's non-study clinical experience. Increases with VPI may have structural basis and be due to general growth, i.e. with general growth over time, the soft palate does not grow in proportion to growth with other tissues, perhaps due to scarring associated with cleft palate repair. However, the fact that increased signs of VPI occurred rapidly with therapy for George, Emma and Grace points to the possibility of another factors at play. It may be that a degree of mild VPI had always been present in these three participants and these participants' lateral and palatal cleft speech characteristics occurred in response to this mild VPI, i.e. lateral and palatal production can be triggered when a patient has more difficulty building up oral pressure to produce high pressure sibilant sounds. Previously, such anterior cleft speech features have been attributed to atypical dentition and malocclusion, rather than VPI (Albery & Grunwell, 1993). Further, it may be that this lateral and palatal production of sibilant sounds serves to hide a mild degree of VPI. Following intervention, Grace and George's turbulence was most evident on typically produced sibilant sounds, perhaps giving support to this suggestion. In Emma's case, nasal turbulence was evident on all oral pressure consonants.

Thus, in this small cohort of patients, age, and severity of patient SSD, did not appear to be important factors in influencing success/progress with therapy. In this study the concomitant difficulty of significant VPI did appear to influence success with therapy.

8.6 Strengths and limitations of the study and implications for further research

This study had a number of strengths. The study adopted a strong within-participant experimental design. The study was evaluated using the PEDro scale (Verhagen et al., 1998). The PEDro scale was developed to give an indication of the quality of study designs, including group and within-participant designs (Verhagen et al., 1998). The authors of this scale acknowledge that it will not be possible to satisfy all scale items for some research designs and some areas of therapy. Using the PEDro scale, the design used in this research achieved a score of 5/11. Points were given for: specification of eligibility criteria; blinding of assessors; outcome measures obtained for more than 85% of the participants; all participants received the treatment condition; and size of treatment effect and measures of variability given for at least one key outcome. It was not possible to satisfy the remaining scale items due to the nature of the study: participants were not randomly allocated to groups, allocation was not concealed, participants and the treating SLT could not be blinded, and between-group statistical comparisons could not be made.

Thus, this study included a number of aspects deemed important for within-participant studies (Tate et al., 2013) including (and as discussed further below):

- Demonstration of treatment effect;
- Target behaviours defined and regularly measured in detail, including generalisation targets;
- Blinding of assessors;
- Report of inter- and intra-reliability.

The ABA multiple baseline design used in this within-participant study is viewed as amongst the strongest design in terms of demonstration of treatment effect (Ebbels, 2017; Tate et al., 2013). In this study, three baseline assessments showed largely unchanging percentage of targets correct prior to intervention for all participants. Each speech target for each participant was treated consecutively. Change in percentage of target correct occurred when each target became the focus of therapy. The exceptions were Grace, where accurate production of /s/ generalised to /z/ production, and Emma, where some change in untreated speech sound production occurred in pseudo-words on EPG assessment. Consequently, for 4/6 participants, one can be more confident that the change seen in participant's percentage of targets correct could be directly attributable to intervention. Further, the theoretic model can explain the across sound class generalisation shown with Grace and Emma, that is, across-sound generalisation occurred through the mechanism of sub-neural activation of similar sounds (Menn et al., 2013).

However, within-participant study designs do have significant limitations. While the within-participant study design allows for in-depth examination of participants' difficulties, together with a detailed analysis of response to intervention, and is thus appropriate for examination of the novel intervention, findings cannot be freely generalised to wider populations (Howard et al., 2015; Pring, 2005). However, this study's use of the effect size statistic may permit a degree of meta-analysis across other within-participant studies.

A further strength of this study was that perceptual speech outcomes were completed by two independent SLTs blind to test time point, and inter- and intra-agreement measures are given. Previous EPG intervention studies have typically not included such independent analysis and agreement measures. As well as EPG and perceptual speech outcomes, this study also reported on holistic outcomes, including speech understandability, speech acceptability and quality of life. Previous EPG studies with this client group have rarely reported such holistic outcomes, despite growing recognition of the importance of such outcomes (Howard & Lohmander, 2011; van Eeden et al., 2019).

Another strength of this research is that intervention included the entire duration of therapy, i.e. participants were treated until lingual speech sounds were accurate in continuous, connected speech in all speech settings, or until no further progress was being achieved. In wider cleft palate speech intervention, and the wider SSD literature, few studies have reported on the total duration of therapy from assessment to discharge (Sugden et al., 2018). A further strength of this study, not included in other cleft EPG intervention, is that it includes a consecutive series of patients put forward for EPG therapy and inclusion and exclusion criteria are clearly reported.

To date, much SSD intervention research has been carried out in university settings, using treatment regimes that are more difficult to replicate in clinical settings (Sugden et al., 2018). The intervention delivered in this study occurred in a clinical setting, with all the constraints of real-life clinical management. As such, findings may be more relevant to clinical cleft caseloads in the UK. A further strength of the study was the intervention's strong theoretical basis. This intervention was driven by a single causal model of phonological development. A drawback of atheoretical treatments (e.g. Van Riper, 1963), or therapies underpinned with more than one theory (e.g. Baker & Williams, 2010; Bowen, 2010) is that reasons for therapy success/failure may be more difficult to disentangle.

In addition to limitations with the research design (in terms of generalisation of findings), this study has a number of other limitations which need consideration when appraising findings. This was a single centre study, with all treatment carried out by a single SLT. This SLT has over 20-years of experience working with this client group and over 10-years of experience with use of EPG as a treatment technique. Future research needs to involve other centres and other SLTs to establish if findings can be replicated. In this study, a well detailed, theoretically motivated, therapeutic technique was delivered. However, the study design would have been strengthened had some sessions been recorded, and these sessions reviewed by an independent person to check for treatment fidelity. A key component of the therapeutic approach was teaching connected speech patterns. However, assessment of connected speech patterns was made through subjective impression (researcher listening to a connected speech sample). Further research is needed to develop a valid and reliable method to assess connected speech patterns.

While improvements with participants' speech occurred following usage-based EPG intervention, thereby providing some support for the theoretical model, a number of uncertainties exist with the technique. Whether or not this technique is more efficacious than other therapy techniques utilising EPG requires further examination. Further research could directly compare the usage-based EPG technique with more standard EPG therapy. As identified in Chapter Two, previous EPG intervention research with cleft palate speech has been composed of traditional Van Riper articulation therapy (Van Riper & Emerick, 1990) and EPG visual biofeedback (Lohmander et al., 2010; Stokes et al., 1995). More recently, this standard approach has been modified by application of motor learning theory (Cleland & Preston, 2021; Gibbon & Wood, 2010; Gibbon et al., 2001). In particular, schema theory has directed such modifications (Maas et al., 2008). Schema theory modifications include requirement of some error in learning, and manipulation of practice schedules and feedback mechanisms (Cleland & Preston, 2021; Preston & Leece, 2021). However, to date, no cleft EPG intervention studies have adopted such motor learning theory modifications. Further EPG intervention research could compare a usage-based approach with a schema motor learning theory approach.

Participants in this study generally acquired target phonemes relatively quickly with EPG feedback, compared to standard articulation therapy (Albery & Enderby, 1984; Gibbon et al., 2001; Van Demark & Hardin, 1986), and this acquisition showed rapid generalisation to untreated words. However, as discussed, functional generalisation, with use of target phonemes in connected speech in all speech settings, took an average of 11 sessions (in addition to sessions taken for lexical generalisation). Further refinement of the technique may be possible to accelerate functional generalisation. In this cohort of patients, generalisation, in part, related to the amount of practice done at home: those participants who carried out very regular practice at home and were supported by family to use target speech sounds in their talking made the most rapid progress with functional generalisation. This finding fits with the theoretical model underpinning therapy. The key mechanism for speech change is high volume speech use, ideally in a range of communicative settings (Bybee, 2001). As well as clearly laying out the requirement for regular home practice, and discussing when and where this will be carried out, additional online, at home, practice sessions could be considered. The recent COVID-19 pandemic has made telemedicine more accessible and widespread and permits changes to service delivery. In addition to weekly face to face therapy, an additional 20-minute practice session with the SLT or SLT assistant at home or at school may increase speed of functional generalisation.

As well as high volume practice, word frequency was posited as important for acquisition and generalisation. Given the design of the current study, the importance of this variable could not be definitively established. As previously discussed, pure volume of production, rather than manipulation of word frequency, may have been what was key to the generalisation shown in this study. Other research examining word frequency provides some support for use of high frequency words in boosting speech sound learning (Gierut & Morrisette, 2012; Gierut, Morrisette, & Hust Champion, 1999; Morrisette & Gierut, 2002), as discussed in section 8.2.1. Further research could manipulate word frequency by comparing use of high frequency word targets with use of low frequency word targets in the generalisation phases of the usage-based treatment approach.

Another lexical variable used in SSD intervention studies is neighbourhood density (Gierut & Morrisette, 2012). Neighbourhood density refers to the number of words that are similar - by one sound - to a given word (Storkel, 2008). For example, neighbours of “seat” include “beat”, “sit”, “seam”, “see”, “sleet”. Words vary in terms of the number of related words; some have high neighbourhood density, with a large number of related words, while others have low neighbourhood density. Gierut and Morrisette (2012) present findings from a within-participant study showing that high neighbourhood density treatment words promoted system-wide change in pre-school children with developmental SSD. Gierut and Morrisette suggest that this effect occurs because high density words may be easier to hold in working memory, thus easing language processing demands and freeing the child to focus on speech sound learning. Applying usage-based theory, high density neighbourhood words will have sub-neural links to many words. Thus, selection of high density target words may serve to promote system-wide generalisation in the later stages of intervention. In the earlier stages of therapy lower density words may be easier to produce, as there is less competition from competing words in terms of retrieval. Goldinger and Van Summers (1989) provide some experimental support for this contention. These authors showed that typical adults produced low-density words more rapidly and accurately than high density words. Further research could test this prediction, through teaching low density words in the early stages of therapy, then high density target words in the later stages of usage-based EPG therapy.

Ultrasound has some advantages over EPG. These include: no dependence on dentition; no requirement for an expensive, custom-made plate; individuals who cannot tolerate a plate in the mouth are not excluded; assessment and treatment can be carried out immediately (Cleland, 2021; Cleland & Preston, 2021). The main limitation of ultrasound is that current ultrasound systems provide less easily interpretable information on tongue-palate contact in the coronal plane. Consequently lingual sibilant speech sounds are less easy to image (Cleland, 2021). Further research needs to establish whether existing ultrasound systems can produce the same kind of results as EPG in the treatment of lingual sibilant sounds, despite current imaging limitations. In addition, development of ultrasound systems to provide more images in the coronal plane may be possible.

In the present study, usage-based phonology was used as the theoretical underpinning for the treatment of this group of patients with persistent cleft palate speech. As discussed throughout this thesis, this connectionist psycholinguistic theory of phonology is in contrast to rule-based, generative linguistic theory which has heavily influenced most existing SSD intervention. The visual feedback provided by EPG was used in the early stages of therapy in this study. Usage-based theory motivated this and subsequent stages. This treatment technique could be used without any provision for visual biofeedback. As it currently exists, the elements of this approach, minus visual feedback, would include: high number of production trials; focus on words rather than single sounds; practice with low frequency words, then high frequency words; facilitation of phonetic gradience; and teaching connected speech patterns. Future research could examine this approach with individuals with SSD traditionally viewed as predominately articulatory in nature, such as cleft palate speech and children with functional articulation difficulties. If effective, research could go on to compare this usage-based therapy with traditional/motor learning articulation therapy.

Although this therapy approach focuses exclusively on production, the basis of this therapy approach is a comprehensive phonological theory. Usage-based phonology surmises that phonological learning and change comes about from phonetic and auditory experiences (Bybee, 2001, 2010; Menn et al., 2013). As such, usage-based therapy can be viewed as a type of “phonological therapy”, and theoretically could be used with all SSDs, as an alternative to existing phonological therapies underpinned by generative theory. Initial, explorative research could examine the usage-based approach, minus EPG, with children traditionally viewed as having “phonological” difficulties, i.e. children who are able to articulate phonemes in certain circumstances, such as production in isolation given instruction, but whose speech difficulties may be described in terms of phonological patterns (McLeod & Baker, 2017). Applying usage-based theory, the phonological patterns shown with these children are due to the operation of sub-network neural representations where, words containing a particular sound are mapped to other articulatory gestures. For example, in the case of a "stopping" pattern, /s/ and /z/ are mapped to [t] and [d] respectively in word production (Menn et al., 2013). As discussed in 8.3.6, the rationale for use of the usage-based technique with this group of patients is as follows: production practice of words containing target sounds produces neuromotor memories for these words. Each time the individual produces a word, other words with motor memories containing similar sounds will be evoked to an extent, thereby producing neural links. With time and speech output, speech sounds in untreated words will be updated, as new, therapy-induced articulatory gestures create a pull away from gestures previously produced atypically. In this way, therapy-induced articulatory gestures will diffuse through the lexicon.

8.7 Clinical and theoretical implications

This study supports other biofeedback intervention studies in suggesting visual biofeedback techniques are particularly helpful for acquisition of target speech sounds for participants with persistent SSD (Gibbon et al., 2001; Gibbon et al., 2003; Martin et al., 2007; Stokes et al., 1995). All participants in this study had previously been unable to produce target lingual speech sounds accurately, despite standard intervention. Five out of the six participants in this consecutive treatment series responded rapidly to the visual biofeedback provided by EPG, and acquired improved speech sound production after just a few sessions. The sixth participant also responded, but more time was needed (11 sessions before target sound acquisition). Thus, SLTs working with patients with persistent cleft palate speech may want to consider using EPG with patients where acquisition of lingual speech sounds has been <u>un</u>achievable with more standard therapy, such as articulation therapy (see below for discussion regarding functional generalisation). This consideration should be done with care, given this type of therapy's existing evidence base (i.e. small number of within-participant studies and small number of small group studies). Also, since this is a specialist technique, requiring specialist therapy skills, SLTs new to the technique should undergo training before delivering this type of therapy. This could be on-the-job training, under the guidance of a therapist experienced in EPG therapy.

This study also supports previous writers who suggest non-EPG activities are required for functional generalisation, once EPG-related acquisition is achieved (Cleland & Preston, 2021). That is, provision of EPG feedback, on its own, is insufficient for this hard-to-treat group of patients. In this study, it was suggested other elements of the usage-based therapy approach were important for functional generalisation. Consequently, the therapy technique used in this study is best described as "usage-based therapy, with use of EPG", rather than "EPG therapy" i.e. EPG was just as one element of the entire usage-based treatment technique delivered in this study. In this study, usage-based teaching episodes identified as important for functional generalisation included: a). word production, rather than single sounds; b). a high amount of drill-type practice, particularly with words occurring with high frequency in the English language; c) facilitation of gradient phonetic change and; d) facilitation of connected speech processes. The clinical application of usage-based phonology is in its very early infancy, and further research is needed. Potential next steps were outlined in the previous section. However, SLTs working with this group of very hard-to-treat patients, may wish to consider the ideas presented in this thesis, when reflecting on their own experience, and when considering treatment for this group of patients.

Three adults participated in this study. As discussed in 8.3.5, previously, the prevailing view has been that adults with persistent cleft palate speech do not respond to therapy (McWilliams, 1984; Noordhoff et al., 1987). Findings from this study go against this view. All three adults showed significant clinical improvement, given therapy. This study adds to previous reports of treatment success with adults with persistent SSD (Martin et al., 2007; Whitehill et al., 1996). Thus, SLTs working with this client group should not necessarily rule out this type of therapy with adults with cleft palate speech. As discussed earlier, the usage-based model sets out a theoretical basis for speech change in adults; intervention with this adult group can further test this theoretical basis.

This study supports other writers who suggest the objective data provided by EPG assessment is an valuable complement to subjective phonetic transcription (Cleland & Preston, 2021; Lee, 2021). In this study, independent SLTs' phonetic transcription showed agreement levels of between 89.75 – 97.07%. Where disagreements occurred, EPG assessment could provide details on exact tongue-palate contact for lingual speech sounds. Such details were important for precision with assessment, and also for directing treatment, such as providing exact phonetic cues. Further, objective data provided by EPG assessment arguably increases confidence with interpretation of study findings. Consequently, SLTs should consider using such instrumental measures for both clinical and research purposes.

9 CONCLUSIONS

To our knowledge, this is the first SSD intervention study for individuals with cleft palate speech that has used usage-based phonology theory as its theoretical underpinning. The motivation for use of this theory was development of an EPG therapy technique that would better promote generalisation of target speech sounds for individuals with persistent cleft palate speech. A further motivation was development of a technique that would be effective for adults. In this study, three school-aged children and three adults with persistent SSDs secondary to cleft palate responded to the novel EPG therapeutic technique, as measured by a within-participant multiple baseline ABA experimental design. Participants were seen for weekly therapy at a regional hospital, and were seen until lingual speech sound production was typical, or no further progress was shown over eight sessions. Improvement in production of speech sound targets in treated and untreated words occurred for all participants. Further, all participants showed generalisation of target speech sounds to continuous, connected speech, though to varying degrees (mean usage in connected speech = 90.66%). Individual participants' response to this usage-based EPG treatment is encouraging and justifies further research involving use of this treatment approach with individuals with persistent cleft palate speech. Directions for further research include: replication with other SLTs/cleft units; further refinement of the novel technique; and whether or not this technique is more effective than an EPG therapy technique based on motor learning theory. Finally, as a single, comprehensive, causal model of phonological development, usage-based phonology theory may show promise for a greater understanding of, and treatment for, child SSDs more widely.

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11 APPENDICES

11.1 Appendix A: Patient information sheets

11.1.1 Adults and children aged 16 – 17 years

<Letter Headed Paper>

Information about the Research

Study Title:

Treatment of cleft palate speech using a modified electropalatography (EPG) therapeutic technique: a case series

Invitation:

We would like to invite you to take part in a research study at the Regional Cleft Unit, Royal Manchester Children's Hospital. This study is being carried out by Kathryn Patrick, Senior Specialist Speech and Language Therapist (who has many years' experience with treating speech difficulties associated with cleft palate) in partial fulfilment of a doctoral research programme of study through the University of Sheffield. Before you decide if you would like to participate, you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Please do not hesitate to ask questions if anything you read is not clear or if you would like more information.

What is the study about?

The purpose of this study is to examine the use of electropalatography (EPG) in treating speech difficulties with people born with cleft palate. EPG is a computer-based system for treating speech sound difficulties involving the tongue. A person using this system has a specially made plate that fits to the roof of his/her mouth as shown in the pictures below.

Picture of an EPG Plate

Therapist using the EPG system with EPG plate in her mouth

This plate contains sensors that detect when the tongue makes contact with the roof of the mouth during speech. The sensors are connected to a computer screen. In this way, tongue / roof of mouth contact can be seen visually on the computer screen. An example of the visual feedback provided by EPG is shown on the picture on the next page.

Picture on the left of the screen showing EPG tongue placement for the "s" speech sound, picture on the right of the screen showing EPG tongue placement for the "t" speech sound

The speech and language therapist uses the EPG system to:

- assess in what way a person is producing a speech sound incorrectly
- help the individual achieve correct placement for tongue / roof of mouth speech sounds produced in error

EPG is typically used with school-aged children and adults where other forms of treatment have been unsuccessful. The usefulness of EPG has been examined and reported in single cases and small group studies. Most individuals are able to change their speech production using EPG therapy. However, one problem identified by previous studies is the difficulty some individuals have in using newly acquired speech sounds in their everyday speech all the time. This study aims to examine patients' response to EPG therapy that has been modified in two ways. These modifications come from new thinking on how individuals learn to speak, and how speech changes in older children and adults. The two modifications to the EPG traditional technique are as follows:

- In regular EPG therapy patients first learn to produce speech sounds in isolation and in syllables. In the modified approach used in this study, less attention will be paid to production in single sounds and syllables. Instead the focus will be on production of problem speech sounds in words because it is thought that speech change may be easier and faster if therapy focuses on words rather than individual sounds.
- In regular EPG therapy less attention is given to use of speech sounds in connected speech. In the modified approach used in this study, participants will receive specific information on how speech sounds in words can change when they are used in sentences, known as connected speech patterns, and participants will practice using connected speech patterns during therapy. It is felt teaching of connected speech patterns may be important to help patients use new speech sounds in their talking in their daily lives.

Why have I been invited to participate?

You have been invited to participate in this study because you have speech difficulties as a result of your cleft palate. These difficulties have not responded to routine speech therapy over many months / years, so EPG therapy has been suggested. This study aims to recruit five to ten individuals aged 7 years and over. If you wish to participate, you will be involved in the study for 6 – 12 months. The entire study will run over a period of 3 - 4 years.

Do I have to take part?

No, it is up to you to decide if you want to participate in this study. We will describe the study and go through this information sheet. If you agree to take part, we will ask you to sign a consent form to show you have agreed to take part. You are free to withdraw from the study at any time, without giving a reason, and this will not affect the standard of care you receive by the Manchester Cleft Team.

What will happen if I agree to take part?

If you decide take part, you will be treated in the same way as any other person receiving specialist EPG speech therapy at the Royal Manchester Children's Hospital, including:

- You will have an initial appointment with the speech and language therapist. An assessment of your speech will be made at this time. This will include a video recording.
- You will have an appointment with one of our orthodontists. The orthodontist will examine your teeth to check if they are suitable to hold a plate. If your teeth are suitable, the orthodontist will take an impression of your upper mouth. This involves a putty-like substance being held to the roof of your mouth to get an impression of your upper teeth and hard palate. This impression is then sent to a dental lab to make a training plate. The training plate is to check that you are able to tolerate having a plate in your mouth for up to 50 minutes (for the duration of therapy appointments). Most people do not have any difficulties wearing these dental plates, and find them comfortable. A very small number find it difficult to wear these plates, can't tolerate having them in their mouth, and therefore are not suitable for this kind of therapy.
- You will then have an appointment to fit the training plate. You will then be asked to practice wearing this plate for short periods of time over a 2 – 3 week period.
- You will then have another appointment with the orthodontist. If you have got on well with the training plate, another impression will be made and this impression will be sent off to make up an EPG plate with sensors.
- The EPG plate will take 6 – 8 weeks to make. Once we have the plate you will be invited back to the hospital and the plate will be fitted. Your speech will be assessed again and therapy will commence. You will only need to wear the plate during therapy and when practicing speech at home.
- You will be invited to attend therapy appointments at the Royal Manchester Hospital every week.

- You will be expected to carry out speech practice at home 5 times / week for 10 -15 minutes duration using a portable EPG machine.
- Treatment will continue until your production of tongue speech sounds is normal, or no further speech gains are being achieved. The duration of therapy will depend on the individual, but might take up to 3 months in less complex cases and up to 10 months (including breaks in therapy) in individuals with very complex speech difficulties involving many speech sounds.

If you decide to take part in this study you will participate in the following **additional** activities:

- You will undergo additional assessments at the beginning, during and at completion of therapy, including:
    - 10 minute longer appointment on initial assessment
    - 10 minute longer appointments on each of the three subsequent appointments (appointments to prepare for the EPG plate)
    - 2 minutes of additional assessment at the end of each 50 minute therapy appointment
    - Up to 5 minutes of additional assessment at the end of every fifth session of therapy
    - 10 minutes of additional assessment at the end of therapy
    - 10 minutes of additional assessment 3 months following completion of therapy
- Your speech will be examined and monitored in more detail by the research speech and language therapist.
- You will receive therapy that is partially modified, compared with routine EPG therapy, as described above.
- Two other specialist speech and language therapist who work with the Cleft Lip and Palate Team will watch videos of your speech made prior to, during and following therapy and make an assessment of your speech. These videos are anonymous (i.e. will not be named using your name).

What are the possible benefits of taking part?

We hope this treatment will improve your speech. We also hope that the information we get from this study will help increase our understanding of, and help improve the treatment of, the small group of individuals who experience persistent speech difficulties due to cleft palate.

Will taking part in the study be kept confidential?

All information collected about you during the course of the research will be kept strictly confidential. Each person participating in the study will have an individual letter-number code, known only to the research team. This number/code replaces your name and will be used throughout the study to identify data relating to you. Use of number / letter codes rather than your name will ensure your anonymity. All assessment data relating to you, including video recordings, will be securely stored on the Central Manchester Foundation NHS Trust’s secure computer server using your letter-number code. The research speech and language therapist will have password access to this server. In addition, two speech and language therapists with the Manchester Cleft Team will have password access to this data for further assessment on your speech. Digital copies of this data will also be backed up to an external hard drive that is encrypted and password protected. This hard drive will be kept in a locked cupboard in the Cleft Office and the Royal Manchester Children's Hospital. Only the research speech and language therapist (Kathryn Patrick) and the lead speech and language therapist (Melanie Bowden) will have access to this data. This hard drive will be wiped on completion of the project. The stored data on the hospital computer server will be retained for 10 years following completion of the study.

Your treatment plans and progress will be documented in your speech and language therapy medical records which are stored securely in the cleft office at the Royal Manchester Children's Hospital.

What will happen if Iparticipate initially but then don't carry on with the study?

If you don't carry on with the study, you can withdraw from the study, but allow us to use the data we have already collected

OR

You can withdraw from the study and we will not use any data we have collected from you towards this study

What will happen to the results of this research study?

The results of this study will be made available to other speech and language therapists and related professionals through written reports and at professional conferences and meetings. You will not be identifiable in any report, publication or oral presentation unless you have given your specific consent for this. A written report will be available for you, should you wish to read this.

Further information / contact details

Further information on this study is available from Kathryn Patrick, Senior Specialist Speech and Language Therapist, 0161 701 9080; kathryn.patrick@cmft.nhs.uk.

Next steps

If you would like to be involved in this study, tell your speech and language therapist, or ring the research speech and language therapist, Kathryn Patrick, on 0161 7019080.

Thank you for taking the time to read this.

Approvals and monitoring

This study has received ethical approval from xxxx . It has also received approval from the NHS Health Research Authority and the Central Manchester University Foundation NHS Trust (CMFT). From time to time CMFT may look at some of the data collected for this research to ensure the study is being managed properly.

Any problems:

If you have a concern about any aspect of this study you should speak to the researcher, Kathryn Patrick, in the first instance, and she will do her best to answer your questions. If you remain unhappy you can speak to the lead speech and language therapist, Melanie Bowden (0161 701 9080; melanie.bowden@cmft.nhs.uk; Cleft Team, Outpatients, Royal Manchester Children's Hospital, Oxford Road, Manchester M13 9WL) or the project supervisor, Dr Silke Fricke, at the University of Sheffield (0114 2222 419; s.fricke@sheffield.ac.uk; Department of Human Communication Sciences, University of Sheffield, 362 Mushroom Lane, Sheffield, S10 2TS) . If you remain dissatisfied, you can complain through the hospital's Patients Advice and Liaison Service (PALS) (0161 276 8686; pals@cmft.nhs,uk) or through the University of Sheffield complaints procedure (0114 222 1100; University Registrar & Secretary, University of Sheffield, Firth Court, Western Bank, Sheffield S10 2TN).

11.1.2 Parents/guardians of children

<Letter Headed Paper>

Information about the Research

Study Title:

Treatment of cleft palate speech using a modified electropalatography (EPG) therapeutic technique: a case series

Invitation:

We would like to invite you and your child to take part in a research study at the Regional Cleft Unit, Royal Manchester Children's Hospital. This study is being carried out by Kathryn Patrick, Senior Specialist Speech and Language Therapist (who has many years' experience with treating speech difficulties associated with cleft palate) in partial fulfilment of a doctoral research programme of study through the University of Sheffield. Before you decide if you would like your child to participate, you need to understand why the research is being done and what it would involve for you and your child. Please take time to read the following information carefully. Please do not hesitate to ask questions if anything you read is not clear or if you would like more information.

What is the study about?

The purpose of this study is to examine the use of electropalatography (EPG) in treating speech difficulties with people born with cleft palate. EPG is a computer-based system for treating speech sound difficulties involving the tongue. A person using this system has a specially made plate that fits to the roof of his/her mouth as shown in the pictures below.

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The speech and language therapist uses the EPG system to:

- assess in what way a person is producing a speech sound incorrectly
- help the individual achieve correct placement for speech sounds produced in error

EPG is typically used with school-aged children and adults where other forms of treatment have been unsuccessful. The usefulness of EPG has been examined and reported in single cases and small group studies. Most individuals are able to change their speech production using EPG therapy. However, one problem identified by previous studies is the difficulty some individuals have in using newly acquired speech sounds in their everyday speech all the time. This study aims to examine patients' response to EPG therapy that has been modified in two ways. These modifications come from new thinking on how individuals learn to speak, and how speech changes in older children and adults. The two modifications to the EPG traditional technique are as follows:

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- In regular EPG therapy less attention is given to use of speech sounds in connected speech. In the modified approach used in this study, participants will receive specific information on how speech sounds in words can change when they are used in sentences, known as connected speech patterns, and participants will practice using connected speech patterns during therapy. It is felt teaching of connected speech patterns may be important to help patients use new speech sounds in their talking in their daily lives.

Why has my child been invited to participate?

Your child has been invited to participate in this study because she/he has speech difficulties as a result of his/her cleft palate. These difficulties have not responded to routine speech therapy over many months / years, so EPG therapy has been suggested. This study aims to recruit five to ten individuals aged 7 years and over. If you wish to participate, your child will be involved in the study for 6 – 12 months. The entire study will run over a period of 3 - 4 years.

Does my child have to take part?

No, it is up to you and your child to decide if you want to participate in this study. We will describe the study and go through this information sheet. If you agree to take part, we will ask you to sign a consent form to show you have agreed for your child to take part. We will also give your child written and verbal information and he/she will be asked to sign a form saying he/she is happy to participate. You are free to withdraw your child from the study at any time, without giving a reason, and this will not affect the standard of care you receive by the Manchester Cleft Team.

What will happen to my child if we agree to take part?

If you decide take part, your child will be treated in the same way as any other person receiving specialist EPG speech therapy at the Royal Manchester Children's Hospital, including:

- Your child will attend an initial appointment with the speech and language therapist. An assessment of your child's speech will be made at this time. This will include a video recording.
- Your child will have an appointment with one of our orthodontists. The orthodontist will examine your child's teeth to check that his/her teeth are suitable to hold a plate. If your child has teeth emerging, or is likely to lose teeth in the coming months, it might not be possible to fit a plate at that particular time. If your child's teeth are suitable, the orthodontist will take an impression of your child's upper mouth. This involves a putty-like substance being held to the roof of the mouth to get an impression of your child's upper teeth and hard palate. This impression is then sent to a dental lab to make a training plate. The training plate is to check that your child is able to tolerate having a plate in his/her mouth for up to 50 minutes (for the duration of therapy appointments). Most children do not have any difficulties wearing these dental plates, and find them comfortable. A very small number find it difficult to wear these plates, can't tolerate having them in their mouth, and therefore are not suitable for this kind of therapy.
- Your child will have an appointment to fit the training plate. Your child will then be asked to practice wearing this plate for short periods of time over a 2 – 3 week period.
- Your child will attend a second appointment with the orthodontist. If your child has got on well with the training plate, another impression will be made and this impression will be sent off to make up an EPG plate with sensors. The EPG plate will take 6 – 8 weeks to make. Once we have the plate you and your child will be invited back to the hospital and the plate will be fitted. Your child's speech will be assessed again and therapy will commence. Your child will only need to wear the plate during therapy and when practicing speech at home.
- Your child will attend therapy appointments at the Royal Manchester Hospital every week.
- Your child will be expected to carry out speech practice at home 5 times / week for 10 -15 minutes duration using a portable EPG machine.
- Treatment will continue until your child's production of tongue speech sounds is normal, or no further speech gains are being achieved. The duration of therapy will depend on the individual, but might take up to 3 months in less complex cases and up to 10 months

(including breaks in therapy) in individuals with very complex speech difficulties involving many speech sounds.

If your child takes part in this study they will participate in the following **additional** activities:

- Your child will undergo additional assessments at the beginning, during and at completion of therapy, including:
  - 10 minute longer appointment on initial assessment
  - 10 minute longer appointments on each of the three subsequent appointments (appointments to prepare for the EPG plate)
  - 2 minutes of additional assessment at the end of each 50 minute therapy appointment
  - Up to 5 minutes of additional assessment at the end of every fifth session of therapy
  - 10 minutes of additional assessment at the end of therapy
  - 10 minutes of additional assessment 3 months following completion of therapy
- Your child's speech will be examined and monitored in more detail by the research speech and language therapist.
- Your child will receive therapy that is partially modified, compared with routine EPG therapy, as described above.
- Two other specialist speech and language therapist who work with the Cleft Lip and Palate Team will watch videos of your child's speech made prior to, during and following therapy and make an assessment of your child's speech. These videos are anonymous (i.e. will not be named using your child's name).

What are the possible benefits of taking part?

We hope this treatment will improve your child's speech. We also hope that the information we get from this study will help increase our understanding of, and help improve the treatment of, the small group of individuals who experience persistent speech difficulties due to cleft palate.

Will taking part in the study be kept confidential?

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Your child's treatment plans and progress will be documented in your child's speech and language therapy medical records which are stored securely in the cleft office at the Royal Manchester Children's Hospital.

What will happen if my child participates initially but then doesn't carry on with the study?

If your child doesn't carry on with the study, you can withdraw from the study, but allow us to use the data we have already collected

OR

You can withdraw from the study and we will not use any data we have collected from your child towards this study

What will happen to the results of this research study?

The results of this study will be made available to other speech and language therapists and related professionals through written reports and at professional conferences and meetings. Your child will not be identifiable in any report, publication or oral presentation unless you have given your specific consent for this. A written report will be available for you, should you wish to read this.

Further information / contact details

Further information on this study is available from Kathryn Patrick, Senior Specialist Speech and Language Therapist, 0161 701 9080; kathryn.patrick@cmft.nhs.uk.

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If you would like your child to be involved in this study, tell your speech and language therapist, or ring the research speech and language therapist, Kathryn Patrick, on 0161 7019080.

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If you have a concern about any aspect of this study you should speak to the researcher, Kathryn Patrick, in the first instance, and she will do her best to answer your questions. If you remain unhappy you can speak to the lead speech and language therapist, Melanie Bowden (0161 701 9080; melanie.bowden@cmft.nhs.uk; Cleft Team, Outpatients, Royal Manchester Children's Hospital, Oxford Road, Manchester M13 9WL) or the project supervisor, Dr Silke Fricke, at the University of Sheffield (0114 2222 419; s.fricke@sheffield.ac.uk; Department of Human Communication Sciences, University of Sheffield, 362 Mushroom Lane, Sheffield, S10 2TS) . If you remain dissatisfied, you can complain through the hospital's Patients Advice and Liaison Service (PALS) (0161 276 8686; pals@cmft.nhs,uk) or through the University of Sheffield complaints procedure (0114 222 1100; University Registrar & Secretary, University of Sheffield, Firth Court, Western Bank, Sheffield S10 2TN).

11.1.3 Children aged 7 – 11 years

<Letter Headed Paper>

Information for young people

Study: How can young people's speech be improved?

We are asking whether you and your parents would like to take part in a research study.

Before you decide if you would like to join in, it is really important that you know about the study, why it is being done, and what it will mean if you take part. Please read this leaflet carefully with your parents. Also talk to your family, friends and your speech and language therapist if you want.

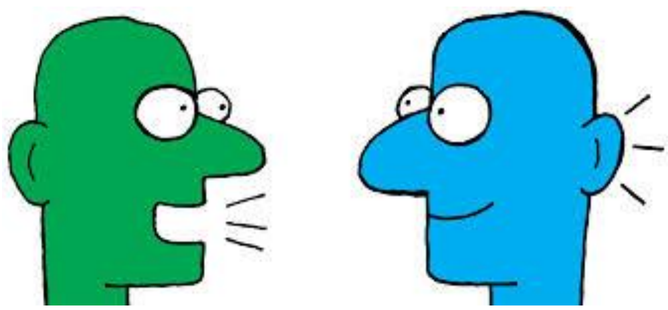
If something doesn't make sense or if you have more questions you can ask your parents to give us a call and we can discuss it with you and your parents.

Thank you for reading this.

What is research? Why is the project being done?

We do research to try and find answers to important questions.

When people are born with a cleft palate/lip (gap in the roof of the mouth/lip) they can have problems with saying some speech sounds. This research is being done to help find ways to improve children's speech.



Interesting facts about speech:

- Over 100 muscles in the lips, tongue, jaw, neck and chest are involved in the production of speech.
- Each spoken word or short sentence has its own pattern of muscular movement. All of the information needed for speech is stored in the brain
- Speech is thought to have appeared around 2.5 million years ago.
- Around 6 000 different languages are spoken in the world today. It is believed these came from one "mother" language.

Why have I been asked to take part?

You have been asked to take part because you have some speech problems due to your cleft palate / lip and you and your parents think your speech is different sounding and sometimes people have difficulty understanding your speech.

Do I have to take part?

No you don't. It is your choice whether you want to take part and you can always change your mind later on.

What will happen to me if I take part?

If you agree to take part we will make a special plate that fits to the roof of your mouth, called an EPG plate, as shown in the picture below.

Picture of an EPG Plate

This plate is connected to a computer screen. The computer screen gives you a picture of what your tongue is doing when you speak. Some of the pictures you get from EPG therapy are shown below. The picture on the left shows what your tongue should do for the "s" sound. The picture on the right shows what your tongue should do for the "t" sound.

EPG Pictures

Once you have your plate you will come and see the speech and language therapist every week. When you come to the hospital you will need to wear your plate and you will practice talking. During some of this speech practice you will play games. Some of the games you might play while practicing your speech are shown in the pictures below.

You will also need to practice your new speech sounds at home for 10-15 minutes most days.

You only need to wear your plate when you are practicing your speech. The plate might feel a bit strange to start, but it shouldn't be uncomfortable.

Will taking part in this research help me?

We hope the speech practice you do at the hospital and at home will mean that your speech improves and that other people will be able to understand your speech better – though we can't promise this for sure.

What will happen to the information gathered about me during this research?

During this study we will give you a "code". This code will be made up of letters and numbers, for example, you might be given the code 22B15. This code will be used instead of your name so that any information we get from you during the research is kept secret. From time to time video recordings will be made of your talking. These videos will be stored safely in the hospital. Only a small number of people involved in this research will be able to look at your videos.

We will let other people know about the findings from this research through written reports and at meetings. Video or audio recordings of your speech will not be used in these reports and meetings unless your parents have given us permission to do so.

What if there is a problem?

If there is a problem you can talk to your parents or your speech and language therapist. Your speech and language therapist will also check that everything is alright each time you see her.

What do I do if I don't want to take part in the research anymore?

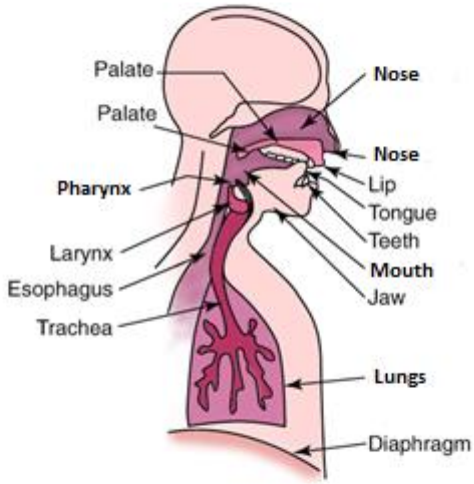
Just tell your parents and your speech and language therapist that you don't want to take part anymore. It is your choice.

Thank you for reading this

Further information / contact details

Further information on this study is available from Kathryn Patrick, Senior Specialist Speech and Language Therapist, 0161 701 9080; kathryn.patrick@cmft.nhs.uk.

The diagram below shows parts of the body that are used for talking. Can you find these words in the word search below?



Word search: the speech mechanism

P	G	U	E	X	W	O	E	Q	U	D	J	K	B	O
M	P	Y	K	D	I	A	P	H	R	A	M	Z	E	B
P	Q	W	M	V	W	D	S	Q	N	F	M	M	P	Q
U	L	N	E	R	Q	A	F	H	B	W	L	S	Y	X
V	H	B	O	J	Z	K	J	T	N	T	Y	U	H	P
E	P	X	V	S	X	X	D	U	E	E	P	G	Q	V
T	I	O	N	L	E	G	V	A	E	H	C	A	R	T
A	L	Z	L	Y	K	A	Y	O	Y	F	X	H	M	Q
L	M	A	L	V	R	V	K	E	X	L	H	P	Q	P
A	X	R	R	B	T	A	U	K	A	U	E	O	E	C
P	B	T	Z	Y	C	G	H	B	E	N	S	S	W	P
X	Z	G	V	K	N	X	Y	P	E	G	R	E	T	E
L	C	Q	M	O	L	X	H	I	C	S	T	O	R	I
V	R	D	T	V	P	I	X	P	I	W	E	T	F	O
M	V	V	U	D	O	N	G	T	E	E	T	H	F	C

PALATE
PHARYNX
LARYNX
OESOPHAGUS
TRACHEA
NOSE
LIP
TONGUE
TEETH
JAW
LUNGS
DIAPHRAM

11.1.4 Young people aged 12 – 15 years

<Letter Headed Paper>

Information for young people

Study: Treatment of speech problems using electropalatography (EPG) therapy

We are asking whether you and your parents would like to take part in a research study.

Before you decide if you would like to join in, it is important that you know about the study, why it is being done, and what it will mean if you take part. Please read this information sheet carefully with your parents. Also talk to your family, friends and your speech and language therapist if you want.

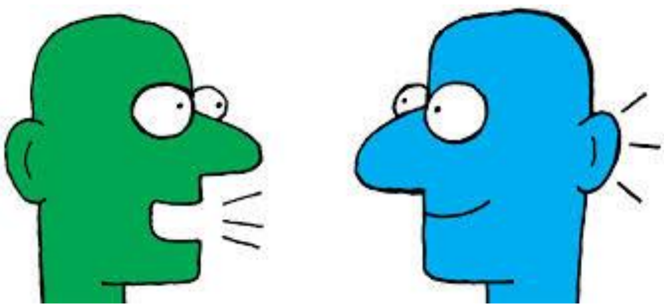
If something doesn't make sense to you or if you have more questions you can ask your parents to give us a call and we can discuss it with you and your parents.

Thank you for reading this.

What is research? Why is the project being done?

We do research to try and find answers to important questions.

When people are born with a cleft palate/lip (gap in the roof of the mouth/lip) they can have problems producing some speech sounds. About half of all people born with a cleft palate/lip will need help from a speech and language therapist at some point in their life. This research is being done to help find out ways to improve speech difficulties in people with repaired cleft palate/lip using a therapy technique called EPG therapy.



Why have I been asked to take part?

You have been asked to take part because you have some speech problems due to your cleft palate / lip and your parents have said that you would like your speech to be clearer.

Do I have to take part?

It is your choice whether you want to take part and you can always change your mind later on.

What is EPG therapy?

EPG therapy is an instrumental technique. It involves having an individually made plate that fits to the roof of the mouth.

An EPG plate contains sensors that detect tongue / roof of the mouth (hard palate) contact. A picture of an EPG plate is shown below (on the left). When the person wearing the plate speaks, a computer screen shows where the tongue touches the hard palate. Examples of the type of pictures produced by EPG therapy are shown in the picture below (on the right). Speech and language therapists use these pictures to help people who have difficulty with sounds involving the tongue, such as the sounds “t”, “d”, “n”, “s”, “z” and “sh”.

Picture of an EPG plate

Picture of visual feedback provided by EPG

How can EPG therapy help my speech?

The speech and language therapist will use the pictures produced by the EPG system to work out in what way you produce speech sounds differently. The therapist and you can then use the EPG pictures to get correct tongue positioning for your speech.

What will happen to me if I take part?

If you agree to take part we will first make you a “training” plate which doesn’t contain sensors. To do this our orthodontist will need to take an impression of your upper teeth and the roof of your mouth. We will ask you to wear this training plate for short periods during the day. Initially the plate may feel a little strange in your mouth, but you should get used to it. If you find wearing this training plate comfortable, we will make you an EPG plate with sensors. The EPG plate takes 6 – 8 weeks to make.

Once your EPG plate is made, you will need to come to the hospital for regular speech and language therapy. The EPG therapy you will have is slightly different to routine (i.e. non-study)

EPG therapy. The research EPG therapy focuses on words and sentences, rather than individual sounds. You will also need to practice your new speech at home for 10 - 15 minutes most days. You will only need to wear your plate when you attend for speech therapy and when you practice at home.

Your speech will also be videoed and assessed at intervals and you will be asked about the effect of your speech on your everyday life. Your speech will be videoed so that your talking can be assessed in detail by 3 speech and languages therapists.

Will taking part in this research help me?

We hope (but can't guarantee) the speech practice you do at the hospital and at home will mean that your speech improves and that other people will be able to understand your speech better. Speech and language therapists have been using EPG for a number of years and other people have been helped by this therapy technique. In the longer term, we hope this study may help improve the effectiveness of EPG therapy and help other people with speech sound difficulties achieve even better results in the future.

What will happen to the information gathered about me during this research?

During this study we will give you a "code". This code will be made up of letters and numbers, for example 22B15. This code will be used instead of your name and will be used to identify any information we gather about you. From time to time video recordings will be made of you talking. These videos will be stored safely on the hospital's computer system and on a portable hard drive (which will be kept in a locked cupboard in the hospital). Only a small number of people involved in this research will be able to look at these videos.

The findings from this study will be available to other people (such as other speech and language therapists working with people with cleft palate) through written reports and at meetings. Video and audio recordings of your speech will not be used in these reports and meetings unless your parents have given us specific permission to do so.

What if there is a problem?

If there is a problem you can talk to your parents or your speech and language therapist. Your speech and language therapist will also check that everything is OK each time you see her.

What do I do if I don't want to take part in the research anymore?

If you decide you no longer want to take part, just tell your parents and/or your speech and language therapist. It is your choice.

Thank you for reading this.

Further information / contact details

Further information on this study is available from Kathryn Patrick, Senior Specialist Speech and Language Therapist, 0161 701 9080; kathryn.patrick@cmft.nhs.uk.

11.2 Appendix B: Great Ormond Street Speech Assessment sentences

Mary came home early.

Bob is a baby boy.

The phone fell off the shelf.

Dave is driving a van.

Neil saw a robin in a nest

A ball is like a balloon.

Tim is putting a hat on.

Daddy mended a door.

I saw Sam sitting on a bus.

The zebra was at the zoo.

Shaun is washing a dirty dish.

Charlie's watching a football match.

John's got a magic badge.

The bell is ringing.

Karen is making a cake.

Gary's got a bag of lego.

Hannah hurt her hand.

This hand is cleaner than the other.

Stuart's hamster scrambled up his sleeve.

We were away all year.

Laura will wear a yellow welly.

11.3 Appendix C: EPG assessment

/ma/ x10 repetitions
/mi/ x10 repetitions
/pa/ x 10 repetitions
/pi/ x10 repetitions
/ba/ x 10 repetitions
/bi/ x 10 repetitions
/fa/ x 10 repetitions
/fi/ x 10 repetitions
/va/ x 10 repetitions
/vi/ x 10 repetitions
/na/ x 10 repetitions
/ni/ x 10 repetitions
/la/ x 10 repetitions
/li/ x 10 repetitions
/ta/ x 10 repetitions
/ti/ x 10 repetitions
/da/ x 10 repetitions
/di/ x 10 repetitions
/sa/ x 10 repetitions
/si/ x 10 repetitions
/za/ x 10 repetitions
/zi/ x 10 repetitions
/ʃa/ x 10 repetitions
/ʃi/ x 10 repetitions
/tʃa/ x 10 repetitions
/tʃi/ x 10 repetitions
/dʒa/ x 10 repetitions
/dʒi/ x 10 repetitions
/ka/ x 10 repetitions
/ki/ x 10 repetitions
/ga/ x 10 repetitions
/gi/ x 10 repetitions
/θa/ x 10 repetitions
/θi/ x 10 repetitions
/ða/ x 10 repetitions
/ði/ x 10 repetitions

Mary came home early.

Bob is a baby boy.

The phone fell off the shelf.

Dave is driving a van.

Neil saw a robin in a nest

A ball is like a balloon.

Tim is putting a hat on.

Daddy mended a door.

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The bell is ringing.

Karen is making a cake.

Gary's got a bag of lego.

Hannah hurt her hand.

This hand is cleaner than the other.

Stuart's hamster scrambled up his sleeve.

We were away all year.

Laura will wear a yellow welly.

11.4 Appendix D: Treated and untreated word lists for all target speech sounds

Untreated	**Treated**
/s/ word initial high frequency Set (CVC) Save (CVC) Say (CV) So (CV)	/s/ word initial high frequency Sit (CVC) Same (CVC) See (CV) Soon (CVC)
/s/ word initial low frequency Sing (CVC) Suck (CVC) Soup (CVC) Sword (CVC)	/s/ word initial low frequency Sail (CVC) Sad (CVC) Soap (CVC) Sock (CVC)
/s/ word final high frequency Horse (CVC) This (CVC) Less (CVC) Force (CVC)	/s/ word final high frequency House (CVC) Face (CVC) Yes (CVC) Pass (CVC)
/s/ word final low frequency Mess (CVC) Boss (CVC) Dose (CVC) Lease (CVC)	/s/ word final low frequency Bus (CVC) Mouse (CVC) Nurse (CVC) Lace (CVC)

<table>
<tr><th>Untreated</th><th>Treated</th></tr>
<tr><td>/z/ word initial high frequency

No high frequency targets available</td><td>/z/ word initial high frequency

No high frequency targets available</td></tr>
<tr><td>/z/ word initial low frequency

Zone (CVC)
Zack (CVC)
Zipper (CVCV)
Zebra (CVCCV)</td><td>/z/word initial low frequency

Zoom (CVC)
Zap (CVC)
Zoe (CVCV)
Zero (CVCV)</td></tr>
<tr><td>/z/ word final high frequency

Does (CVC)
Was (CVC)
His (CVC)
As (VC)</td><td>/z/ word final high frequency

Days (CVC)
Those (CVC)
Is (VC)
Has (VCV)</td></tr>
<tr><td>/z/ word final low frequency

Toes (CVC)
Cheese (CVC)
Keys (CVC)
Shoes (CVC)</td><td>/z/ word final low frequency

Nose (CVC)
Knees (CVC)
Bees (CVC)
Rose (CVC)</td></tr>
</table>

Untreated	**Treated**
/tʃ/ word initial high frequency Choose (CVC) Chance (CVCC) Charge (CVC) Check (CVC)	/tʃ/ word initial high frequency Choice (CVC) Child (CVCC) Church (CVC) Change CVCC)
/tʃ/ word initial low frequency Chip (CVC) Chew (CV) Chase (CVC) Cheek (CVC)	/tʃ/ word initial low frequency Chin (CVC) Chair (CV) Cheese (CVC) Chain (CVC)
/tʃ/ word final high frequency Reach (CVC) Such (CVC) March (CVC) Each (CV)	/tʃ/ word final high frequency Teach (CVC) Much (CVC) Watch (CVC) Which (CVC)
/tʃ/ word final low frequency Punch (CVCC) Rich (CVC) Touch (CVC) Fitch (CVCC)	/tʃ/ word final low frequency Lunch (CVCC) Beach (CVC) Search (CVC) Pitch (CVCC)

Untreated	Treated
/ʃ/ word initial high frequency Short (CVC) Share (CV) Show (CV) Shall (CVC)	/ʃ/ word initial high frequency Shop (CVC) She (CV) Shown (CVC) Should (CVC)
/ʃ/ word initial low frequency Ship (CVC) Shape (CVC) Shut (CVC) Shock (CVC)	/ʃ/ word initial low frequency Sheep (CVC) Shine (CVC) Shed (CVC) Shirt (CVC)
/ʃ/ word final high frequency Wish (CVC) Publish (CVCCVC) British (CCVCVC)	/ʃ/ word final high frequency Fish (CVC) English (VCCVC) Finish (CVCVC) (no other high frequency words available)
/ʃ/ word final low frequency Harsh (CVC) Rush (CVC) Punish (CVCV) Ash (VC)	/ʃ/ word final low frequency Wash (CVC) Bush (CVC) Polish (CVCV) Cash (CVC)

Untreated	**Treated**
/dʒ/ word initial high frequency James (CVCC) John (CVC) June (CVC) January (CVCCVCVCV)	/dʒ/ word initial high frequency Just (CVCC) Job (CVC) Join (CVC) July (CVCV)
/dʒ/ word initial low frequency Jet (CVC) Jaw (CV) Joke (CVC) Jam (CVC)	/dʒ/ word initial low frequency Jane (CVC) Jar (CV) Juice (CVC) Jump (CVCC)
/dʒ/ word final high frequency Charge (CVC) Stage (CCVC) Range (CVCC) Manage (CVCVC)	/dʒ/ word final high frequency Large (CVC) Page (CVC) Change (CCVC) Village (CVCVC)
/dʒ/ word final low frequency Judge (CVC) Merge (CVC) Rage (CVC) Age (VC)	/dʒ/ word final low frequency Hedge (CVC) Lodge (CVC) Ridge (CVC) Cage (CVC)

Untreated	**Treated**
/s/ consonant cluster word initial high frequency Smile (CCVC) Star (CCV) Step (CCVC) Space (CCVC)	/s/ consonant cluster word initial high frequency Small (CCVC) Stay (CV) Start (CCVC) Speak (CCVC)
/s/ consonant cluster word initial low frequency Sleeve (CCVC) Score (CCV) Spin (CCVC) Smoke (CCVC)	/s/ consonant cluster word initial low frequency Sleep (CCVC) Sky (CCV) Spell (CCVC) Snake (CCVC)
/s/ consonant cluster word final high frequency Must (CVCC) Task (CVCC) Rest (CVCC) Used (CVCC)	/s/ consonant cluster word final high frequency Best (CVCC) Last (CVCC) Most (CVCC) It's (VCC)
/s/ consonant cluster word final low frequency Least (CVCC) Mix (CVCC) Dust (CVCC) Knives (CVCC)	/s/ consonant cluster word final low frequency Missed (CVCC) Pigs (CVCC) Lost (CVCC) Laughs (CVCC)

Untreated	**Treated**
/t/ word initial high frequency Tell (CVC) Town (CVC) Take (CVC) Time (CVC)	/t/ word initial high frequency Ten (CVC) Top (CVC) Talk (CVC) Two (CV)
/t/ word initial low frequency Tin (CVC) Ton (CVC) Toe (CV) Tea (CV)	/t/ word initial low frequency Tail (CVC) Tap (CVC) Tall (CVC) Tie (CV)
/t/ word final high frequency Light (CVC) But (CVC) Meet (CVC) Put (CVC)	/t/ word final high frequency Let (CVC) That (CVC) It (CV) Might (CVC)
/t/ word final low frequency Nut (CVC) Bite (CVC) Boat (CVC) Hurt (CVC)	/t/ word final low frequency Hut (CVC) Bat (CVC) Boot (CVC) Dot (CVC)

Untreated	Treated
/d/ word initial high frequency Done (CVC) Door (CV) Do (CV) Dog (CVC)	/d/word initial high frequency Date (CVC) Day (CV) Did (CVC) Dark (CVC)
/d/ word initial low frequency Deep (CVC) Doll (CVC) Dig (CVC) Dock (CVC)	/ d/ word initial low frequency Dip (CVC) Dot (CVC) Dish (CVC) Duck (CVC)
/ d/ word final high frequency Need (CVC) Good (CVC) Would (CVC) Bad (CVC)	/d/ word final high frequency Did (CVC) Could (CVC) Made (CVC) Bed (CVC)
/d/ word final low frequency Mad (CVC) Bid (CVC) Card (CVC) Fade (CVC)	/d/ word final low frequency Mud (CVC) Bird (CVC) Code (CVC) Feed (CVC)

Untreated	**Treated**
/l/ word initial high frequency Lie (CV) Late (CVC) Learn (CVC) Live (CVC)	/l/ word initial high frequency Lay (CV) Lead (CVC) Lot (CVC) Leave (CVC)
/l/ word initial low frequency Lap (CVC) Lid (CVC) Lock (CVC) Loud (CVC)	/l/ word initial low frequency Lamb (CVC) Lane (CVC) Log (CVC) Lawn (CVC)
/l/ word final high frequency Feel (CVC) Well (CVC) Goal (CVC) Feel (CVC)	/l/ word final high frequency Fill (CVC) Tell (CVC) Call (CVC) Fill (CVC)
/l/ word final low frequency Tall (CVC) Ball (CVC) Bull (CVC) Tail (CVCVC)	/l/ word final low frequency Doll (CVC) Bowl (CVC) Dull (CVC) Said (CVCVC)

Untreated	Treated
/n/ word initial high frequency New (CV) Name (CVC) Nice (CVC) Note (CVC)	/n/ word initial high frequency No (CV) Night (CVC) Nine (CVC) Not (CVC)
/n/ word initial low frequency Nurse (CVC) Noise (CVC) Nut (CVC) Neat (CVC)	/n/ word initial low frequency Nose (CVC) Nerve (CVC) Net (CVC) Near (CV)
/n/ word final high frequency An (CV) Fine (CVC) Than (CVC) When (CVC)	/n/ word final high frequency On (CV) One (CVC) Then (CVC) Man (CVC)
/n/ word final low frequency Born (CVC) Bean (CVC) Fun (CVC) Burn (CVC)	/n/ word final low frequency Bone (CVC) Bin (CVC) Fan (CVC) Pan (CVC)