UNIVERSITY OF SOUTHAMPTON

FACULTY OF HEALTH SCIENCES

A feasibility study of visual feedback speech therapy for nasal speech associated with velopharyngeal dysfunction

by

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Thesis for the degree of Doctor of Clinical Practice

March 2013

UNIVERSITY OF SOUTHAMPTON

ABSTRACT

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A FEASIBILITY STUDY OF VISUAL FEEDBACK SPEECH THERAPY FOR NASAL SPEECH ASSOCIATED WITH VELOPHARYNGEAL DYSFUNCTION

by Ginette Maria Phippen

Nasal speech associated with velopharyngeal dysfunction (VPD) is seen in children and adults with cleft palate and other conditions that affect soft palate function, with negative effects on quality of life. Treatment options include surgery and prosthetics depending on the nature of the problem. Speech therapy is rarely offered as an alternative treatment as evidence from previous studies is weak. However there is evidence that visual biofeedback approaches are beneficial in other speech disorders and that this approach could benefit individuals with nasal speech who demonstrate potential for improved speech. Theories of learning and feedback also lend support to the view that a combined feedback approach would be most suitable.

This feasibility study therefore aimed to develop and evaluate Visual Feedback Therapy (VFTh), a new behavioural speech therapy intervention, incorporating speech activities supported by visual biofeedback and performance feedback, for individuals with mild to moderate nasal speech. Evaluation included perceptual, instrumental and quality of life measures.

Eighteen individuals with nasal speech were recruited from a regional cleft palate centre and twelve completed the study, six female and six male, eleven children (7 to 13 years) and one adult, (43 years). Six participants had repaired cleft palate and six had VPD but no cleft. Participants received 8 sessions of VFTh from one therapist.

The findings suggest that that the intervention is feasible but some changes are required, including participant screening for adverse response and minimising disruptions to intervention scheduling. In blinded evaluation there was considerable variation in individual results but positive changes occurred in at least one speech symptom between pre and post-intervention assessment for eight participants. Seven participants also showed improved nasalance scores and seven had improved quality of life scores.

This small study has provided important information about the feasibility of delivering and evaluating VFTh. It suggests that VFTh shows promise as an alternative treatment option for nasal speech but that further preliminary development and evaluation is required before larger scale research is indicated.

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Declaration of Authorship

I, Ginette Maria Phippen declare that the thesis entitled:

A feasibility study of visual feedback speech therapy for nasal speech associated with velopharyngeal dysfunction

and the work presented in the thesis are both my own, and have been generated by me as the result of my own original research. I confirm that:

- this work was done wholly or mainly while in candidature for a research degree at this University;
- where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
- where I have consulted the published work of others, this is always clearly attributed;
- where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
- I have acknowledged all main sources of help;
- where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
- parts of this work are to be published as:
- Phippen, G., (ed.) (In press, 2013) The Confident Clinician: Speech therapy in cleft palate and velopharyngeal dysfunction'). J&R Press, Guildford, UK.

Signed: ……..

Date:……

Acknowledgements

I would like to gratefully acknowledge the support and guidance provided by my supervisors throughout my clinical doctorate and the writing of this thesis. They are Professor Mandy Fader, who helped me to see the 'big picture' and Dr Helen Rushforth who made sure I covered the detail too. I am also grateful to my third 'guest' supervisor, Dr Chris Markham, who gave me added confidence in my work.

Sincere thanks to all the participants and their families for taking part in the study. They all brought something different to the therapy sessions that helped me to understand how VFTh might work.

I am truly indebted to all my speech and language therapy colleagues in Salisbury and Oxford for their consistent encouragement and support, in particular Cindy, Sandra, Fiona and Benny – I really could not have achieved this without your help. Special thanks go to Dr Lisa Hirst, SLT head of service, for allowing me to work flexibly and giving much needed encouragement.

I am grateful both to Mr Steve Robinson on behalf of CLAPA Wessex and the SLT Trust fund for their generous contributions to buy therapy equipment for my research. I also gratefully acknowledge the financial support for my post from the Spires Cleft Centre and Salisbury NHS Foundation Trust.

Finally, I need to thank my family, but especially David, Izzy and Josh for their patience, tolerance and acceptance of my need to do this, along with too many other things at the same time. It would not have been possible without their support.

Glossary

Articulation
The process of forming speech sounds using the lips, tongue and palate.

Behavioural speech therapy intervention
An approach used to effect a positive change in an individual's communication functioning using behavioural techniques.

Biofeedback
A behavioural technique which provides sensory information regarding performance to an individual via a signal from an external source, which enables them to improve performance by using information from his or her own body.

Feedback
A behavioural technique designed to reinforce a desired behaviour. In speech therapy feedback can be verbal, visual, and tactile.

Cleft Lip and Palate
A cleft is a disruption in the growth and development of a baby's face and mouth while in the womb. It is a relatively common condition, occurring in approximately 1 in 700 babies born within the UK.

Cleft Centre
A regional hospital centre providing multidisciplinary care for all children with cleft lip and/or palate born within a catchment area.

Consonants
Speech sound produced by some obstruction of the breath stream, such as bringing the lips together e.g. for 'p' and 'm'.

Hypernasality
A quality of voice characterised by excessive emission of air through the nose resulting in poor intelligibility of speech.

Hyponasality
A quality of voice characterised by insufficient resonance of air in the nasal cavity, so that speakers sound as if they have a cold.

Nasal Speech
This refers to an excessively nasal tone of voice, often associated with speakers who have a repaired cleft palate or severe hearing impairment. Also referred to as hypernasality or nasality.

Nasal Emission
An abnormal flow of air through the nose accompanying speech. Usually indicative of an incomplete seal between the mouth and nose by the soft palate.

Nasendoscopy
A procedure where a small camera is passed through the nose to view the surface and movement of the soft palate during speech.

Nasometry
An instrumental measure of the nasal quality of speech giving a percentage score known as **nasalance.** The higher the nasalance score the more nasal speech is perceived to be.

Palate
The roof of the mouth including the bony portion at the front (hard palate), and the soft part at the back (the soft palate, or velum).

Palatal Investigation Clinic (PIC)
A specialist clinic, usually based in a Cleft Palate centre which offers assessment of speech disorders associated with palatal dysfunction.

Pharyngeal Flap or Pharyngoplasty
A surgical procedure to correct nasal speech in which a flap of the lining of the throat is used to close most of the opening between the soft palate and the back of the throat.

Phonation
Vibration of the vocal folds (cords) against air pressure from the lungs causing the release of air into the vocal tract.

Prosthetic Speech Appliance
A removable acrylic appliance (similar to a dental brace), which provides a structural means of achieving palate closure to correct nasal speech. This is also known as a palatal lift, speech bulb or obturator depending on exact design.
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Respiration
The breath stream provided by the lungs and converted into an acoustic signal through movements of the larynx and mouth.

Resonance
Voice quality associated with the vibration of air in the mouth and nose.

Soft Palate
The soft tissue attached to the back of the hard part of the palate, crucial to swallowing and speech. It contains muscles whose function results in the closure of the mouth cavity from the nose cavity, preventing the escape of air during speech and food/drink during swallowing.

Speech Therapy
A broad term used within the speech and language therapy profession to describe an eclectic range of techniques and approaches across a wide range of client groups with communication disorders. The terms treatment and intervention are also widely used.

Velopharyngeal Dysfunction (VPD)
A global term used to describe a range of problems with the function of the soft palate which prevents it from sealing against the back of the throat as normal during speech. Other terms include velopharyngeal incompetence, inadequacy and insufficiency.

Non-cleft VPD, As above but in the absence of a diagnosis of cleft palate.

Videofluroscopy
An investigation using X-rays to assess the movements of the soft palate during speech usually performed at between 5 -10 years of age.

Vowels
Speech sounds produced without any obstruction of the outgoing breath from the articulators e.g. ‘a’ and ‘e’.

List of Journal Abbreviations

AJSLP	American Journal of Speech-Language Pathology
Aphas	Aphasiology
B&SR	Behaviour and Self-Regulation
BJDC	British Journal of Communication Disorders
CL&Ph	Clinical Linguistics and Phonetics
CPJ	Cleft Palate Journal
CPCJ	Cleft Palate-Craniofacial Journal
Dev Neuro	Developmental Neurorehabilitation
FPL	Folia Phoniatrica et Logopaedica
IJCD	International Journal of Communication Disorders
IJLCD	International Journal of Language & Communication Disorders
IJPO	International Journal of Pediatric Otorhinolaryngology

IJSLP	International Journal of Speech-Language Pathology
JABA	Journal of Applied Behaviour Analysis
JAHS&P	The Internet Journal of Allied Health Sciences and Practice
JHTR	Journal of Head Trauma Rehabilitation
JMS	Journal of Maxillofacial Surgery
JoV	Journal of Voice
JPD	The Journal of Prosthetic Dentistry
JSHD	Journal of Speech & Hearing Disorders
JSHR	Journal of Speech & Hearing Research
LPV	Logopedics Phoniatrics Vocology
PRS	Plastic & Reconstructive Surgery

'Legend has it that, in times past, a superior race of SLTs roamed the earth. They were confident, well-dressed ladies who believed all was well with the world and that language disorders were no match for the force of their personality. Evolution caught up with them. Evidence-based healthcare proved a hostile environment for this life form, and their descendants are an altogether more timid species'.

Pring (2004), Chapter 14, p 21

Chapter One: Nasal Speech

1.1 Introduction

Nasal speech describes a speech disorder where there is an imbalance in the resonant quality of the speaker's voice. When the voice sounds excessively nasal (hypernasal), this may be caused by an underlying defect or dysfunction of the soft palate, known as velopharyngeal dysfunction (VPD). This is most commonly seen in individuals born with cleft palate but also in other conditions affecting the function of the soft palate during speech. Whatever the underlying cause, individuals with nasal speech are often perceived negatively by others and experience reduced quality of life at home, school and at work.

Clinical experience reveals that in some individuals the amount or degree of nasal speech varies, suggesting that soft palate function is inconsistent. In such cases this variability may indicate potential for reducing or eliminating nasal speech through the use of a behavioural speech therapy intervention, thus possibly avoiding surgery in some individuals. However as the efficacy of such interventions in reducing nasal speech is uncertain, they are not routinely considered for these individuals.

This thesis explores the background to this view and the lack of evidence available. It describes a feasibility study designed to explore a behavioural speech therapy intervention using visual biofeedback and performance feedback, for individuals with mild to moderate nasal speech associated with VPD.

1.1.1 Study context

This study has been undertaken in fulfilment of a clinical doctorate programme of study. This programme offers the opportunity to develop a combination of research and professional skills appropriate for a lead clinician role, in my case in the field of

speech and language therapy (SLT). The decision to develop and evaluate a SLT intervention has been informed by the broad political and specific professional and clinical drive for evidence; as clinicians and other stakeholders increasingly desire to know which interventions work. The impetus for the study arises from the current clinical context where specialist SLTs like myself have a well-defined role in assessment and also in the treatment of articulatory disorders relating to cleft palate and velopharyngeal dysfunction, but where our role in the treatment of nasal speech symptoms is unclear and the existing evidence weak. This raises the question of whether there may be more that SLT can offer to individuals as an acceptable alternative to surgical treatment. A well designed intervention study would therefore contribute to the body of knowledge and debate regarding the feasibility of behavioural speech therapy interventions for nasal speech.

1.1.2 Thesis overview

The thesis is divided into 7 chapters. From this point Chapter One explains the background and rationale for this study. The chapter defines nasal speech and discusses its presentation, causes and impact on the individual. Treatment options are considered, both surgical and non-surgical, including speech therapy and feedback approaches.

Chapter Two critically reviews the literature and evidence available for non-surgical treatments for nasal speech. Within this review, previous studies using visual biofeedback are identified, with positive results for some individuals. Chapter Two considers the broader context of performance feedback and visual biofeedback approaches used across speech and language therapy. This leads to the formation of the research questions.

Chapter Three outlines the development of the feedback intervention used in the study as informed by the findings of the literature review in Chapter Two, and underpinned

by theories of learning and feedback. The selection of an appropriate research design and outcome measures is also discussed.

Chapter Four describes the methodology. This includes ethical considerations and full description of study methods, including sampling and plans for analysis.

Chapter Five presents a summary of the data collected during the study, and includes graphical presentation and analysis of the main findings in relation to the research questions.

Chapter Six discusses the results as they relate to each of the research questions, as well as the theoretical frameworks outlined in Chapter Three. The results are also considered in relation to previous research and the strengths and limitations of the methodological design. Finally the implications for future research are discussed.

Chapter Seven concludes the thesis and summarises the contribution of the research including recommendations for future work.

1.2 Background

1.2.1 Speech

Speech, as a form of communication, can be seen as a core life skill. The World Health Organisation (WHO, 2003) defines life skill as the ability for adaptive and positive behaviour that enables an individual to deal effectively with the demands and challenges of everyday life. Communication is one of ten life skills proposed by the WHO as being essential for the healthy development of children and adolescents. Similarly, Bercow (2008) describes the ability to communicate as being 'at the core of all social interaction.' The Bercow Report is a recent government review of services for children and young people with speech, language and communication needs (SLCN) and the commonly accepted definitions of these terms are as follows:

- **Speech**: using sounds to communicate words, with a clear, fluent, expressive voice.
- **Language**: using words to build sentences and conversations and understanding and making sense of what people say.
- **Communication**: interacting with others, both verbally and non-verbally.

SLCN therefore encompasses a wide range of difficulties related to all aspects of communication, including difficulties with forming sounds and words, speaking fluently, formulating sentences, understanding what others say, and using language socially. These difficulties may be congenital (present at birth), developmental (occur at some stage in a child's development) or acquired at any stage of childhood or adulthood as the result of disease or trauma affecting the organs of speech (Frattali, 1998).

The Bercow report estimates that approximately 7% of five year olds entering school in England have significant difficulties with speech and/or language and approximately 1% have the most severe and complex SLCN. According to ICAN (2006), a specialist UK children's communication charity, 1 in 10 children have communication difficulties that require specialist help, equating to two or three children in every classroom. There is no definitive data about the prevalence of SLCN in UK adults but conservative estimates range from 1% to 2% of the population (Adult Communication Coalition England, 2009). This currently represents over 1 million adults who have difficulty communicating effectively without support. International prevalence data for SLCN is also patchy with limited reports available, mostly from developed countries. Accurate data is difficult to gather as some developmental SLCN are transient whilst others persist into adulthood. In Australia, McLeod et al (2007) reported up to a quarter of 4,983 four to five year old children identified by parents and/or teachers as having SLCN and 16% receiving professional help. The American Speech and Hearing Association (ASHA, 2008) also reported that SLCN is common in children with

disabilities in the United States; with around 25% receiving services. However this estimate does not include children with SLCN secondary to other conditions such as autism or hearing impairment and is therefore likely to be an underestimate of true prevalence.

The impact of SLCN is well documented; a systematic review of relevant papers over a period of 10 years by McCormack et al. (2009) revealed that speech impairment in childhood may be associated with limitations or restrictions in a wide range of domains including reading, writing, thinking, relationships and work. There are reports of bullying (Knox & Conti-Ramsden, 2003), withdrawal (Fujiki et al., 2001) and poor academic achievement (Lewis et al., 2004). Children whose impairments involve speech alone are generally thought to be less at risk than those whose impairments involve language, particularly understanding what others say (Law et al., 1998, Young et al., 2002). However Lewis and Freebairn (1992) also found evidence that early difficulties with phonology (speech sounds) are associated with poor performance in reading and spelling in adolescents and adults. Similarly, children with voice disorders may be perceived more negatively than children with normal voice (Hooper, 2004).

In relation to adults, studies show a range of effects resulting from communication impairment including negative impact on personal development, social engagement, activity, employment and productivity (Ramig & Verdolini, 1998, Resnick et al., 1997, Hayhow et al., 2002, Krischke et al., 2005). There is also evidence that childhood communication impairment is a risk factor for mental health difficulties later in life (Baker & Cantwell 1987) and individuals with SLCN are significantly over-represented in the young offender and prison populations (The Communication Trust, 2010). In addition it appears that the degree of impact on communicative confidence and self-esteem is not necessarily correlated with the severity of the communication impairment (McLeod, 2004). It has been postulated that this variability may relate to psychological resilience whereby people differ in their ability to cope with negative

events, including health difficulties, depending on individual traits as well as the influence of external factors (Tugade et al., 2004).

1.2.1.1 Speech production

Speech production is a complex process under the control of the central nervous system. It involves the integration of many physiological mechanisms and functions including respiration (breathing), phonation (voice production), and articulation (speech sound formation), (Leahy & Stemple, 1989).

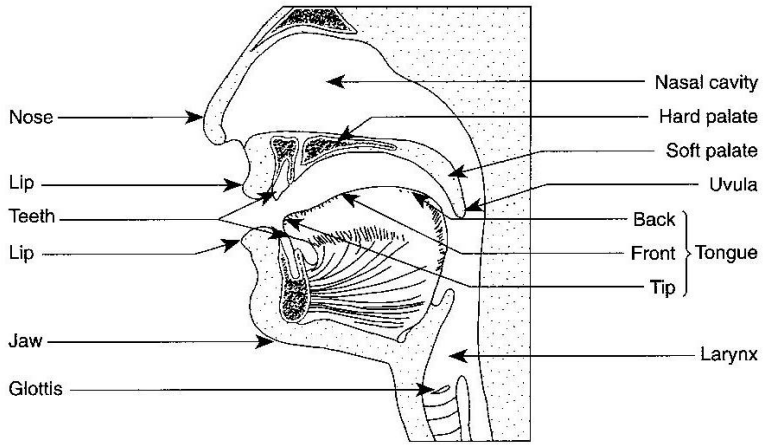

Figure 1-1: The organs of speech

Reproduced from 'Coping with Stuttering', Louw (1996)

Original source Byrne (1991) Talking about Stammering, British Stammering Association, reproduced with permission.

Figure 1-1. illustrates how air moves from the lungs up the trachea (windpipe), through the larynx (voice-box) and towards the vocal tract (throat, nose and mouth). As the air passes through the larynx it causes the vocal cords to vibrate resulting in the generation of sound. This sound passes through the vocal tract which acts as a resonating tube and is modulated by the velum (soft palate) and the articulators (tongue, lips and jaw) to produce the sounds in speech.

1.2.1.2 Nasal Speech

The term 'nasal' may be used to describe speech where there is too much or too little air resonating in the nasal cavity (Sell et al., 1999). In all speakers there is a balance of acoustic energy (sound) from air flowing from the lungs through the vocal cords into the cavities of the mouth and the nose (Jones 2006). This is perceived by the listener as resonance, which is a quality of voice (Mathieson, 2001). There is a spectrum of resonance from hypernasality to hyponasality. If there is too much nasal resonance, caused by excess air passing through the nose, this is perceived as hypernasality. Conversely a reduction in nasal resonance results in a hyponasal voice quality (Sweeney, 2011). Both hypernasality and hyponasality may be clinically significant as they can interfere with the clarity of speech. However hyponasality is generally considered to have less of an impact on speech as it primarily affects only the nasal class of consonants (speech sounds) in English speakers i.e. 'm', 'n' and 'ng'. Hypernasality on the other hand affects all vowel sounds, results in weak consonants (Harding & Grunwell, 1996) and has been reported as more negatively perceived by the listener (Lallh & Rochet, 2000).

This study is concerned with hypernasality as a clinical symptom of velopharyngeal dysfunction (VPD). The term 'nasal speech' is used herein to encompass one or both of the following speech characteristics; hypernasality (excessively nasal voice quality) and nasal emission (excessive nasal airflow accompanying speech sounds). In addition the term nasal turbulence is used to describe a noisier form of nasal emission, perceived as a 'buzz' or 'snort' type sound (Sell et al., 1999). Figure 1-2. shows how the excess flow of air from the lungs into the nose results in nasal speech.

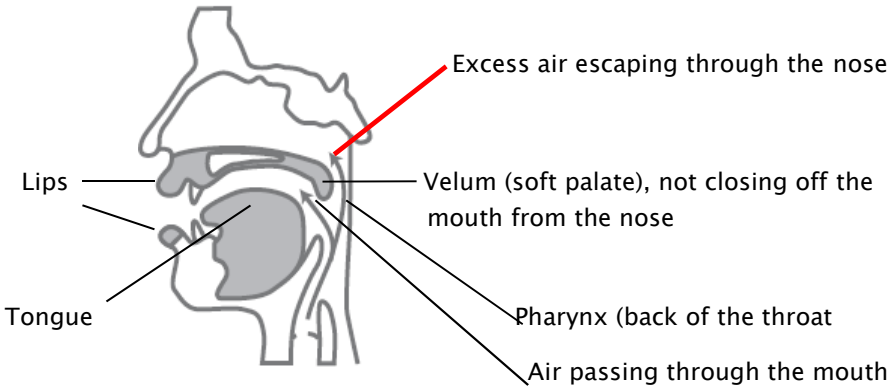

Figure 1-2: Airflow during speech resulting in nasal speech

1.2.2 The velopharyngeal mechanism in speech

The velopharyngeal (VP) mechanism consisting of the velum (soft palate) and the pharynx (throat) has been extensively explored. In an early study Skolnick et al. (1975) note the marked variability in VP function between individuals with multiple patterns of movement involving the velum, and lateral and posterior pharyngeal walls. Stal & Lindman (2000) examined palate muscle characteristics and identified distinct morphology suggesting high aerobic capacity, fatigue resistance and a special proprioceptive (position-movement) control system. In addition Shimokawa et al. (2005) present an anatomical study of the nerves involved in VP function and comment on the variation in descriptions of VP muscle innervation. Nohara et al. (2006) later investigated fatigue of palate muscles in small groups of speakers with and without repaired cleft palate and found greater fatigue in those with cleft palate. These studies contribute to current understanding of normal and abnormal function of the VP mechanism.

Videoradiograhic and endoscopic studies have confirmed that despite variability, during speech, muscle activity causes the soft palate to elevate and retract which, along with movement of the muscles in the throat, results in the soft palate making a

seal with the throat, described as velopharyngeal closure (Moon & Kuehn, 2004). This occurs for the majority of English speech sounds and serves to seal the mouth off from the nose in order to achieve the correct balance of oral and nasal resonance. Only for the sounds 'm', 'n' and 'ng' does the soft palate remain lowered to allow air to resonate in the nose. In some languages there may also be incomplete VP closure during the production of vowel sounds ('a, e, ai' etc.). Irrespective of accent, dialect or language it is clear that rapid, automatic and coordinated movement of the soft palate is needed during conversational speech to achieve the required balance of resonance (Bzoch, 2004).

1.3 Velopharyngeal dysfunction/VPD

Velopharyngeal dysfunction or VPD is a global term used to describe a range of problems with the function of the soft palate which prevents it from sealing against the back of the throat as normal during speech. It is one of several terms in the literature used to describe a structural or functional problem with VP closure that results in nasal speech (Albery & Russell, 2005). Other terms include velopharyngeal incompetence, and insufficiency. Velopharyngeal incompetence generally refers to an underlying problem with neuromotor control and the term velopharyngeal insufficiency is mainly used when the cause is believed to be anatomical. Peterson-Falzone et al. (2006) discuss the use of such terminology in relation to underlying aetiology and adopt velopharyngeal inadequacy (VPI) as the preferred umbrella term but accept VPD as an alternative. VPD is the term used throughout this thesis as it is in common usage in UK cleft centres.

1.3.1 Causes of VPD

The cause of VPD may be structural, neurogenic or mechanical, or the result of mislearning as summarised in Table 1-1. In some individuals who present with nasal speech associated with VPD there is a combination of aetiological factors, for example cleft palate (structural) and dyspraxia (neurogenic). Cleft palate is the most common

structural cause of VPD (Johns et al., 2003). Where VPD is the result of something other than cleft palate this is generally subsumed under the heading of non-cleft VPD. In the UK this term is used to describe a problem with the structure or function of the soft palate in the absence of a diagnosis of cleft palate. This covers VPD resulting from conditions such as cerebral palsy, stroke, traumatic brain injury and a wide range of degenerative neurological conditions (Specialised Services National Definitions Set, Number 15, 2010).

Table 1-1: Causes of VPD (Adapted from Peterson-Falzone et al., 2006)

Causes of VPD – Cleft and Non-Cleft	
Structural	**Mechanical**
Cleft palate	Enlarged tonsils
Short palate/Large nasopharynx	Posterior pillar webbing
Post-adenoidectomy	
Post- ablative surgery (e.g. cancer)	
Neurogenic	**Mislearning**
Cranial nerve damage/dysfunction	Speech sound error (Active nasal
Dysarthria (muscle weakness)	fricative)
Dyspraxia (muscle incoordination)	Hearing impairment

1.3.1.1 Structural causes of VPD

Cleft palate is the primary structural cause of VPD. A cleft literally means 'a separation' or 'split' (CLAPA, 2009) and can involve the lip on one or both sides as well as the palate. Its causes are not entirely understood and are likely to be multifactorial (Watson, 2001). There is good evidence from studies of embryology and genetics of the heterogeneity of the cleft palate population. Spriestersbach et al. (1973) demonstrate that isolated cleft palate and cleft lip are distinct in origin and environmental factors are likely to be of significance only in relation to cleft palate In

this early comprehensive review of cleft palate, Spriestersbach reports evidence of both genetic factors (e.g. chromosome defects and mutant genes) and environmental factors (e.g. anti-emetic & anti-epileptic drugs and folic acid deficiency) and also discuss the likelihood of interaction between the two in many cases. More recently Stanier and Moore (2004) report on advances in identifying the genes and gene pathways associated with clefting. They acknowledge the difficulty in relating experimental studies to the complex human situation but conclude that there should be optimism about further pinpointing the contributory factors.

Approximately 1000 babies are born each year in the UK with a cleft lip, cleft palate or combination of the two (CRANE database, 2009). Worldwide the birth prevalence ranges from 1 in 1000 to 2.69 in 1000 births (McLeod et al., 2004) making cleft lip and/or palate one of the most common congenital anomalies. In 80% of these babies, a cleft of the palate will be present. Cleft lip and palate is more common in males, whilst isolated cleft palate occurs more in females (Stanier & Moore, 2004). Cleft lip and palate can occur in isolation or present as part of a complex disorder, and is described as a feature in over 400 known syndromes (Mossey et al., 2009).

In the UK a cleft lip is usually surgically repaired at three months of age and a cleft palate at six to nine months of age (Watson, 2001). The timing of this surgery continues to be the subject of international debate. Rohrich et al. (1996, 2000) discuss how the controversy centres on the apparent conflict between a need for early palate repair for optimum speech development versus delayed hard palate repair to allow for undisturbed facial growth. Supporters of early palate repair (12 months of age and under) argue that the nature of speech development as a learned behaviour indicates that anatomy should be restored as soon as possible (Estrem & Broen, 1989; Russell & Grunwell, 1993). In an attempt to resolve this question a randomised controlled trial (RCT) is currently underway involving cleft centres in the UK, Scandinavia and Brazil (Timing of Primary Surgery for Cleft Palate [TOPS], in progress).

This study is aiming to determine whether surgery for cleft palate, using a specified technique, at age 6 months, when compared to surgery using the same technique at age 12 months, affects speech development outcomes at ages 3 and 5 years.

Notwithstanding timing of surgery, as cleft palate is a structural defect it is hoped that the initial surgical palate repair will result in adequate soft palate function so that during speech air flows through the mouth rather than the nose. If this does not happen then air will escape into the nose and a child's developing speech may have a nasal tone. This may be because the repaired soft palate is too short, or does not move well, or there is a residual hole in the palate known as a fistula (Mercer & Pigott, 2001). At least 20% of children will continue to experience escape of air down the nose after the primary cleft repair resulting in nasal speech (Witt et al., 1997).

1.3.1.2 Other structural causes of VPD

In addition to cleft palate as a structural cause of VPD other individuals present with structural non-cleft VPD that is the result of a congenital disproportion between the structures of the palate and the throat. The cause of this is generally unknown and in such cases the palate does not reach to close against the back of the throat because it is too short or the space at the back of the throat is too large (Stewart et al., 2002, Peat et al., 1994).

Adenoidectomy can also result in structural VPD and in some cases may reveal an underlying palatal abnormality such as submucous cleft palate (Saunders et al., 2004). The adenoids are lymphoid tissues near the posterior pharyngeal wall and, in some individuals VP closure is achieved by moving the soft palate against the adenoid bed itself. Therefore when the adenoids are removed there can be a transient VPD and in a small proportion of patients permanent VPD occurs (Kummer, 2004).

1.3.1.3 Mechanical causes of VPD

This refers to interference with the function of the VP mechanism in speech due to the presence of enlarged tonsils or less frequently tonsillar webbing (Trost-Cardamone,

1989). In particular it is thought that enlarged tonsils can impede the soft palate movement by preventing it from moving effectively to seal against the back of the throat, resulting in hypernasal speech (Ren et al., 1995). One study of 20 patients by Shprintzen et al. (1987) reported resolution of hypernasality following tonsillectomy and no other treatment.

1.3.1.4 Neurogenic causes of VPD

This category encompasses a wide range of conditions with an underlying neurological component that can impact on many aspects of physiological functioning including the VP mechanism. The disruption to this mechanism may be caused by weakness, paralysis or lack of coordination resulting from congenital or acquired conditions such as cerebral palsy, dyspraxia and traumatic brain injury (Johns et al., 2003).

1.3.1.5 Mislearning

Finally, VPD can also result from the mislearning of speech sounds whereby specific speech sounds (usually 's' and 'z') are produced with air directed down the nose, often perceived by the listener as a nasal 'snort'. This has been described as Phoneme Specific VPI (Kummer & Lee, 1996) and is an articulation error. Sell et al. (1999) use the term 'Active Nasal Fricative' which perhaps more accurately describes the process by which specific sounds are replaced by turbulent nasal airflow in the absence of generalised palatal dysfunction.

1.3.2 The impact of VPD and nasal speech

1.3.2.1 Impact on speech

The primary impact of VPD (cleft and non-cleft) is on speech, although nasal regurgitation of food and drink and nasal or facial grimacing can also be features (Bradbury, 2001). A nasal speech quality caused by VPD sounds noticeably abnormal to the listener making speech weak, indistinct and difficult to understand. This can include the substitution of nasal sounds for their oral counterparts so that 'b' is perceived as 'm' and 'd' as 'n' (Grunwell, 1993). This significantly reduces speech

clarity, for example the word 'baby' sounds like 'mamy' and 'daddy' sounds like 'nanny'. Similarly, the noisy escape of air accompanying speech, through a small gap between the soft palate and back of the throat (nasal turbulence) is intrusive in conversation (Wyatt et al., 1996).

As well as adversely affecting speech clarity VPD can also interfere with the actual development of speech sounds (articulation) (Harding & Grunwell, 1996; Riski & Delong, 1984). Substituted sounds may develop as a compensatory mechanism due to the structural inadequacy of the VP mechanism, which prevents the required build-up of intra-oral air pressure to form consonant sounds such as 'b' 'd' and 'g'. Russell and Harding (2001) explain how these articulation errors can become established at an early age in children with cleft palate resulting in a significant impact on speech clarity.

The severity of nasal speech and/or articulation errors resulting from VPD varies between individuals, with a mildly nasal voice quality at one end of the spectrum and severely unintelligible speech at the other. The terms mild, moderate and severe are widely used in healthcare to indicate symptom severity. These descriptive terms have no absolute values although health researchers have attempted to clarify and quantify the boundaries between them, for example in defining degrees of pain (Serlin et al., 1995). The terms are also seen in definitions of severity and levels of impairment in speech disorders (Bowen, 1998, ASHA, 2010). In relation to nasal speech these relative descriptors are also widely used in speech assessment following cleft palate repair (Vogel et al., 2009, Henriksson et al., 2005, Lee et al., 2003).

1.3.2.2 Impact on quality of life

It is accepted that nasal speech often results in a negative impact on quality of life for children and adults (Lallh & Rochet, 2000; Barr et al., 2007; Deary et al., 2003). The concepts of activity and participation are central in considering quality of life. These form part of the International Classification of Functioning, Disability and Health (ICF) framework proposed by the World Health Organisation (WHO, 2001). In this

framework activity and participation feature alongside body structures and functions, in attempt to focus on impact and not simply disorder. McLeod (2004) has reported preliminary work in applying these concepts to speech impairment, using the following definitions:

Activity	Participation
-Being intelligible (clarity of speech)	-Being able to take part in family life/events
-The ability to hold a conversation	-The ability to form relationships with peers
-The ability to express needs and wants	-The ability to interact with the community
-Being able to read and spell	-Being able to achieve goals in life

As Havstam and Lohmander (2011) state, the implication is that any assessment of nasal speech should consider the impact on the individual and their 'communicative participation'. They also point out that the effect of nasal speech on the individual may not necessarily correlate with its severity; in some individuals a relatively mild nasal voice quality can lead to low self-esteem and lack of participation in educational and social situations.

1.3.3 Treatment for nasal speech

Treatment for nasal speech (hypernasality/nasal emission/nasal turbulence) associated with VPD can take the form of surgery, prosthetics, speech therapy or any combination of these approaches. Management is generally delivered by a multidisciplinary team including a speech and language therapist and plastic surgeon as key team members (Sell & Grunwell, 2001).

1.3.3.1 Surgical treatment

Surgery is considered the appropriate treatment where there is a structural basis for the nasal speech. In the context of cleft palate management this is often known as secondary speech surgery. The aim is to restore the function of the velopharyngeal mechanism and in doing so improve the ability to communicate (Boseley & Hartnick, 2004).

There are broadly two types of surgery routinely offered in the UK; palatoplasty and pharyngoplasty. Palatoplasty, or palate re-repair, involves dissection and repositioning of the muscles in the soft palate to produce optimum muscle function (Sommerlad, 2003). Pharyngoplasty involves rearranging tissue at the back of the throat, usually by creating flaps, in order to make it easier for the palate to close (Riski, 2004).

Outcomes of speech surgery are variable and reports of success can range from 22% to 95% depending on the measurement tools and parameters (Boseley & Hartnick, 2004). Success would generally be taken to mean a good perceptual speech outcome as judged by trained listeners but could also include rates of surgical breakdown or fistulae. Any surgical intervention of this type has associated risks including anaesthetic complications, bleeding, infection and tissue breakdown (Paradise, 1996). In the management of VPD specifically, pharyngoplasty has also been reported to be associated with obstructive symptoms and sleep apnoea, although this does depend on the exact nature of the surgery undertaken (Mercer & Pigott, 2001). Although in this context the surgical risks are relatively small, nevertheless the decision to proceed requires careful consideration.

1.3.3.2 Prosthetic treatment

Prosthetic treatment may be offered for individuals with nasal speech where surgery is contraindicated or declined (Sell et al., 2006). A removable prosthesis such as a palatal lift can help by elevating an immobile soft palate or a speech bulb can partially block a gap between the tissues of the soft palate and the back of the throat (Figures

1-3 and 1-4). The decision to recommend prosthetic treatment is often due to the failure of previous surgery (Sell et al., 2006). It requires a high level of motivation, is only suitable for specific patients and can place a high burden of care on the individual in terms of hospital attendance for fitting and adjustment. However when successful it may facilitate improvements in speech and quality of life that have proved elusive.

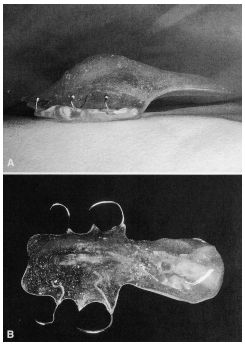

Figure 1-3: Palatal Lift

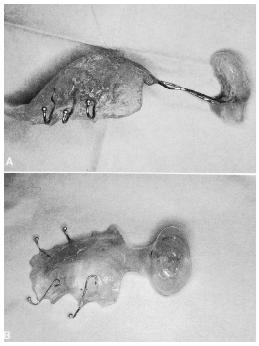

Figure 1-4: Speech Bulb

1.3.3.3 Speech Therapy and other non-surgical treatments

There is a lack of consensus about the role of speech therapy and other non-surgical treatments in treating nasal speech. The past fifty years has seen significant developments in the frameworks for assessing the complex speech mechanism and identifying the contributory components in speech disorders associated with VPD (Kuehn & Moller, 2000). There is some evidence that speech therapy combined with surgery can be the most effective approach (Mercer and Piggott, 2001), generally to address compensatory articulation patterns, but the use of speech therapy as a primary treatment for nasal speech is not widely supported.

Nevertheless speech therapy treatment of nasal speech continues to attract interest and debate as there is very limited evidence of efficacy. Kuehn & Moller (2000) summarise the trends and outline three phases in opinion relating to the effectiveness of speech therapy treatment, as earlier described by Tomes and colleagues in 1997. In

phase one (1940s to 1960s) the accepted view was that speech therapy was effective. In phase two (1960s to 1970s) the view was that speech therapy does not work for nasal speech but can improve articulation and speech intelligibility generally. The third phase (1970s to 2000) is that speech therapy might work for nasal speech but that more data is needed on the efficacy of specific treatments.

The ultimate goal of any intervention for nasal speech must be a functional improvement in speech, with no adverse effects (Johns et al., 2003; Boseley & Hartnick, 2004). A variety of interventions have been offered including palate exercises, stimulation, resistance training, visual biofeedback, articulation therapy and voice therapy. These type of behavioural interventions may be defined as a speech therapy intervention or not but all operate on the premise that new speech behaviours can be learnt; as long as there is sufficient palate tissue to close the mouth off from the nose. In addition there must be adequate muscle and nerve function for movement of the soft palate. Where speech therapy and other non-surgical interventions are described it is therefore vital to consider the underlying mechanisms which may be operating, as well as the aim of the intervention, which may be:

- To change the velopharyngeal muscles (strength/mass/endurance)?
- To change control of the velopharyngeal muscles (co-ordination/consistency)?
- To change other speech factors to reduce nasal speech quality (respiratory/laryngeal/articulatory)?

Cole (1979) proposes the terms **direct** and i**ndirect** to differentiate between approaches designed either to stimulate or to influence velopharyngeal function. The former term refers to an intervention that involves direct sensory or motor stimulation of palate function and includes palate exercises, prosthetics, stimulation of the soft palate and resistance training. **Direct** interventions are based on the premise that muscle training or stimulation techniques can increase the activity of the muscles involved in

velopharyngeal closure. In direct treatment approaches, there is therefore an assumption that changes will occur in palate muscle function, for example increased strength, tone or elasticity. In contrast an i**ndirect** intervention refers to an approach designed to influence velopharyngeal function but one that does not focus specifically on muscle activity. With indirect approaches, the change is a behavioural one with increased voluntary control over palate movement and associated nasal speech. Speech therapy, including the use of feedback, generally falls into this category.

Ruscello (1982) refers to Cole's definitions in his review of palatal training approaches and it is a useful distinction in that it allows consideration of the mechanisms underlying the intervention. This refers to the mechanism by which functional improvement in speech might occur. The direct versus indirect definitions are also used in other related but distinct contexts. In relation to the assessment of velopharyngeal function, direct assessment refers to techniques that allow for visualisation of VP activity (nasendoscopy and videofluoroscopy) whilst indirect techniques provide inferences about VP function, usually through instrumental or perceptual measures (Johns et al., 2003, Kuehn & Moller, 2000). The terms are also used to describe speech and language interventions more generally. Direct interventions are those delivered by a speech and language therapist whilst indirect interventions are usually delivered through a third party, for example a parent/carer or teaching assistant. In this study the terms are used with reference to Cole's (1979) definitions outlined above.

Exercises are a direct intervention using stimuli such as blowing, sucking, swallowing and gagging. They have been used to treat nasal speech and are classed as non-speech or oromotor exercises (Bowen 2005). This is in contrast to speech therapy approaches that use speech stimuli (speech sounds and words) to influence parameters such as rate, clarity and fluency. The use of non-speech exercises is based on physical therapy principles of exercise, involving practice and repetition (Palmer et

al., 2007) and includes active muscle exercise and passive muscle exercise. The former term applies to exercises involving strength training and stretching whilst passive exercise is the movement of a muscle or muscle group assisted by another person or a machine. The focus is generally on active exercise in relation to palate function. Several authors hypothesise regarding the effect of such exercises on the palate, for example Witt et al., (1995) suggests that a muscle tends to assume the length in which it is habitually maintained so that actively exercising the palate muscle could result in increased stretch or extension during speech.

The use of non-speech tasks to influence palate function or improve speech has been largely discredited (Kuehn and Moller, 2000; Bunton, 2008; Bowen, 2005). Ruscello (2006, 2008) comprehensively reviews this area and concludes that muscle treatments are not an effective approach for most individuals with VPD. The rationale for this is that the underlying mechanisms controlling speech and non-speech palate function differ, with different patterns of palate closure evident in each. Bunton's paper (2008) in particular, effectively challenges the assumption that movement characteristics and task demands are similar for speech and non-speech oral behaviours. The author goes on to assert that this is supported by motor learning theory of task specificity as well as differences in neural organisation demonstrated by MRI studies in healthy individuals.

Palate Stimulation is also a direct intervention based on physiological principles. It aims to increase blood supply to the palate in order to reduce fatigue and increase muscle bulk, tone and stretch (Kuehn et al., 2002). Approaches have tended to use either electrical or tactile stimulation, or a combination of both. Peterson (1974) hypothesised that electrical stimulation might result in increased firing of nerve fibres, therefore constituting a type of 'active' exercise for the palate in order to stimulate movement.

Resistance Training describes the requirement of muscles to move against an opposing force, usually provided by equipment of some kind (Fleck and Kraemer 2004). The aim of this direct approach is to create resistance between the device and muscular forces in the palate and so improve velopharyngeal function. Approaches have included bespoke inflatable devices, nose-holding to introduce positive pressure into the oral cavity and the use of pressurised air with Continuous Positive Airway Pressure (CPAP) equipment.

Prosthetic Training is a direct intervention that involves the speaker wearing a bespoke dental appliance in the mouth that fixes on to the upper teeth and extends back to touch the soft palate (Ruscello, 1982). A reduction programme is employed to stimulate the muscles and nerves involved in palate closure, with the premise that duration of using the prostheses and its size is gradually reduced as compensation of velopharyngeal musculature occurs. Prosthetic palatal appliances are currently routinely used as a treatment option for VPD/nasal speech but not as a direct intervention designed to stimulate the palate. Rather, prosthetic appliances take the form of a palatal lift or bulb (page 14) that is designed to compensate for inadequate palate movement and/or insufficient palate tissue rather than to stimulate improved sensation or function (Sell et al., 2006).

The lack of recent or multiple studies in most of the above areas reflects a move away from the theoretical perspective of treating the palate muscles as any other muscle in the body i.e. as amenable to training to improve strength, range, speed or precision of movement. However it should be noted that such regimes have not been systematically evaluated in large numbers of individuals and with effective control groups. Nevertheless, it is generally accepted in clinical practice that these direct approaches are not appropriate in treating VPD (Peterson-Falzone et al., 2006).

Biofeedback is an indirect intervention which provides information regarding speech performance to the individual via a signal from an external source. This enables the individual to attempt to volitionally modify their performance by using information from his or her own body (Earles et al., 2003; Laures & Shisler, 2004). Biofeedback approaches to treating nasal speech have taken two forms; direct visualisation of the velopharyngeal structures using a rigid or flexible nasendoscope and visual biofeedback generated by specialised instrumental equipment. Brunner et al., (2005) give a useful theoretical breakdown of nasendoscopic biofeedback into four areas as follows, highlighting the learning process involved:

- Introduction to structures and function
- Experimentation – self monitoring with guidance
- Coupling – anchoring motor and auditory perception
- Automatization – overlearning

They propose that the concepts of improved self-monitoring and improved sensory control are central to the process of nasendoscopic biofeedback and that in order for palate closure to be improved there must be evidence that this is achievable.

The same principles can be seen to apply to instrumental biofeedback where physiological output, in this case airflow associated with speech, generates a visually displayed signal. This allows the speaker to monitor their speech and attempt to modify it according to defined parameters or thresholds.

Articulation therapy is a generic terms for a range of indirect approaches used to treat speech disorders relating to abnormal structure and or faulty learning of speech sounds (Bowen, 2009). Articulation therapy is essentially based on behavioural principles of shaping a desired response through modelling and reinforcement. In

practice however, as Bowen explains, clinicians draw upon a number of theoretical learning frameworks including those relating to speech at the cognitive-linguistic level.

Articulation therapy is widely used to treat errors associated with VPD, including mislearnt speech sounds (Dixon-Wood, 2004; Golding-Kushner, 2009). The aim is to establish the correct patterns of articulation required for a developmentally appropriate range of speech sounds. Articulation therapy is also used to treat VPD when a child presents with absent development of oral speech sounds (Sweeney, 2011). This is with the aim of assessing potential for improvement in speech prior to surgery and is known as diagnostic therapy or stimulability testing (Kuehn & Moller, 2000). The primary focus in this case is on establishing whether correct oral articulation patterns can be elicited in the presence of VPD.

As with articulation therapy in **Voice Therapy** the aim is to modify speech production, predominantly through behavioural shaping and reinforcement. Treatment programmes vary but can include a focus on increasing vocal awareness, postural adjustment, targeted relaxation, breathing and patterns of voice production based on physiological principles (Cavalli, 2011). Many voice therapy programmes will also pay attention to psychosocial aspects that may be impacting on the physiology of the voice (Ramig & Verdolini, 1998).

Cavalli (2011) states that voice therapy may be considered appropriate for individuals who have abnormal voice associated with VPD e.g. dysphonia characterised by hoarseness, breathiness and strain. It is not widely used for treating individuals with nasal sounding speech, despite this also being a perceived characteristic of voice. This is likely to be because, as with other non-surgical treatments, there is an assumption that such an approach is ineffective. However its use has not been extensively investigated with this population and therefore evidence of efficacy is lacking.

1.4 Summary

Despite a resurgence of interest in non-surgical treatments for nasal speech in the late 1970s, speech therapy was not widely re-introduced as a primary treatment due to the general belief that this approach was ineffective. Unsurprisingly this view has persisted over the last 30 years (Shprintzen & Marrinan, 2009; Russell, 2006; Golding-Kushner, 2001; RCSLT, 2005) as there is insufficient supporting evidence. This applies to both direct and indirect approaches and equally in cases where surgery or prosthetic treatment is not indicated or is declined, or where palate function is inconsistent. This means that in a proportion of cases the individual is offered no treatment at all.

However, Kummer (2004) proposes that speech therapy could be considered as an appropriate treatment if the nasal speech characteristics are mild and inconsistent, if the individual can achieve reduction of the characteristic (is stimulable), or if the characteristic is caused by faulty use (mislearning) of the velopharyngeal structure or associated with oral motor dysfunction. These criteria could therefore be defined as the individual demonstrating the necessary potential for velopharyngeal closure. Similarly, several other authors acknowledge the role of speech therapy for selected individuals; usually those with mild and/or inconsistent symptoms or where there is evidence of potential for closure of the palate against the back of the throat (Ruscello, 2008; Ysunza et al., 1997). Jones et al., (2004) also describes a category of patients with 'borderline' velopharyngeal competence and again it could be argued that in such cases variation in nasal speech may indicate potential for improvement using speech therapy.

1.5 Conclusion

Nasal speech is a speech disorder, with a spectrum of severity, caused by dysfunction of the soft palate (VPD). As speech is considered to be a core life skill, nasal speech can significantly impact on quality of life in both children and adults, for some resulting in low self-esteem, bullying and withdrawal. This chapter has outlined the

available treatment options including surgery, prosthetics and other non-surgical approaches, including speech therapy, and highlights the fact that some patients face the prospect of no treatment if surgery is contraindicated or declined.

The lack of evidence for speech therapy as a primary treatment for nasal speech has been discussed but also the view that there may be a subgroup of patients who do demonstrate potential for improvement in speech without surgical treatment. This may be an important consideration for individuals when surgery is not an option or even as a less invasive alternative to surgery, in some cases. The following chapter will therefore describe a series of literature reviews of the evidence relating to speech therapy and other non-surgical treatments for nasal speech, as well as the wider context of the use of feedback approaches in speech and language therapy treatment.

Chapter Two: Speech Therapy and Nasal Speech

2.1 Non-Surgical interventions for nasal speech

2.2. Introduction

The previous chapter describes the nature of nasal speech, its potential for negative impact on the individual and the range of treatment options available. It may be that there is greater scope for the use of speech therapy and other non-surgical treatments, and this chapter examines the existing evidence for these approaches. The second section of the chapter then reviews the evidence from the wider speech and language therapy literature, specifically in relation to the use of feedback and visual biofeedback. The findings are considered along with the methodological issues affecting the research to date, in order to determine the outstanding questions in this field and the most appropriate methods of addressing them.

2.3 The literature review

A broad inclusion criterion was employed in this literature review as there are few studies in this field at higher levels of the evidence hierarchy. This is not surprising and reflects the research context across the speech & language therapy profession as a whole (Hayhow 2011; Reilly, 2004; Lewison & Carding, 2003) and in relation to nasal speech.

The most up to date evidence source potentially comes from a systematic review carried out by Bessell et al. (2013) which examined the evidence for speech therapy in children with cleft palate. The inclusion criteria of the Bessell review was any speech therapy approach, at any age, time and setting. However, single case studies and studies with five participants or less were excluded, as were studies that included subjects other than children with cleft palate and those with no reported speech

outcome. Only six papers were therefore identified from 1241 citations as meeting these criteria and none related to interventions for nasal speech. This approach to the consideration of intervention research has been criticised by Pring (2004) who argues that systematic reviews often omit the detail of interventions and discourage the necessary small scale, staged approach appropriate to speech therapy research. Garrett and Thomas (2006) challenge Pring's assumption that systematic reviews only address effectiveness questions and experimental studies. They argue for a broader view of their value with the emphasis on identifying what is, and what is not known using appropriate methods of synthesis. Roulstone et al. (1999) and Reilly (2004) acknowledge that the practicalities of investigating speech therapy interventions are complex whilst Turner et al. (2007) comment on the growing importance of evidence based practice as clinicians and other stakeholders desire to know which interventions work. Similarly Garrett and Thomas (2006) note that it is challenging to work out the state of the art and to ensure that an appropriate study design and previous knowledge are used. This requires the development of skills in literature searching and critical appraisal (Straus, 2007) but also awareness that the process of making sense of evidence is not straightforward and may be subject to professional bias and social prejudice (Cohen & Gureje, 2007).

In view of the factors outlined above the approach taken in this literature review was to allow for the inclusion of *all* relevant studies reporting on speech therapy and other non-surgical treatment approaches for nasal speech whilst acknowledging the risks relating to weak methodology. Studies were graded using the Australian National Health and Medical Research Council (NHMRC, 2009) framework for ranking evidence from case series (level IV) through to systematic review (level I) (Appendix A). The NHMRC framework is one of a number of frameworks available for ranking research evidence, for example the Oxford Centre for Evidence Based Medicine levels of evidence (2009 and 2011) and the Canadian Interior Health traditional research evidence hierarchy (2006). Historically such frameworks have focused on the validity

of the results as determined by the research design. The NHMRC framework was selected as it does allow for the evaluation of a range of studies and methodologies including those that would not be considered in a systematic review but are representative of the existing evidence base. Evans (2002) supports this broader perspective and cautions that the quality of research must also be evaluated, irrespective of the research design.

2.3.1 Search strategy

An initial literature review was conducted as outlined in Table 2-1.

Table 2-1: Literature review search strategy (nasal speech)

Online Database	**Specific Journals** (Electronic search)	**Hand Search**
CINAHL Ovid OldMedline Ovid Medline Ovid Embase Classic Journals@Ovid Cochrane Clinical Trials	Cleft Palate-Craniofacial Journal International Journal of Language & Communication Disorders Journal of Speech, Language and Hearing Research	Personal Files Cleft Palate texts Speech and Language Therapy texts
Primary search term = 'nasal speech' **Secondary search terms** = 'velopharyngeal' 'hypernasality' 'speech therapy' 'cleft palate' and 'treatment'. Truncation and Boolean 'AND' were applied to maximise the relevance of the search results.		
Inclusion criteria= Intervention: Speech therapy or another non-surgical treatment approach designed to eliminate nasal speech and/or improve palate function for speech.		

2.4 Results

A total of sixty-four citations were identified from initial searches of the electronic databases after duplicates were removed, with an additional eight papers from hand searching. Of these, thirty-five fulfilled the criteria of an intervention paper relating to speech therapy or other non-surgical treatment for nasal speech (Table 2-2 and 2-3) and thirty papers which did not meet these criteria were excluded. Studies involving prosthetic appliances were only included if the aim was to directly influence palate function and or reduce nasal speech through a reduction programme. The remaining seven papers are descriptive accounts of treatment approaches and guidelines and these are summarised in Table 2-4. The intervention papers are presented according to Cole's direct versus indirect intervention taxonomy as described in Chapter One (Cole, 1979), as far as this is clear from the information given. Where an intervention includes both direct and indirect approaches this is described as a combined intervention. There are fourteen direct intervention studies, eighteen indirect intervention studies and three combined (direct/indirect) intervention studies.

Full details of all the intervention studies are presented in Table 2-2 and 2-3. including date and source, population studied, intervention details, outcome measures, results and NHMRC level. The studies are grouped according to the type of intervention used; exercises, palate stimulation, resistance training, prosthetic training, nasendoscopic biofeedback, instrumental biofeedback and speech therapy approach. Full listing of abbreviated journal titles is included on page xxiii. The evidence from the studies is then discussed in depth.

Table 2-2: Direct Intervention Studies

Exercises

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
1	Massengill & Quinn	1974 CPJ	Single case study Pre~ post treatment comparison	n=1 18 yr old male Non cleft VPD	Sucking exercises 10 mins per day	No objective measures.	Reported improvement only. Sucking exercise not described.	IV
2	Massengill et al.	1968 CPJ	Case series Pre~ post treatment comparison	n=13 Cleft palate VPD Age range 8 to 18 yrs.	Non-speech exercises: Gp A: Blowing Gp B: Sucking Gp C: Swallowing 20 mins per day for 27 days	Outcome measure = size of VP gap, no discussion of validity. No perceptual measure.	Reduction in VP gap for swallowing group only. Concurrent speech therapy.	IV
3	Powers & Starr	1974 CPJ	Case series Pre~ post treatment comparison	n=4 Cleft palate VPD Age range 8 to 11 yrs No control group	Non-speech exercises, blowing, sucking, gagging, swallowing – 6 wks (4x day, 5x week)	VP gap measurements from lateral X-ray. Perceptual measure 12 trained listeners (students) – blinded assessment.	VP closure had to be achievable prior to intervention. Palate length not found to be a good indicator of potential for VP closure. Non-significant results.	IV

Table 2-2: Direct Intervention Studies continued

Palate Stimulation

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
4	Peterson	1974 CPJ	Controlled quasi-experiment - pre and post comparison	Gp A= 5 subjects cleft palate VPD Gp B= 5 cleft palate subjects, no VPD Gp C=10 subjects Control- no cleft or VPD	Tactile versus electrical stimulation of the palate	Lateral X-ray. Evaluation of palatal elevation by consensus (2 judges).	One subject selected for 'Tactile' stimulation only but rationale not given. Variable results. Control group had no palatal pathology but did have speech 'defects' - not defined.	III
5	Weber et al.	1970 PRS	Case series Pre~ post treatment comparison	n=34 Age range 4 to 50 yrs 23 cleft palate VPD 11 non cleft VPD No control group.	Electrical stimulation of palate (Muscle stimulation using electrical current)	VP closure from lateral X-ray. Perceptual and instrumental outcome measures.	Only 7 of the 34 subjects followed up. Only 1 subject showed reduction in hypernasality. 16 subjects had concurrent therapy using 'speechometer' to provide visual feedback.	IV

Table 2-2: Direct Intervention Studies continued

Resistance Training

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
6	Lubit & Larsen	1971 JSHD	Case series **Pre~ post treatment comparison**	n=4 Cleft palate **Age range 8 to 12 yrs**	Palatal Stimulator: inflatable palate exerciser Treatment duration from 2 months to 2 yrs	Lateral X-ray. No perceptual measure	Discussion of 2 types of palatal stimulator. Reported changes in soft palate thickness and function No discussion of speech outcomes.	IV
7	Lubit & Larsen	1969 CPJ	Single case study Pre~ post treatment comparison	n=1 9 yr old girl Cleft palate VPD	Palatal Stimulator: inflatable palate exerciser 6 months of therapy, 5-6 sessions per day	Lateral X-ray. Perceptual and instrumental outcome measures. No discussion of reliability. No blinding	Reported improvement in articulation but not hypernasality – attributed to inadequate rating scale. Increase in palate length and change in contour.	IV
8	Fisher	2004 JAHS&P	Case series Pre~ post treatment comparison	n=5 Non cleft VPD Age range 10-44yrs No control group	Prolonged Nasal Cul-De-Sac with High Pressure Speech Acts (P.I.N.C.H.) 6 sessions over 3-4 weeks	Instrumental outcome measure. No perceptual assessment.	Reduction in post treatment nasalance scores for 3/5 subjects. Theoretical basis for treatment described. Long term follow up 2/5 subjects.	IV

Table 2-2: Direct Intervention Studies continued

Resistance Training

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
9	Cahill et al.	2004 JHTR	Case series Pre~ post treatment & follow up comparison	n= 3 Adults with traumatic brain injury & nasal speech	Continuous Positive Airway Pressure (CPAP) 4 times per week for 4 wks	Perceptual and instrumental outcome measures. Reliability reported. Assessors blinded.	2/3 some improvement; reduction in nasality or increase in intelligibility. Improvement in VP function inferred.	IV
10	Kuehn et al.	2002 CPCJ	Case series Pre~ post treatment comparison	n=43 Cleft palate VPD Age range 3 to 23 yrs No control group	Continuous Positive Airway Pressure (CPAP) 8 weeks treatment, 6 times per week	Perceptual and instrumental outcome measures. Blinded assessment.	Intervention well described. Poor correlation perceptual measure and instrumental measure. Net reduction in hypernasality but variability.	IV

Table 2-2: Direct Intervention Studies continued

Prosthetic Training

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
11	Tachimura et al.	1999 CPCJ	Cohort study	n=8 Cleft palate VPD Age range 11-38yrs. No control group	Prosthetic palatal appliance Gp 1:palatal lift Gp2: palatal bulb	Palate muscle activity measured (EMG) during blowing.	Change in muscle activity observed.	IV
12	Weiss	1971 CPJ	Retrospective case series	n=20 14 cleft VPD 6 non cleft VPD	Prosthetic palatal appliance -reduction programme	Descriptive only.	No detailed results.	IV
13	Witt et al.	1995 CPCJ	Case Series Pre~ post treatment comparison	n=25 12 cleft VPD 13 non cleft VPD Age range 4 to 45 years	Prosthetic palatal appliance Mean duration 4.4 months	Pre & post endoscopy ratings of VP closure. Reliability of blinded raters poor to good.	70% no change, 15% better, 15% worse. Concurrent speech therapy.	IV
14	Wolfaardt et al.	1993 JPD	Case Series Pre~ post treatment comparison	n=32 VPD (aetiology not specified)	Prosthetic palatal appliance reduction program	Outcome measures only reported with palatal appliance not reduction phase.	Minimal subject information. 66% reported to have benefitted from program. 47% went on to surgery to correct VPD.	IV

Table 2-3: Indirect Intervention Studies

Nasendoscopic Biofeedback

No.	Author	Date/ Source	Design	Population	Intervention (Indirect)	Outcome Measures	Results	NHMRC Level
1	Brunner et al.	2005 CPCJ	Case series. Pre~ post treatment & follow up comparison.	n= 11 Cleft palate VPD Age range 7 to 30 yrs. 6 subjects had phoneme specific VPD. No control group.	Nasendoscopic biofeedback. Range from 2 to16 sessions. VP closure had to be achievable prior to intervention.	Nasendoscopy: binary rating of VP closure by 3 raters. Reliability reported. Perceptual assessment by patient questionnaire only.	Significant improvement VP closure from 5% pre intervention to 91% post intervention and 86% at 6month follow up. 3 subjects had concurrent speech therapy.	IV
2	Shelton et al.	1978 CPJ	Case studies Pre~ post treatment comparison	n=2 17yrs cleft palate VPD. 12yrs non-cleft VPD. No control group	Nasendoscopic biofeedback. Range from 6 to15 sessions	Nasendoscopy; rating of palatal closure. No perceptual assessment. Blinding of assessors.	Improvement in VP closure reported but not maintained. Training limited to sounds & syllables only.	IV
3	Siegel-Sadewitz & Shprintzen	1982 CPJ	Single case study	n=1 'normal' adult female	Nasendoscopic biofeedback	Nasendoscopy; categorisation of VP closure. Blinding of assessors.	Demonstrated that subject could voluntarily control VP closure.	IV
4	Witzel	1989 CPJ	Case studies	n=3 Adults with cleft palate VPD	Nasendoscopic biofeedback.	Nasendoscopy; evaluation of VP closure.	Descriptive results only – improvement in all cases. 2 subjects received speech therapy between sessions.	IV

Table 2-3: Indirect Intervention Studies continued

Nasendoscopic Biofeedback

No.	Author	Date/ Source	Design	Population	Intervention (Indirect)	Outcome Measures	Results	NHMRC Level
5	Witzel et al.	1988 IJPO	Single case study Pre~ post treatment comparison	10 year old girl Phoneme specific nasality (non-cleft VPD)	Nasendoscopic biofeedback.	Nasendoscopy; evaluation of VP closure.	1 session only required VP closure for all speech sounds including /s/	IV
6	Yamaoka	1983 JMS	Case series Pre~ post treatment comparison	n=59 Age range 8 to 45 yrs. Cleft palate VPD	Nasendoscopic biofeedback	Nasendoscopy; rating of VP closure. No blinding of assessors.	Results unclear as categories not explained.	IV
7	Ysunza et al.	1997 IJPO	Cohort study 2 groups	n=17 Cleft palate VPD Age range 11 to 13 yrs	Speech therapy with or without nasendoscopic biofeedback. ST 3 x per week Biofeedback additional 2x week	Nasendoscopy; lateral pharyngeal wall movement.	Unclear results. Mixed methods – treating articulation and VPD. No definition of compensatory articulation. All subjects also underwent surgery for VPD.	IV

Table 2-3: Indirect Intervention Studies continued

Instrumental Biofeedback

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
8	Daly & Johnson	1974 JSHD	Case series Pre~ post treatment comparison	n=3 Children with learning disability & nasal speech. Cleft palate1 1 Down's Syndrome unspecified 1. Age 13-14yrs. No control group.	The Oral Nasal Acoustic Ratio (TONAR). Visual biofeedback. 2 subjects 30 sessions, 1 subject 15 sessions over 3 weeks	Perceptual and instrumental outcome measures. No blinding of assessors in perceptual assessment.	3/3 reduction in hypernasality immediately post treatment. No follow up assessment. Visual and verbal feedback in intervention.	IV
9	Fletcher	1972 JSHD	Case studies Pre~ post treatment comparison	n=2 23yrs Non-cleft VPD. 15yrs Cleft palate VPD No control group	TONAR II. Visual biofeedback	Instrumental outcome measure. No perceptual assessment.	Reduction in % nasality for both subjects. Visual and verbal feedback in intervention.	IV

Table 2-3: Indirect Intervention Studies continued

Instrumental Biofeedback continued

No.	Author	Date/ Source	Design	Population	Intervention (Direct)	Outcome Measures	Results	NHMRC Level
10	Fletcher & Higgins	1980 JSHD	Case series Pre~ post treatment comparison	n=12 Students with hearing impairment and nasal speech Age range 10 to 18 yrs No control group	TONAR II Visual biofeedback 14 sessions (maximum)	Instrumental outcome measure. No perceptual assessment.	Variable results; 5 subjects significant reduction in nasalance, 3 subjects moderate reduction, 4 subjects little or no change. Discussion of possible factors underlying variability in this population.	IV
11	Kunzel	1982 FPL	Case Series	n=4	Visual feedback Velograph	Timing of VP movement. No perceptual assessment.	Subjects improved ability to modify timing of velopharyngeal movement in response to feedback.	IV

Table 2-3: Indirect Intervention Studies continued

Instrumental Biofeedback

No.	Author	Date/ Source	Design	Population	Intervention (Indirect)	Outcome Measures	Results	NHMRC Level
12	Main et al.	1999 IJLCD	Single case study Crossover design	Single case study 52 yr old male with acquired VPD secondary to tumour removal	6 weeks of conventional weekly ST compared with 6 weeks of SNORS visual biofeedback	Perceptual outcome measure was intelligibility rating. Instrumental outcome measure using SNORS. No blinding of assessors.	'Conventional' therapy included non-speech tasks. SNORS therapy described as significantly more effective than conventional therapy but reliability and validity of SNORS unclear. Study carried out by SNORS designers.	IV
13	Moller et al.	1973 JSHD	Single case study Pre~ post treatment comparison	n=1 12 yr old with cleft palate VPD	Visual biofeedback relating to velar elevation 15 sessions	Pre and post lateral X-rays. Perceptual assessment.	Production of one vowel sound only studied. Velar elevation increased but no change in VP gap or perceptual assessment. Procedure well described.	IV

Table 2-3: Indirect Intervention Studies continued

Instrumental Biofeedback continued

No.	Author	Date/ Source	Design	Population	Intervention (Indirect)	Outcome Measures	Results	NHMRC Level
14	Roll	1973 JABA	Case study Crossover design Pre~ post treatment comparison	n=2 Cleft Palate VPD 10 & 13 yrs No control group	Differential visual feedback (feedback versus no feedback) 5 x per week	Vibration of walls of nasal cavities measured, validity not discussed. Perceptual assessment.	Decrease in nasalized sound productions in both cases. Concurrent speech therapy	IV
15	Ruscello et al.	1991 JSHR	Single case study Pre~ post treatment comparison	n=1 Adult with phoneme specific nasality (non-cleft VPD)	Visual biofeedback	Perceptual and instrumental measures. Blinding of assessors.	Correct articulation of 's' achieved, nasal emission eliminated. Discussion of approach in relation to VPD.	IV

Table 2-3: Indirect Intervention Studies continued

Instrumental Biofeedback

No.	Author	Date/ Source	Design	Population	Intervention (Indirect)	Outcome Measures	Results	NHMRC Level
16	Shprintzen et al.	1975 JSHD	Case Series Pre~ post treatment comparison	n=4 3 – cleft palate 1 – non-cleft VPD. Age range 4 to 19 yrs. No control group.	Instrumental visual biofeedback – 'Scape-Scope' Range 22-36 sessions	No perceptual assessment. Descriptive evaluation only.	2 consistent VP closure, 1 inconsistent VP closure, 1 did not complete study. Concurrent speech therapy	IV

Speech Therapy Approaches

17	Shelton et al.	1969 JSHD	Case series Pre~ post treatment comparison	n=17, 6 cleft palate & 3 non-cleft VPD. Age 6-12 yrs. Control group (8)-no therapy	Articulation therapy 2x per week over 6 months	Measurement of VP gap and tongue position from lateral X-ray. No perceptual assessment.	Small sample but group means presented. No effect on VP closure but improvements in articulation reported.	III
18	Wenke et al.	2010 IJCD	Randomised Controlled Trial	n=10 (subset of larger study) Adults with non-progressive dysarthria and nasal speech.	Gp 1: Lee Silverman Voice Therapy (LSVT) (5) Gp2: Traditional dysarthria therapy (5)	Perceptual and instrumental outcome measures. Blinded assessors.	Findings comprehensively discussed. Inconclusive results, some reduction in hypernasality. Traditional therapy with multiple strategies.	II

Table 2-4: Combined approach (Direct/Indirect) Intervention Studies

Combined Intervention

No.	Author	Date/ Source	Design	Population	Intervention (Combined)	Outcome Measures	Results	NHMRC Level
1	Tash et al.	1971 CPJ	Cohort study 3 groups	n=6 4: normal VP closure. 2: non-cleft VPD Age range 4-8 years.	Direct/indirect intervention. Training pharyngeal wall movement. Visual/performance Feedback	Outcome measures questionable (sensitivity & specificity).	Increased pharyngeal wall movement but not increased VP closure.	IV
2	Tudor & Selley	1974 BJDC	Case series	n=24 15 children; 4 with cleft palate & 11 non-cleft VPD. 9 dysarthric adults with nasal speech	Direct/indirect intervention. Visual feedback with palatal appliance	Descriptive only.	VP closure in some cases. No detailed results.	IV
3	Yules & Chase	1969 PRS	Case series Pre~ post treatment comparison	n=30 21 cleft palate VPD 9 non cleft VPD Age range 4 to 50 years.	Direct/indirect intervention - Electrical stimulation and visual feedback	Instrumental (airflow) No perceptual assessment. No blinding of assessors.	60% eliminated hypernasality, 80% reduced nasal airflow post intervention. Concurrent speech therapy	IV

Table 2-5: Treatment of nasal speech/VPD: Descriptive papers

No.	Author/ Date	Source	Title	Comments
1	Bivati, M.J 2002	eMedicine	Velopharyngeal Insufficiency	**Descriptive** review defining VPD, and outlining assessment and treatment approaches. States that primary indication for surgical treatment is a structural palate problem. Surgical complications are discussed but not clearly referenced.
2	Dworkin et al. 2004	Language, Speech and Hearing services in Schools	Velopharyngeal Dysfunction: Speech characteristics, Variable Etiologies, Evaluation Techniques, and Differential Treatments	**Descriptive** review. Describes indications for surgical and speech therapy treatment. No reference to evidence base for therapy approaches.
3	Johns et al. 2003	Plastic and Reconstructive Surgery	Velopharyngeal Incompetence: A Guide for Clinical Evaluation	**Descriptive** guide. Aims to aid assessment and differential diagnosis. Describes clinical and instrumental evaluation.
4	Kuehn & Moller 2000	Cleft Palate & Craniofacial Journal	Speech and Language Issues in the Cleft Palate Population: The State of the Art	**Literature** review. Assessment and management in cleft palate population. Comprehensive descriptive review.

Table 2-5: Treatment of nasal speech/VPD: Descriptive papers continued

No.	Author/ Date	Source	Title	Comments
5	Kummer 2004	Current Opinion in Otolaryngology & Head and Neck Surgery	Velopharyngeal dysfunction: current thinking on the cause, effect, assessment and treatment	**Descriptive** review. Clearly states indications for speech therapy treatment in VPD but reference only to one small study of treatment efficacy.
6	Ruscello 2006	Journal of SLP & Applied Behaviour Research	Treatment of velopharyngeal closure for speech: discussion and implications for management	**Descriptive** review. Comprehensive review of treatment studies. Discussion of theoretical rationale for 'muscle' treatments including concepts of 'plasticity' and 'flexibility'.
7	Wyatt et al. 1996	British Journal of Plastic Surgery	Cleft palate speech dissected: a review of current knowledge and analysis	Descriptive review.

2.5 Discussion

2.5.1 Design

A range of study types were identified in this review including single case studies, prospective cohort studies and prospective case series; one randomised controlled trial (Wenke, 2010) was also identified. The majority of studies are case series with pre and post-intervention designs and are therefore at the lowest level of the NHMRC hierarchy. This framework does not differentiate between the quality of the studies at this level but gives an indication of the relative weakness of the evidence. The most recent study dates from 2005 but only five studies were identified in the last ten years and nineteen studies are thirty years old or more. This both reflects and informs the lack of support within the speech therapy profession for speech therapy treatments for nasal speech.

2.5.1.1 Sample

Many of the studies reviewed have limited sample sizes; twenty-one studies had less than ten subjects and of these seven are single case studies. There are nine larger studies, involving twenty subjects or more, with the largest study having fifty-nine subjects (Yamaoka, 1983). However four of these larger studies had inadequate outcome measures (Weiss 1971; Tudor & Selley, 1974; Yamaoka 1983; Wolfaardt, 1993), another three were confounded by concurrent additional speech therapy treatment (Yules & Chase, 1969; Weber et al., 1970; Witt et al., 1995), and only one had a control or comparison group (Peterson, 1974) but in this study assessors were not blinded.

In eleven of the thirty-five studies the subjects had nasal speech associated with cleft palate VPD compared to seven studies where subjects had nasal speech and non-cleft VPD. A further nine studies included heterogeneous subjects but not necessarily in comparison groups. There were also three studies involving subjects with nasal speech associated with hearing impairment, traumatic brain injury or acquired

dysarthria. The heterogeneity of subjects makes it very difficult to evaluate whether the interventions studied are equally suitable for different types of VPD (i.e. cleft or non-cleft). Positive results were reported for individuals in eighteen studies, irrespective of the aetiology of their nasal speech. This could be seen as a positive indicator of the need for further investigation were it not for the fact that methodological weaknesses seriously undermine the results in all of the studies. However, a well-designed study using a pre and post-test design could allow for subject variability by comparing each individual with themselves before and after the intervention.

2.5.1.2 Outcome Measures

The outcome measures employed in the studies reviewed were variable. Only fourteen out of thirty-five studies included both perceptual and instrumental measures. A perceptual measure is a rating scale used to judge speech across a variety of parameters depending on the focus of the measure. The emphasis is on measures that are considered to be objective i.e. measurement of velopharyngeal gap size or rating of velopharyngeal closure using lateral X-ray or nasendoscopy, as well as the use of instrumental measures to give a numerical result, such as nasometry and manometry. Twenty-eight out of thirty-five studies included at least one objective measure but the validity and reliability of the measures is often not reported and blinding of assessors was stated in only eight studies. This introduces the risk of substantial bias in the majority of the studies.

2.5.1.2.1 Assessment of speech (Perceptual)

Only eleven studies clearly reported the use of a perceptual speech measure, despite the aim of the interventions being to treat nasal speech. Perceptual measures are by their very nature subjective as they involve a judgement based on what is heard in speech, by expert or lay listeners or a combination of the two. Despite this subjectivity perceptual measures are considered to be the 'gold standard' in the assessment of disordered speech (Kuehn & Moller, 2000; Bhuta et al., 2004; Sell, 2005). It is clear that acoustic and perceptual measures cannot speculate on the effect of any

intervention on velopharyngeal function, therefore any study of treatment for nasal speech must include a perceptual speech assessment. Sell and Sweeney (2001) go as far as to state that perceptual judgements of speech production are in fact central to the interpretation of other analyses. This is especially important as the correlation between different types of speech outcome measures can be poor and this view is supported by Kuehn (2002) who emphasises the clinical importance of nasal speech based on perceptual evaluation.

2.5.1.2.2 Subjective outcomes

It is also of note in this review that only one study, from Brunner et al. (2005) included a subjective or 'patient reported' outcome measure, in the form of a questionnaire. There has been a noticeable increase in the use of such measures in speech therapy research over the last few years and a growing trend of triangulating research methods in response to the multifaceted nature of health and health services (Bowling, 2002; Parahoo, 2006). An important aspect of future studies will be the patient's perception of their speech symptoms and any functional impact of the intervention.

2.5.2 Interventions

All but one of the thirty-five primary studies was concerned with the evaluation of an intervention for nasal speech. The remaining study aimed to evaluate the response to nasendoscopic biofeedback in a 'normal' adult speaker. The majority of studies adopt a quasi-experimental design in terms of aiming to evaluate the effect of an intervention. However, only five of the thirty-five studies included a control or comparison group.

Finally, a major confounder in the reported studies is concurrent 'conventional' or 'traditional' speech therapy treatment; this was evident in eight of the studies reviewed. Therefore, although an attempt is made to evaluate the effect of a specific intervention a clear treatment effect cannot be established. In the context of efficacy research confounding is a form of bias that involves the distortion, or mixing, of

effects (Rothman, 1976). In the studies reviewed the inclusion of another, related but uncontrolled intervention in the form of speech therapy treatment means that any positive findings cannot be attributed to the intervention of interest.

A comparison of the key quality criteria for all thirty-five studies is shown in Table 2-6., with the studies presented chronologically, from most recent to oldest. The quality criteria selected are informed by the CONSORT (2010) statement and checklist for clinical trials (Appendix B). The use of this checklist has been shown to be associated with improved reporting of trials in a systematic review by Plint et al. (2006). In the context of this literature review the key criteria are considered to be:

Methods:

- Intervention: was there a control or comparison group?
- Intervention: was speech therapy controlled?
- Outcomes: were outcomes adequate and clearly defined?
- Outcomes: were assessors blinded?

Results and interpretation:

- Results: were results fully reported?
- Results: were variable or negative results discussed?

Table 2-6: Key quality criteria for intervention studies reviewed

No.	Main author	Date	n= ≥10	Control or comparison group	Adequate Outcome Measures*	Results reported	Blinding of assessors reported	Speech therapy controlled
1	Wenke	2010	✓	✓	✓	✓	✓	✓
2	Brunner	2005	✓	x	✓	✓	?	x
3	Cahill	2004	x	x	✓	✓	✓	✓
4	Fisher	2004	x	x	x	✓	?	✓
5	Kuehn	2002	✓	x	✓	✓	✓	✓
6	Main	1999	x	x	✓	✓	x	✓
7	Tachimura	1999	x	x	x	✓	?	✓
8	Ysunza	1997	✓	x	x	x	?	✓

*Minimum of instrumental and perceptual measure reported, but not validity or reliability

Table 2-6: Key quality criteria for intervention studies reviewed continued

No.	Main author	Date	n= ≥10	Control or comparison group	Adequate Outcome Measures*	Results reported	Blinding of assessors reported	Speech therapy controlled
9	Witt	1995	✓	x	x	✓	✓	x
10	Wolfaardt	1993	✓	x	x	✓	?	✓
11	Ruscello	1991	x	x	✓	✓	✓	✓
12	Witzel	1989	x	x	x	x	?	x
13	Witzel	1988	x	x	x	✓	?	✓
14	Yamaoka	1983	✓	x	x	x	x	✓
15	Kunzel	1982	x	x	x	✓	?	✓
**16	Siegel-Sadewitz	1982	x	x	✓	✓	✓	✓
17	Fletcher & Higgins	1980	✓	x	x	✓	?	✓

*Minimum of instrumental and perceptual measure reported, but not validity or reliability

** Study of one 'normal' subject

Table 2-6: Key quality criteria for intervention studies reviewed continued

No.	Study	Date	n= ≥10	Control or comparison group	Adequate Outcome Measures*	Results reported	Blinding of assessors reported	Speech therapy controlled
18	Shelton	1978	x	x	x	✓	✓	✓
19	Shprintzen	1975	x	x	x	✓	?	x
20	Daly & Johnson	1974	x	x	✓	✓	x	✓
21	Massengill & Quinn	1974	x	x	x	x	x	✓
22	Peterson	1974	✓	✓	✓	✓	x	✓
23	Powers & Starr	1974	x	x	✓	✓	✓	✓
24	Tudor & Selley	1974	✓	x	x	x	?	✓***
25	Moller	1973	x	x	✓	✓	?	✓
26	Roll	1973	x	x	✓	✓	?	x
27	Fletcher	1972	x	x	x	✓	?	✓

*Minimum of instrumental and perceptual measure reported, but not validity or reliability

***Combined direct & indirect intervention

Table 2-6: Key quality criteria for intervention studies reviewed continued

No.	Study	Date	n= ≥10	Control or comparison group	Adequate Outcome Measures*	Results reported	Blinding of assessors reported	Speech therapy controlled
28	Lubit & Larsen	1971	x	x	x	x	?	✓
29	Tash	1971	x	✓	x	✓	?	✓***
30	Weiss	1971	✓	x	x	x	?	✓
31	Weber	1970	✓	x	✓	x	?	x
32	Lubit & Larsen	1969	x	x	✓	✓	x	✓
33	Shelton	1969	✓	✓	x	✓	?	✓
34	Yules & Chase	1969	✓	x	x	✓	x	x***
35	Massengill	1968	✓	✓	x	x	x	x

*Minimum of instrumental and perceptual measure reported, but not validity or reliability

***Combined direct & indirect intervention

It is clear from Table 2-6 that methodological issues seriously compromise the findings presented in these thirty-five studies and limit the conclusions that can be drawn from them (Fathalla & Fathalla, 2004). Only five studies had a control or comparison group and in eight studies concurrent speech therapy was delivered. There was considerable variation in outcome measures used; as a minimum studies were evaluated according to whether both perceptual and instrumental measures were included. Only fourteen studies reported this and this does not take account of the quality of the measures used in terms of validity and reliability.

2.5.2.1 Definition of interventions

The use of Cole's direct versus indirect classification of interventions (Cole, 1979) allows for consideration of the aim of the intervention and the underlying theory of how it might have an effect, although this is often not made explicit in the papers reviewed. Whilst some studies clearly state the rationale for the treatment approach others are less clear and in some it is evident that both direct and indirect approaches are employed. Cahill et al. (2004), for example do clearly state the aims of their intervention as 'to exercise and strengthen the palate in the context of motor impairment' and also 'to increase speech quality and functional intelligibility'. As Ruscello (1982) comments, it can be difficult to make a clear distinction between approaches reported in the literature and to establish the goal of the intervention under investigation. This conceptualisation is key but is clearly lacking in many previous studies. Only one study in the review comprehensively evaluated an indirect intervention (using speech tasks and feedback techniques) designed to reduce nasal speech in a defined population, with some positive results. This randomised controlled trial by Wenke (2010) allocated patients to two groups receiving different therapy approaches. The aim of the study was clearly stated as to investigate the short and long-term effects of the two approaches on hypernasality in speakers with dysarthria. Valid outcome measures were used (perceptual rating and instrumental measure), reliability reported and evaluation was blinded. This degree of clarity and

methodological rigour was not evident in any of the other studies reviewed and further highlights the need for a well-designed intervention study in this area.

2.5.2.2 Direct Interventions

Fourteen of the thirty-five intervention studies describe the use of a direct intervention alone and three others describe a direct approach used in combination with an indirect approach (Tables 2-2 and 2-4)

2.5.2.2.1 Exercises

Three early studies report on the use of exercises to stimulate palate function. Powers and Starr (1974) studied four subjects with repaired cleft palate undergoing a 6 week programme of palate exercises (blowing and sucking). They found no significant differences in velopharyngeal gap or hypernasality immediately following the programme. In contrast Massengill (1968) and Massengill and Quinn (1974) reported improvements in a total of fourteen individuals using sucking and swallowing exercises. However in both studies subjects received concurrent articulation therapy so it is not possible to attribute the results to the intervention.

2.5.2.2.2 Palate stimulation

Stimulation of the palate to improve VP function for speech, not with exercise, but using an external device has been studied with mixed results in three studies. Weber (1970) describes the use of electrical stimulation in a study of thirty-four subjects, twenty-three with cleft VPD and eleven with non-cleft VPD. However only seven subjects were followed up with one reported to show a reduction in hypernasality. Peterson (1974) also attempted to evaluate the effect of electrical stimulation on palate movement and to compare this with tactile stimulation. This study did have a control group of subjects without VPD but with unspecified speech pathology. In addition only one subject was selected to receive tactile as opposed to electrical stimulation but the rationale for this was not given. In an even earlier study Yules and Chase (1969) used a combination approach of electrical stimulation and visual feedback with thirty subjects and reported a 'reduction' in nasal airflow in 80% of subjects and complete

elimination of hypernasality for 60%. Unfortunately the inclusion of both direct (electrical stimulation) and indirect (visual feedback) components means that it is not possible to interpret a treatment effect.

It is important to note that the above studies are over forty years old and since then the use of palate stimulation has been largely abandoned. The evidence for stimulation from these few studies is weak and the positive results reported by Yules and Chase (1969) are further compromised by the delivery of concurrent speech therapy as well as the lack of blinding in the assessment of outcomes.

2.5.2.2.3 Resistance training

Five studies employ the principles of resistance training to improve palate function. Fisher (2004) describes the use of positive intra-oral pressure and reports decreased hypernasality (nasal speech) in five subjects, using an instrumental outcome measure but no perceptual measure of speech. A different technique was adopted by Lubit and Larsen (1969, 1971) who conducted two small studies (one a single case study) using an inflatable device. The device consisted of an inflatable bag in an acrylic bite block designed to exert pressure on the palate and back of the throat. They reported a change in palate length and contour but no reduction in hypernasality. However only the 1969 study included a perceptual speech measure and assessors were not blinded. Both studies do attempt to use objective measurement of changes in the palate but the validity of the measures is unclear and both studies are very small.

Following similar principles of resistance training to these earlier studies, Cahill et al. (2004) report a small study of continuous positive airway pressure treatment (CPAP) where air pressure was delivered through a nose mask during speech tasks. There was a positive effect of the intervention for two out of three subjects. A larger study of CPAP by Kuehn et al. (2002) also found a net reduction in hypernasality, although results were variable across the forty-three subjects, with eight subjects showing an increase in hypernasality. However seventeen of the twenty-five subjects assessed at follow-up had a reduction in blinded hypernasality rating immediately after CPAP. This

type of approach therefore appears to offer potential benefit for some individuals. Participants in the studies described above included adults with nasal speech resulting from traumatic brain injury as well as children and adults with cleft palate. This suggests that more research is needed to establish clearer candidate suitability.

2.5.2.2.4 Prosthetic training

Five studies describe the use of prosthetic training to improve palate function. Weiss (1971) describes a palatal appliance (prosthetic) reduction programme involving twenty subjects. Weiss outlines use of a variety of perceptual and instrumental measures but the results are descriptive only. However the approach continued to attract interest and Wolfaardt (1993) carried out a study involving a prosthetic reduction programme with thirty-two subjects with VPD. Minimal subject information was given and the underlying aetiology of the VPD was not stated. However 66% were reported to have benefited from the programme although despite these positive results 47% went on to have surgery to correct the VPD.

A study by Witt et al. (1995) with twenty-five subjects found no evidence to support the use of palatal prostheses in stimulating velopharyngeal activity. Of the subjects 70% showed no improvement and 15% worse VP function after an average of four months use of the prostheses. However this study did not include a perceptual speech measure and subjects received concurrent speech therapy meaning that once again the effect of the prosthetic intervention cannot be evaluated. In contrast to these findings, Tachimura et al. (1999) did report increases in palate muscle activity as objectively measured using electromyography in eight subjects, but this was observed during blowing and not speech and therefore no direct inference can be made about an effect on speech.

Finally Tudor and Selley (1974) reported 'encouraging 'results in their study which examined the effect of two intra-oral palatal appliances in treating hypernasal speech. This intervention was a combination approach as the appliances were designed to

increase tactile feedback and provide visual biofeedback via a light activation unit in order to stimulate the palate muscles and reduce hypernasality. The appliances were used with four children with repaired cleft palate, eleven children with non-cleft VPD and nine adults with acquired dysarthria. Unfortunately despite clearly describing the appliances and procedures the authors present limited descriptive results. It is not clear which of the two types of appliance was used with which subjects and no individual results are presented. Tudor and Selley state that sensory perception of the soft palate was increased and subjects could control movements voluntarily but there is no description of the outcome measures used.

The limited evidence available from the studies described suggests that the use of prosthetics to actively stimulate palate function and reduce nasal speech is not effective. However as with the other types of direct interventions included in this review it is difficult to draw firm conclusions due to the methodological weaknesses of previous studies.

2.5.2.3 Indirect interventions

There are eighteen studies in the review that solely describe the use of an indirect intervention i.e. one designed to influence palate function but without specifically targeting velopharyngeal muscle function (Table 2-3). As previously described three further studies describe an indirect approach used in combination with a direct approach (Table 2-4). Indirect interventions include nasendoscopic biofeedback, instrumental biofeedback and speech therapy approaches.

2.5.2.3.1 Nasendoscopic biofeedback

Seven of the studies describing indirect interventions involve the use of nasendoscopic biofeedback. Results of this approach have generally been positive but study designs are weak. A single case study by Siegel-Sadewitz and Shprintzen (1982) demonstrated the ability of the subject to voluntarily control palate movement but the subject had normal palate anatomy and therefore the results cannot be applied to any clinical population. Shelton (1978), Witzel (1988, 1989) and Ysunza et al. (1997) all present

positive findings using nasendoscopic biofeedback with reports of improved velopharyngeal closure and/or speech. The Shelton (1978) study included only two adolescent subjects but did involve blinded rating of palate closure. Improvement in VP closure was reported in both cases but not maintained. Two studies by Witzel et al. (1988, 1989) were also very small with three and one subjects respectively. Once again outcome measurement took the form of rating of palate closure although variable rating systems makes comparisons across studies impossible. The later study by Ysunza et al. (1997) similarly reported rating of palate closure and was a larger study, with seventeen subjects with cleft related VPD. However assessors were not blinded making the outcomes susceptible to bias. Yamaoka (1983) also presents positive findings in a larger study of fifty-nine subjects with repaired cleft palate ranging from eight to fort-five years of age. This study evaluated palate function during speech and non-speech tasks but is again weakened by the lack of independent rating of outcomes and perceptual speech assessment.

The most recent study is from Brunner et al. (2005), once again presenting positive results but in a mixed group of subjects with cleft palate VPD and phoneme specific VPD. The latter, as previously described in Chapter One, describes the substitution of a nasal sound for an oral speech sound such as 's', caused by mislearning rather than an underlying structural deficit. However the potential impact of the study is seriously limited by the design weaknesses. Most importantly, in all seven studies, subjects received concurrent 'conventional' speech therapy and therefore the positive outcomes cannot be attributed to the visual biofeedback intervention.

2.5.2.3.2 Instrumental biofeedback

Nine studies in the review involved biofeedback approaches other than nasendoscopy but where the visual feedback is again the key mechanism in the intervention. All nine studies were small with samples ranging from one to twenty-four subjects, but with some positive findings. Daly & Johnson (1974) and Fletcher (1972) both report reductions in hypernasality for a small number of subject with nasal speech using an

instrumental system called TONAR which measured sound levels from oral and nasal channels. However a later study by Fletcher & Higgins (1980) using TONAR produced less consistent results. This could be related to the different nature of the subjects in this study, who were students with hearing impairment, or the total training time which was less than five hours for all subjects compared to fourteen to thirty sessions in the previous studies. Shprintzen et al. (1975) also produced variable results in three subjects using a low-tech instrument (Scape-Scope), which provides visual biofeedback of nasal air escape through a piece of plastic tubing placed in the nostril connected to a cylinder containing a foam piston. Unfortunately subjects in this study also received speech therapy. The Velograph is another instrumental visual biofeedback approach described by Kunzel (1982) and in this study all four subjects showed the ability to modify the timing of VP movement in response to the feedback.

Three of the remaining studies are single case studies. The earliest of these is Moller et al. (1973) who carried out a pre and post-treatment comparison of palate elevation using lateral X-rays in a twelve year old with nasal speech associated with cleft palate. Post-treatment assessment followed fifteen sessions of visual biofeedback and showed that palate elevation increased although there was no change in perceptual speech assessment. Main et al. (1999) describe an instrumental system called SNORS using visual biofeedback, which they compared with standard speech therapy. However there was no blinding of the assessors, who were also the designers of the SNORS (super nasal oral ratiometry system) system increasing potential for bias. The final case study is Ruscello et al. (1991) who used visual biofeedback to successfully treat an adult with phoneme specific VPD, further reinforcing the idea that in order for palate closure to be improved there must be evidence that this is achievable.

Roll (1973) is the only study in this category to use a crossover design, comparing treatment of nasal speech with and without visual feedback in two cases. However no

conclusions can be drawn about the intervention, or the feedback component, due to the delivery of concurrent speech therapy.

In summary, sixteen of the eighteen studies involving an indirect intervention used visual biofeedback. A further three studies describe combined interventions where visual biofeedback was used alongside a direct approach; muscle training, prosthetic appliance and electrical stimulation (Tash et al., 1971; Tudor & Selley, 1974; Yules & Chase, 1969). Results across all twenty-one studies, where visual biofeedback is a component, are variable but positive results are reported for some subjects in fifteen studies. This suggests that visual biofeedback may offer potential as a treatment modality for nasal speech. However there has been only one study using nasendoscopic visual biofeedback undertaken in the last ten years (Brunner et al., 2005) despite nasendoscopy being routinely available at regional cleft palate/VPD treatment centres and more speech and language therapists now independently undertaking this procedure (Sell et al., 2008). This suggests that there is potential for further investigation of this treatment approach. Furthermore there are existing instrumental devices e.g. nasometry, which can provide visual biofeedback specifically in relation to nasal speech and which are also widely available at cleft palate treatment centres. The literature review describes the use of similar instrumentation in the past, in the form of TONAR (Daly & Johnson, 1974; Fletcher, 1972) and the Velograph (Kunzel, 1982), with some evidence of positive effects. More rigorous investigation of either or both of these visual biofeedback approaches could be undertaken with the focus on a clearly defined protocol and comprehensive perceptual and instrumental assessment and analysis.

2.5.2.3.3 Speech Therapy approaches: Articulation and voice therapy

Two specific types of speech therapy treatment are reported as indirect approaches aiming to influence palate function and reduce nasal speech; articulation therapy and voice therapy. The use of articulation therapy specifically to influence palate function is described in one very early study by Shelton (1969). This found no effect of

articulation therapy on velopharyngeal closure in children, although articulation itself was improved. The specific techniques used in the articulation therapy are not described and no rationale given for how the approach might work.

In a number of later studies articulation therapy is provided alongside other interventions for nasal speech (Witzel, 1989; Ysunza et al., 1997; Brunner et al., 2005; Roll, 1973; Shprintzen et al., 1975). The specific effect of articulation therapy in treating nasal speech is therefore unclear, although the consensus is that there is no evidence to suggest articulatory treatment results in improvement in velopharyngeal function (Kuehn & Moller, 2000). It is clear however that there is a relationship between articulatory development and VPD (Pamplona, 2000), with certain articulation disorders considered to be compensatory behaviours secondary to the palate dysfunction. Therefore any attempt to evaluate a treatment for nasal speech will need to control for other related and potentially confounding treatments such as articulation therapy.

Voice therapy has been used, albeit minimally, to modify nasal speech, again using behavioural techniques. The study of Lee Silverman Voice Therapy (LSVT) by Wenke (2010) is the only randomised controlled trial in the review. Ten subjects were presented as a subset of a larger study of adults with non-progressive dysarthria, a condition which affects the rate, clarity and rhythm of speech. In the Wenke study subjects were randomly allocated to one of two groups receiving either LSVT (using voice therapy techniques) or traditional dysarthria therapy, to reduce nasal speech. The LSVT group involved the subjects using increased vocal loudness and maximum physiological effort whilst the traditional dysarthria therapy group involved multiple strategies to target specific impairments. The study is small so the results are inconclusive and the boundaries between LSVT and traditional dysarthria therapy somewhat blurred. However in three out of five subjects receiving LSVT nasal speech was reduced compared to one subject receiving traditional therapy. Further research is

needed in this area with larger groups and different therapy techniques in order to fully investigate the effect of behavioural voice therapy.

2.6 Summary of literature findings

The limitations highlighted by this literature review mean that there is insufficient evidence to support an immediate change in practice at this point. To date no study has provided unequivocal support for the use of a particular speech therapy or other non-surgical treatment for nasal speech and the majority of approaches outlined in this review have not been studied rigorously. Consequently although there is consensus that speech therapy and other non-surgical approaches alone cannot address a structural palate defect (Kummer, 2004; Dworkin et al., 2004; Ruscello, 2006), uncertainty remains about the possible benefits of some interventions for individuals with nasal speech. This is unsatisfactory as it means that individuals with nasal speech may be presented with the option of invasive surgery or prosthetic treatment or no treatment at all.

Despite the weak evidence overall the literature does lend support to the use of visual biofeedback and resistance training approaches, in that observable changes in palate function and reduction of nasal speech characteristics have been reported for some individuals. These promising findings suggest that further investigation is warranted and that some individuals do have the potential for functional speech improvement without surgery by using behavioural techniques.

The potential for the use of visual biofeedback has been highlighted, featuring in over half of the thirty-five studies reviewed, and it is also clear that this has not been fully explored to date. This type of intervention is supported by research in other areas of speech therapy including articulation and voice disorders and dysphagia (Marchant et al., 2008; Hagg & Larsson, 2004; Roy et al., 2003) and this related literature provides a stronger evidence base for the use of such techniques, albeit in different conditions.

The following section of this review will therefore explore this related evidence and discuss how this leads to the specific research questions and development of the intervention in this study.

2.7 Feedback in speech therapy

2.7.1 Background

The previous section has highlighted the use of visual biofeedback in the treatment of nasal speech associated with VPD, with positive results for some individuals but without conclusive evidence of efficacy. Feedback techniques are described across a wide range of speech and language therapy client groups and disorders, where feedback may be given verbally by the speech therapist in response to the individual's communication behaviour in relation to specified targets (performance feedback) or visually through the use of instrumentation and computer technology in response to physiological stimuli from the individual (visual biofeedback). This section examines the use of such feedback techniques in other conditions in order to determine whether further investigation of feedback treatment for nasal speech is justified, and if so what methods should be used.

2.7.2 Search strategy

In 2010 an initial search of the Cochrane database yielded fifteen systematic reviews, one protocol and one registered title using 'speech therapy' as the primary search term and 'feedback' and 'biofeedback' as secondary search terms (Appendix C). Previously, in 2006 El Dib and Atallah identified thirteen systematic reviews and two protocols covering a diverse range of disorders, including five systematic reviews related to speech and language therapy management of swallowing disorders (dysphagia) as well as three relating to hearing impairment. The remaining five reviews cover treatments for aphasia and dysarthria in adults and apraxia, cerebral palsy and speech and language delay or disorder in children. None of the systematic reviews' primary focus was on speech therapy treatments using feedback techniques. All concluded that the

evidence for the treatment under review was weak and that further large scale research was needed.

A further literature review was therefore carried out in order to identify individual studies relating primarily to the use of feedback in speech therapy, across all client groups and disorders. Studies were searched for in the following electronic databases, extending back over a twenty year period to 1990 (Table 2-7). Broad inclusion criteria were applied as in the preceding literature review in order to capture studies at the lower end of the research framework hierarchy, including single case studies. Once again the rationale for this approach relates to the current status of the speech therapy evidence base.

Table 2.7: Search strategy (Speech therapy/feedback)

Online Database	Specific Journals	Hand Search
CINAHL Ovid OldMedline Ovid Medline Ovid Embase Classic Journals@Ovid	Journal of Speech, Language and Hearing Research	Personal Files International Journal of Language & Communication Disorders (from 1989) Speech and Language Therapy texts

Primary search term = 'speech therapy'.

Secondary search terms = 'feedback' and 'biofeedback'.

Truncation and Boolean 'AND' were applied to maximise the relevance of the search results.

2.8 Results

A total of twenty-four citations were identified from initial searches of the electronic databases after duplicates were removed. Three systematic reviews were identified, two relating to speech therapy for dysarthria in children and for adults with Parkinson's disease and one relating to electropalatography for articulation disorders associated

with cleft palate. Within these reviews only three studies met the rigorous inclusion criteria specified. One paper is a descriptive review of speech therapy using ultrasound. A further twenty primary studies were identified as fulfilling the criteria of a speech therapy study using feedback or visual biofeedback. The studies cover a range of disorders and client groups with the focus on speech disorder (voice and articulation) and aetiologies including cerebral palsy, hearing impairment, dysarthria (muscle weakness), cleft palate, Down's syndrome and vocal misuse. Fourteen of the studies are paediatric (children or adolescents) and ten relate to adult subjects.

Full details of all the speech therapy studies are presented (Table 2-8) including date and source, population studied, intervention details, outcome measures, results and NHMRC level. The studies are grouped according to the type of intervention used; visual biofeedback (electropalatography, ultrasound, electromyography, spectral feedback, electroglottography, Respitrace, electromagnetic articulograph, transnasal flexible laryngoscopy), performance feedback and speech therapy (including feedback). A full listing of abbreviated journal titles is included on page xxiii. The evidence from the studies is then discussed in depth.

Table 2-8: Speech and language therapy feedback studies

Visual biofeedback: Electropalatography (EPG)

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
1	Lee et al.	2009 Cochrane	Systematic Review	EPG for articulation disorders associated with cleft palate	Visual biofeedback: Electropalatography	Included RCTs comparing EPG to no treatment or other SLT treatment	One RCT identified (Michi, 1993). Conclusion= evidence supporting EPG is not strong	I
2	Bacsfalvi et al.	2007 IJSLP	Case series No control	n=3. Adolescents with hearing impairment.	Visual biofeedback: Electropalatography & ultrasound	Perceptual and instrumental outcome measures.	Changes towards adult English targets in all subjects (vowels).	IV
3	Fujiwara	2007	Case series No control	n=5. Children with articulation disorder associated with cleft palate. Age range 8 to 13 yrs	Visual biofeedback: Electropalatography (home training using portable training unit)	Instrumental outcome measure. No perceptual measure reported.	4/5 subjects showed improvements in articulatory patterns after 7 to 9 months of home training.	IV
4	Nordberg et al.	2011 CL&Ph	Case series No control	n=5 Children with dysarthria and cerebral palsy	Visual biofeedback: Electropalatography	Perceptual and instrumental outcome measures.	Increase in anterior articulatory placement for some sounds.	IV
5	Gibbon et al.	2003a IJLCD	Single case study No control	n= 1 10 year old girl with Down's Syndrome	Visual biofeedback + Performance feedback. Electropalatography x12 over 14 wks + home practice	Range of outcome measures. Lack of independent assessment – potential for bias.	Improvement in target sounds. Positive evidence that cognitive impairment not barrier to use of EPG.	IV

Table 2-8: Speech and language therapy feedback studies, continued

Visual biofeedback: Electropalatography (EPG)

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
6	Gibbon et al.	2003b CL&Ph	Single case study No control	n=1 8 year old boy with mild cerebral palsy	Visual biofeedback: Electropalatography	Perceptual and instrumental outcome measures.	Improvement in target sounds.	IV
7	McAuliffe & Cornwell	2008 IJLCD	Single Case study No control	n=1 11 year old girl with persistent /s/ misarticulation	Visual biofeedback: Electropalatography /Intensive motor learning approach x12 over 4 weeks	Perceptual and acoustic outcome measures. Assessors blinded	Improvement in /s/ on both perceptual and acoustic measures.	IV
8	Pantele-midou et al.	2003 CL&Ph	Single case study No control	n=1 Child cochlear implant user	Visual biofeedback: Electropalatography	Perceptual and instrumental outcome measures.	Articulatory changes and generalisation to untaught words.	IV
9	Scobbie et al.	2004 CL&Ph	Single case study No control	n=1. child with articulatory disorder associated with cleft palate	Visual biofeedback: Electropalatography	Perceptual and instrumental outcome measures.	Positive articulatory changes	IV

Table 2-8: Speech and language therapy feedback studies, continued

Visual biofeedback: Ultrasound

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
10	Bernhardt et al.	2005 CL&Ph	Descriptive review	n/a	Visual biofeedback: Ultrasound	Range of outcome measures reported	All studies reported are single case – adults and adolescents. Variability within subjects acknowledged.	IV

Visual biofeedback: Electromyography (EMG)

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
11	Allen, Bernstein & Chait	1991 JBT& EP	Single case study No control	n=1. 5 year old boy hyperfunction dysphonia	Visual biofeedback: EMG biofeedback	Perceptual and instrumental outcome measures.	Resting and speaking EMG levels reduced from baseline at 3m and 6 month follow up.	IV

Table 2-8: Speech and language therapy feedback studies, continued

Visual biofeedback: Electromyography (EMG) continued

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
12	Marchant et al.	2008 Dev Neuro	Single case study Crossover design	n=1. 13 year old child with spastic dysarthria	Phonetic placement therapy versus EMG visual biofeedback therapy	Perceptual and instrumental outcome measures.	No perceptual changes following either treatment. Small acoustic changes.	IV
13	Blood	1994 AJSLP	Single case studies Crossover design	n=2 Adults with hyperfunction dysphonia	Computer assisted visual biofeedback (EMG)	Perceptual and instrumental outcome measures.	Improvements in voice but no difference in effect with biofeedback.	IV
14	Sime & Healey	1993 B&SR	Single case study No control	n=1 Adult male age 45 years with dysphonia	Voice therapy + EMG visual biofeedback + cognitive behavioural therapy	Perceptual and instrumental outcome measures.	Reductions in muscle activity. Improved breathing and voice onset.	IV

Visual biofeedback: Spectral Feedback

15	Laukkanen et al.	2004 LPV	Random allocation to groups	n=12 Student actors (6m + 6f) No speech pathology	Vocal exercising with and without spectral visual biofeedback	Reliability reported. Considered relationship between perceptual and acoustic parameters,	Voice quality improved in both groups. Biofeedback provided concrete goals but may be associated with hyperfunction.	IV

Table 2-8: Speech and language therapy feedback studies, continued

Visual feedback: Electroglottography (EGG)

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
16	Chernobel -sky	2002 LPV	Case series No control	n=4 Adolescents with hearing impairment	Visual biofeedback: Electroglottography	Perceptual and instrumental outcome measures.	All subjects achieved vocal fold vibration at target pitch.	IV

Visual biofeedback: Respitrace

17	Murdoch et al.	1999 Dev Neuro	Single case study Crossover design No control	n=1 12 year old boy with dysarthria (traumatic brain injury)	Traditional therapy versus Respitrace visual biofeedback for breath support for speech	Validity of physiological and perceptual measures not discussed Lack of independent assessment.	Greater improvements in speech breathing patterns with biofeedback treatment. Improvement on all measures.	IV

Visual biofeedback: Electromagnetic Articulograph (EMA)

18	Katz et al.	2007 Aphas	Single case study No control	n=1 65 year old male – left CVA (buccofacial apraxia)	Visual biofeedback: Electromagnetic articulograph (EMA)	Assessors blinded. Reliability reported. Pass/fail outcome measure, validity not discussed.	Feedback increased effectiveness but different oral gestures were targeted with and without feedback.	IV

Table 2-8: Speech and language therapy feedback studies, continued

Visual biofeedback: Transnasal Flexible Laryngoscopy (TFL)

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
19	Rattenbury et al.	2004 JoV	RCT	n=50 Adult subjects with dysphonia	Traditional voice therapy ~ biofeedback: transnasal flexible laryngoscopy (TFL)	Perceptual, instrumental and QOL measure; patient report	All subjects improved. TFL therapy took less time.	II

Performance Feedback

20	Holmberg et al.	2001 JoV	Case Series No control	n=11 Female adult subjects with vocal nodules and dysphonia	Performance feedback: Structured behavioural voice therapy protocol	Perceptual and physiological outcome measures. Assessors blinded. Reliability reported.	10/11 reduced nodule size and reduced oedema following intervention. Decrease dysphonia.	IV
21	Palmer et al.	2007 IJLCD	Crossover trial	n=7 Adults with stable dysarthria	Performance feedback: Traditional versus computerized therapy	Speech assessment (intelligibility)	Computer therapy as effective as traditional therapy.	III
22	Dromey et al.	1995 JSHR	Single case Study No control	45 year old male with Parkinson's disease	Performance feedback: Voice exercises And sensory feedback training (Lee Silverman)	Instrumental outcome measures – acoustic and phonatory.	Increase in vocal intensity as well as articulatory changes.	IV

Table 2-8: Speech and language therapy feedback studies, continued

Systematic reviews

No.	Author	Date/ Source	Design	Population	Intervention	Outcome Measures	Results	NHMRC Level
1	Pennington et al.	2010 Cochrane	Systematic Review	Children with acquired dysarthria before age 3 years	Speech therapy interventions, some including feedback	Included RCTs and quasi-experimental designs but no studies identified.	Conclusion = no firm evidence for ST for children with acquired dysarthria	I
2	Deane et al.	2009 Cochrane	Systematic review	Adults with dysarthria in Parkinson's disease	Speech therapy (ST) interventions, some including feedback	Only RCTs included in review – 2 studies identified	Conclusion = insufficient evidence for speech therapy in Parkinsons disease	I
3	Lee et al. (See above)	2009 Cochrane	Systematic review*		Electropalatography			I

* Systematic review (SR): Electropalatography, listed with other EPG studies that did not meet SR inclusion criteria

2.9 Discussion

2.9.1 Study Design

The majority of the studies are single case reports or small studies. There is just one randomised controlled trial (Rattenbury et al., 2004) and three systematic reviews which include speech therapy treatments involving feedback. Overall the evidence generated by the studies is mixed. In some feedback made no difference to the outcome, when compared to therapy without feedback. In others the outcome is positive in terms of reducing the symptom of interest, though only six studies include a control or comparison group and just three report random allocation to groups.

Treatment approaches fall broadly into two categories; visual biofeedback and performance (verbal) feedback. Seventeen primary studies, one systematic review (EPG) and the descriptive review (ultrasound) involve visual biofeedback techniques either using nasendoscopy or specially designed instrumentation, whilst three primary studies utilise performance feedback as the central component of the treatment. The two remaining systematic reviews focus on speech therapy interventions for a specific speech disorder, dysarthria, some of which include feedback.

2.9.2 Interventions using visual biofeedback

Electropalatography (EPG) is the most commonly reported instrumental visual biofeedback treatment, with eight studies and one Cochrane systematic review identified in this review. EPG is an instrument for visually displaying tongue palate contact during speech to allow for modification of articulatory patterns (Gibbon et al., 2003a, b). It has been used with both children and adults with cleft palate and a variety of other speech disorders, in the UK and internationally. Despite this, the systematic review by Lee et al. (2009) concluded that the evidence for its use for treating articulatory disorder associated with cleft palate is not strong. This reflects the predominance of single case study research in this area, with only one, methodologically flawed randomised controlled trial meeting the inclusion criteria of

Lee's review. It could be argued however, that the body of EPG case study evidence across speech disorders in fact paves the way for larger, comparative studies and as Lee et al. (2009) suggest, constitutes the pre-clinical/theoretical phase of investigating a complex intervention as defined in the Medical Research Council Guidelines on complex interventions (2000).

This view is borne out in the results from the eight EPG studies that were not part of Lee's systematic review (Bacsfalvi et al., 2007; Fujiwara, 2007; Gibbon et al., 2003a, 2003b; McAuliffe & Cornwell, 2008; Nordberg et al., 2011; Pantelemidou et al., 2003; Scobbie et al., 2004). All report positive changes to articulation (speech sound formation) following the intervention, in children from eight to sixteen years and with a range of conditions; hearing impairment, cleft palate, cerebral palsy and Down's syndrome. As previously stated, single case studies are the predominant design (five out of eight studies) and the remaining studies are small, with between three and five subjects. However all eight studies were reported within the last ten years and half within the last five years; a very different picture to the dated studies relating to nasal speech previously described.

The remaining studies refer to a range of other visual biofeedback approaches; ultrasound (1), electromyography (4), spectral biofeedback (1), electroglottography (1), Respitrace (1), electromagnetic articulograph (1), and transnasal flexible laryngoscopy (1), (Table 2-9). All are instrumental visual biofeedback approaches apart from transnasal flexible laryngoscopy which is nasendoscopic. The majority of the studies are also single cases, once again limiting the generalisability of the findings.

There are two larger studies; the first by Rattenbury et al. (2004) is an RCT of fifty subjects with dysphonia (voice disorder). The study prospectively compared the use of traditional voice therapy with treatment assisted by visual biofeedback provided by transnasal flexible laryngoscopy (TFL). TFL is another name for nasendoscopy and in

this case allows direct visualisation of the vocal cords in the larynx (voice box) during speech, as opposed to visualisation of the soft palate as previously described in 2.5.2.2.1. The principle of shaping a behavioural response in response to visualisation of speech anatomy and function is the same however. The Rattenbury (2004) study found that subjects improved in both groups, but that the TFL assisted therapy was significantly more efficient. This quality of study is rare in the speech and language therapy literature and therefore adds weight to the evidence base relating to nasendoscopic biofeedback.

The second larger study in the group, by Laukkanen et al. (2004), is also well designed although with only twelve subjects randomly allocated to groups to compare the effect of vocal exercises with and without spectral biofeedback (visual display of sound properties). In this case the subjects studied were student actors and so the results cannot be applied to any clinical population but the results do give an indication of the differential effect that visual biofeedback can have on behaviour.

Table 2-9: Biofeedback approaches used in speech therapy

Biofeedback Approach	Process /Physiological Stimuli	Number of studies in review
Electropalatography	A plate worn in the mouth records tongue palate contact to give a real time visual display of speech.	8 +1 systematic review
Ultrasound (Sonography)	Sound waves give information about oral structures in real time.	1 review paper
Electromyography (EMG)	Electrical activity from the muscles is recorded using probes and displayed visually.	4
Spectral Feedback	Speech sounds are recorded to create an image displaying specific spectral features (time, frequency, amplitude).	1
Electroglottography (EGG) (also known as Laryngograph)	Electrical resistance between two electrodes placed on the neck is measured to give information about vocal cord closure during speech.	1
Respitrace	External movements of the chest and abdomen are measured to monitor breathing during speech.	1
Electromagnetic Articulograph (EMA)	Sensors are placed on the face or within the mouth to measure movements during speech.	1
Transnasal Flexible Laryngoscopy (also known as flexible nasendoscopy)	Movements of the voice box during speech are directly visualised (and recorded) using a flexible endoscope passed through the nose.	1

2.9.3 Interventions using performance feedback

Three studies in the review primarily investigate the use of performance feedback (Table 2-8). This is likely to be a significant underestimate of the use of performance feedback which is central to speech therapy clinical practice. For example, therapy to correct sound production errors involves describing, modelling and reinforcing target sounds (Albery & Russell, 1990) and aphasia therapy uses a variety of performance feedback techniques such as cueing and facilitation (Horton, 2006). These techniques are such an intrinsic part of speech therapy that they are not necessarily made explicit in descriptions or evaluation of therapy (Pring, 2005, Horton, 2006). It may be that in the context of speech therapy it is impossible to completely separate feedback from therapy tasks and from the client-therapist interaction. This is the view proposed by Damico et al. (1995) who describe a process of task, feedback and interaction that ebbs and flows through a therapy session. Horton (2006) supports this view and states that the complex interaction that takes place in most therapy sessions cannot simply be described as a process of elicitation – response – follow-up.

Nevertheless, some researchers have sought to examine the effect of performance feedback and the need to define the critical or active components of therapy is well accepted (Hayhow, 2011; Law & Forsyth, 2006; Pring, 2004). Despite the limited evidence available, the three studies reviewed do give an indication of how performance feedback is used in speech therapy and how it can be evaluated. Two of the studies are limited by having no control or comparison group but the small crossover trial by Palmer et al. (2007) compares two different modalities of delivering performance feedback to seven adults with speech difficulties. Traditional therapy is compared with computer based therapy and the two are found to be equally effective in improving speech clarity, with the added benefits of increased flexibility and accessibility of the computer based therapy. The two other studies also report positive results although these should be interpreted with caution due to the methodological limitations. Dromey et al. (1995) report improvements in voice and articulation in a single case study using Lee Silverman voice exercises and more recently Holmberg et

al. (2001) report both physiological (vocal nodule reduction) as well as perceptual speech improvements in eleven adults following a structured voice therapy programme. Both of these studies involved the use of specific performance feedback in the intervention.

2.10 Summary of literature findings

This review summarises the evidence relating to speech therapy using feedback to treat a variety of speech disorders and provides further evidence to support this approach. The majority of studies identified reported on visual biofeedback rather than performance feedback approaches. Visual biofeedback tends to be used in more specialist settings, where equipment such as electropalatography is available. This may also have influenced the number of studies generated in this area as specialist units and academic institutions will have resources and research facilities that are not available in other clinical settings.

Performance feedback, on the other hand, is an intrinsic part of most speech therapy interventions. This does not make it less deserving or requiring of investigation, perhaps even more so, and the drive for efficacy research in the field of speech and language therapy is leading to renewed attempts to define the precise nature of therapy approaches being used in clinical studies (Pring, 2004). In the past the focus in the literature has predominantly been on describing the nature of speech and language disorders and 'there is relatively little that addresses the therapy process, its impact and acceptability' (SLTRU, 2011).

Seventeen of the twenty-one speech therapy studies reviewed reported positive results following intervention using feedback of some kind. There are however serious methodological limitations which mean that this can only be taken as indication of a trend rather than evidence of efficacy. As in the previous literature review in this chapter, many of the studies are single case studies or small case series with no

control or comparison so that any positive results cannot be attributed to the intervention. However this is in part ameliorated by the inclusion of both perceptual and instrumental repeated measures (triangulation) in seventeen studies, combined with blinding of assessors.

Finally, the three systematic reviews all highlight the practical difficulties and methodological challenges facing researchers in speech therapy which is borne out by the lack of larger, high quality studies. It is essential however that this is recognised for what it is, a lack of evidence, rather than evidence of a lack of efficacy.

2.11 Conclusion

Results from the literature reviews tentatively suggest that some individuals can benefit from non-surgical treatment for nasal speech associated with velopharyngeal dysfunction (VPD). A small number of studies report positive effects of interventions using visual biofeedback techniques or continuous positive airway pressure (CPAP), but the evidence is weak and the study samples small. However, positive findings have also been reported, in more recent and better quality studies, for the use of visual biofeedback in treating other speech disorders, suggesting that this approach appears worthy of further investigation. The use of performance feedback as an intervention or component of an intervention is less well described. It is acknowledged however that it forms a critical part of therapist-client interaction in any behavioural intervention and as such warrants greater attention in the evaluation of interventions than it has received to date.

Taken as a whole, the evidence for a behavioural approach using visual biofeedback and performance feedback in treating nasal speech and other speech disorders, suggests that an intervention that includes both of these components is likely to benefit individuals with nasal speech. In total there were forty studies identified as using feedback to treat speech disorder and in thirty-two of these positive changes were reported for some individuals. However the weak nature of the existing evidence

indicates that any further research in this area should be at a level designed to test feasibility as a first step, and that any intervention be investigated systematically using valid and reliable outcome measures.

This study asks whether a combination of visual biofeedback and performance feedback could be used to develop a feasible intervention for nasal speech. The gaps in knowledge highlighted by the literature review are addressed by attempting to answer the following question:

What is the feasibility of a newly developed speech therapy intervention combining visual biofeedback and performance feedback for adults and children with nasal speech associated with VPD?

The study aims to:

1 Develop an intervention for nasal speech combining visual biofeedback and performance feedback, based on theoretical models.
2 Assess the feasibility of delivering a behavioural intervention for nasal speech.
3 Explore a range of measurable and relevant outcome parameters.
4 Consider the value of information to be gained from further research.

Finally, evaluation of previous studies in this review has revealed that in many cases the interventions employed are underpinned by theories of learning in relation to speech production. Exploration of this theoretical perspective will therefore provide valuable insight into the mechanisms by which a behavioural feedback intervention for nasal speech might work. The following chapter discusses how these theories contribute to the development of the intervention and the selection of the appropriate study design and outcome measures.

Chapter Three: Development of a Feedback Intervention

3.1 Introduction

The previous chapter identified various types of visual biofeedback that have been used in the treatment of nasal speech and other speech disorders, including nasendoscopic biofeedback and instrumental biofeedback techniques. These approaches have demonstrated some positive but variable results and are therefore not universally supported (Peterson-Falzone et al., 2006). However, as Ruscello (2006) argues there is a subset of patients who appear to benefit from treatment using visual biofeedback, possibly because they have the neurophysiology and structural capacity to do so. This indicates a need for further research to develop understanding of the nature of a response to this type of behavioural treatment, as studies to date have made little reference to theory resulting in limited evaluation of the nature of interventions. This chapter outlines the development of the intervention used in this study, as informed by theories of feedback and biofeedback and their application in speech therapy. Evaluation of the intervention is then discussed including selection of an appropriate study design, methods and outcomes.

3.2 Development of the study intervention: Principles of feedback and learning

The development of the intervention in this study is informed by the theoretical models of feedback, learning and speech production. Over the last 100 years social science research has attempted to elucidate the relationship between feedback, learning and behaviour (Baker & Buckley, 1996). In the context of skill acquisition or behaviour change, feedback broadly refers to information that is generated in the process of learning (Annett, 1969). Different theories have placed different emphasis

on the nature and role of feedback in learning (Glassman & Hadad, 2008). Behaviourism dominated up until the 1950s with the emphasis on the relationship between directly observable stimulus, response and reinforcer. In contrast, the humanist and cognitive theories that followed acknowledged the internal aspects of the learning process and feedback mechanisms i.e. mental processes and development of self.

Despite the differences in emphasis and terminology between various theories of behaviour and learning there are commonalities. It is likely that different learning situations call for a variety of feedback mechanisms and it is generally accepted that feedback and its role in learning are strongly linked to the concepts of self-regulation and motivation. For example feedback may be used simply to describe a technique for changing behaviour (Michie et al., 2008) or as Carver et al. (1990) argue, is central to the process of moving toward various goals. The role of feedback can therefore be viewed in three ways, as reinforcement, self-regulation and evaluation.

3.3 Feedback

3.3.1 Feedback as reinforcement

From a strictly behaviourist perspective, most well known in relation to the seminal works of Pavlov, Watson and Skinner (Glassman & Hadad, 2008), feedback functions simply as reinforcement following a response to a stimulus. Learning is defined as change in behaviour resulting from experience involving this stimulus-response process. Behavioural theories also describe how voluntary behaviours change over time as a function of their consequences, and this key concept is known as operant conditioning. Furthermore, behaviourists argue that complex behaviours, which do not emerge fully-formed, can be shaped by reinforcing a series of simpler behaviours and successful approximations.

The concept of reinforcement whilst central to behaviourist psychology is also evident in both humanism and cognitive theory. Carl Rogers (Rogers 1951), as one of the

founders of humanistic psychology believed that humans are motivated to 'self-actualize', that is to fulfil one's potential in life. He also proposed that positive regard is essential to human growth and distinguished between conditional and unconditional forms of this. Conditional positive regard includes praise and attention for specific behaviours which can be seen as a reinforcer. However the difference between this concept and the behavioural term lies in the humanistic focus on the subjective experience of the individual. Similarly a cognitivist view differs in its assertion that feedback (or 'knowledge of results') is not only reinforcing but is necessary for revision and error correction (Hartley, 1998).

3.3.2 Feedback as self-regulation

Cybernetic theory is an example of a cognitive theory where behaviour and learning are viewed as self-regulating with built-in 'circular' control mechanisms (Murray, 2006). A basic cybernetic model is shown below (Figure 3-1). According to this model, discrepancies in performance are identified and corrective action is taken to modify behaviour (Pratt, 1978). Cognitive theories are therefore concerned with thinking and mental processes i.e. perception, language and memory, and not just observable behaviours (Glassman & Hadad, 2008).

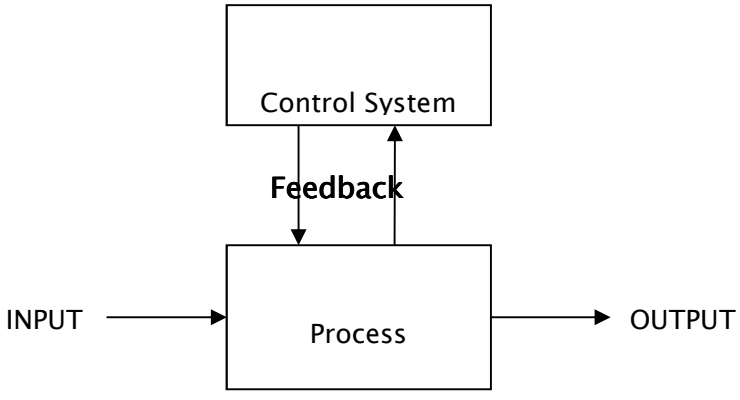

Figure 3-1: Basic cybernetic model, adapted from Pratt (1978)

3.3.3 Feedback as evaluation

Evaluation at a conscious level, as opposed to self-regulation at an automatic or semi-automatic level, is considered essential to the learning process according to humanist theory (Rogers, 1985). In this way humanism rejects the concept of determinism, where behaviour has specific causes, in favour of individual choice and the 'actualizing tendency'. This refers to motivation or the desire to enhance one's capabilities, and is also known from Maslow's influential work as 'self-actualisation'.

3.4 Learning

3.4.1 Learning theory and speech therapy

Kamhi (1989) outlines the central place of learning theory in speech therapy clinical practice, with a significant emphasis on behaviourism and operant conditioning methods. Early speech therapy texts demonstrate the basic premise of therapy as designed to change behaviour (West et al., 1937, Morley, 1966). In the broadest sense then the clinical process of speech therapy can be described as;

'the methods and actions used to effect positive change in people's communication functioning'

Bray et al. (1999): page viii

As previously discussed in Chapter Two, the underlying theoretical perspectives of interventions in speech therapy clinical practice are not generally made explicit in the selection of an approach, but appear to become internalised as part of the clinical decision making process (Horton, 2006). Behavioural approaches are widely but not exclusively employed, with the therapist providing contingent responses to a patient or client in a variety of forms. The feedback in this context can provide reinforcement in a classically behaviourist sense but can also involve shaping or cueing to promote error correction, self-regulation and ultimately adaptation or automatisation. The contribution of paradigms such as cognitivism and humanism can be clearly seen in

descriptions of therapy for adults with aphasia and stammering for example, where the concepts of self-acceptance, therapeutic partnership and realising potential are as important as behavioural change (Chapey, 1994; Stewart & Brosh, 1997).

3.4.2 Motor learning and speech therapy

Motor learning is another central concept described in early seminal speech therapy texts such as Morley (1967). It continues to be seen as having applicability to learning or regaining movements for speech with feedback as a critical element (Adams & Page, 2000; McCauliffe & Cornwell, 2008; Palmer et al., 2007). Maas et al., (2008) review the theoretical principles and discuss application to the treatment of motor speech disorders. They refer to the definition of motor learning from Schmidt and Lee (2005);

'a set of processes associated with practice or experience leading to relatively permanent changes in the capability of movement.'

Maas et al. 2008: Page 302

Katz et al. (2007) give a detailed analysis of motor learning, making reference to Schmidt's Schema Theory (1975). This assumes that by practising a movement an individual creates a schema (set of rules) which can be used to generate different versions of the movement. The implication is that new patterns of movement can be developed with practice and shaping from appropriate feedback. This suggests that without the feedback element new desirable behaviours cannot be generated and reinforced. Maas et al. (2008) support this view and make an important distinction between performance and learning. Performance is a temporary state, observed during practise, and learning refers to changes in performance over time, where retention or transfer of a motor skill is seen.

The key constructs of Schmidt's Schema theory are generalised motor programmes (GMPs) and parameters. GMPs are set motor programmes that contain abstract

information about relative timing and force of a movement. Parameters are values assigned to a GMP that allow for adjustment to meet specific environmental demands. The development of a 'schema' will depend on a number of variables:

- Type of feedback
- Amount of practice
- Practice schedule
- Supporting cognitive mechanisms (memory, attention, motivation)

The above factors emerge frequently from the rehabilitation literature. Palmer et al. (2007) state that in order to be effective, treatment using motor based approaches should be frequent and consistent, and again should include feedback. Similarly frequent practice ('use it or lose it') is the first of ten principles of neural plasticity proposed by Kleim and Jones (2008), which also cover stimulus selection and feedback, (Table 3-1).

Table 3-1: Principles of neural plasticity (Kleim and Jones, 2008)

Use it or lose it
Use it and improve it
Plasticity is experience specific
Repetition matters
Intensity matters
Time matters
Age matters
Transference
Salience matters
Interference

3.4.3 Neural plasticity

The concept of neural plasticity is important in considering how a biofeedback intervention might work. The basic premise is the ability of the brain to change, in response to external influence, leading to associated behavioural changes. Robbins et al. (2008) discuss these principles and their clinical application in swallowing disorders, including the possibility that behavioural change could result from compensatory brain activity as well as plasticity. Ludlow et al. (1998) also examine the concept in relation to the motor control of speech. They report supporting evidence from experimental studies which have shown that motor training can alter neural signalling pathways, but question whether this can lead to long term changes in motor performance. They also suggest that speech production may differ from other movements in the effect of practice. Critically though they do report that neural plasticity can occur over the entire lifespan.

Borden (1980) also discusses the plasticity of neural networks in relation to speech production and how this may change from infancy into adulthood. She argues that adult speakers perceive speech in accordance with their experience and so will be more inhibited in modifying new patterns than children. This view is supported by Thelen (1991) who argue that attractor states (stable articulatory movements) are more difficult to disrupt as children get older. This relates to Dynamic Systems Theory (Kamm et al., 1990) which proposes a relationship between instability and variability of motor function in speech development that involves a series of changes of relative stability and instability rather than a linear progression towards stability. Disrupting factors occur in typically developing children, such as anatomical changes, increasing motor control and development of perceptual skills, and these influence existing attractor states. Gibbon et al. (2003a) refer to DST in relation to children with speech disorders and propose that instrumental visual feedback could act as a powerful disrupting force in treating habitual speech errors.

Finally Maas et al. (2008) make an important point in stating that knowledge relating to motor learning is largely based on evidence from non-speech tasks and in individuals with intact central nervous systems. The focus on non-speech tasks in the clinical application of motor learning principles is one of the most debated aspects in this area (Clark, 2003; Bunton, 2008; Bowen, 2005; Williams et al., 2006). A systematic review of oro-motor exercises for drooling and swallowing disorders in children found insufficient evidence to support this motor approach (Arvedson et al., 2010). However the authors do argue for the importance of theoretical frameworks in the absence of unequivocal evidence of efficacy, a view reiterated by Weismer (2006) and Robbins et al. (2008).

3.4.4 Summary: Learning theory and speech therapy

The degree to which the theoretical perspectives of learning and motor learning are addressed in the speech therapy efficacy literature is highly variable. This means that the mechanisms by which behavioural change does or does not take place in the context of speech therapy are inconsistently explored although there is a growing awareness of the need to make explicit the active components of therapy. One area that is generally acknowledged as a prerequisite for behavioural change in relation to speech disorders is the concept of frequent practice or repetition (McAuliffe & Cornwell, 2008; Marchant et al., 2008; Caruso & Strand, 1999). However it is not entirely clear how this frequency is determined. There are references to short and distributed, or intensive practise being more effective than longer sessions (Ramig & Verdolini 1998; Strand & Skinder, 1999), and texts often recommend daily short practice (Rosenbek, 1985). However, despite a consensus that there is a need for greater integration of theory into clinical practice and research, it is likely that frequency of speech therapy treatment is more often determined by service delivery constraints than motor learning principles.

3.5 Models of speech production

The changes produced by speech therapy may be poorly understood (Sommerlad, 1981) but models of speech production suggest that there is potential for change at a number of levels. A number of models exist that focus on different aspects of this complex mechanism i.e. physiological (feedback), acoustic and psycholinguistic. Aspects of these models have already been discussed earlier in this chapter in relation to motor learning and the role of feedback and several focus on feedback as a central mechanism of the speech production process.

3.5.1 Feedback models of speech production

Borden (1980) presents a closed loop model of speech production, where control continually relies on feedback (Figure 3-2).

Three levels of motor organisation are proposed, after Evarts' model (1971):

- Internal feedback
- Response feedback
- Knowledge of results/external feedback

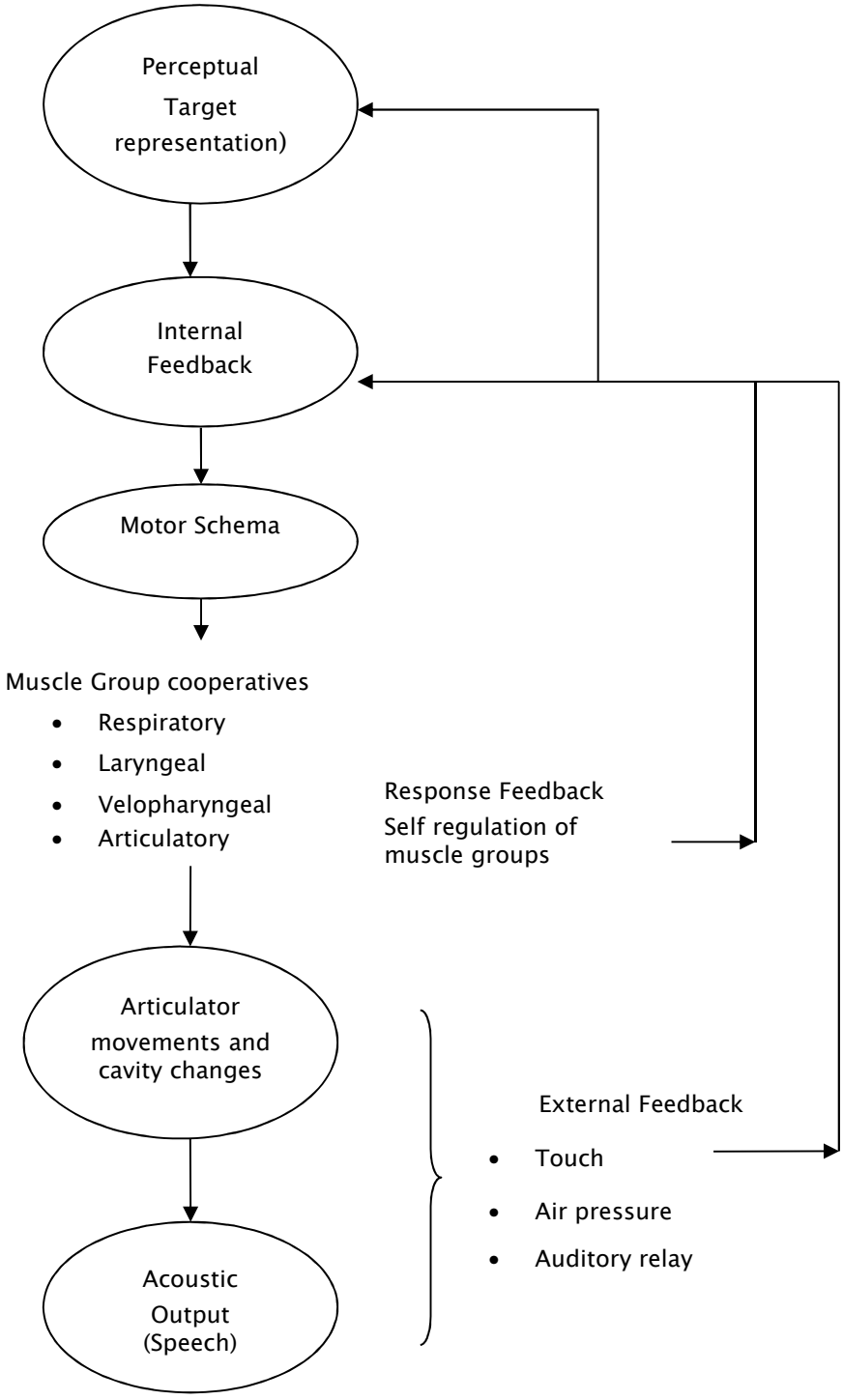

Figure 3-2: Feedback model of speech production, Borden (1980)

Internal feedback takes place at a cortical level, response feedback at the level of muscles and joints and external feedback through sensory and kinaesthetic experience.

Burzynski and Starr (1985) also refer to the physiologically based open-loop and closed loop theories of speech production. Early proponents of the open-loop model see speech as the result of the pre-programmed organisation and execution of movements (Peterson & Shoup, 1966) whilst in a closed-loop system, such as the Borden model above, speech originates in the brain but control relies on and is sensitive to continual sensory feedback. It is generally accepted that the process is complex with different systems operating to control different aspects of speech.

Some aspects of speech may be under the control of or influenced by feedback whilst others are more pre-programmed (Garber & Moller, 1979). Casserly and Pisoni (2010) go further still in arguing that the processes of speech production and speech perception are intrinsically connected and interdependent, and Kent & Read (2002) suggest that it is the acoustic (sound) signal that is the intermediary between the two processes as illustrated below (Figure 3-3).

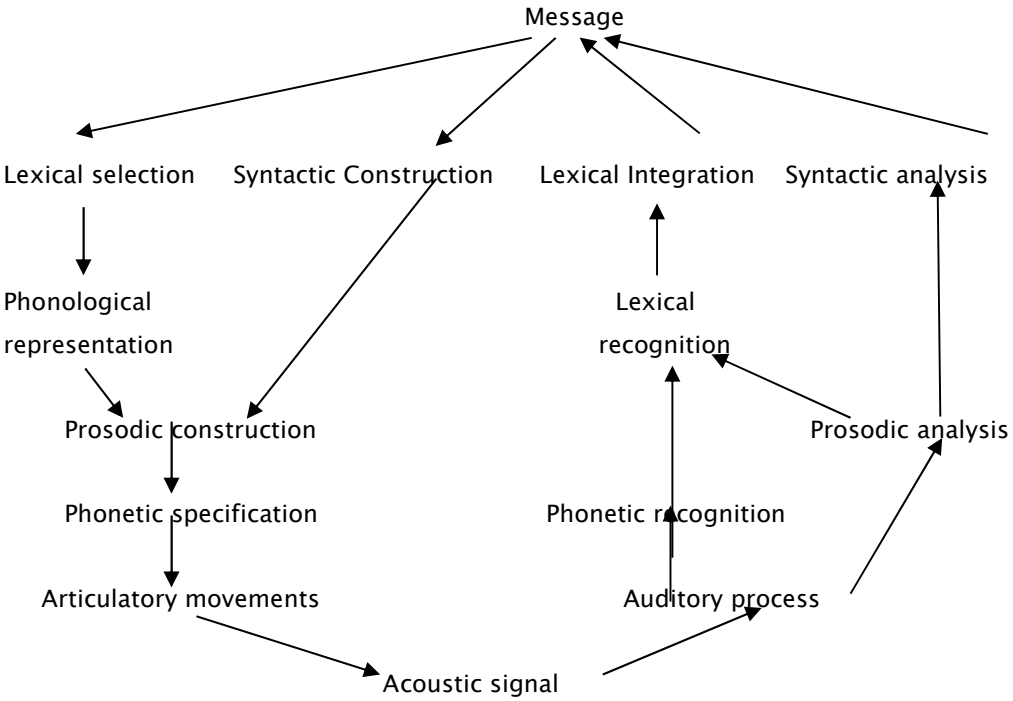

Figure 3-3: Diagram of speech production & processing where the acoustic signal is the intermediary between these two processes. Kent and Read (2002) pg15. From Kent / Read. *Acoustic Analysis of Speech*, 2E. © 2002 Delmar Learning, a part of Cengage Learning, Inc. Reproduced by permission. www.cengage.com/permissions

3.5.2 Acoustic models of speech production

As illustrated above in the Kent and Read model (2002), the acoustic (sound) signal simultaneously represents the output of the speech production system and the input of the speech processing system. Source-Filter theory is the major model of speech acoustics. In this model speech involves a source function and a subsequent filtering process (Fant, 1981). The energy source is the vibrating vocal cords in the larynx (voice box) and the vocal tract is the filter radiating and modifying the acoustic energy output to form speech. This model allows the essential properties of speech sounds to be described in terms of how the acoustic signal is modified as it passes through the vocal tract. For example, the essential property of nasal consonants (/m/, /n/, and /ng/) and nasalised vowels is that the velum (soft palate) is lowered, allowing sound energy to pass through the nasal tract (nose) for nasal consonants and both the nasal and oral tracts (nose and mouth) for nasalised vowels. Acoustic theory therefore allows prediction of how coupling between the nasal and oral tract will influence the sound perceived (Chen, 1997).

Although influential, Source-Filter theory has limitations in explaining the complex features of speech production (Kent & Read, 2002; Titze et al., 2008). These relate to underlying assumptions of linearity and independence. Titze et al. (2008) argue that the process is not linear or independent as there is interaction between the acoustic source and the vocal tract filter. This is particularly true for female and child speakers where the range of fundamental frequency (the basic tone of sound produced at the larynx) is greater than for male speakers. It has also been demonstrated that it is possible to exploit this interaction to achieve various vocal qualities (Kent & Read, 2002) highlighting the potential for clinical application of the process.

Teager and Teager (1990) also challenge the traditional mathematical perspective of Source-Filter theory and argue for an aerodynamic rather than acoustic model where non-linear phenomena can take place in response to changes in the vocal tract. These authors reference quantal theory (Stevens, 1972) where acoustic patterns may change

from one quasi-steady state to another as articulatory parameters vary, again contrasting with the linearity of Source-Filter theory.

Despite the limitations of Source-Filter theory it is clear that the study of acoustics provides a sound basis for understanding the process of speech production. In reality though acoustics cannot be separated from the physiology or the perception of speech and therefore a connectionist view is appropriate. This is illustrated in the diagram below which shows the relationship between perceptual features of speech and their physical correlates (Figure 3-4).

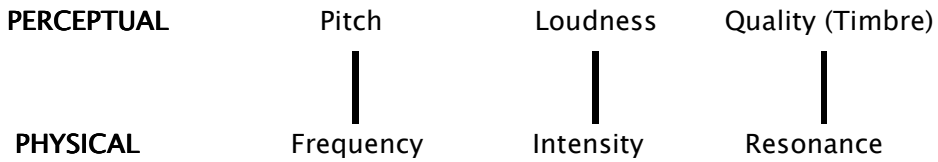

Figure 3-4: Physical correlates of perceptual speech features

3.5.3 Psycholinguistic models of speech production

Psycholinguistics is the process of mapping a message a speaker intends to communicate onto its form (Goldrick, 2007). Psycholinguistic models therefore offer a connectionist approach to analysing speech as an output of language beyond the level of the acoustic signal. Levelt's model of lexical access (1993) is the most well-known theory, describing the process from conceptualisation to generation of a phonetic representation (spoken word) with specific articulatory gestures and timing. Treiman et al., (2003) emphasise the importance of the 'mental lexicon' where speech production represents the retrieval of words from this lexicon, along with their syntactic (grammar) and morphological (units of meaning) properties, a view also supported by Altmann (2001).

Psycholinguistic models tend not to go beyond consideration of the phonological form of a word i.e. the sequencing of sounds to create a word. However it is possible that the concepts of storage and retrieval also apply to the suprasegmental features or acoustic correlates of the speech sounds. This would imply that the selection of specific articulatory gestures, including velopharyngeal closure, could form part of the mental lexicon for a speaker.

3.6 Clinical application of speech production models

Feedback and motor learning models indicate that the speech production system is sensitive to external influences, for example Burzynski and Starr (1985) report studies of pitch and nasalisation which suggest that these features are influenced by external sensory cues. This is supported by studies of individuals with hearing impairment where the lack of auditory feedback can result in nasal sounding speech in spite of apparent capacity for adequate velopharyngeal closure.

Acoustic models offer an insight into the physical properties of speech sounds and the effect of small changes in the system, whether viewed as linear or non-linear. For example Jones (2006) reports that the perception of nasality may be influenced by articulatory transitions that affect the oral nasal balance. This suggests that changes in the one aspect of the speech mechanism may have an effect on another.

In addition, psycholinguistic theory offers a connectionist perspective of the speech mechanism as a whole, from perception to production. Stackhouse and Wells' (1997, 2002) work is perhaps most well known in speech and language therapy. Their speech processing model mapped onto the speech chain from ear to mouth (Figure 3-5) allows for consideration of the speaker's stored 'lexical representations' of sounds and sound properties and the possibility that these can be replaced with different representations.

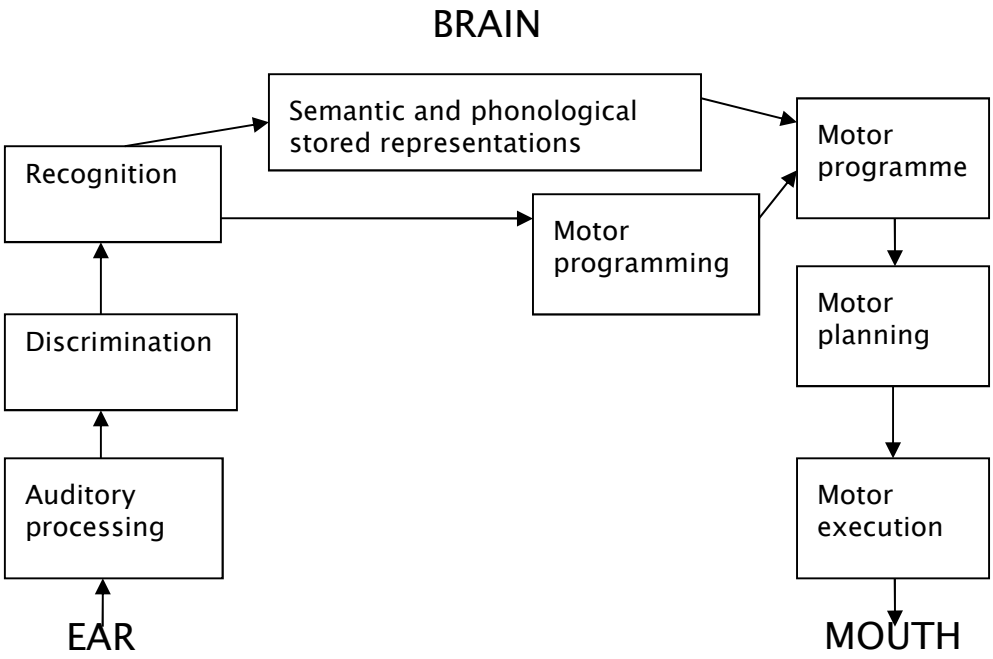

Figure 3-5: Psycholinguistic model, adapted from Stackhouse & Wells (1997)

In an earlier work Greene and Mathieson (1989) refer to the idea of habitual 'voice settings', which the speaker is not aware of but which can be modified. They acknowledge that there is an underlying voice quality, unique to each speaker, determined by the dimensions of the vocal tract and the resonating system. However they also refer to learned muscular tension adjustments and habitual levels of volume and pitch. This is what makes individual voices recognisable whilst also potentially variable. From a psycholinguistic perspective this supports the view of voice as part of the structure of pronunciation at the level of the phoneme (speech sound) i.e. as an integral part of each phoneme and therefore part of the speaker's repertoire of stored representations.

The concept of voice settings proposed by Greene and Mathieson would account for the fact that nasalised vowels are not used in English but are found in around 20% of the world's languages (Hombert, 1986). Nasalised vowels are produced by lowering

the velum (soft palate) during the production of the sound so that air resonates in both the nose and mouth. In certain languages they are used to differentiate meaning and so are learnt and stored as a unit, with the appropriate nasal voice quality and associated articulatory movement of the velum. Greene and Mathieson also refer to the concept of 'contamination' whereby adjacent nasal sounds influence vowel quality. This concept is extended to 'decontamination' in voice therapy by alternating vowels and consonants in order to increase the oral balance of the vowels.

3.7 Summary of theory underpinning the study intervention

The sensitivity of the speech mechanism to the presence or absence of feedback coupled with the possibility of habitual voice settings lends support to the idea that a speaker may be able to modify his or her nasal speech quality in response to different types of feedback. Learning theory, and in particular motor learning models, also suggests that it is possible for a speaker to generate new movement patterns through practice and repetition. The neural mechanisms for such modification remain unclear; it may be that new neural pathways are developed or existing unused ones recruited (Huang et al., 2006). The principle of neural plasticity appears to support this hypothesis (Ludlow et al., 1998) although it is not clear whether changes in speech as a result of training or new learning will lead to long term changes in performance. Nevertheless, the key components of feedback, practice and reinforcement can be identified as central to an intervention designed to influence speech production.

3.8 A model for Visual Feedback Therapy

A model of how a visual biofeedback and performance feedback could work in a speech therapy intervention for nasal speech is presented in Figure 3-6. This is a physiologically based model, with consideration given to the interaction between the acoustic (sound) source and the filter function provided by the vocal tract, as previously discussed. The VFTh model is circular reflecting the central role of feedback, as described in closed-loop speech models, where control of the speech

mechanism is reliant on and sensitive to feedback generated and received at different levels.

The contribution of the nasal speaker and the speech therapist are included in the model in order to illustrate the learning process involved, by which a change in speech behaviour is identified and reinforced using two different types of feedback, visual biofeedback and performance feedback. Furthermore the VFTh model draws on learning theory in identifying the role of conscious evaluation of speech performance in conjunction with the more in-built self-regulatory function of feedback as described in cybernetic theory.

There are three core levels within the VFTh model described as **capacity**, **learning** and **comparator**, leading to the generation of feedback and subsequent speech modification attempts. These levels relate directly to the theories and models discussed previously in this chapter.

Capacity

The term capacity is used to describe the different aspects contributing to the potential for the nasal speaker to use less nasal speech. This includes physiological and cognitive aspects. The physiological aspects are vocal tract dimensions, motor skills and settings and neural plasticity. The cognitive aspects are motivation and ability to utilise the visual biofeedback and performance feedback. Correspondingly for the speech therapist the capacity level includes knowledge of the mechanisms of speech production and behaviour modification techniques i.e. capacity to provide meaningful performance feedback.

Learning

The learning level describes the way in which the speech therapist makes explicit the desired speech behaviours and associated mechanisms for change. The nasal speaker

can then explore how to achieve these desired behaviours. The need for practice and repetition is also established at this level as a pre-requisite for motor learning and behavioural change.

Comparator

This level describes how the biofeedback tools and performance feedback measure or identify discrepancies in speech behaviours. The term comparator is used to reflect the idea that the nasal speaker is able to generate a new motor speech pattern (schema) and associated acoustic signal which can be directly compared to habitual speech patterns. This is supported both by dynamic systems theory and psycholinguistic theory; which allow for the possibility that existing or stored speech patterns can be destabilised and replaced with new motor speech representations and programmes.

At this comparator level speech modification attempts by the nasal speaker involve self-monitoring supported by the visual biofeedback and the contiguous reinforcement and shaping (performance feedback) from the speech therapist. The performance feedback is immediate and specific verbal feedback about the nature of speech in relation to the specified goals identified at the learning level. Reinforcement, evaluation and self-regulation, as central components of learning theory, can all be seen to be at work at this level.

It is proposed therefore that the combination of visual biofeedback and performance feedback in VFTh allows the nasal speaker to consciously attend to speech output in order that existing voice settings can be subjected to disrupting or destabilising factors. Consequently, the potential for new voice settings can be explored and reinforced.

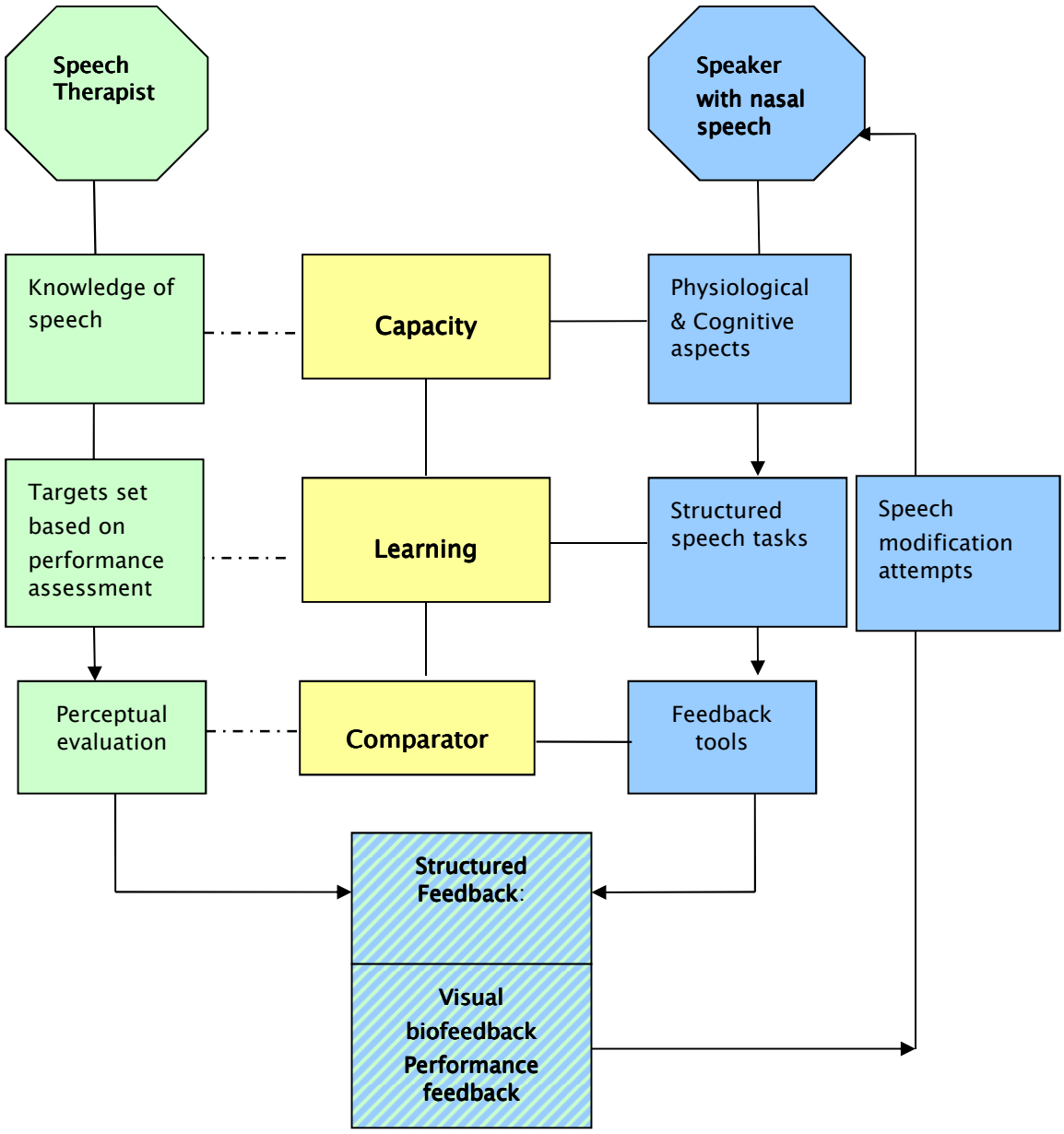

Figure 3-6: A Model of Visual Feedback Therapy

3.9 Development of the study intervention

3.9.1 Intervention: Goal

The term Visual Feedback Therapy (VFTh) has been adopted to describe the study intervention which involves the use of techniques designed to increase a speaker's voluntary control over one aspect of their speech i.e. the degree of nasal tone of voice perceived by the listener. As shown in the above model, VFTh is a behavioural treatment. In the context of treatment for nasal speech this is defined as 'not surgical, not prosthetic, not instrumental' (Whitehill, 2002a).

The goal of the intervention is to improve the perception of nasal speech i.e. to reduce the degree nasal tone perceived by the listener and the participant and so maximise communication effectiveness. As this is considered to be the primary goal of VFTh there is no attempt to directly evaluate any effect on the velopharyngeal mechanism. In this context communication effectiveness encompasses both clarity and perceived quality of speech, as these are known to be impacted by excessive nasality (Barr et al., 2007; Grunwell, 1993). This intervention goal mirrors that described by Ramig and Verdolini (1998) in their review of behavioural voice treatments generally; to maximise vocal effectiveness given the existing disorder and to reduce the 'handicapping effect' of the voice problem.

3.9.2 Intervention: Components

VFTh been developed to include two distinct but interrelated components; the central component is visual biofeedback supported by the addition of performance feedback. Visual biofeedback is provided in response to the nasal escape of air during speech through the use of three feedback tools. Performance feedback is direct and targeted verbal feedback given by a speech and language therapist in response to speech behaviour. The rationale for these components of the intervention is given below.

3.9.2.1 Visual Biofeedback component

Visual biofeedback was selected as a component of VFTh in response to the tentatively positive results of the literature reviews and based on theories of feedback. The use of visual biofeedback in this way allows covert physiological processes to be made more overt (Huang et al., 2006), so that the participant can actually see the result of excessive nasal airflow or resonance in a concrete visual form. Huang et al. also propose that visual biofeedback serves to heighten sensory cues and also to allow adaptive strategies. This is supported by Brunner et al. (2005) who reports that after visual biofeedback treatment using endoscopy subjects were able to describe kinaesthetic features of speech e.g. tongue placement. In addition Brunner asserts that visual information provides a strong stimulus to change existing speech patterns and this is supported by theories of feedback and learning as previously discussed.

3.9.2.1.1 Visual Biofeedback tools

The visual biofeedback tools used in VFTh are the Nasometer™ II 6450, See-Scape™, and a nasal mirror (Figure 3-7). These are readily available in UK regional cleft palate centres. The **Nasometer**™ is a form of nasometry, an instrumental tool that gives specific visual feedback of the oral versus nasal balance of speech output via a computer screen. It has been selected as the main biofeedback tool as it provides immediate visual feedback about the degree of nasal speech, in picture and graph form and in the form of a percentage nasalance score. Similar tools have been identified in the literature review (TONAR, Velograph) but an important additional feature of the Nasometer™ II is the colourful and motivating games, which again provide immediate visual feedback if speech production reaches a pre-set target, for example below a certain percentage nasalance threshold. The overall objective for the participant is to reduce the level of the graph (lower indicates less nasal) during speech.

Nasometry See-Scape Nasal mirror



Figure 3-7: Biofeedback tools used in VFTh

See-Scape™ provides visual feedback about the amount of air escaping down the nose during speech. If air flows into the tube through the nose-piece placed in the nostril this causes a coloured polystyrene float to rise in the tube. The overall objective for the participant is to limit or stop the float rising in the tube during speech. The **nasal mirror**, a low-tech feedback tool, is placed under the nose and detects nasal escape of air during speech, indicated by fogging of the mirror. The overall objective for the participant is to reduce or eliminate fogging of the mirror during speech.

3.9.2.2 Performance feedback

The rationale for incorporating performance feedback alongside biofeedback in the intervention primarily relates to the goals of the intervention and also the feedback tools used. In the motor learning literature performance feedback is a term used to describe feedback related to the nature of the movement of interest (Schmidt & Lee, 2005). This feedback may be generated by the individual themselves or given verbally by another person monitoring the movement. In the context of speech production therefore, performance feedback can be used by the speech therapist to give immediate and specific verbal feedback about the nature of speech in relation to specified goals. It therefore functions as a conditioned reinforcer of desired speech behaviours whilst also allowing them to be explicitly described, or 'externalised', and shaped.

In addition Hooper (2004) comments that most voice therapy programmes use insight and discussion and include a focus on the self-perception of the 'good voice'; the role played by performance feedback in this study's intervention. This is true of the majority of speech therapy approaches using biofeedback as explanations and monitoring are provided alongside the visual biofeedback component. In fact Pantelemidou et al. (2003) argue that this is the aspect that differentiates an approach such as electropalatography from traditional auditory speech therapy, as the individual takes an 'active' role in discovering the relationship between tongue placement and speech sound production. In the same way a feedback treatment for nasal speech could allow someone to actively explore their ability to modify their voice quality in response to visual and performance feedback.

Performance feedback is used in several ways primarily to support the participant in making sense of the visual feedback display by checking concepts and revising goals. In addition descriptive terms are used to label the nasometry visual feedback display e.g. 'towers', 'chimneys', 'tall/short', 'invisible sounds'. These descriptions may be generated by the participant themselves and then developed and reinforced in subsequent sessions.

3.9.3 Intervention: Content and phases

VFTh uses speech tasks for maximum relevance and the participant progresses in a graded way through the tasks from practice of single speech sounds to sound combinations, single words, word combinations and finally sentences. This sequence represents a hierarchy of complexity as described by Albery and Russell (2005) and which is routinely used in speech therapy.

Preliminary target selection is based on initial speech measures and progression then individually tailored to the participant. The tailoring of the VFTh does introduce variability and Carding (2000) notes that in studies of voice therapy the treatment for

different patients in the same study is likely to be dissimilar. Each step is therefore described for each participant in treatment session notes in order to identify as clearly as possible the precise components of VFTh. This detailed description is presented in the case study section of Chapter Five.

Finally, VFTh, although individually tailored in delivery, does consist of a number of phases commonly employed in speech and voice therapy programmes (Hooper, 2004) as shown below. This provides a common underpinning framework for the tailored VFTh intervention.

- General awareness of speech behaviour; in this case oral versus nasal voice quality and airflow.
- Specific awareness of speech behaviour to change; e.g. production of target sounds without nasal emission or nasal resonance.
- Direct speech production activities; repetitive practice of target sounds and words.
- Generalisation activities; e.g. conversational speech.

3.9.4 Intervention: Delivery

As well as the theoretical modelling previously described in this chapter, development of the intervention involved trying out the tools and procedures in the clinical population. Prior to the study this involved using VFTh with two patients, both children, under the umbrella of 'diagnostic' therapy. Diagnostic therapy is a common approach undertaken either when a patient is waiting for surgery or as part of the decision making process for surgery (Albery & Russell, 2005; Trost, 1981). It more usually relates to treatment for articulation errors rather than nasal speech but the same principle of testing potential for improvement applies. This developmental work demonstrated that the children could understand the concept of 'nasal' speech, were able to understand and were willing to use the feedback tools in order to try to modify their speech production.

3.9.5 Intervention: Frequency and intensity

There is a lack of consensus in speech and language therapy practice generally about the optimal frequency and intensity of treatment (Law et al., 2004) and in relation to specific treatments for individuals with cleft palate/VPD (Vallino-Napoli, 2011). The frequency and duration of VFTh is based on the premise that any behavioural change should take place quickly, which is taken to mean within weeks rather than months or longer time periods (Whitehill, 2002a, Ruscello, 2006). This is based on the consensus in the literature that frequent repetition is needed for behavioural change to occur (Kleim & Jones, 2008, McAuliffe & Cornwell, 2008). In addition Law et al. (2004) tentatively suggest that interventions of more than eight weeks are more effective than those of less than eight weeks.

3.10 Aim of the study

This is a preliminary study in preparation for a possible explanatory trial in the future. The overall aim of this study is to evaluate the novel intervention outlined above in terms of feasibility and effect, in order to decide whether a full clinical trial should be pursued. If the results of this study are promising then it will be important to undertake a trial to assess whether VFTh is the best treatment for selected individuals with nasal speech. Only then will clear evidence be available to show that VFTh should be considered as a treatment approach. The primary aim of this study is therefore to explore the feasibility of a behavioural speech therapy intervention for nasal speech and the study will ask:

What is the feasibility of a trial of a newly developed visual feedback speech therapy intervention for adults and children with nasal speech associated with VPD?

The study aims to:

- Assess the feasibility of developing and delivering a behavioural speech therapy intervention (VFTh) for nasal speech.
- Explore a range of measurable and relevant outcome parameters.
- Consider the value of information to be gained from further research.

The most important parameters in this study relate to the intervention itself; the clinical outcomes, feasibility and acceptability of the intervention. It therefore fulfils the criteria of a feasibility study as defined by the National Institute for Health Research (NIHR) Evaluation, Trials and Studies Coordinating Centre (NETSCC, 2008);

'Feasibility Studies are pieces of research done before a main study in order to answer the question "Can this study be done?'' They are used to estimate important parameters that are needed to design the main study'.

2008 NETSCC (www.netscc.ac.uk/glossary)

This is distinct from a pilot study which NETSCC define as a time and/or size restricted version of a main study that is run to test whether the components of the main study can all work together. In reality the boundaries between these definitions are often blurred and there is overlap in the use of the terms in the literature along with a lack of formal guidance about what clearly constitutes one or the other (Lancaster et al., 2004). Thabane et al. (2010) argue that the main focus of a pilot study should be the assessment of feasibility. In contrast Arain et al. (2010) make a clear distinction between the specific requirements for a pilot and a feasibility study and recommend use of the NETSCC definitions for clarity. In relation to this study, there is currently no planned follow-up trial and therefore it does not represent a defined pilot stage in the route of progression to a substantive study. In addition, the emphasis is on assessment of the intervention rather than power calculations, sample size estimation and procedure testing, as would commonly be expected in a pilot study. This study is therefore a feasibility study, the main aim of which is to explore the components and

effect of a specific intervention in a specified population, with progression to a larger study dependent upon the progress of the study and the results obtained (Hayhow, 2011). The study also meets the criteria proposed by Bowen et al. (2009), that there are few previously published studies in the field but there is positive evidence for the intervention in different settings.

3.11 Research models and frameworks

3.11.1 Phased clinical research

The suitability of a feasibility study for assessing VFTh is further indicated by reference to several research models and frameworks. The first of these is a five-phased research model of clinical outcome research (Figure 3-8.) as described by Robey and Shultz (1998).

The model firmly frames this study as a preliminary study at Phase 1 to 2; attempting to detect a therapeutic effect in a selected group of subjects. The aim at this stage is therefore not to demonstrate efficacy but to adhere to the recommended standards of clinical outcome testing (Mackenzie & Lowit, 2007). This is appropriate given the context of the existing evidence base which is weak and methodologically flawed, as discussed in Chapter Two.

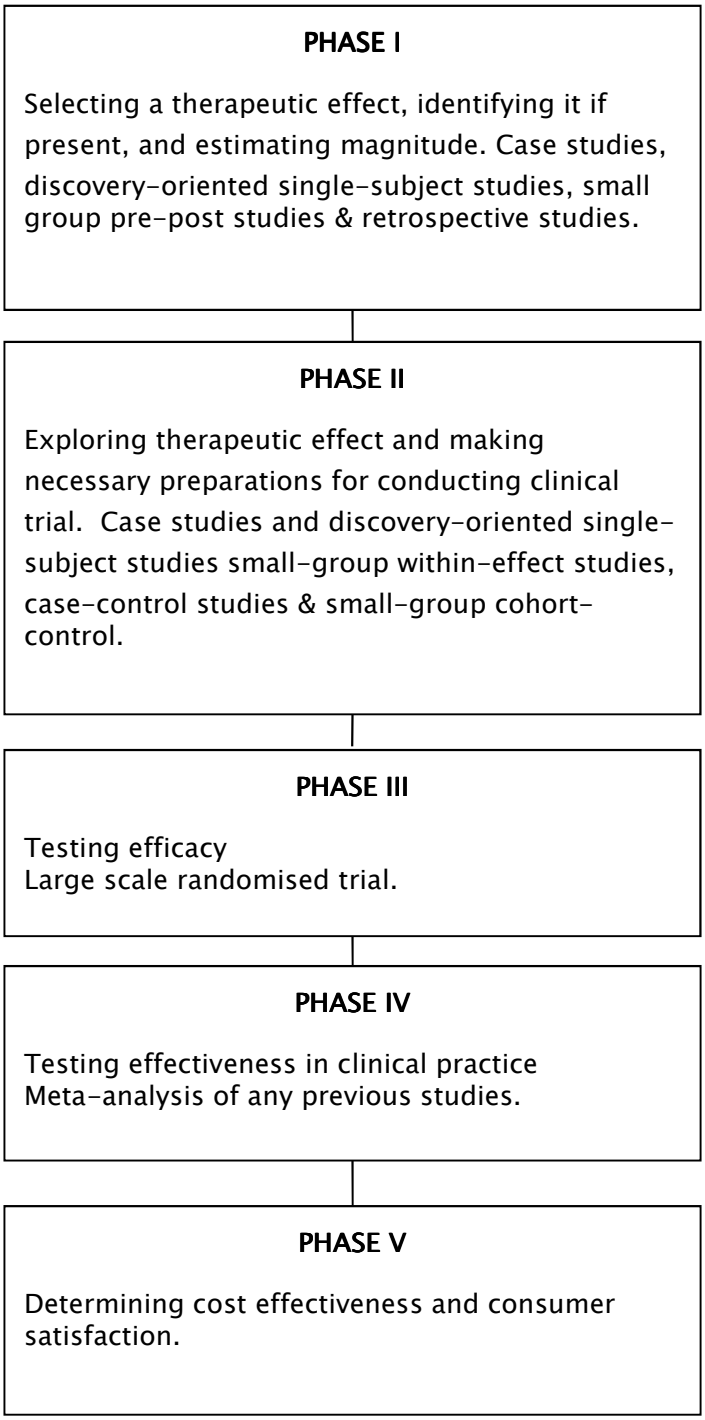

Figure 3-8: Phased clinical research model. Adapted from Robey (2004)

3.11.2 Complex Interventions

More recently the Medical Research Council has updated its guidelines on Complex Interventions, originally published in 2000, (MRC, 2008) and this provides a useful complementary framework to consider the design and aims of this study (Figure 3-9).

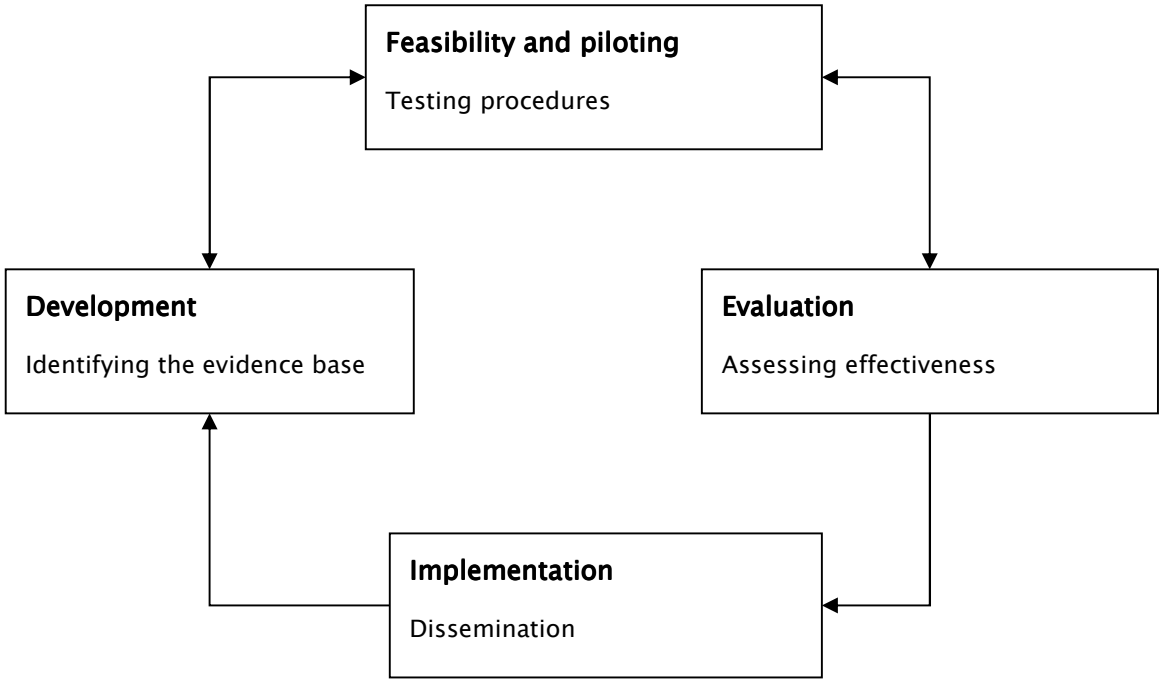

Figure 3-9: Complex interventions: Key elements of the development and evaluation process (Reproduced from Craig et al., BMJ 2008)

VFTh can be seen as a complex intervention as there are a number of interacting elements and outcomes to consider (Craig et al., 2008, Carding & Hillman, 2001). For example, although the main focus of the study is the effect of VFTh on speech there is also the nature of the client-therapist relationship to consider and the possibility that any change in behaviour is the result of a response to therapeutic attention. Therapy

is a dynamic process where the subtleties of the client-therapist interaction may be as important to the outcome as the techniques used (Horton, 2006).

The key objectives of this study involve examining the theoretical principles of VFTh and discussing how individual outcomes may relate to the intervention and may vary between individuals. This reflects the developmental nature of the study. In parallel the study aims to explore the feasibility of the intervention; highlighting the integration of process and outcome evaluation proposed in the MRC guidelines (Figure 3-9). This reflects a move away from earlier linear models such as Robey & Schultz (1998) and the previous MRC guidelines (2000). The updated MRC guidelines emphasize the importance of early phase developmental work. This study conforms to these guidelines and recognises the following crucial aspects in the development of the study intervention in order to justify further evaluation:

1. Identifying existing evidence.
2. Developing a theoretical understanding of the likely process of change.
3. Modelling: designing the intervention and identifying suitable measures.

3.11.3 Pre-experiment model

Finally, the approach taken in this study could also be described as a pre-experiment (Bowling, 2002). This captures features of a true experiment, i.e. systematic observation and the before and after measurement of a variable in relation to an intervention, whilst acknowledging that such an approach is more susceptible to bias due to the lack of randomisation and/or control group.

The question of whether observed changes could have occurred without VFTh cannot be fully answered by this study. It cannot demonstrate a causal relationship between the VFTh and speech outcomes but it may be possible to tentatively propose whether a relationship exists between the two variables. It is therefore important to view the study in the context of the available evidence and of speech and language therapy research as a whole, as highlighted in Chapter Two.

3.12 Selection of study design

In order to address the questions of feasibility and effect this study uses a within subjects case series design. This pre-test post-test design is appropriate given the current limited evidence base and the feasibility aims of the study. A randomised controlled trial is not appropriate at this point as the intervention has not yet been developed to a stage where it could 'reasonably be expected to have a worthwhile effect' (MRC, 2008). In addition the question of whether it would be possible to use the intervention needs to be addressed before committing time and funds to a substantive trial.

3.12.1 Pre-test post-test design

The pre-test post-test design is selected to answer the questions of this study in relation to the effect of the intervention. The study has been designed to maximise the advantages of the within-subjects design; participants meet specific inclusion and exclusion criteria (pg. 121-22) so that they share certain characteristics and they act as their own perfectly matched controls. The major strength of the design is therefore a reduction in error variance associated with individual differences (Hall, 1998; Bowling, 2002; Parahoo, 2006) as each participant serves as his or her own baseline. Carryover effects are not a concern as there is no comparison intervention or separate control group. In addition a practice effect or learning is actually a focus of the study.

3.13 Development of protocol: Assessment of recruitment potential

Prior to the main study there was an initial phase of work in order to develop the study protocol. This involved a review of existing patient records at the clinical sites where the research study would be recruiting, to confirm the existence of the proposed study population i.e. individuals with mild to moderate nasal speech associated with VPD who are seeking treatment for their nasal speech. At the time of this records review

there was incomplete information available due to the lack of a centralised database so that data was available in relation to individuals with non-cleft VPD only and not individuals with cleft VPD. In the period 2003-2007 there were 39 referrals recorded for individuals with nasal speech associated with non-cleft VPD, in 2008 there were 32 referrals recorded. Severity of nasal speech ranged from borderline to severe during both periods. The outcomes of the referrals are displayed in Table 3-2:

Table 3-2: Non- cleft VPD referrals and outcomes 2003-2007 and 2008

Referral period	Number of referrals	Surgical treatment	Speech therapy (articulation)	Prosthetic treatment	Watchful waiting
2003-2007	39	14 (36%)	12 (31%)	1 (2.5%)	12 (31%)
2008	32	11 (34%)	7 (22%)	1 (3%)	13 (41%)

A hand search was also undertaken of records of attendance at the Palatal Investigation Clinic (PIC) between January 2006 and December 2007. PIC is the main assessment clinic for individuals with nasal speech. It is an established clinic which takes place monthly, with six to eight patients (children and adults) attending each clinic. During the stated period 68 patients attended; 28 with repaired cleft palate and 40 with non-cleft VPD. Again the full range of severity of nasal speech was noted.

At this time there was limited comparative information available about referral rates and treatment options for individuals with nasal speech at cleft centres throughout the UK. Information was available about the caseload at one other cleft centre in 2000 (Sell, 2003). Sell reported that 76% of patients referred with nasal speech and/or articulation difficulties associated with VPD went on to have surgery, 15% had speech therapy (for articulation only) and 9% were treated with prosthetics.

This scoping work, whilst acknowledging the limitations of incomplete data, confirms the existence of the target population for the study i.e. individuals (children and adults) with mild or moderate nasal speech associated with VPD. In the most recent data from 2008, 36% of individuals identified at the research clinical sites (with non-cleft VPD) went on to have surgical treatment for nasal speech. This is considerably lower than the 75% figure reported by Sell (2003). This figure is likely to have been higher if data for individuals with cleft related VPD had also been available. Approximately one third of individuals in the same time period were offered speech therapy for articulation, another third were put into a watchful waiting group. This means that in 62% of cases surgery was not considered the appropriate treatment option at that time, in spite of the presence of nasal speech symptoms. The reasons for this decision in each case are not presented and it may be that some of these individuals went on to have surgery at a later date. However clinical experience indicates that in a proportion of cases surgery will not be offered or will be declined and this group could benefit from an alternative speech therapy treatment for their nasal speech.

Finally it is important to note that surgery rates and the rates of the other treatment options will to a certain degree reflect the particular treatment protocol of different centres. This is validated by later unpublished data provided by all UK cleft centres (Lead Cleft SLT Forum, 2010) which shows variation in referral rates and treatments offered for nasal speech between centres.

3.14 Development of protocol: Selection of outcome measures

A multiple outcome measure approach is considered essential in this study in view of the complex nature of the intervention, as described earlier in this chapter (Carding, 2000; Pring, 2005). As Carding (2000) recommends, the selection of measures is also informed by consideration of participant comfort as well as validity and reliability. The measures were therefore selected to be non-invasive and to include both perceptual

and instrumental assessment as well as evaluation by the participant. This is in contrast to the majority of previous studies which present a single measure only or use measures with no report of validity or reliability.

3.14.1 Perceptual assessment

Perceptual assessment is regarded as the 'gold standard' for assessment of disordered speech (Kuehn & Moller, 2000; Bhuta et al., 2004; Sell, 2005). It involves a listener or listeners judging a sample of speech on various parameters using a rating scale of some kind. Many such assessments have been developed using a variety of parameters and scales (Lohmander & Olsson, 2004), highlighting the difficulties in standardising approaches to speech assessment, particularly across linguistic barriers.

The Great Ormond Street Speech Assessment (GOS.SP.ASS, Sell et al., 1999) is the routine clinical assessment used in the UK for patients with cleft palate and VPD. The Cleft Audit Protocol for Speech-Augmented (CAPS-A, John et al., 2006) was developed with reference to GOS.SP.ASS and was selected as the perceptual measure in this study as it is a validated tool specifically designed for use in reporting speech outcomes for children with cleft palate, a common cause of nasal speech (Appendix D). It is in routine use for capturing speech outcomes across UK cleft centres and its reliability and validity is well documented which facilitates wider applicability as a tool for audit and research (John et al., 2006). In addition, an extensive training programme has been delivered in the UK over the past 5 years resulting in a bank of specialist speech and language therapists (SLTs) who are trained in the use of CAPS-A and whose reliability has been assessed (Sell et al., 2009). Finally, ongoing maintenance of listening skills through programmes of regular consensus listening is well established in UK speech therapy clinical settings. Five CAPS-A trained SLTs were used as expert listeners in this study and their intra-rater reliability results obtained as part of the training process are presented in Chapter Five.

CAPS-A consists of ten sections each covering a separate and defined speech parameter (intelligibility, voice, hypernasality, hyponasality, audible nasal emission, nasal turbulence, grimace, cleft speech characteristics, non-cleft speech errors, and speech-and-language therapy intervention). Each parameter has a rating scale with between two and five points, depending on the nature of the parameter. The parameters selected for rating in this study are **hypernasality**, **audible nasal emission** and **nasal turbulence** as these are the speech symptoms that contribute to the perception of nasal speech.

Hypernasality is rated using a five point scale, where 0 = absent and 4 = severe: evident on vowels and voiced consonants). Audible nasal emission and nasal turbulence are rated using a three point scale, where 0 = absent on pressure consonants and 2 = frequent: pressure consonants affected >10% of the sample (judged as highly pervasive or highly distinctive). The term pressure consonant refers to English speech sounds that involve the build-up of air pressure in the mouth which is then released by the tongue or lips to produce a sound known as either a stop (e.g. 'p' 't' 'k') or fricative (e.g. 'f' 's' 'sh') (Harding & Grunwell, 1996).

The standard CAPS-A speech sample is used in this study and includes:

1. Spontaneous speech 'Tell me about……'
2. Automatic speech: Counting 1-20 and 60-70.
3. Repetition of 22 sentences; representing all English consonants (Appendix D).

3.14.2 Instrumental assessment

Instrumental assessment is considered to be a valuable adjunct to perceptual assessment (Sweeney & Sell, 2008; Dalston et al., 1991; Bressman et al., 2006). The Nasometer ™, an instrumental device available since 1986 (Kay Elemetrics, Lincoln Park, NJ), is used to carry out measurement and analysis of the ratio of oral and nasal airflow during speech (nasometry). It measures the acoustic correlate of nasality and

results are expressed as a percentage nasalance score (nasalance = nasal/ [oral + nasal] x 100, Watterson et al., 1998).

Nasometry (current Nasometer™ II model 6450) was chosen as the instrumental assessment in this study as again it is routinely used in clinical assessment in UK cleft centres, particularly before and after palate surgery to improve speech. It is also used in routine clinical care to supplement perceptual assessment as required.

There have been concerns about performance variability relating to dialect and gender (Seaver et al., 1991) but a study by Dalston et al. (1991) demonstrated good levels of specificity and sensitivity for this tool. In a later study Keuning et al. (1999) did not find evidence of good correlation between perceptual ratings of nasal speech and nasalance scores. However, Sweeney et al. (2004) comment on the methodological differences between studies that report variable correlations between the two types of measure. They found good test-retest reliability for the Nasometer™ and in a later study (Sweeney & Sell, 2008) a strong relationship between perceptual ratings of hypernasality and instrumental nasometry scores.

The procedure for nasometry involved the participant wearing a headset comprising of a sound separator and an oral and nasal microphone on either side. The Nasometer™ was calibrated according to the manufacturer instructions prior to each assessment. Participants were asked to repeat a series of 16 standard stimulus sentences read by the CI (Appendix E). The stimulus sentences are those used in the studies by Sweeney et al. (2004) and Sweeney & Sell (2008) and are adapted from the Great Ormond Street Speech Assessment (GOS.SP.ASS, Sell et al., 1999). They are very similar to the sentences used in CAPS-A in that they are presented in groups according to consonant types (high pressure, low pressure, mixed, nasal). They are also used in routine clinical practice to support perceptual assessment.

Finally, nasometry was also used in this study as one of the VFTh feedback tools (see page 101).

3.14.3 Quality of Life assessment (QoL)

Quality of life assessment was included in the battery of measures in order to capture the subjective impact of VFTh for each participant. This reflects a growing trend of evaluating the impact of speech disorders on affected individuals, both children and adults (Carding, 2000; McLeod & Bleile, 2004; Threats and Worral, 2004; Markham & Dean, 2006). McLeod and Bleile (2004) propose the use of the International Classification of Functioning, Disability and Health (ICF World Health Organisation, 2001) as a goal setting framework for children with speech impairment. The key concepts are activity and participation in facilitating health and well-being, regardless of the degree of impairment affecting body structure and/or function. This is in contrast to the traditional aetiology and impairment based models used in speech and language therapy (Bowen, 2009). In an earlier paper McLeod (2003) describes the use of a series of professional workshops with 200 speech pathologists to develop a tool designed to capture the impact of speech difficulties on children's lives. The Speech Participation and Activity of Children (SPAA-C) tool was designed to be used flexibly with the child and significant others. The SPAA-C remains un-validated and therefore has not been selected for use in this study. However this type of work reflects a significant shift in speech assessment and goal setting where greater consideration is given to areas previously seen as informal.

In relation to adults QoL tools have also been increasingly used with a range of groups, for example adults with aphasia (Ross & Wertz, 2003), traumatic brain injury (Horneman et al., 2005), and voice disorders (Fang et al., 2008). Cruice (2008) evaluates the contribution of the ICF to quality of life concept development in adult communication disorders. Whilst acknowledging the conceptual importance of the framework, Cruice argues that the ICF model can lead to quality of life being

'compartmentalised' and overly focused on functioning so that subjective well-being is neglected. However Cruice is clear that;

'improvements in quality of life are an outcome of health and health care, and require measurement just as frequency, severity and consequences of diseases are measured.'

Cruice (2008: 43)

3.14.3.1 Selection of QoL measures

Three quality of life questionnaires were selected to cater for both child and adult participants (Table 3-3. and Appendix F). The **Peds QL 4.0** measure, a generic measure of quality of life, was initially selected for use with children in the study as it is well validated. Locker et al. (2005) argue that generic measures fail to capture condition specific concerns. They describe the use of the Child Perceptions Questionnaire (CPQ, 11-14), a component of the Child Oral Health Quality of Life Questionnaire. This measure does specifically relate to the appearance and function of the mouth and teeth but does not include speech concerns so was not appropriate for this study. The **Ped SaL QoL** (Markham, 2008) was then identified as a more valid measure for children with speech difficulties and a decision taken to include this alongside the generic measure as it may be more sensitive to the features of interest. This measure has been developed and validated in populations of children and young people with speech, language and communication difficulties and so is condition specific and is likely to offer greater validity in the evaluation of quality of life in the study population.

For adults, the literature related to voice disorders offered the most appropriate potential measures as abnormal resonance, (i.e. where an oral nasal imbalance is perceived), is a feature of voice and there was no tool available specifically for individuals with nasal speech or VPD. There are a number of validated voice related tools available including the VR-QOL (Hogikyan & Sethuraman, 1999), the Voice Handicap Index (Jacobson et al., 1997) and the Voice Symptom Scale (VoiSS, Deary et

al., 2003). The VoiSS was selected for use in this study as it best represents a well validated, patient reported measure of outcome for disorders of voice.

These questionnaires provide a comprehensive measure of QoL for this study which combined with the perceptual and instrumental measures of speech fulfil the objective of multidimensional outcome assessment.

Table 3-3: QOL measures selected for the study

1. The Pediatric Quality of Life Inventory (Peds QL 4.0):
23 items in four sections:
Health and Activities
Feelings
Getting on with others
School
2. The Paediatric Speech and Language Quality of Life Scale (Ped SaL QoL):
37 items in five sections:
Satisfaction
Communication and Feelings
Independence and Participation at school
Support at school
Activities
3. The Voice Symptoms Scale for adults (VoiSS):
30 items, covering subjective perception of:
Voice
Feelings
Function
Health

3.15 Conclusion and research questions

In this study the intervention being explored is Visual Feedback Therapy (VFTh), a behavioural speech therapy treatment incorporating visual biofeedback and

performance feedback. This chapter defines the components of VFTh and outlines its development with reference to existing empirical evidence and theory. This study is therefore unique in presenting a conceptual model of the intervention of interest. The study is informed by the positive findings for some subjects in previous studies, as well as their methodological limitations as discussed in Chapter Two. A combination of outcome measures have been selected, including self-reported quality of life, an area largely overlooked in previous studies. In summary, this study contributes in four main ways to the understanding of whether individuals with nasal speech might benefit from a VFTh intervention:

1. It examines the feasibility of the intervention in a series of individuals with nasal speech.
2. It proposes a theoretical model to describe how the intervention might work.
3. It employs a valid and acceptable multidimensional framework for outcome assessment; including perceptual, instrumental and quality of life measures
4. It is delivered exclusively in order to avoid the confounding effect of other speech therapy interventions.

This feasibility study therefore contributes to the body of knowledge generally regarding speech therapy using visual biofeedback, and specifically in the treatment of nasal speech. This has several important clinical implications; the first being that it addresses the ongoing need for the identification of evidence-based treatments. In addition it presents the possibility that an individual's speech disorder, and the associated impact on quality of life, can be resolved in a non-invasive way or addressed where there had previously been no appropriate therapeutic option. This chapter concludes with the emergent research questions, representing the appropriate next step in evaluating VFTh, and the following chapter outlines the study methods.

3.15.1 Research questions:

What is the feasibility of a newly developed speech therapy intervention combining visual biofeedback and performance feedback for children and adults with nasal speech associated with VPD?

Does VFTh improve nasal speech and quality of life as measured by a range of perceptual, instrumental and patient-reported outcome measures?

Chapter Four: Methods

4.1 Introduction

This study uses a Visual Feedback Therapy (VFTh) intervention, and examines its feasibility and the mechanisms by which it may effect change. The study takes a combination of measures, including perceptual, instrumental and quality of life assessment, in order to provide a comprehensive evaluation of speech characteristics before and after VFTh. This chapter reiterates the aims and objectives of the study and describes the standardised assessment protocol and procedures involved.

4.2 Aims and objectives

As outlined in Chapter Three, this study aims to address the following research questions relating to feasibility and effect:

What is the feasibility of a newly developed speech therapy intervention combining visual biofeedback and performance feedback for adults and children with nasal speech associated with VPD?

Does VFTh improve nasal speech and quality of life as measured by a range of perceptual, instrumental and patient-reported outcome measures?

4.2.1 Study objectives: Feasibility

The study is designed to answer the following specific questions relating to the feasibility of the intervention:

1 How feasible is it to develop and deliver a behavioural speech therapy intervention (VFTh) for nasal speech?

2 What are the most relevant outcome parameters and appropriate measures?

3 What valuable information could be gained to inform further research?

4.2.2 Study objectives: Effect of VFTh

The study is designed to answer the following specific questions relating to the effect of the intervention:

1. Is there any change in nasal speech symptoms (hypernasality, nasal emission and nasal turbulence), before and after VFTh, as measured by the perceptual and instrumental measures?
2. If there is change in nasal speech symptoms, is it still detectable at three month follow-up, as measured by the perceptual and instrumental measures?
3. Is there any change in quality of life reported by the participant before and after VFTh, as measured by the quality of life measures?

4.3 Methods

4.3.1 Setting

The clinical context of this study is a designated regional cleft centre offering specialist multidisciplinary care from birth, or referral, to individuals with cleft lip and palate and/or VPD. The centre consists of two clinical sub-centres with twin clinical teams, sited 75 miles apart. Approximately 160 individuals each year attend across the two sites for assessment of nasal speech and palate function; this includes individuals with cleft palate related VPD as well as those with non-cleft VPD.

4.3.2 Ethics

This study has NHS Research Governance and National Research Ethics Service (NRES) ethical approval (Appendix G). It has been undertaken as part of a Clinical Doctorate programme at the University of Southampton and is therefore also subject to the University's research governance process. Confidentiality of participants' personal

data was assured along with anonymity of all research data, as per Good Clinical Practice research governance guidelines (1996).

There were several areas requiring specific ethical consideration. First of all this study includes children and young people from the age of 7 years. This required that that both parental consent, and assent or consent from the child, was gained. Young people aged 16 or 17 years are presumed competent to give consent for themselves (Department of Health, 2001). Clear and appropriate verbal and written explanations of the study aims and procedures were provided and written consent or assent documented (Appendix H & J). Clinical experience indicates that in the case of children there can be a conflict between the motivation of the child and the parent/s to undergo therapy. As cooperation with treatment is required it was essential that all participants were willing to comply. A child was only accepted as a participant if both he/she and the parent/s were in agreement and all participants were advised of their right to withdraw from the study at any time without prejudice to their ongoing clinical care.

Access to VFTh was a potential issue as participants could live up to one hour's travel distance away from the treatment centre, more if public transport was used, and were required to attend for twelve sessions. This could have resulted in a significant impact on school attendance for children and work/college for adults, as well as the financial expense. The need to travel to access VFTh and to commit to a series of twelve appointments could also introduce selection bias. In view of these issues a bid was successfully submitted to the speech therapy Trust Fund to purchase a portable Nasometer™. This allowed VFTh to be delivered at locations away from the cleft centre and closer to participants' homes, widening both access and recruitment. In addition travel expenses were reimbursed with support from the local branch of the Cleft Lip and Palate Association (CLAPA).

4.3.3 Sample

The recruitment of participants from the Chief Investigator's (CI) treatment centre introduced the potential for selection bias. This was addressed by using other members of the centre's speech therapy team to recruit participants as they presented to the clinical service. The team of seven SLTs were given a recruitment pack containing the research proposal, inclusion/exclusion criteria and study information sheets (see Appendices). They were asked to identify potential participants according to the inclusion/exclusion criteria and to introduce the study and give the written information sheets. Participants could agree to participate at this point if they wished but were given the option to take the information away to allow time for decision making. In this instance they were asked for permission for the CI to contact them in one week to discuss recruitment (Table 4-1). Participants were also advised that they were free to change their mind about participating at any time. The full research protocol is included in Appendix G.

4.3.4 Sample size

Participants were recruited consecutively from the start date of the study as they presented to the clinical service, at either of the two centre sites. It was not appropriate to carry out sample size calculations due to the feasibility nature of the study. As the Chief Investigator was the sole SLT delivering the intervention, a target of twenty study participants was set. The recruitment target was greater than in many previous studies in the literature review, where nearly two thirds had ten subjects or less and of this one third were single case studies. It was also important that the sample was representative of the population of interest, hence the preliminary caseload scoping work and consecutive recruitment strategy.

Table 4-1: Recruitment process

1. Child or adult attends routine multidisciplinary cleft clinic, as new referral or follow-up. 2. Routine speech assessment is carried out by specialist SLT (not the CI) 3. Potential research participant is identified by the assessing specialist SLT, according to inclusion criteria. 4. Information is given about the research project, verbally and in information sheets. 5. Child (with parent/carer agreement) or adult is invited to participate. 6. Child (with parent/carer agreement) or adult agrees to participate, is formally recruited and consent/assent forms* completed. Confirmation letter is sent within two weeks.
Or
1. Child or adult chooses to have more time to make their decision. 2. CI contacts child (parent/carer) or adult by telephone one week after clinic attendance to invite participation. 3. Child (with parent/carer agreement) or adult agrees to participate and is formally recruited. Confirmation letter and consent/assent forms* are sent within two weeks.
Or
1. Child (parent/carer) or adult declines to participate and continues with routine follow-up/care.

*consent/assent forms Appendix J.

4.3.5 Inclusion and exclusion criteria

The inclusion and exclusion criteria for participant selection were as follows:

Inclusion:

- Minimum age 7 years, with no upper age limit.
- Presenting with mild/moderate hypernasality/nasal emission/nasal turbulence as rated in routine speech assessment using the Great Ormond Street Speech Assessment [GOS.SP.ASS]).
- Evidence of inconsistency of speech symptoms as rated in routine speech assessment using GOS.SP.ASS
- Diagnosis of velopharyngeal dysfunction confirmed by routine palatal investigation (videofluoroscopy/nasendoscopy).
- Evidence of potential for velopharyngeal closure during speech confirmed by routine videofluoroscopy and/or nasendoscopy.
- Participants, and where appropriate parent/s, express a desire to change their existing speech symptoms.

Exclusion:

- Routine videofluoroscopy and/or nasendoscopy confirm the diagnosis of velopharyngeal dysfunction but with no evidence of potential for velopharyngeal closure during speech.
- Presenting with severe hypernasality/nasal emission/nasal turbulence indicative of an underlying structural velopharyngeal defect.
- Presence of moderate or severe hearing impairment which would prevent cooperation with the therapy programme and home practice schedule.
- Presence of a diagnosed learning disability severe enough to prevent cooperation with the therapy programme and home practice schedule.

4.3.5.1 Rationale for inclusion/exclusion criteria

The lower age limit for recruitment was set to allow for understanding of and cooperation with VFTh as a behavioural intervention. This is informed by

developmental theory and clinical experience. Piaget's cognitive developmental framework (Atherton, 2010) describes a period (concrete operations) from around the age of 7 to 12 years characterised by increasingly organised, logical thought and concrete problem solving ability as opposed to earlier intuitive and more egocentric thinking. As Atherton (2010) states, this stage theory can be viewed as somewhat rigid and younger children may be able to understand and respond to VFTh successfully. However the stages of attention control proposed by Cooper et al. (1978) also support the minimum age of 7 years, as by this age attention is well established and maintained for up to sixty minutes.

Potential for velopharyngeal closure was established using **videofluoroscopy** and **nasendoscopy**, which was carried out in routine palatal investigation clinics. Videofluoroscopy is an investigation using X-ray to assess the movement of the soft palate during speech. This gives a lateral view of palate movement in relation to the back of the throat as shown in Figure 4-1.

Figure 4-1: Videofluoroscopy image of soft palate movement

Nasendoscopy is a procedure where a small camera is passed through the nose to view the surface and movement of the soft palate during speech as shown in Figure 4-2

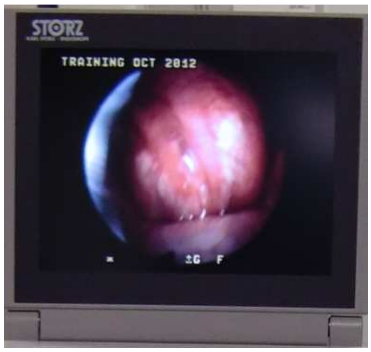

Figure 4-2: View of the velum (soft palate) and the adenoids (throat) using nasendoscopy

These instrumental techniques highlight any variability in palate function across a range of speech tasks (sounds, words, sentences). Inconsistent velopharyngeal closure must be observed and was taken as potential for improved function. This was supported by evidence of inconsistently nasal speech as rated on the GOS.SP.ASS perceptual speech assessment. The above measures are all routine clinical practice as part of the assessment process for individuals requesting treatment for nasal speech and therefore do not form part of this research intervention.

4.4 The Intervention (VFTh)

The development of the Visual Feedback Therapy intervention is fully described in Chapter Three. It comprised:

- A VFTh session once a week for 45 minutes over eight consecutive weeks from the Chief Investigator (CI) only, a specialist SLT.
- Home practice for 10-15 minutes each day during the study period using sound and/or wordlists generated during each intervention session.
- No other speech therapy intervention for the duration of the study.

The two active components of VFTh were:

- Instrumental visual biofeedback: relating to levels of nasal speech.
- Performance feedback: relating to the participant's attempts to modify nasal speech.

4.5 Outcomes

4.5.1 Perceptual assessment (CAPS-A)

Participants provided an audio speech recording at each of the four assessment points. This consisted of the standard CAPS-A speech sample (Appendix D) recorded using a Phillips Digital Voice Recorder Model TCD-D8. The recorder was positioned approximately 15 cm from the speaker's mouth, as per the manufacturer's instructions for optimum recording quality. All assessments were completed by the CI and took place in a quiet clinic room. Recordings were assigned a participant number and transferred to a secure computer in the SLT department. Data linking participant numbers to participant identity was stored on a separate secure computer to the audio recordings, again in the SLT department. All information identifying participants was edited from the recordings prior to being randomised for assessment point and copied to an encrypted audio CD for expert rating.

Speech recordings were assessed by a group of five specialist CAPS-A trained SLTs (expert listeners) who work in the same clinical setting as the CI but who were not involved in the clinical care of the study participants. The procedure for expert listening is included in appendix D. The expert listeners blindly rated the nasal speech symptoms using the CAPS-A scale. It is widely accepted that blind independent analysis of speech data by specialist SLTs should be the gold standard methodological approach when reporting audit and research outcomes (Sell, 2005). In addition a consensus rating was used in line with recommended practice in assessing speech outcomes (Sell et al., 2009). The expert listeners rated the speech symptoms independently and then discussed the ratings until agreement was reached. This method is supported by Carding (2000) who comments that:

'Unanimous judgement by a group of listeners may provide one of the most convincing measures of the effectiveness of therapy.'

Carding (2000:46)

Listening conditions were standardised through the use of a standard location and equipment. This comprised: Genelec 1029A Active 2 way monitors, Marantz PDM331 CD Player and Mackie 1402VLZ mixing desk. Recordings were randomised and coded as to assessment point and participant, to control for bias from these two factors. The content of the recordings provided no clues as to the assessment point. A reference sample of non-nasal speech was also inserted between participant recordings to minimise direct comparison between consecutive recordings. This sample was provided by volunteers with no history of nasal speech. The adult sample was matched for age and gender (adult male) and the child sample was matched for age only as gender differences in speech are not significant in the age group of the participants studied.

4.5.2 Instrumental assessment (Nasometry)

Nasometry assessment was carried out at each of the four assessment points. Participants were recorded repeating the sixteen standard nasometry sentences (Appendix E) while wearing the nasometry headset. The recorded data and analysis were saved on each occasion on a secure computer in the SLT department or on the password protected portable Nasometer™ II. The nasometry scores at each assessment were also recorded in the participant's clinical file in case of electronic data loss.

4.5.3 Quality of life assessment

Quality of life assessment was carried out at two assessment points only, A1 (first baseline pre-intervention) and A4 (follow-up). It would not have been appropriate to administer the questionnaires at all four assessment points due to the length of these questionnaire measures.

4.6 Study procedure and data collection

Each participant attended four assessment and eight VFTh sessions; a total of twelve appointments over 6 months as shown below (Table 4-2). The four assessment points are A1 (first baseline pre-intervention), A2 (second baseline pre-intervention), A3 (post-intervention), and A4 (follow-up). A2 is included to examine stability of speech symptoms and A4 to examine any continuation of effect at three month follow-up.

Table 4-2: Study timeline

Time	Session	Procedures	Outcome or Intervention
Week 1	A1 (Pre-intervention)	Perceptual – CAPS-A Instrumental – Nasometry QoL – PedsQL, Ped SaL QoL, VoiSS	Outcome
Week 3	A2 (Pre-intervention)	Perceptual – CAPS-A Instrumental – Nasometry	Outcome
Week 5-12 (inclusive)	Intervention	Visual Feedback Therapy	Intervention
Week 13	A3 (Post-intervention)	Perceptual – CAPS-A Instrumental – Nasometry	Outcome
Week 25	A4 (Follow -up)	Perceptual – CAPS-A Instrumental – Nasometry QoL – PedsQL, Ped SaL QoL, VoiSS	Outcome

4.6.1 Results and data analysis

Analysis of the results of this study primarily involved evaluation of the feasibility aspects as previously discussed. Outcome data analysis then took the form of descriptive summary statistics and detailed description of individual participant's response to the intervention. At this stage the emphasis was not on establishing

statistical significance but rather to evaluate the intervention and analyse any differences between participant's performances before and after the intervention, in keeping with the feasibility aims. Analysis therefore focused on the difference in perceptual speech ratings, instrumental nasometry scores and quality of life questionnaire scores from pre-intervention to post-intervention assessment and follow-up at 3 months.

A comparison of the two pre-intervention assessments (A1 and A2) was undertaken to examine the stability of nasal speech symptoms. This was important in terms of assessing any differences observed at the assessment points following the intervention (A3 and A4).

Analytical statistics were employed, but in a way appropriate to the feasibility focus of the study and the repeated measures design. Non-parametric analysis was used as no assumptions could be made about the distribution of the data and the sample size was small (Petrie & Sabin 2000). Statistical tests were selected to evaluate the differences between the paired measurements taken for each individual, before and after the intervention (IBM SPSS Statistics 19).

4.7 Conclusion

The study aims and methods are outlined in this chapter. This study aimed to examine the feasibility and effect of Visual Feedback Therapy (VFTh) using a pre-test post-test design. The multidimensional outcome measures described are considered valid and reliable, are in common usage and were selected to provide a comprehensive and complementary range of outcomes. Both perceptual and instrumental assessments were included and in addition to these observed outcomes the study also used quality of life questionnaires to capture participant perceived outcomes. Details of the data analysis are given and any alternative or subsequent analysis will be described in the next chapter along with the study results.

Chapter Five: Analysis and Results

5.1 Introduction

The analysis and results of the feasibility study are presented in this chapter in order to determine whether further evaluation of VFTh is practicable or worthwhile. The focus is primarily on the feasibility issues of recruitment and retention of participants alongside delivery of and participant response to the intervention. Individual and group outcome data is also examined, and results presented graphically together with descriptive summary statistics. For completeness and to further explore the effects of the intervention, preliminary inferential analysis of the results is reported for all outcomes, with full details contained in Appendix L. The chapter concludes with detailed case descriptions of all the study participants' outcomes and their individual response to the VFTh intervention.

5.1.1 Research questions

Feasibility: What is the feasibility of a newly developed speech therapy intervention combining visual biofeedback and performance feedback for adults and children with nasal speech associated with VPD?

Effect: Does Visual Feedback Therapy (VFTh) improve nasal speech and quality of life as measured by a range of perceptual, instrumental and patient-reported outcome measures?

5.2 Participants

Participants were recruited to the study from a specialist treatment centre for cleft palate/velopharyngeal dysfunction (VPD). A total of twenty-four individuals fulfilling the inclusion criteria, as described in Chapter Four (page 128), were approached

during the time period of eighteen months available for completion of the study, see adapted CONSORT flow-chart below (Figure 5-1).

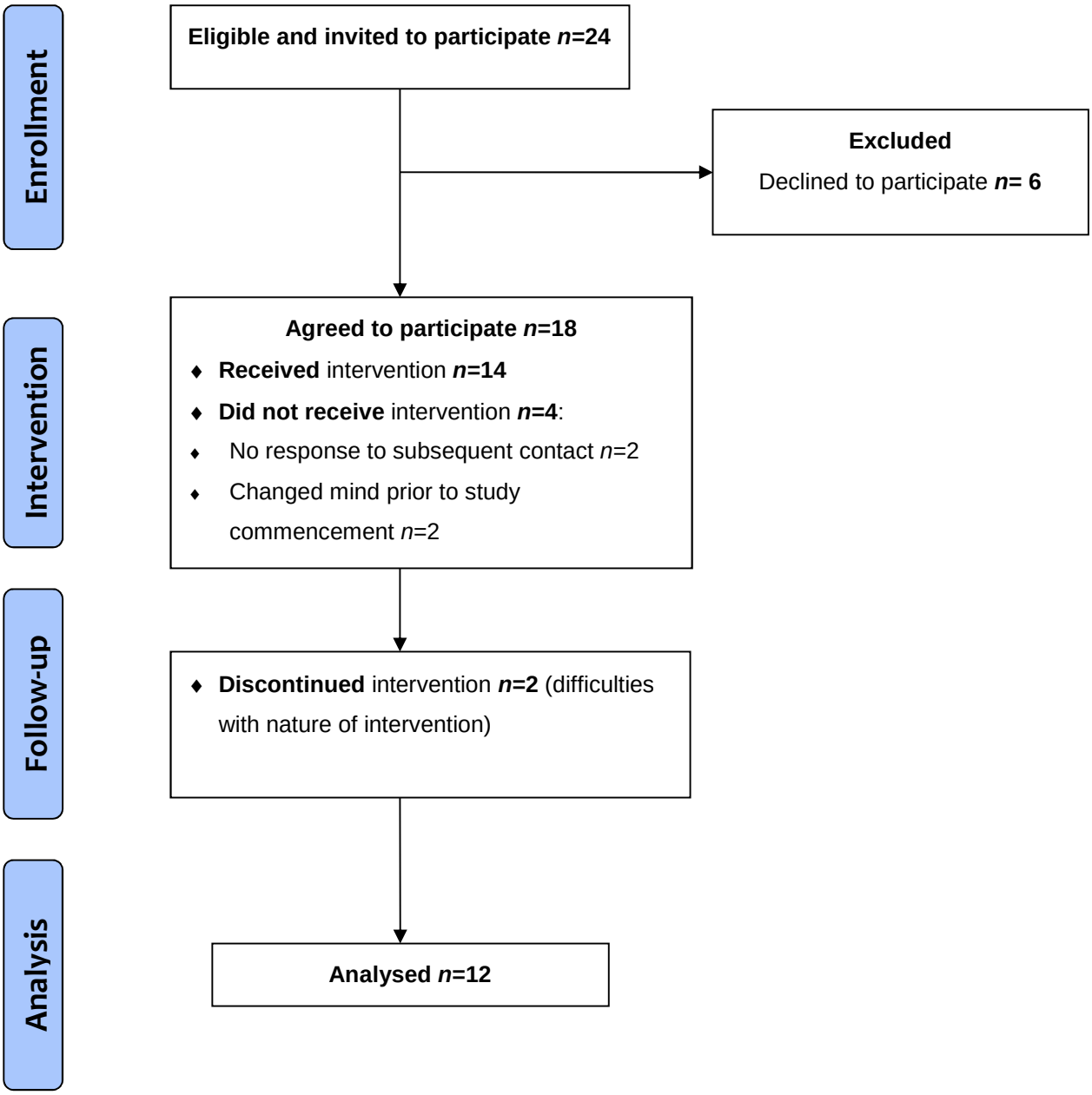

Figure 5-1: Adapted CONSORT flow-chart

5.2.1 Eligibility

Twenty-four individuals were identified as having nasal speech by a specialist speech and language therapist at routine clinical assessment, met the inclusion/exclusion

criteria and were invited to take part. This routine assessment data was used to assess eligibility for inclusion in the study and this information played no further part in the data analysis for participants who completed the study. The specific nasal speech symptoms observed in each participant at this point are summarised in Appendix L, Table L-1.

5.2.2 Individuals who declined to participate

Twenty-four individuals were initially invited to participate and six declined; details of the six patients who declined are shown in Table 5-1, including any reason given for not participating in the study.

5.2.3 Individuals who initially agreed to participate but did not commence the study

Table 5-2 provides details of four individuals who initially agreed to participate but did not commence the study. Two individuals did not respond to further written contact. One individual changed their mind about taking part as they realised regular travel to the centre would be too difficult, and at that time the portable nasometer had not yet been purchased. Another had been invited to take part whilst waiting for palate surgery. This individual subsequently decided to wait until after surgery. None of these individuals received any study assessment or intervention sessions.

Table 5-1: Eligible patients who declined to take part in the study (*n*=6)

Patient	Gender	Age	Aetiology	Reason for declining
1	Female	8	Non-cleft VPD	Not known
2	Male	10	Cleft palate	Impact on school attendance
3	Female	13	Non-cleft VPD	Impact on school attendance
4	Male	14	Cleft palate	Not known
5	Male	17	Cleft palate	Not concerned about mild nasal speech symptoms identified at routine cleft review
6	Male	9	SMCP	Not concerned about mild nasal speech symptoms identified at routine cleft clinic review

VPD, velopharyngeal dysfunction; SMCP, submucous cleft palate

Table 5-2: Individuals who initially agreed to participate but did not commence study (n=4)

Patient	Gender	Age	Aetiology	SLT history	Reason for not commencing intervention
1	Male		Cleft palate	Yes- CSCs	No response to written contact
2	Female		Non-cleft VPD	Yes- Language development	No response to written contact
3	Male	11	Non-cleft VPD	Yes – Articulation	Travel difficulties
4	Male	10	Non-cleft VPD	Yes – Language & articulation	Deferred until after surgery

VPD, velopharyngeal dysfunction; CSCs, cleft speech characteristics

5.2.4 Individuals who discontinued the study

Two participants completed the study assessments and commenced the intervention but discontinued due to difficulties with the intervention itself (M and N, Table 5-3). Participant M, a 17 year old girl, found it difficult to be presented with visual feedback of her speech and what she described as 'getting it wrong all the time'. This was despite careful explanation of the aims and procedure for the intervention and reassurance that variation in performance was not a negative outcome. This participant attended four intervention sessions before it was agreed that she should

withdraw from the study. The second participant to withdraw after starting the intervention was an 8 year old boy who was highly distractible and unable to sufficiently attend to the visual feedback being presented, even with a high level of performance feedback. This was discussed with the boy's mother and he was withdrawn from the study after five sessions. In both of these cases there was potential for the intervention to adversely affect the participant, (see discussion, Chapter Six). Several other participants also required careful management of the feedback components of the intervention, as described later in the individual case studies, in order to maximise success and maintain motivation. The implications of this for future delivery of the intervention are discussed in the following chapter.

Table 5-3: Individuals who discontinued the study (*n*=2)

Patient	Gender	Age	Aetiology	SLT history	Reason for discontinuing
M	Female	17	Cleft palate	Yes - CSCs	Difficulties with intervention
N	Male	9	SMCP	Yes - CSCs	Difficulties with intervention

SMCP, submucous cleft palate; CSCs, cleft speech characteristics

5.2.5 Individuals who completed the study

Twelve individuals went on to complete the study; six female and six male participants, representing a 66.6% completion rate (twelve completers from eighteen individuals who initially agreed to participate), full details are given in Table 5-4. Eleven of the participants were children between 7 and 13 years of age and there was one male adult aged 43 years. The mean age of the children was 9.2 years.

Table 5-4: Participants who completed the study (*n*=12)

ID	Gender	Age	Aetiology	SLT history
A	Female	9	Cleft palate	Yes – CSCs
B	Female	7	Cleft palate	Yes – CSCs
C	Male	9	Non-cleft VPD	No
D	Male	7	SMCP	No
E	Male	8	Non-cleft VPD	Yes – S&L delay
F	Female	7	Non-cleft VPD	Yes – S& L delay
G	Male	13	Non-cleft VPD	No
H	Female	8	SMCP	Yes – CSCs
I	Male	43	Cleft palate	No
J	Male	10	Non-cleft VPD	Yes – Dyspraxia
K	Female	12	SMCP	No
L	Female	11	Non-cleft VPD	Yes – Articulation

VPD, velopharyngeal dysfunction; SMCP, submucous cleft palate; CSCs, cleft speech characteristics; S&L delay, speech & language delay

Six participants had a history of cleft palate, three of whom had a submucous cleft palate. The remaining six participants had a diagnosis of non-cleft VPD i.e. dysfunction of the soft palate in the absence of an overt cleft palate or submucous cleft palate. Seven participants had received individual speech therapy intervention

before recruitment, for a variety of speech and or language difficulties as indicated, but not for nasal speech.

Prior to admission to the study (at eligibility assessment, pre-recruitment) as one of the inclusion criteria, all twelve participants were rated as hypernasal; six participants were additionally rated with audible nasal emission and two with nasal turbulence, one of whom had both. This differs from the profile of the speech symptoms subsequently identified at the baseline (pre-intervention) speech assessments (A1and A2) where only eleven participants were rated as hypernasal. Nine participants differed in one or more nasal speech symptoms between pre-recruitment and pre-intervention and the implications of this difference are discussed in Chapter Six.

In addition, at routine assessment (pre-recruitment) six participants were noted as having active nasal fricative (see Chapter Four) as an inconsistent feature of their speech, which was in addition to the nasal speech symptoms measured in this study (Appendix L, Table L-1). This feature is not the focus of this study, however it is potentially confounding in relation to the outcome measures used in the study and this aspect will also be discussed in Chapter Six.

5.3 The intervention

Three of the twelve participants received eight VFTh intervention sessions at weekly intervals, as per the study protocol. The remaining nine also received eight sessions but not over consecutive weeks due to illness (participant or chief investigator), or participant holiday, school or work commitments (Table 5-5). Four participants had one delayed session (range 2 to 4 weeks) but in three cases there were two delays (2 to 3 weeks) and in two cases three delayed sessions (3 to 4 weeks). This variation from the study protocol is another potential confounder and will be discussed in Chapter Six.

Table 5-5: Participants who had disruptions to the study protocol

Participant	Gender & Age	Number of delayed sessions (maximum 8)	Maximum delay (weeks)
A	Female, 9yrs	1	4 (incl. Easter)*
B	Female, 8yrs	1	2 (incl. Half Term)
D	Male, 8yrs	3	3 (incl. Christmas)
E	Male, 8yrs	2	2 (incl. Christmas)
F	Female, 7yrs	2	3 (incl. Christmas)
G	Male, 13yrs	3	4 (incl. Chris. & Half Term)
I	Male, 43yrs	1	4**
J	Male, 10yrs	2	3 (inc. Easter & Half Term)
L	Female,13yrs	1	4 (incl. Summer holiday)

*delay between session seven and eight ** delay between session five and six

None of the participants had previously received speech therapy to address their nasal speech symptoms and no other intervention was received during the period of the study.

Home practice was planned as part of the study protocol with a requirement to record a daily session of 10-15 minutes in a diary. This diary was designed to serve only as a record of practice completed and was not analysed further. Poor compliance with diary completion meant that compliance with home practice as instructed could not be accurately evaluated alongside the other study outcomes. Often practice was verbally reported but the diary was not brought to the session or was not filled in. This means that it is unclear whether some participants did practice at home as instructed but

simply did not record this practice whilst others may not have complied with either the practice or the diary completion. The implications of this are discussed in Chapter six.

Three visual biofeedback tools were utilised (Nasometer™ II 6450, See-Scape™ and nasal mirror, Chapter Three, page 98). The exact selection of tools for each participant was based on information gained at baseline assessment and the individual profile of nasal speech symptoms (see Chapter Four and individual case studies, this chapter). Nasometry was the only visual biofeedback tool to be used with all twelve participants.

5.4 Outcome measures

The study included a range of outcome measures; perceptual, instrumental and quality of life (see Chapter Four). The perceptual measure, CAPS-A, uses a rating scale of defined speech parameters (hypernasality, audible nasal emission and nasal turbulence). Perceptual outcomes were obtained from blind rating of recorded standard speech assessments, by expert listeners. The instrumental measure, nasometry, produces a percentage nasalance score obtained directly from the calibrated Nasometer™ II 6450, based on a standard speech sample. Finally, quality of life scores were calculated from the validated questionnaires using numerical likert scales, according to published scoring procedures. The scales and maximum total scores for each outcome measure are summarised below;

Table 5-6: Study outcome measures scales and scores

Outcome	Perceptual (CAPS-A)			Instrumental (Nasometry)	Quality of Life		
	Hyper	ANE	NT		PedsQL4.0	PedSaLQoL	VoiSS
Scale	0-4	0-2	0-2	0-100%	100 maximum total score	160 maximum total score	25 maximum total score
Severity	Higher rating indicates increased severity			Higher score indicates increased severity	A higher score is associated with better quality of life for Peds QL & PedSal QoL A lower score is associated with better quality of life for VoiSS		

5.4.1 Data handling

In light of the feasibility nature of this study, the decision was taken to present the results descriptively as individual and group differences between ratings and scores, before and after the intervention. There were four assessment points in total, two pre-intervention (A1 and A2) and two post-intervention (A3 and A4). The perceptual and instrumental outcomes were measured at all four points and the quality of life outcomes only at A1 and A4, as previously outlined in Chapter Four. Two baseline (pre-intervention, A1 and A2) assessments were carried out for the perceptual and instrumental measures to examine the stability of nasal speech symptoms and two post-intervention assessments (A3 and A4) to evaluate the stability of any change.

As expected there was variability evident between A1 and A2 for both perceptual (CAPS-A) ratings and instrumental (nasometry) scores (see Chapter Four) and the issue of variability as a potential confounder is discussed in detail in Chapter Six. The two samples of A1 and A2 are strongly associated (correlation >0.8) and statistically have the same mean value, indicating that either rating could be representative of the

baseline perceptual and instrumental measurements taken before the intervention. However, it could be argued that a better representation of these two baseline measures is their average for each case (participant), particularly in view of the small sample size, and following statistical advice, perceptual and instrumental results from the two baseline assessments (A1 and A2) were therefore combined for the purpose of analysis (Appendix L, Table L-3, L-6). The possible implications of this approach are discussed in the next chapter.

5.4.2 Missing data

Despite the small number of participants in the study there was missing outcome data. This is summarised in Table 5-6. Eight participants attended all four assessment sessions as per the study protocol but two participants did not attend the first post-intervention assessment (A3). This resulted both in missing CAPS-A speech recordings and nasometry scores for these two participants at A3.

Technical failures resulted in the loss of perceptual speech data (CAPS-A recordings) for four participants at the second baseline assessment (A2). Technical failure also resulted in the loss of one CAPS-A recording at the first post-intervention assessment (A3) and for the nasometry results of a different participant at A3.

Table 5-7: Data available (*missing*) at the four assessment points

Assessment point	CAPS-A recording	Nasometry score	QoL measure		
			PedsQL4.0	PedSaLQoL	VoiSS
A1 (baseline 1)	12	12	9 (*-2*)	11	1
A2 (baseline 2)	8 (*-4*)	12			
A3	9 (*-3*)	9 (*-3*)			
A4	12	12	9 (*-2*)	11	1

There is also missing data for the quality of life outcomes. There are results for all eleven children in the study for PedSaLQoL and nine of the children from PedsQL4.0, at both A1 and A4. The remaining two completed the PedSaLQoL at the first assessment point (A1) but could not maintain their concentration to then complete the PedsQL. They were therefore only invited to complete the PedSaLQoL questionnaire at the post-intervention assessment (A4). Apart from the paediatric QoL measures there was complete perceptual and instrumental data available for all twelve participants from the first baseline (A1) and final follow-up assessment (A4).

Implications of the missing data are discussed in subsequent chapters but for the purposes here of presentation and analysis of the results a simple ad hoc imputation approach was employed for both perceptual (CAPS-A) and instrumental (Nasometry) missing data. This approach uses the last value carried forward (LVCF), also known as last observation carried forward (LOCF) and has been commonly used as a conservative approach to managing missing data in clinical trials (Hamer & Simpson 2009). The imputation of the last available measurement to the missing data point (A2 or A3) was possible in this study as all participants had baseline data available from baseline A1 and this therefore allowed for the use of methods of presentation and analysis as for complete data. This does however introduce the potential for bias in terms of making assumptions about the data that may represent either an overestimation or underestimation. The possible implications of this for a feasibility study are discussed in Chapter Six.

5.5 Individual results across all outcome measure

The comparison of pre and post-intervention results for all participants across all outcome measures (perceptual, instrumental and quality of life) is shown in Table 5-8. Pre-intervention results relate to the average of the combined baseline assessments (mean A1+A2) and post-intervention results relate to the follow-up assessment (A4), three months after the final intervention session. Results from immediately post-

intervention (A3) are not presented in Table 5-8 as these are intermediate results and therefore cannot show whether any change has been sustained after the intervention. Although the two baseline assessment results were combined for analysis this was not appropriate for the post-intervention results as they represent two different outcome points measured three months apart. Full results from all assessment points (A1, A2, A3, and A4) are presented later in this chapter.

Results in Table 5-8 are colour-coded to indicate whether there has been a positive change (green), no change (amber/yellow) or negative change (red), between the pre and post-intervention assessment points. This traffic-light system of colour coding was originally devised by Harland (1996) and subsequently developed for use with the Cleft Audit Protocol for Speech (CAPS: Harding et al.,1997) and the revised CAPS-A (John et al., 2006). The principle of the traffic light system in CAPS-A was to give an indication of outcome and future treatment need as follows;

Green: a satisfactory result with no intervention needed
Yellow: a need to monitor progress because there may be a requirement for some intervention
Red: an unsatisfactory result requiring further detailed speech assessment, structural investigations, and probable surgical intervention and/or speech therapy intervention.
John et al., (2006) page 275

In Table 5-8., it is used to highlight change between the baseline (mean A1+A2) and final follow-up (A4) outcomes but does not necessarily indicate that this change is either clinically or statistically significant. Where a green outcome is shown this indicates a reduction in rated severity of nasal speech symptom, a reduction in nasalance or an improved quality of life score (increased for paediatric measure and decreased for adult measure). Amber indicates no change in the outcomes, including where a nasal speech symptom is rated as zero (absent) both pre and post intervention. An amber colour-coding is also used where there is less than a five point

change in percentage nasalance or quality of life score as this may reflect within-subject variability as discussed in Chapter Six. Red indicates an increase in nasal speech symptom severity, increased nasalance or worse quality of life score.

Finally, blue shading is used in the participant column to indicate those participants who deviated from the study protocol in frequency of the intervention sessions.

Table 5-8: Individual results pre (mean A1+A2) and post (A4) VFTh intervention for all outcome measures

	Perceptual (CAPS-A)						Instrumental		Quality of Life Total Score					
	Hyper		ANE		NT		% Nasalance*		PedSalQoL**		PedsQL**		VoiSS***	
Partic.	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
A	1	1	0	0	1	2	24	24	138	156	90.65	84.78		
B	2.5	1	0.5	0	0	0	28	45	116	134	79.21	65.22		
C	0.5	0	0	0	0	0	37	39•	112	109	81.30	69.57		
D	3	2	0	0	0.5	0	51	45	103	110	54.35	59.78		
E	0	1	0	0	0	2	42	43•	124	122				
F	0	0	0	1	0	1	41	37•	144	134				
G	0	1	1	0	1	0	50	43	102	123	71.74	72.83		
H	2	1	0	0	1.5	1	48	48	108	122	73.91	84.78		
I	2.5	1	0	0	0	0	43	24					22	11
J	0	0	0	0	0	0	25	24•	118	130	72.83	69.57		
K	2.5	0	0	0	1	0	54	30	145	140	73.91	80.43		
L	2	1	0.5	0	0.5	1	62	28	107	119	79.35	73.91		

Participant colour coding: Blue= disrupted delivery of intervention; White= intervention delivered as per study protocol

Outcome colour coding: Green= positive change; Amber= no change; Red= negative change

•< than 5% change in nasalance or 5 point change in QoL score; *nasalance norms=26-32%;

child participants only, higher score=improved QoL; * adult participant only, lower score=improved QoL

5.6 Perceptual assessment results (CAPS-A)

Results are presented for the three perceptual (CAPS-A) parameters relating to nasal speech quality: hypernasality (HYPER), audible nasal emission (ANE) and nasal turbulence (NT), as blindly rated by five expert listeners. For all three speech symptoms a higher CAPS-A rating is indicative of greater severity. The differences between ratings at all four assessment points are summarised for each speech symptom. The findings of subsequent inferential analysis are included with full details given in Appendix L.

5.6.1 Expert listener reliability

Analysis of inter-rater reliability was not required in this study as a consensus rating procedure was used to assess the speech recordings (see Chapter Four). However in order to demonstrate the level of consistency of the individual expert raters, intra-rater reliability results were obtained, with consent, from their CAPS-A training records. Intra-rater reliability results are issued to all specialist SLTs who undertake CAPS-A training and are presented as percentage agreement and weighted kappa values (Altman 1991, Landis & Koch 1977). A percentage agreement of 80% is considered a good standard and interpretation of the weighted kappa statistic is shown in Table 5-9. Full results for all raters are presented in Table 5-10. The limitations of this approach are discussed in Chapter Six.

Table 5-9: Interpretation of KAPPA statistic

KAPPA value	Strength of agreement
<0.20	Poor
0.21 – 0.40	Fair
0.41 – 0.60	Moderate
0.61 – 0.80	Good
0.81 – 1.00	Very good

Table 5-10: Intra-rater reliability results for all raters (% agreement and KAPPA)

Rater	HYPER % agreement	KAPPA	ANE % agreement	KAPPA	NT % agreement	KAPPA
SLT 1	96.25	0.86	95.00	0.88	85.00	0.62
SLT 2	96.53	0.89	90.00	0.74	71.50	0.46
SLT 3	97.92	0.91	72.50	missing value*	82.50	0.50
SLT 4	89.38	0.65	83.33	0.41	86.11	0.53
SLT 5	95.83	0.84	82.50	0.59	85.00	0.61

*K value missing for one rater

Intra-rater reliability was over 80% for all raters on the parameter of hypernasality (HYPER) with weighted KAPPA values ranging from 0.65 (good) to 0.91 (very good). The agreement for audible nasal emission (ANE) was over 80% for four out of five of the raters; KAPPA range 0.41 (moderate) to 0.88 (very good). Similarly for NT ratings agreement was over 80% for four out of five raters; KAPPA range 0.50 (moderate) to 0.62 (good).

5.6.2 Perceptual (CAPS-A): Individual and group results

The individual participant changes for each perceptual CAPS-A nasal speech parameter, across all four assessment points pre and-post intervention are first given in Table 5-11 and then illustrated in Figures 5-2, 5-3 and 5-4. Curved lines and line markers are used to ensure that all participants' data is visible across four assessment points and within the five point (HYPER) and three point (ANE and NT) rating scales.

The graphical presentation of the results from all four assessment points in this way gives an indication of the variability of the nasal speech symptoms for some participants, including those who showed an apparent reduction in one or more speech symptoms at follow-up assessment (A4). The implications of this variability in relation to the outcome measures used in the study are discussed in Chapter Six.

Group results are not included here in the main results chapter due to the small study sample size but can be found in Appendix L.

Table 5-11: Perceptual (CAPS-A) results across all assessment points

P.ID	A1			A2			A3			A4		
	HYPER	ANE	NT	HYPER	ANE	NT	HYPER	ANE	NT	HYPER	ANE	NT
A	1	0	1	1*	0*	1*	1*	0*	1*	1	0	2
B	2	1	0	3	0	0	2	0	1	1	0	0
C	1	0	0	0	0	0	0*	0*	0*	0	0	0
D	3	0	0	3	0	1	2	0	0	2	0	0
E	0	0	0	0	0	0	0	0	0	1	0	2
F	0	0	0	0*	0*	0*	0	0	1	0	1	1
G	0	1	1	0*	1*	1*	1	0	0	1	0	0
H	2	0	1	2	0	2	1	0	2	1	0	1
I	3	0	0	2	0	0	2	0	0	1	0	0
J	0	0	0	0*	0*	0*	0	0	0	0	0	0
K	2	0	0	3	0	2	1	0	0	0	0	0
L	2	1	1	2	0	0	2*	0*	0*	1	0	1

*imputed result (LVCF)

5.6.2.1 Hypernasality results

Figure 5-2 shows results for HYPER for all twelve participants with a trend towards reduction in severity. All twelve participants are included as all presented with hypernasality pre-recruitment as an eligibility requirement, even if they subsequently received zero (absent) HYPER ratings pre-intervention at A1 and A2 (participants E, F, G & J). However pre-recruitment ratings are not presented here or included in the analysis.

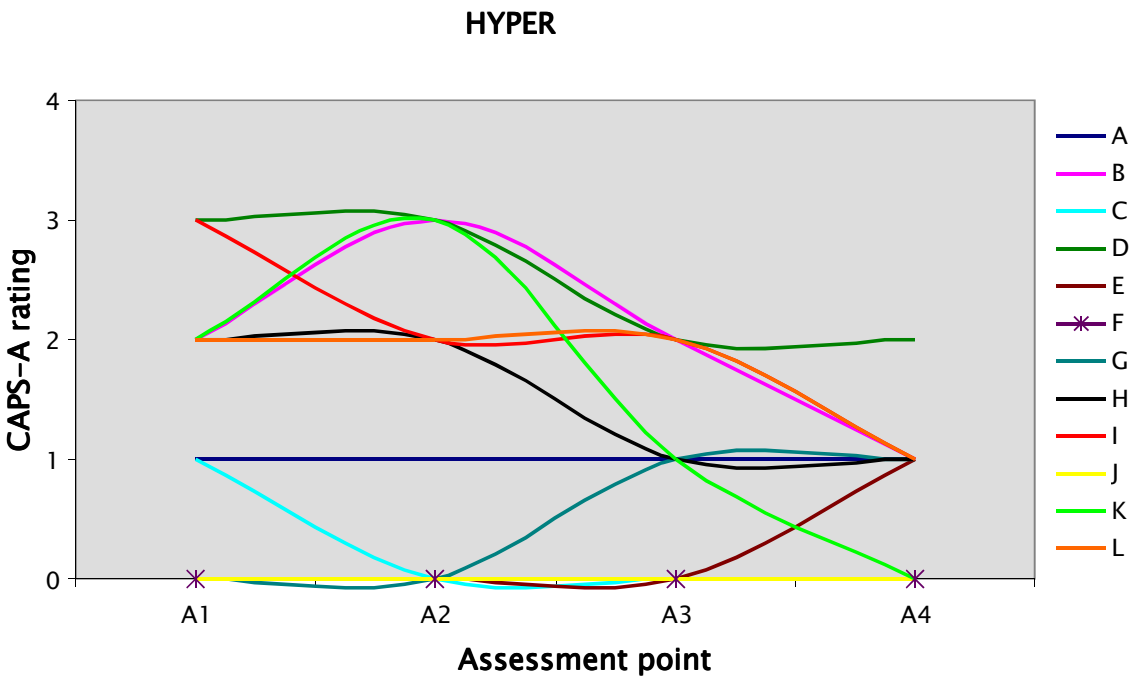

Figure 5-2: Individual results of HYPER ratings across all assessment points
*Starred line used for participant F to aid visibility as partially obscured by participants D and J

Data was missing at post-intervention (A3) for three participants (A, C, L) and the rating was carried forward from the previous available assessment point.

At the follow-up assessment point (A4) hypernasality results were available for all twelve participants. As blindly rated by the expert listeners, seven participants showed

a reduction in hypernasality severity rating from combined baseline (mean A1+A2), three no change and two showed an increase in severity rating. Two of the three participants who showed no difference between combined baseline and follow-up were rated zero (absent) for hypernasality at A1, A3 and A4 but had imputed data at A2.

The Wilcoxon Signed Ranks test was used to analyse the differences between the pre and post-intervention points and showed a difference approaching significance for hypernasality (HYPER) from combined baseline (mean A1+A2) to immediately post-intervention (A3), p=0.085 and significant at follow up (A4), p=0.040, (Table L-4, Appendix L).

5.6.2.2 Audible nasal emission results

The results for ANE are shown in Figures 5-3. This graph is included here alongside the other perceptual (CAPS-A) results for completeness but should be viewed with particular caution due to the small number of participants included. Figure 5-3 shows changes in audible nasal emission for the four participants who presented with this speech symptom either pre-intervention (B, G, L) or post-intervention (F).

The remaining eight participants (A, C, D, E, H, I, J, K) are not included in Figure 5-3 as they had zero (absent) ratings for ANE at all study assessment points including pre-intervention, although J had missing data at A2, C had missing data at A3 and A had missing data at both A2 and A3. Imputed ratings were again used at these missing data points where the last available rating was carried forward.

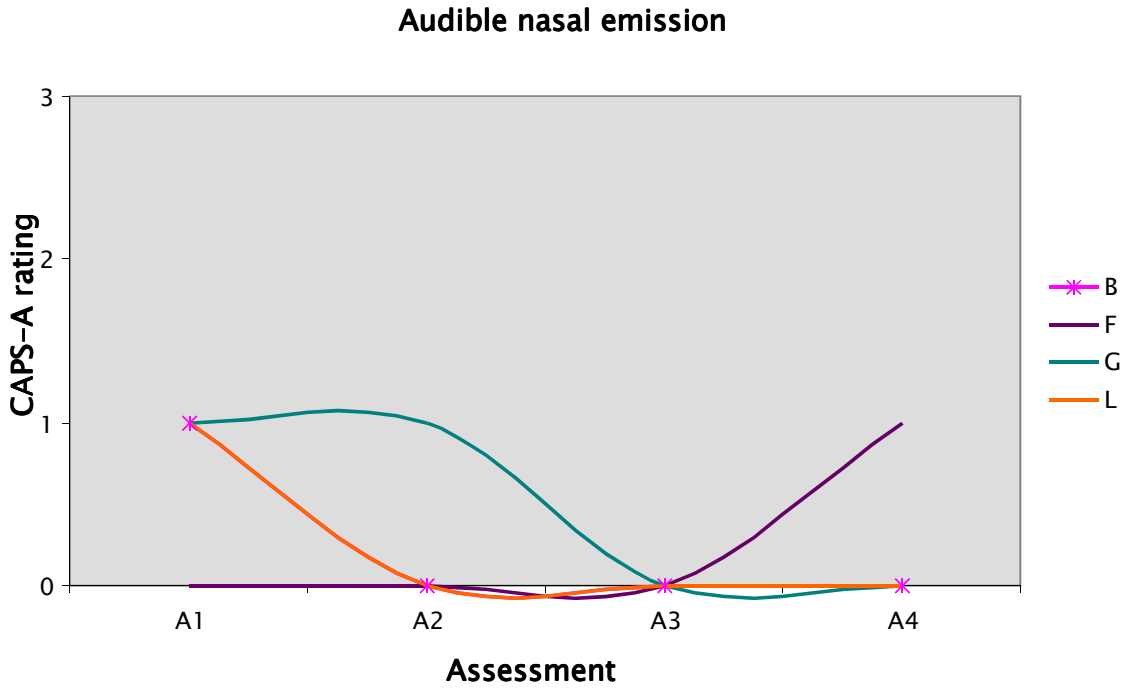

Figure 5-3: Individual results of ANE rating across all assessment points for the four participants who presented with this speech symptom.

Of the four participants two (B, G) showed a reduction in ANE severity rating immediately post-intervention (A3) compared with combined baseline (mean A1+ A2), and two showed no change (F, L). However F had imputed data at A2 and L at A3.

At the follow-up assessment point (A4) ANE results were available for all twelve participants, including the four who presented with ANE at any of the study assessment points. Of these four participants three showed a reduction in severity rating (B, G, L) and one showed increased ANE severity (F). The remaining eight participants all had zero (absent) ANE ratings at A4.

The Wilcoxon Signed Ranks test was used to analyse the differences between pre and post-intervention points for all twelve participants and showed no significant difference for ANE from combined baseline (mean A1+A2) to immediately post-

intervention (A3), p=0.09, and follow-up (A4), p=0.258 (Table L-5, Appendix L). Again this result should be viewed with caution given the small number of participants

5.6.2.3 Nasal turbulence results

Figure 5-4 includes the results for nine participants presenting with NT either pre-intervention (D, G, K) or post-intervention (B, E, F) or both pre and post (A, H, L). The remaining three participants (C, I, J) are not included in Figure 5-4 as they had zero (absent) ratings for NT at all study assessment points including pre-intervention, although J had missing data at A2 and C had missing data at A3, therefore imputed ratings were used.

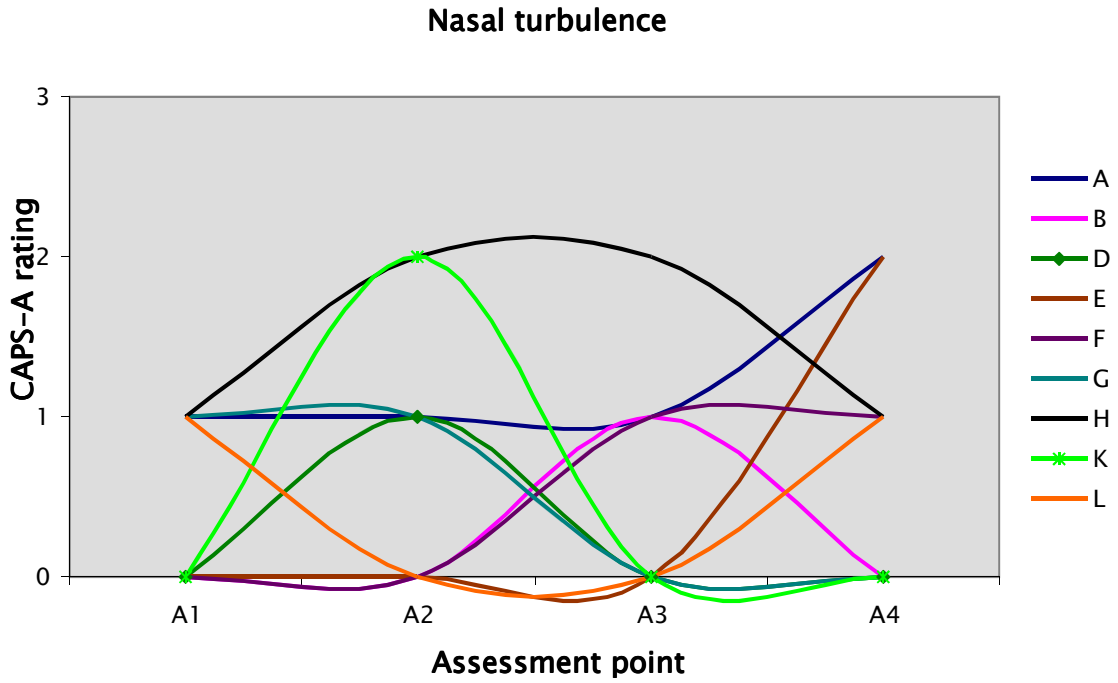

Figure 5-4: Individual results of NT ratings across all assessment points

Immediately post intervention (A3) three participants (D, G, K) showed a reduction in NT severity rating compared with combined baseline (mean A1+A2), three an increased rating (B, F, H) and six showed no change (A, C, E, I, J, L).

At follow-up (A4) four participants showed a reduction from their combined baseline (mean A1+A2) severity rating (D, G, H, K), four no change (B, C, I, J) and four participants showed an increase in NT severity (A, E, F, L). Of the four participants who showed no change at A4, three were rated as zero (absent) for NT at all assessment points (C, I, J) but C had imputed data at A3 and J at A2. Finally participant B had zero (absent) ratings at A1, A2 and A4 but an increased severity rating of 2 at A3.

The Wilcoxon Signed Ranks test was used to analyse the differences between the pre and post-intervention points and showed no significant difference for NT from combined baseline (mean A1+A2) to immediately post-intervention (A3) $p=0.5$, and to follow-up (A4), $p=0.334$,(Table L-6, Appendix L).

5.6.3 Summary of differences in perceptual (CAPS-A) results from combined baseline (mean A1+A2) to follow-up (A4)

Taking the combined baseline (mean A1+A2) and final assessment points (A4) differences were present in nine out of twelve participants for hypernasality (seven reduced, two increased, three no change), four participants for audible nasal emission (three decreased, one increased, eight no change) and eight participants for nasal turbulence (four decreased, four increased, four no change), as shown in Table 5-11. These results should be viewed in the context of the variability evident in perceptual ratings of speech symptoms, even between baselines A1 and A2 and this will be discussed in Chapter Six.

Table 5-12: Change between pre and post-assessment point ratings (mean baseline A1+A2 and follow-up A4)

Participant	Hypernasality	Audible nasal emission	Nasal turbulence
A	↔	↔ (absent)	↑
B	↓	↓	↔
C	↓	↔ (absent)	↔ (absent)
D	↓	↔ (absent)	↓
E	↑	↔ (absent)	↑
F	↔	↑	↑
G	↑	↓	↓
H	↓	↔ (absent)	↓
I	↓	↔ (absent)	↔ (absent)
J	↔ (absent)	↔ (absent)	↔ (absent)
K	↓	↔ (absent)	↓
L	↓	↓	↑

↓reduction in severity ↔no change in severity ↑increase in severity ↔ (absent) no change in severity (absent pre and post)

Eight participants (B, C, D, G, H, I, K, L) showed an improvement (reduction in severity) in one or more speech symptom from combined baseline (mean A1+A2) to follow-up (A4). Two of these participants (G and L) showed a reduction in two symptoms but an increase in the third, hypernasality and nasal turbulence respectively.

A further three participants showed an increase in severity of one or more speech symptoms post-intervention (A, E, F) without a reduction in any other symptom. Participant J showed no change across all three speech symptoms between pre and post-assessment, but was given zero (absent) ratings at all assessment points. This

may reflect the very mild degree of nasal speech that was identified prior to recruitment to the study and this will be discussed in Chapter Six.

5.7 Instrumental assessment results (Nasometry)

Results are presented as percentage nasalance scores with a higher percentage score indicative of greater severity of nasal speech. The differences between nasalance scores at all four assessment points are summarised for each participant. The findings of inferential analysis are then summarised, with full details provided in Appendix L.

5.7.1 Instrumental (Nasometry): Individual and group results

The individual participant results for nasometry across all four assessment points pre and-post intervention are shown in Table 5-13 below. A difference in nasalance between combined baseline (mean A1+A2) and immediately post-intervention (A3) was present for all twelve participants; traffic light colour-coding is again used to highlight the direction of change. For eight participants the difference was a reduction (improvement) in nasalance but in three of these participants (C, H, L) the reduction was less than 5% and one participant (L) had missing data at A3 so had the score at A2 carried forward, as previously described.

Table 5-13: Comparison of individual nasalance scores pre and post-intervention

	Nasalance %					
Participant	**A1**	**A2**	**A1 + A2 (mean)**	**A3**	**A4**	**Difference between A1+A2 (mean) & A4 %**
A	24	22	23	32	24	+1
B	28	32	30	41	45	+15
C	37	33	35	33*	39	+4
D	51	51	51	45	45	-6
E	42	30	36	21	43	+7
F	41	41	41	29	37	-4
G	50	48	49	41	43	-6
H	48	53	50.5	50	48	-2.5
I	43	45	44	45*	24	-20
J	25	40	32.5	25	24	-8.5
K	54	42	48	56	30	-18
L	62	57	59.5	57*	28	-31.5

* imputed value

Outcome colour coding: Green= positive change; Amber= no change or less than 5% change; Red= negative change

Four participants (A, B, I, K) showed an increase in nasalance from combined baseline (mean A1+A2) to immediately post-intervention (A3). In one participant (I) the increase was less than 5% but this participant had data missing at A3.

At follow-up (A4) eight participants showed a reduction (improvement) in nasalance from combined baseline (mean A1+A2). In two of these participants the reduction was less than 5% (F, H). Four participants showed an increase in nasalance (A, B, C, E), with two of these increasing their scores by more than 5% (B, E). The difference between combined baseline (mean A1+A2) and follow-up (A4) nasalance scores ranged from a reduction of 31.5% to an increase of 15%. The mean reduction was 12.06% and mean increase 6.75%.

It interesting to note that three participants showed initial improvement (decreased nasalance) at A3 and then increased nasalance at A4 (C, E, F), although the variation in nasalance for participant C was less than 5% at both assessment points and the A3 value was imputed from A2. Conversely participant K showed increased nasalance at A3 but improvement at A4 and this variability between A3 and A4 results will be discussed in Chapter Six.

The Wilcoxon Signed Ranks test was used to analyse the differences between the pre and post-intervention points and showed no significant difference for nasometry scores from combined baseline (mean A1+A2) to immediately post-intervention (A3) $p=0.271$ but approaching significance at follow-up (A4), $p=0.073$,(Table L-7, Appendix L). Group mean nasalance scores at all assessment points are given in Appendix L, Table L-8.

The change in nasometry scores for all participants across all four assessment points is illustrated in Figure 5-5, with each participant effectively acting as his or her own control. The dotted lines indicate the range of nasometry scores that would be considered 'normal' in an English speaking population, i.e. between 26% and 32% (Sweeney 2011). The relationship of the participant's nasometry scores in relation to these published norms is also included in Appendix L (Figure L-4) and is discussed in Chapter Six.

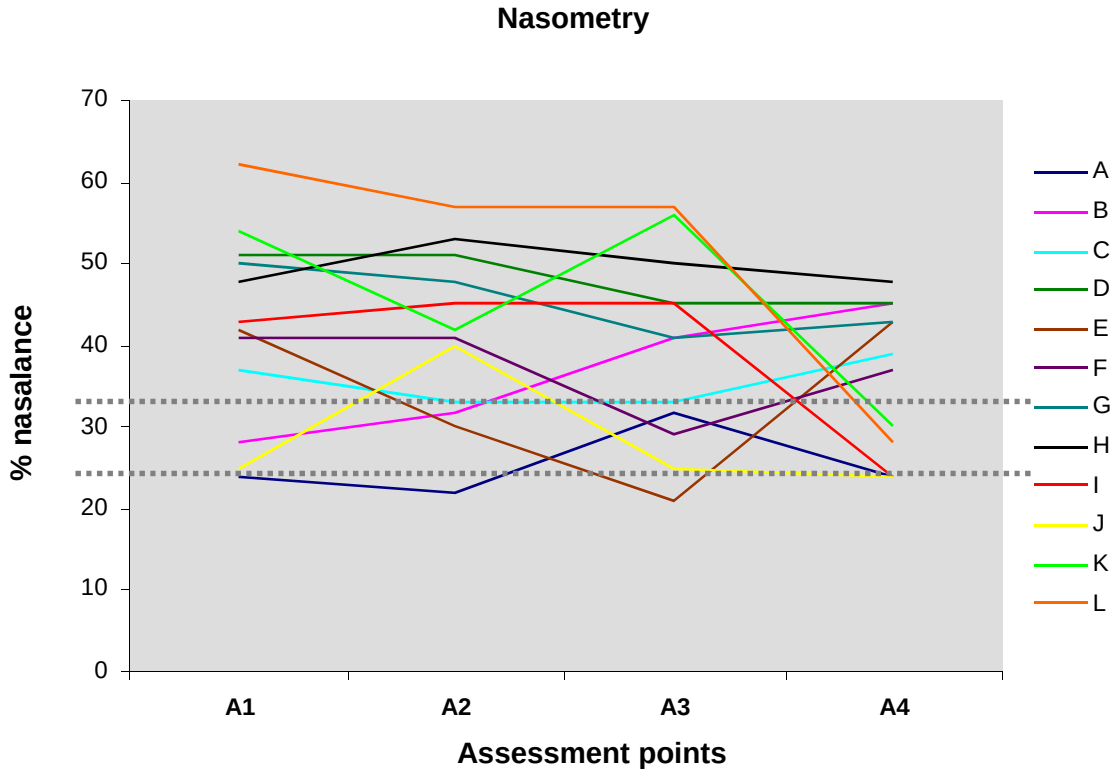

Figure 5-5: Individual nasometry results across all assessment points

5.8 Quality of Life assessment results

Results are presented as total and subsection scores for all quality of life measures; PedSaLQoL, PedsQL4.0 and VoiSS at the two assessment points pre-intervention A1 and post-intervention A4. PedSaLQoL is the communication specific measure for children and PedsQL4.0 the generic measure. VoiSS is the communication measure used with the adult in the study (Appendix F). Results from the VoiSS are presented for the sole adult participant. For the paediatric measures a higher score is associated with increased quality of life whilst in contrast lower scores on the VoiSS are better.

5.8.1 Ped SaL QoL individual and group results

The maximum possible Total Score for the PedSal QoL is 160. Participants A, B, D, G, H, J, and L all increased their PedSaL QoL Total Score post-intervention whilst participants C, E, F and K had a decreased score, Figure 5-6.
There is a significant positive difference in the Communication & Feelings section (increased by 5.2, p=0.012, Wilcoxon Signed Ranks test) and Total Score (increased by 7.5, p=0.025), before and after the intervention (Table L-9, Appendix L). There was a reduction in mean score on the Support at School section, but this was not significant, p=0.444.

Mean group scores for all PedSaL QoL subsections, mean total scores and standard deviations pre and post-intervention (A1 and A4) are given in Appendix L, Table L-11, and illustrated in Figures L-5 and L-6.

5.8.2 PedsQL4.0 individual and group results

The maximum possible Total Score for the PedsQL 4.0 is 100. Participants D, G, H and K showed small increases in their PedsQL4.0 Total Score but A, B, C, J and L all had decreased scores, participants E and F did not complete this assessment due to fatigue, Figure 5-7

There was an increase in the School section score which approached significance, p=0.076, but none of the changes were significant with non-parametric testing, Hact p=0.448, Feel p=0.451, Geton p=0.100, Phys p=0.475, Total score p=0.260 (Table L-9, Appendix L.).

Mean group scores for all PedsQL 4.0 subsections, mean total scores and standard deviations pre and post- intervention (A1 and A4) are given in Appendix L, Table L-12, and illustrated in Figures L-7 and L-8.

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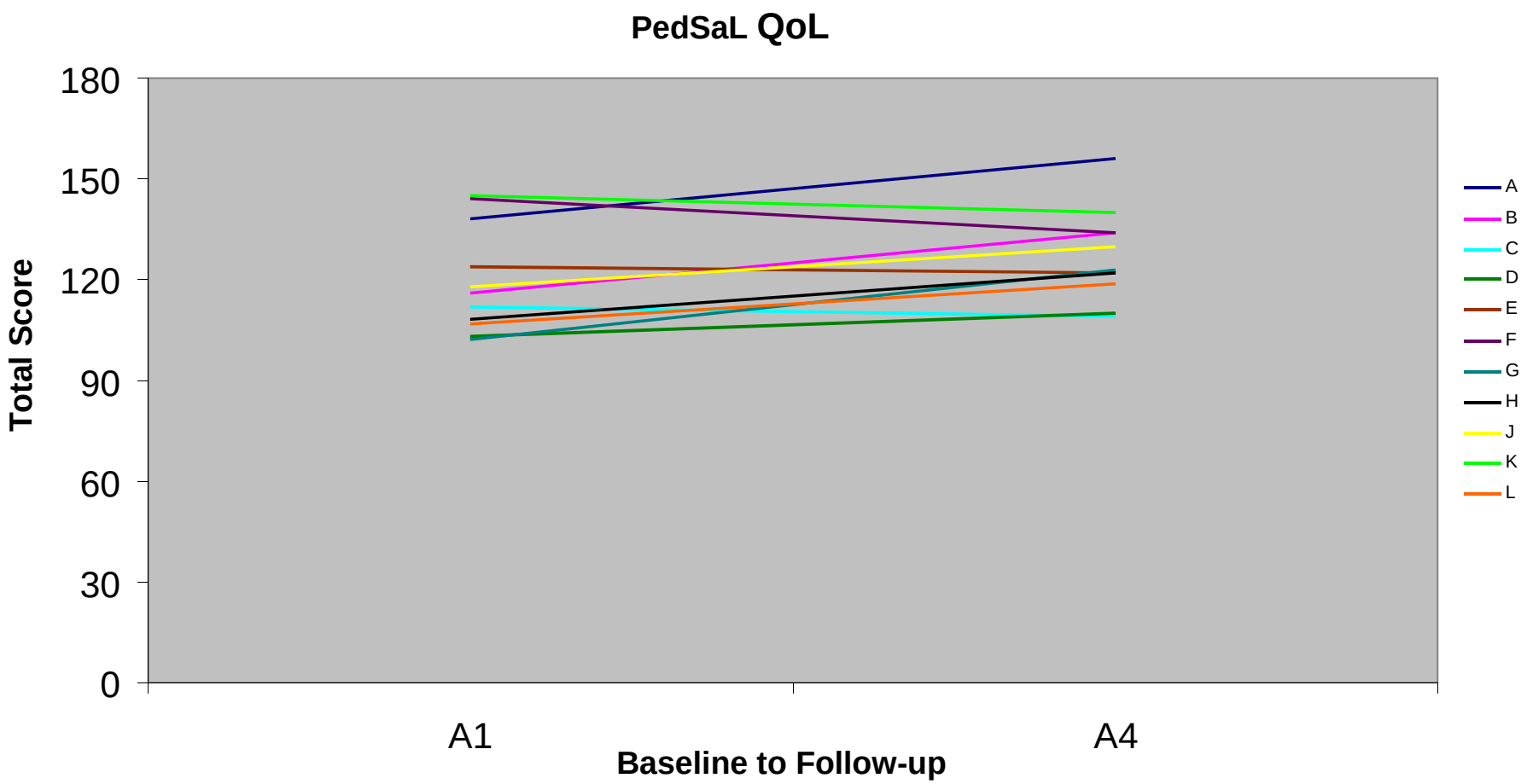

Figure 5.6: PedSaL QoL: individual results (Total Score) from baseline (A1) to follow-up (A4) (A4)

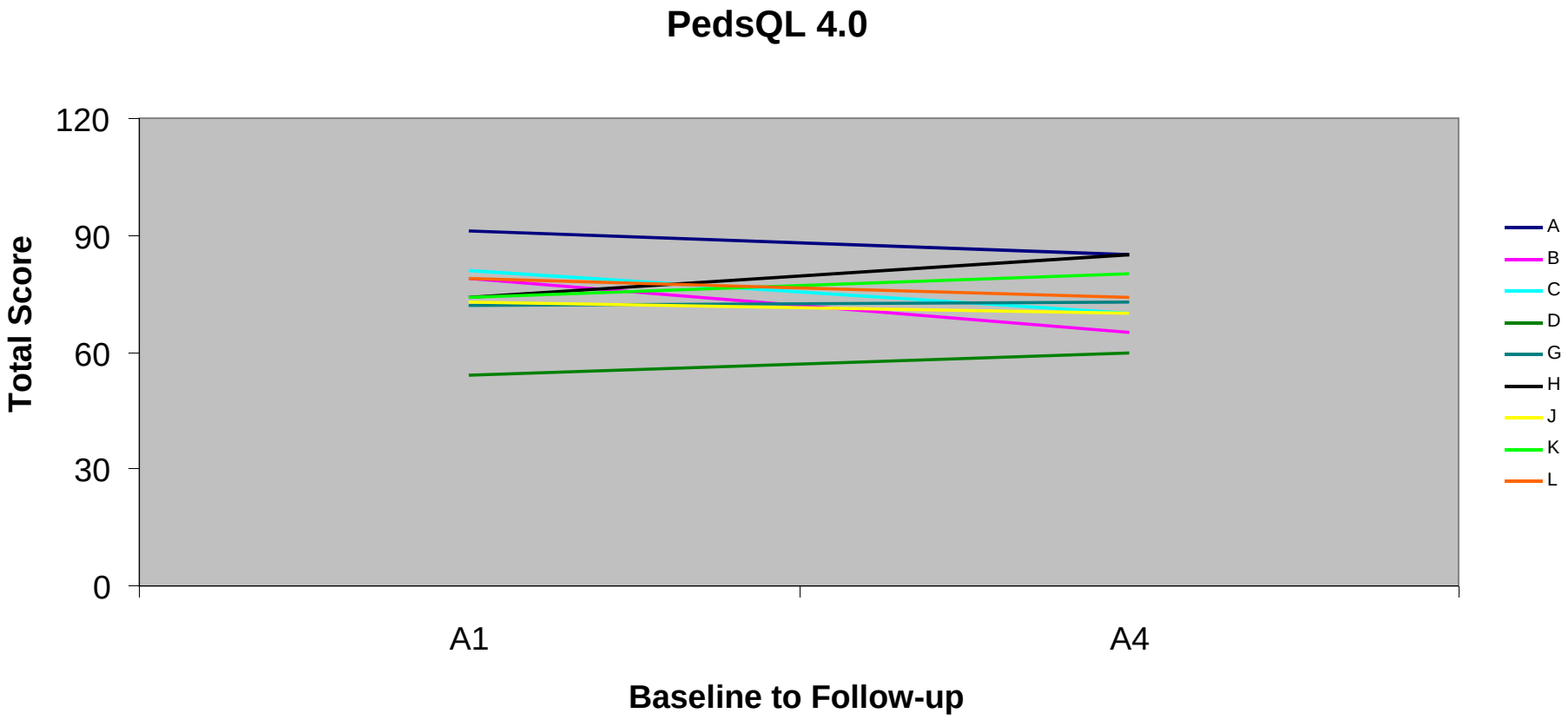

Figure 5.7: PedsQL 4.0: individual results (Total Score) from baseline (A1) to follow-up (A4)

5.8.3 Ped SaL QoL and PedsQL4.0: Comparison of individual results

There was variability in Total Score results between individuals and between the two quality of life measures (Table 5-14). Only three of the nine participants who completed both measures had increased scores on both post-intervention (D, G, H) and possible reasons for this difference are discussed in Chapter Six.

Table 5-14: Comparison of individual results (Total Score) for PedSaLQoL and PedsQL4.0

Participant	PedSaLQoL Total Score	PedsQL4.0 Total Score
A	↑	↓
B	↑	↓
C	↓	↓
D	↑	↑
E	↓	Not completed
F	↓	Not completed
G	↑	↑
H	↑	↑
J	↑	↓
K	↓	↑
L	↑	↓

↑ increased total score ↓ decreased total score

5.8.4 VoiSS: Individual results

The results for VoiSS pertain to the sole adult in the study (participant I) and are included here for completeness, as the results cannot be compared with the child

participant's quality of life results or any other adult results. A lower score on this measure is associated with increased quality of life. Participant I showed a reduction in all three subsections of the VoiSS measure; Impairment (IMP), Physical (PHYS) and Emotional (EMOT) and a corresponding reduction in total score, Figure 5-8. Note that the post-intervention score for EMOT was zero.

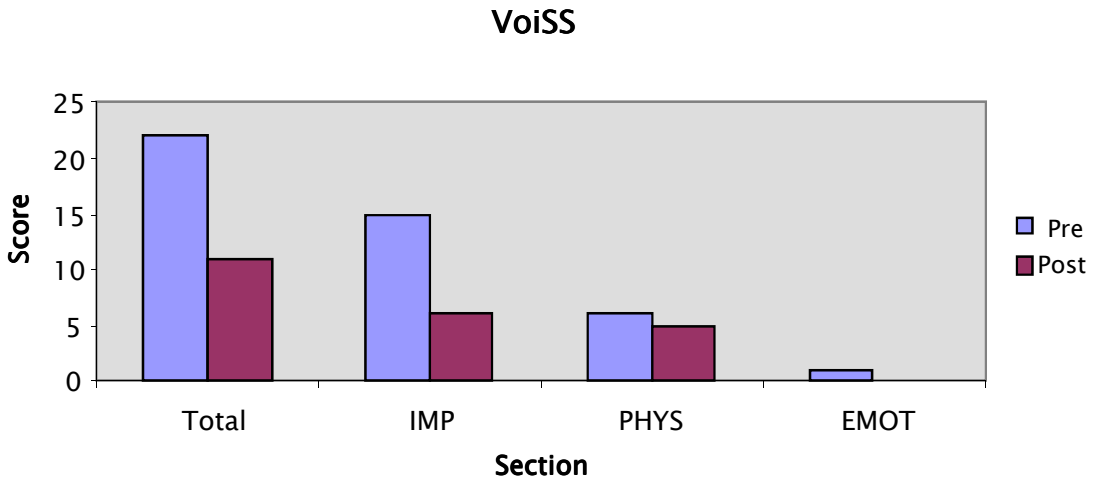

Figure 5-8: VoiSS scores for adult participant pre and post-intervention

5.9 Individual case studies: Response to the VFTh intervention

This section summarises the detailed session notes that were kept by the chief investigator (CI) for all participants for the duration of the intervention. This allowed for in-depth exploration of individual participants' response to VFTh and is a crucial aspect of the results of this feasibility study. The individual outcomes are also reported for each participant with traffic light colour-coding as before. This highlights the variability in outcomes for some participants and this is discussed in detail in the following chapters.

Participant A (PA)

Participant A was a girl, aged 9 years at the start of the research. She was born with a cleft of the soft palate and repair was carried out at age 6 months at a UK cleft centre.

PA's speech was characterised by mild and variable hypernasality, inconsistent nasal turbulence and a distinctive 'husky' voice quality. She had received speech therapy since the age of 6 years, previously aimed at eliminating an active nasal fricative sound used as a substitution for the sounds /s/ and /z/.

Outcomes		
Perceptual:	Hypernasality Nasal turbulence	no change increased severity rating
Instrumental:	Nasalance	no change pre-intervention: score below normal range post-intervention: score below normal range
QoL:	PedSaLQoL PedsQL4.0	increased score (better) decreased score (worse)

Details of intervention:

The feedback tools used with PA were performance feedback, nasal mirror, See-Scape™ and nasometry. She received a total of eight 45 minute sessions at the clinic but not over consecutive weeks due to school commitments.

The tailored therapy objectives for PA were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. no misting of the mirror, blue foam indicator kept at the bottom of the See-Scape™ tube, nasometry games achieved below agreed nasalance threshold.
3. To progress from single sound production to words and short sentences beginning with low pressure sounds /w,l,y/, then introducing high pressure sounds /p,t,s,f/.

Response to intervention:

The key words/phrases for performance feedback agreed with PA were 'smooth', 'soft voice not creaky frog voice', and 'super shiny not foggy mirror'.

PA carried out regular home practice with one of her parents using the mirror and the See-Scape™. On average five practice sessions per week were recorded in her weekly diary. PA was very aware of perceived failure in using the feedback tools despite reassurance about just looking and noticing. Objectives were revised within sessions in response to her performance and sensitivity to failure. For this reason the nasalance threshold was set high initially and reduced in small increments of 2%-3% to allow her to feel successful.

Participant B (PB)

Participant B was a girl, aged 8 years at the start of the research. PB was born with a cleft of the soft palate. Her cleft palate repair was carried out at age 6 months at a UK cleft centre. PB's speech was characterised by mild to moderate variable hypernasality and nasal substitutions for some oral sounds; /b/ and /d/. PB had received previous speech therapy since the age of 5 years aimed at eliminating articulatory cleft speech characteristics. These were backing (replacing front of mouth sounds /t/ and /d/ with back of mouth sounds /k/and /g/) and active nasal fricative which she used as a substitution for /s/ and /z/.

Outcomes		
Perceptual:	Hypernasality	**decreased severity rating**
	Audible nasal emission	**decreased severity rating**
	Nasal turbulence	**no change**
Instrumental:	Nasalance	**increase of 17% (worse)** pre intervention score: within normal range post-intervention score: above normal range
QoL:	PedSaLQoL	**increased score (better)**
	PedsQL4.0	**decreased score (worse)**

Details of intervention:

The feedback tools used with PB were performance feedback, nasal mirror, See-Scape™ and nasometry. She received a total of eight 45 minute sessions at the clinic over consecutive weeks.

The tailored therapy objectives for PB were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.

2. To establish feedback targets i.e. no misting of the mirror, blue foam indicator kept at the bottom of the See-Scape™ tube, nasometry games achieved below agreed nasalance threshold.
3. To contrast the oral sounds /b/ and /d/with their nasal counterparts /m/ and /n/.

Response to intervention:

The key words/phrases for performance feedback agreed with PB were 'mouth and nose sounds' and 'super shiny not foggy mirror'. Home practice was minimal. PB needed a great deal of verbal prompting to look at the visual feedback provided by the feedback tools. The initial nasalance threshold was set high at 50% and reduced in 5% increments to 30% at session two and to 25% at session four, achieved with low pressure sounds (w, y, l). Over the course of the sessions PB was able to demonstrate awareness that the feedback had changed on the nasometry screen but was unable to consistently modify her speech in response to this feedback.

Participant C (PC).

Participant C was a boy, aged 9 years at the start of the research who presented with nasal speech associated with non-cleft VPD. PC had undergone tonsillectomy and adenoidectomy at age 8 years. His speech was characterised by mild variable hypernasality.

Outcomes		
Perceptual:	Hypernasality	decreased severity rating
Instrumental:	Nasalance	increase of 2% (same/worse) pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	decreased score (worse)
	PedsQL4.0	decreased score (worse)

Details of intervention:

The feedback tools used with PC were performance feedback, mirror, See-Scape™ and nasometry. He received a total of eight 45 minute sessions at the clinic, over consecutive weeks.

The tailored therapy objectives for PC were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. blue foam indicator kept at the bottom of the See-Scape™ tube, nasometry games and graphs achieved below agreed nasalance threshold.

Response to intervention:

The key words/phrases for performance feedback agreed with PC were 'soft sounds', 'shiny mirror' and 'tall and short towers' (nasometry). PC completed some home practice using low pressure words with the See-Scape™. The frequency of this varied from two to five times per week recorded in his weekly diary. The use of the mirror was discontinued after session two as slight misting gave only fleeting feedback of audible nasal emission. By session seven PC was successfully producing oral plosive sounds

without generating nasometry screen feedback and had achieved vowel production consistently below a 25% nasalance threshold.

Participant D (PD)

Participant D was a boy, aged 8 years at the start of the research. PD was diagnosed with a submucous cleft palate at the age of 5 years and his palate repair was carried out at a UK cleft centre. His speech was characterised by mild to moderate hypernasality and nasal turbulence.

Outcomes		
Perceptual:	Hypernasality	decreased severity rating
	Nasal turbulence	no change
Instrumental:	Nasalance	decrease of 6% (better) pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	increased score (better)
	PedsQL4.0	increased score (better)

Details of intervention:

The feedback tools used with PD were performance feedback and nasometry. Mirror and See-Scape were also explored but not used as the main speech symptom was hypernasality affecting vowel sounds. He received a total of eight 45 minute sessions at the clinic, but not over consecutive weeks, due to family illness.

The tailored therapy objectives for PD were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.

Response to intervention:

The key words/phrases for performance feedback agreed with PD were 'light and heavy sounds' (light = oral, heavy = nasal), 'tall and short towers' (nasometry) and 'smooth sounds', as well as 'foghorn, cow, Frenchman' (nasalised vowels). Home practice was carried out and recorded consistently five times weekly in his diary. Over the course of the sessions PD demonstrated a developing awareness of vowel quality and the difference between an oral vowel and a nasalised vowel. He progressed from being unable to discriminate oral versus nasalised vowels to 100% success with this. Fricative consonant sounds (/f/, /s/, /sh/) were introduced to 'decontaminate' the adjacent vowels i.e. reduce their nasalance. Initially PD produced the vowel 'aw' with nasalance of between 40 and 50%. By session three he achieved the sound under a 40% threshold and introduction of a prolonged 'f' before the vowel reduced nasalance to 6%.The vowel 'ee' began at around 60% nasalance and was also reduced when combined with 'f'. At Session six PD reduced 'ee' from 82% to 30% nasalance. Nasometry games were used as a reinforcer with the threshold set at 40% and PD achieved the vowels 'aw' and 'ah' at 25%-30% nasalance.

During the sessions PD spontaneously adopted various strategies in response to the visual feedback in an attempt to reduce hypernasality. This included raising the pitch of his speech, using whispered speech and also adjusting his posture. These were discussed openly and the relationship between the strategy and its effect on speech was examined so that ineffective or potentially unhelpful strategies could be discarded.

Participant E (PE)

Participant E was a boy, aged 8 years at the start of the research presenting with nasal speech associated with non-cleft VPD. PE's speech was characterised by mild to moderate variable hypernasality and nasal turbulence and inconsistent active nasal fricative.

Outcomes		
Perceptual:	Hypernasality	**increased severity rating**
	Nasal turbulence	**increased severity rating**
Instrumental:	Nasalance	**increase of 1% (same/worse)** pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	**decreased score (worse)**
	PedsQL4.0	not completed

Details of intervention:

The feedback tools used with PE were performance feedback, nasal mirror and nasometry. See-Scape™ was not sensitive to the variable nasal emission and turbulence. He received a total of eight 45 minute sessions at the clinic over consecutive weeks.

The tailored therapy objectives for PE were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games nasometry and graphs achieved below agreed nasalance threshold
3. To achieve stability of sound productions and reduction of variability.

Response to intervention:

The key words/phrases for performance feedback agreed with PE were 'soft sounds', 'shiny mirror' and 'tall and short towers' (nasometry). PE completed minimal home practice. From session one PE showed understanding of the visual feedback from nasometry but he had difficulty responding to performance feedback and modelling. By session four he was able to alternate between a nasalised and oral vowel sound using the nasometry visual feedback. Masking his speech helped i.e. the CI producing the target sounds at the same time. Nasometry game thresholds were reduced by 5% nasalance increments over the course of the sessions from 40% to a minimum of 30%.

Participant F (PF)

Participant F a girl age 7 years at the start of the research presenting with nasal speech associated with non-cleft VPD. PF had a diagnosis of cerebral palsy and her speech was characterised by mild to moderate variable hypernasality, audible nasal emission and nasal turbulence. She had received previous speech therapy aimed at improving breath support for speech, improving articulatory precision and eliminating active nasal fricative as well as language development targets.

Outcomes		
Perceptual:	Hypernasality	no change
	Audible nasal emission	increased severity rating
	Nasal turbulence	increased severity rating
Instrumental:	Nasalance	decrease of 4% (same/better) pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	decreased score (worse)
	PedsQL4.0	not completed

Details of intervention:

The feedback tools used with PF were performance feedback and nasometry. She received a total of eight 45 minute sessions at the clinic but not over consecutive weeks, due to her illness.

The tailored therapy objectives for PF were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.

Response to intervention:

The key words/phrases for performance feedback agreed with PF were 'tall spikes/chimneys and short spikes/chimneys' (nasometry), 'mouth and nose sounds', 'smooth sounds' and 'soft sounds'. PE completed regular home practice, alternating nasal and not nasal sounds documented in her weekly diary. PF was motivated by the feedback tools and nasometry in particular. In session one she reduced the nasalance threshold for the syllable 'fa' to below 30%, then to below 25%. Similarly with the syllable 'faw' the threshold was reduced from 20% to below 15%. At session two PE achieved the sounds 'ah' in isolation below 8% nasalance threshold and 'aw' below 7% whilst /p/ and /t/ were achieved below 10%. She was also able to respond to performance feedback to soften her sound production attempts rather than using a forced voice. From session three each session started with a nasometry game as a motivator. By session five PF was consistently achieving smoother vowels in isolation and was successful in alternating between higher and lower nasalance for vowels.

Participant G (PG)

Participant G was a boy, aged 13 years at the start of the research. PG's speech was characterised by mild to moderate variable hypernasality, audible nasal emission and

nasal turbulence associated with non-cleft VPD. He had received speech therapy previously aimed at eliminating an active nasal fricative sound used as a substitution for the sounds /s/ and /z/.

Outcomes		
Perceptual:	Hypernasality	increased severity rating
	Audible nasal emission	decreased severity rating
	Nasal turbulence	decreased severity rating
Instrumental:	Nasalance	decrease of 7% (better) pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	increased score (better)
	PedsQL4.0	increased score (better)

Details of intervention:

The feedback tools used with PG were performance feedback and nasometry. He received a total of eight 45 minute sessions at the clinic over consecutive weeks.

The tailored therapy objectives for PG were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.
3. To monitor voice quality, notice creakiness and replace with smooth voice.

Response to intervention:

The key words/phrases for performance feedback agreed with PG were 'barcode' (creaky voice), 'nasal', 'smooth sounds' and 'tall and short towers'. Home practice was

inconsistent, on average twice per week. An agreed target of three times per week was only achieved once. The threshold on nasometry for PG was initially set at 30% nasalance for the vowels 'aw' and 'ah'. By session three he was using the visual feedback independently to modify his speech and reduce nasalance. At session five he was able to alternate between more and less nasal vowels, although at times he inserted an 'ng' sound in an attempt to produce a nasal vowel. The nasometry games threshold was initially set at 50% for success and over the course of the sessions reduced to 20% with the vowels 'aw' and 'ah'.

Participant H (PH)

Participant H was a girl, aged 8 years at the start of the research. PH was diagnosed with a submucous cleft palate at age 4 years. Her palate repair was carried out at age 5 years at a UK cleft centre. She also had a diagnosis of 22q11 deletion syndrome. Her speech was characterised by variable hypernasality, nasal turbulence and inconsistent active nasal fricative.

Outcomes		
Perceptual:	Hypernasality	decreased severity rating
	Nasal turbulence	no change
Instrumental:	Nasalance	no change pre-intervention score: above normal range post-intervention score: above normal range
QoL:	PedSaLQoL	increased score (better)
	PedsQL4.0	increased score (better)

Details of intervention:

The feedback tools used with PH were performance feedback, See-Scape™ and nasometry. She received a total of eight 45 minute sessions at the clinic over consecutive weeks.

The tailored therapy objectives for PH were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. blue foam indicator kept at the bottom of the See-Scape™ tube, nasometry games and graphs achieved below agreed nasalance threshold.
3. To consistently use the oral sounds /s/ and /z/.

Response to intervention:

The key words/phrases agreed for performance feedback with PH were 'tall and short chimneys/spikes' and 'invisible sounds' ('s', 'sh', 'z' produced with zero nasalance). Home practice involved the use of See-Scape™ with 's' and 'z' sounds. Regular practice was recorded in the diary some weeks but was inconsistent. From session one PH was able to increase the nasalance of vowels when imitating the CI's model, although prompting was needed to attend to the visual feedback on the nasometry screen. PH was sensitive to perceived failure so the nasometry games were used as motivator, with a high nasalance threshold. At session two PH achieved a reduction in vowel nasalance using fricative 'decontaminator' consonants, then the vowels in isolation. At session five she was able to produce 's' and 'z' without nasal emission using the See-Scape™. In addition the threshold was reduced from 40% to 35% on the nasometry game. There was within session variability, at session six PH achieved the vowels 'ah' and 'aw' below thresholds of 18% and 25% respectively when preceded by 'f' and 's'. However when attempting to replicate this level for the vowel in isolation nasalance rose to between 45% and 60%. In session seven the nasometry game threshold was

initially set at 40% initially with target syllables and was successfully reduced to 25% in 5% increments.

Participant I (PI)

Participant I was an adult male, aged 43 years at the start of the research. PI had a cleft palate, repaired in infancy. He underwent further palate surgery to improve his speech in 2009. His speech was characterised by mild to moderate hypernasality.

Outcomes		
Perceptual:	Hypernasality	**decreased severity rating**
Instrumental:	Nasalance	**decrease of 19% (better)** pre-intervention score: above normal range post-intervention score: below normal range
QoL:	VoiSS	**decrease in all 3 subsection scores and Total Score (better)**

Details of intervention:

The feedback tools used with PI were performance feedback and nasometry. He received a total of eight 45 minute sessions but not over consecutive weeks due to his work commitments and illness.

The tailored therapy objectives for PI were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.
3. To hear and produce a clear contrast between nasalised and oral vowels.

Response to intervention:

The key words/phrases for performance feedback agreed with PI were 'soft sounds', 'nasal and non-nasal'. PI completed some home practice, increasing and decreasing the nasal quality of vowels by ear, but did not record this in this diary.

Initially PI found it difficult to alter the vowel sounds, but using nasometry feedback by session three he reduced the nasalance of 'ee' from 50% to 12%-14%. At session five all vowels were achieved with a reduction in nasalance of 10%-20% using nasometry visual feedback. At session six PI used the nasometry threshold bar set at 30% for 'ah and 'aw'. He was able to increase and decrease the nasalance for these sounds but could not bring it below the threshold. However using a preceding oral 'decontaminator' consonant facilitated a reduction in nasalance to 25%.

Participant J (PJ)

Participant J was a boy, aged 10 years at the start of the research presenting with nasal speech associated with non-cleft VPD. His speech was characterised by mild variable hypernasality. This was noted at routine assessment prior to recruitment to the study. However the ratings of hypernasality from both pre and post-intervention assessments were absent and therefore essentially normal, at least at these assessment points and for the speech tasks involved. However there was evidence of mild inconsistent hypernasality during the intervention sessions and PJ was aware of this.

Outcomes		
Perceptual:	Hypernasality	rated as absent pre and post-intervention
Instrumental:	Nasalance	decrease of 1% (same/better) pre-intervention score: within normal range post-intervention score: below normal range
QoL:	PedSaLQoL PedsQL4.0	increased score (better) decreased score (worse)

Details of intervention:

The feedback tools used with PJ were performance feedback and nasometry. He received a total of eight 45 minute sessions at the clinic but not over consecutive weeks, due to CI illness.

The tailored therapy objectives for PJ were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.
3. To hear and produce a clear contrast between nasalised and oral vowels.

Response to intervention:

The key words/phrases agreed for performance feedback with PJ were 'high and low sounds' (nasometry), 'nasal and not nasal sounds'. PJ completed some home practice, alternating nasal and not nasal vowels at reduced speed, by ear, but this was not recorded in his diary. At session one PJ's nasometry feedback showed variability in vowel sounds, some were nasalised and some oral and he had difficult stabilising this in response to the visual feedback. By the end of session two PJ could increase and

decrease nasalance for the vowels. At session three variability was evident again; nasalance for 'ah' ranged between 6% and 40% with a similar picture for 'ee' and 'aw'. The nasometry threshold bar was set at 60% for 'ee' and reduced in 5% increments to 45%. At session four 'ee' was achieved consistently below a 35% threshold and 'ah' below 15%. Introducing prolonged 'f' as a 'contaminator' sound preceding vowel facilitated reduced nasalance. At session five 'ah' and 'aw' were consistently achieved below a 10% threshold in isolation and with a preceding consonant as low as 3%. This was repeated at session six. Production of 'ee' was more variable but PJ achieved this sound inconsistently below a 20% threshold. By session six PJ was also spontaneously using strategies to reduce nasalance i.e. increasing then decreasing nasalance and using oral consonants to influence vowel nasalance.

Participant K (PK)

Participant K was a girl, aged 12 years at the start of the research. PK had a submucous cleft palate identified and repaired at the age of 11 years. Her speech was characterised by mild to moderate hypernasality and nasal turbulence.

Outcomes		
Perceptual:	Hypernasality	decreased severity rating
	Nasal turbulence	no change
Instrumental:	Nasalance	decrease of 24% (better) pre-intervention score: above normal range post-intervention score: within normal range
QoL:	PedSaLQoL	decreased score (worse)
	PedsQL4.0	increased score (better)

Details of intervention:

The feedback tools used with PK were performance feedback, See-Scape™ and nasometry. She received a total of eight 45 minute sessions over consecutive weeks.

The tailored therapy objectives for PK were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. blue foam indicator kept at the bottom of the See-Scape™ tube, nasometry games and graphs achieved below agreed nasalance threshold.
3. To hear and produce a clear contrast between nasalised and oral vowels.

Response to intervention:

The key words/phrases agreed with PK for performance feedback were 'soft sounds', 'nasal and not nasal sounds' ' wobbly sounds' (See-Scape™) and 'tall and short towers' (nasometry). PK completed home practice use the See-Scape™ with the sounds 'f', 's', 'sh', 'z'. Three to four sessions per week were recorded in her diary. In session one PK was able to reduce nasalance associated with nasal emission through the use of 'soft sounds' for 'f','s' and 'sh'. At session two she was able to increase and decrease the nasalance of vowels using nasometry visual feedback, but this was inconsistent. At session three PK practised increasing and decreasing vowel nasalance by ear as well as using nasometry feedback and was able to differentiate between the CI's models and her own attempts. PK reduced the nasalance of 'ah' from 42% to between 29%-33% but was unable to modify 'aw'. However producing 'ah' before 'aw' facilitated the lowering of nasalance. At session four PK was able to increase vowel nasalance but the minimum level was 35% even with a preceding oral 'contaminator' consonant.

Participant L (PL)

Participant L was a girl, aged 13 years at the start of the research presenting with nasal speech associated with non-cleft VPD. Her speech was characterised by mild hypernasality, audible nasal emission and nasal turbulence.

Outcomes		
Perceptual:	Hypernasality	decreased severity rating
	Audible nasal emission	decreased severity rating
	Nasal turbulence	no change
Instrumental:	Nasalance	decrease of 34% (better) pre-intervention score: above normal range post-intervention score: within normal range
QoL:	PedSaLQoL	increased score (better)
	PedsQL4.0	decreased score (worse)

Details of intervention:

The feedback tools used with PL were performance feedback and nasometry. She received a total of eight 45 minute sessions at the clinic but not over consecutive weeks, due to bad weather and school commitments.

The tailored therapy objectives for PL were:

1. To experiment with nasal and oral airflow using the feedback tools, recognising the contrasting feedback for oral and nasal sounds.
2. To establish feedback targets i.e. nasometry games and graphs achieved below agreed nasalance threshold.

Response to intervention:

The key words/phrases agreed with PL for performance feedback were 'soft sounds', 'nasal and not nasal'. PL completed some home practice alternating nasal and not

nasal vowels, by ear, but this was not recorded in her diary. At session one PL showed variability in vowel nasalance; the sound 'ah' ranged between 8% and 60% and 'aw' between 10% and 30%. In this first session PL was successful in achieving a consistent level for each vowel below a 15% threshold. Prompting was needed to slow her speech rate and to use increased nasalance as a benchmark. In the same session syllable sound production was introduced and the syllable 'fah' was achieved below 11% with 'faw' achieved below a 13% threshold. At session two variability in vowel production was again noted initially but consistency was quickly achieved and syllable targets were all achieved below 15%. Nasometry games were used for reinforcement practice with the nasalance threshold set initially at 20% and reduced to 10% successfully for syllable repetition. By session four PL was using strategies to reduce nasalance without prompting i.e. increasing nasalance as a contrast if her production attempt resulted in an increase in nasalance.

5.10 Summary

This chapter presented the study results in terms of the feasibility and effect of VFTh. This includes recruitment and attrition results plus the results of the intervention at an individual, group and case study level.

The results showed that the intervention is feasible but some changes are required, particularly around participant screening, if further research is planned. In terms of effect, in blinded evaluation, positive changes were observed in at least one speech symptom between pre and post-intervention assessment for eight participants. Eight participants showed improved nasalance scores and seven had an improved total score on the condition-specific quality of life measure.

The above results must be viewed with caution as there was also variability evident between the two pre-intervention (baseline) repeated measures for perceptual and

instrumental outcomes and also variability between the different outcomes (perceptual, instrumental and quality of life).

The next chapter will consider the intervention and the results in relation to the feasibility methodology and research questions. The variability of results observed across pre and post intervention points will be given particular attention. The theoretical model of the intervention will also be discussed in the light of the results, with reference to previous studies in the field.

Chapter Six: Discussion

6.1 Introduction

This study investigated the use of Visual Feedback Therapy (VFTh) in twelve participants with nasal speech symptoms associated with velopharyngeal dysfunction (VPD). To the author's knowledge, this is the first study that has set out to assess the feasibility of a behavioural speech therapy treatment for patients with nasal speech, using a range of outcome measures.

The primary aim was to investigate the feasibility of the intervention; including recruitment and retention, theoretical modelling and the practicalities of delivery. The secondary aim was to investigate effect related outcomes using perceptual and instrumental measures of speech along with self-reported quality of life

The results are discussed in this chapter as they relate to the above aims as well as the theoretical frameworks outlined in Chapter Three. The results are also considered in relation to previous related research and the strengths and limitations of the methodological design. Finally the implications for future research are discussed.

6.2 VFTh: Feasibility

A feasibility approach was considered appropriate to provide information about the delivery of and response to the intervention in this study. This preliminary stage is necessary in order to evaluate a complex intervention such as VFTh, as discussed in Chapter Three .

The following research question has been addressed:

What is the feasibility of a newly developed speech therapy intervention combining visual biofeedback and performance feedback for children and adults with nasal speech associated with VPD?

6.2.1 Recruitment

Eighteen participants were recruited over an 18 month period to receive the VFTh intervention from one speech therapist (the Chief Investigator). An additional six eligible participants were approached at the start of the study but declined to participate. Reasons given for not taking part included the need to travel to the hospital site to receive the intervention and the need to attend appointments during school hours. The intervention itself was problematic for two participants and the decision was made to discontinue intervention in order to minimise the potential for harm i.e. loss of self-esteem and or motivation. This important aspect is discussed further in subsequent sections.

Twelve participants completed the study, representing a 67% (n=12) completion rate of the 18 recruited, and 50% of the eligible participants identified completing the study. The overall dropout rate was therefore high at 33% (n=6).

The wide geographical area served by the regional cleft centre may inevitably have impacted on recruitment and retention. Eligible participants could live at least one hour's drive from the centre, longer on public transport. Purchase of portable nasometry equipment six months into the study, after initial recruitment difficulties, did facilitate additional recruitment; allowing the intervention to be delivered at a location closer to home, in the case of six participants. It should be noted however that the action taken to increase recruitment by travelling to participants did have resource and financial implications i.e. the cost of the portable nasometer and time and travel costs of the chief investigator. These costs were not anticipated in the planning of the study, in particular researcher time required to travel and deliver the

intervention away from the main site. This may, in fact, have negatively impacted on recruitment overall.

It is possible that the drop-out of at least three of the six participants who did not complete could have been prevented by the earlier availability of the portable nasometer. These were participants who gave travel issues or impact on school attendance as the reason for not taking part. A further four eligible participants who declined or did not respond to written contact may also have responded differently had the option been available for use of the portable nasometer at a location closer to home.

These are important considerations in planning and funding future research as potential participants are likely to be recruited from one or more regional cleft palate centres, with similar travel requirements.

6.2.2 The participants

The sample size in this study was small but opportunistically included equal numbers of males and females. Nasal speech is not gender specific and therefore this aspect did not present any particular issues relating to recruitment or delivery of the intervention.

Participants were recruited from two sites of a specialist treatment centre for cleft palate/VPD where the majority of patients are under 18 years of age but with a proportion of adults returning to seek further treatment, so in this sense were representative of the population presenting with nasal speech. Two adults were recruited but only one completed the study. It is not possible to draw conclusions about the feasibility of the intervention in adults based on this sole adult participant; however a positive aspect of the inclusion of an adult participant in the study was that the individual expressed strong motivation to participate and to understand how he could modify his speech production. He was able to articulate the process involved in

responding to the visual feedback and this provided valuable supporting information about the intervention and how it might work.

Motivation to change nasal speech symptoms was an inclusion criterion for recruitment to the study judged through discussion with eligible individuals and their parents, where appropriate. Two of the twenty-four individuals who met the initial eligibility criteria of presenting with nasal speech actually declined to participate as they were not concerned about the mild nature of their speech symptoms. The requirement for motivation means that the sample was potentially biased towards participants who were motivated to improve. However this is accepted as a necessary requirement of programmes involving an element of behavioural change (Maas et al., 2008) and it was important that the intervention was tested under optimum conditions for change or learning to take place. Varying levels of motivation were observed among the child participants, on a session by session basis, although all were judged to be motivated to participate following initial discussion at recruitment. The subsequent variation in motivation was not formally recorded or evaluated but could be an interesting addition to a future study in relation to both recruitment/retention and outcomes.

6.2.2.1 Variation in participant profiles following recruitment

It is noted that there was variability evident between the pre-recruitment nasal speech symptoms and the subsequent pre-intervention (baseline) assessments for the twelve participants who completed the study (see Chapter Five, Table 5-12 and Appendix L, Table L-1). All eighteen participants who were invited to participate were rated with inconsistent mild to moderate nasal speech symptoms at routine assessment pre-recruitment. Nine participants differed in one or more nasal speech symptoms between pre-recruitment and pre-intervention (A1). This presents various challenges in this study and for future research. First of all there is the possibility that a participant identified with nasal speech symptoms pre-recruitment could subsequently present with no symptoms at pre-intervention assessment. It could be argued that

this participant should not then commence the intervention as the inclusion criteria are not strictly met. In the same way, a potential participant could present with no nasal speech symptoms 'on the day' at routine assessment (pre-intervention) and would therefore not be invited to take part in the study, even though they could be reporting nasal speech symptoms.

It is interesting to consider possible explanations for the variability observed and the implications for future recruitment. One possibility is that it reflects the inconsistency of nasal speech symptoms known to be a feature of all participants' speech. In fact, evidence of inconsistency was stated as one of the study inclusion criteria as an indication of potential for less nasal speech. Therefore, it could be argued that although problematic in terms of evaluating the outcome measures used and any effect of the intervention, this variability was to be expected. If this is accepted, it would also imply that recruitment procedures need to take account of potential variability, so that an individual who presents for routine assessment with concern about current nasal speech symptoms, but no observed symptoms 'on the day' can be considered as a potential participant. Similarly if nasal speech is identified pre-recruitment but is not then evident pre-intervention this should not necessarily preclude recruitment. Some modification to the recruitment procedure would be needed to clarify the exact circumstances supporting inclusion or not.

The variability in profiles between assessment points could also be influenced by the relatively mild severity of the nasal speech symptoms observed for some participants. This could mean that participants who are rated with borderline or mild nasal speech symptoms on one occasion and are inconsistent in the presentation of these symptoms, could be rated as less severe or even absent on another occasion. Again this presents a challenge in recruitment and in measuring the effect of the intervention in this study.

An alternative explanation for the difference between pre-recruitment and pre-intervention profiles is that this relates to the different assessment tools used, GOS.SP.ASS and CAPS-A. This will be discussed in a later section in relation to outcome measures and expert rating.

6.2.3 The intervention

An important strength of this study relates to the development of VFTh, informed by clear principles of speech acoustics and learning theory. To date no other study has fully explored the theoretical rationale for such an intervention or fully described the intervention itself although the importance of theoretical frameworks in the absence of unequivocal evidence of efficacy is widely supported (Arvedson et al., 2010; Robbins et al., 2008; Weismer, 2006). The theoretical model proposed in Chapter Three (page 96) is further strengthened by the positive outcomes for some participants combined with the individual responses to the visual biofeedback and performance feedback procedures.

In this study the intervention, VFTh, was not completely standardised. It followed specific principles and used standard equipment but was tailored to the needs of individual participants both in content and progression. This is accepted practice in speech and language therapy where therapists take an 'eclectic view' in constructing an individual's therapy programme (Roulstone et al., 1999). Objectives were tailored for each individual in relation to their nasal speech symptoms. At each session understanding of the objectives was checked, including the meaning of the visual feedback provided by the feedback tools. Key words and phrases were used as part of performance feedback to conceptually reinforce the objectives. These were often generated by the participants themselves when asked to describe the visual feedback e.g. 'foggy mirror', 'wobble' or 'jump' (See-Scape) or 'tall tower' (nasometry graphs). Otherwise key words/phrases were offered by the therapist (CI) and adopted according to the response of the participant.

It was particularly interesting that the adult participant was able to describe what he could feel happening in his mouth when he alternated between nasalised and oral vowel sounds. This is similar to the findings of Brunner et al. (2005) who report that after visual biofeedback treatment using endoscopy, subjects were able to describe kinaesthetic features of speech e.g. tongue placement. The adult in the current study also spontaneously described attempting to find the 'right setting' for the sounds when he practised them, which once achieved, he was able to repeat. This lends support to the concept of 'voice settings' (Greene & Mathieson, 1989), as discussed in Chapter Three. The ability of participants to modify their speech in response to biofeedback, particularly the nasalance of vowels as demonstrated through nasometry, suggests that they may have developed a habitual vowel setting in the presence of previously existing or ongoing VPD. Fletcher (1972) also refers to a 'strong internalized speech production model' which develops as a result of VPD but which may be modifiable.

This ties in with a 'bottom-up' model of speech processing (Stackhouse & Wells 1997) whereby the speaker's stored 'representations' of sounds and sound properties can be replaced with different representations. In the present study several participants were able to progress from reducing the nasalance level of vowel sounds in isolation and in syllables, using the nasometer, to making this sound modification by ear, i.e. without the presence of visual feedback, and in some cases maintained this new 'setting' at three month follow-up. Others were able to reduce audible nasal emission accompanying speech sounds using nasometry feedback indicating that their habitual or internalised model of speech was modifiable. To date no other study has considered the concept of habitual voice settings in relation to nasal speech in this way. This is clearly an area worthy of further exploration, both theoretically and experimentally.

The tailored approach does present a challenge in being able to comprehensively 'understand, describe and detail the components of therapy' as advocated by Carding

et al. (1998). It does however reflect real life practice and the fact that the intervention is ultimately designed for clinical practice and not just research (Adams et al., 2012). Nonetheless, in order to limit this element of variability at this early stage of evaluation, the intervention was based on core principles of behavioural intervention and was delivered by one speech therapist (CI). In addition there were no co-interventions for any of the participants during the study period. This was an important difference between this study and some previous research where at least one quarter of the studies in the literature review included concurrent speech therapy alongside the biofeedback intervention being studied.

6.2.3.1 Frequency of intervention

All participants received eight intervention sessions as planned but not necessarily over consecutive weeks due to participant or CI illness, participant work or school commitments or bad weather. The frequency of therapy sessions was an unplanned variable in this study. The aim was for participants to receive a session of forty-five minutes duration once a week for eight weeks, based on the accepted view of frequency and consistency as important principles of motor learning (see Chapter Three, page 88).

Only three participants received the intervention sessions over consecutive weeks due to illness (participant or chief investigator), or participant holiday, school or work commitments (see Chapter Five, page 143). Eight of the nine participants who had a disrupted intervention schedule were children and for all of them one or more school holidays contributed to the delays, as did availability of a clinic room in one locality to maximise the number of children seen on one day. For the adult participant it was work and family commitments which resulted in a delay of 4 weeks between session five and six. The issues with scheduling and managing other commitments are pertinent for all participants. The experience of this study suggests that there is a need for the behavioural aims of the intervention to be made explicit, in order that the

requirements of frequency and consistency of practice are met (Kleim & Jones, 2008; Palmer et al., 2007). Based on models of learning it is also suggested that disruptions to scheduling should be kept to a minimum (Maas et al., 2008; Schmidt & Lee, 2005).

Although difficulty with scheduling is typical of speech and language therapy intervention in clinical practice it is a potential confounder in evaluating efficacy. It is likely that session frequency in relation to treatment effect, for a behavioural intervention such as VFTh, varies in importance between individuals. Some of the participants who had disruptions reported continuing with home practice until the next session and others did not. Two participants (E and F) who showed little or no positive response to the intervention also had difficulties with attention and learning which may have made it more important for their intervention sessions to be frequent and regular. Both participants had two disruptions (maximum delay 2 weeks and 3 weeks respectively). However, two participants (D and G) who had the most disruptions (three each, ranging from three to four weeks) showed improvements across all three outcome measures.

To overcome the potential confounding caused by schedule variation a future study should aim to avoid disruptions as far as possible. As school holidays were a major factor in the disruptions it would seem sensible to schedule a maximum of five or six sessions depending on half term lengths. This would be primarily a pragmatic decision though as the optimum number of sessions for benefit from the intervention remains unclear. If it is accepted that any behavioural change should take place quickly, i.e. within weeks rather than months, (Whitehill, 2002a; Ruscello, 2006), a course of weekly sessions over five or six weeks should allow for any change to be seen. Alternatively a course of eight sessions could be delivered as in the current session but with the addition of a two week planned break to coincide with school holidays. There would also be the option of increasing the intensity of the interventions, perhaps offering twice-weekly sessions over a shorter intervention period. Of course this would be dependent on overcoming any travel and time

constraints identified in planning a future study. A further recommendation is for a cap to be placed on the maximum delay so that any cancelled session is rescheduled within two weeks. Although there will inevitably be disruptions in an intervention scheduled over several months these steps could help to reduce intervention variability when results are analysed.

6.2.3.2 The intervention tools

The biofeedback tools used in the study were readily available and easy to use. The Nasometer™ II was the main biofeedback tool, with See-Scape™ and nasal mirror used as supporting tools for those participants with nasal airflow errors (ANE and NT). Participants did not have the opportunity to use all of the feedback tools in between sessions. The See-Scape™ and nasal mirror were available for loan to all participants as they are relatively inexpensive but these were the least useful of the tools as they provided only a gross and transient indication of nasal airflow during speech. They were used with some participants only to contrast and reinforce the concepts of oral and nasal airflow and this is how they are generally used in clinical practice.

A future study could focus on the Nasometer alone as it has been demonstrated in the current study to provide the majority of participants with visual biofeedback in an accessible, sustained and motivating format. In this way nasometry can be seen to fulfill the same function as nasendoscopy; which has been reported with some weak evidence of beneficial effect in previous studies. Both approaches generate visual information to provide a strong stimulus to change existing speech patterns (Brunner et al., 2005), but the advantage of nasometry is that it is much less invasive.

Unfortunately there was only a single portable Nasometer™ available in the study so this could not be loaned for additional practice between scheduled sessions. This could be difficult to address in a future study due as each nasometer costs around £5000. It would be possible to carry out a study where participants receive the

intervention sequentially and so are able to use nasometry for home practice but this would probably be unrealistic in terms of timescale. Alternatively funds could be sought to purchase additional portable Nasometer™ units for home practice. However it may be that a simplified home practice version of the equipment could be developed at a lower cost, perhaps through sponsorship by the manufacturers Kay Pentax. At present there is only one alternative commercially available as the market is relatively small. This is called the Nasality Visualisation System (NVS), produced by a US company, Glottal Enterprises. This system can be used on a laptop or PC (Windows or Mac) and has a hand held mask or oral/nasal separator plate, in contrast to the headset used with the Nasometer™. This system also costs around £3500. However, to the author's knowledge the NVS system is not used in the UK and would require testing to establish validity and reliability for clinical and research use.

6.2.3.3 Home Practice

The intervention was deliberately tailored to meet individual participant needs, including worksheets for daily home practice but the procedure for monitoring home practice was unsuccessful. Participants were asked to keep a diary of home practice and to bring back worksheets with target sound/word lists to subsequent sessions. Compliance with diary completion was extremely poor; the majority of participants reporting that they forgot to complete or bring back the diary, although some gave a verbal report of practice sessions. This means that it is not possible to assess compliance with the home practice schedule and therefore the role that home practice may have played in the intervention.

Other studies have reported the use of different methods to record home therapy practice including diary sheets (Carter & Edwards, 2004), audio recordings (Holmberg et al., 2001) and even teleconferencing (Fujiwara, 2007), with variable success. A future study would either need to exclude home practice altogether or look at ways of increasing compliance with and validation of reporting. This would be the preferred

option as amount of practice is accepted as an important variable in the development of new motor programmes (Schmidt 1975) and frequent practice a requirement of motor rehabilitation (Palmer et al., 2007). The use of audio recording technology such as a voice recorder, smart phone or tablet computer, could facilitate the monitoring of actual home practice completed and allow for this aspect of the intervention to be included in analysis. These items are becoming more widely available in speech therapy departments and are often available in patient's own homes. Checking audio recordings of home practice sessions would provide the most reliable record of home practice completed and this would be feasible for sessions lasting a maximum of 15 minutes each. The content of the audio recordings could also provide additional data for analysis, subject to available resources.

6.2.3.4 Feedback

Most participants quickly understood the visual displays from the feedback tools in relation to their speech behaviours. Nasometry targets were based on best performance rather than norms with participants aiming to maintain resonance at a level below a set threshold, as described in Chapter Three. This is known as a low goal ratio and Fletcher and Higgins (1980) identify this as potentially problematic because subjects are used to a high goal ratio generally being desirable in learning tasks; for example increasing performance in some way in order to reach a pre-set target. Some participants in the present study did require additional revision of the low goal objectives during the course of sessions but this did not present a significant barrier in any one case.

The feedback element of VFTh in itself did prove to be a barrier for several participants. These individuals did not find it easy to see their speech displayed, particularly when they were not able to modify it or achieve the target level of nasometry, or other target related to See-Scape™ and nasal mirror. This was despite a clear aim on the part of the CI to explicitly distinguish between the person and their action, as Rogers (1985) would advocate. One young adult participant withdrew from

the study for this reason, although age and educational commitments may also have played a part. In other cases it was possible to talk about speech performance as external to the individual but under their control and to set achievable thresholds to allow experience of success.

Despite the positive response of the majority of participants, the possibility of a negative reaction to the intervention became evident during the course of the study. This potential for adverse effects is a very important aspect of the feasibility evaluation of this study. Roulstone et al. (1999) draw attention to this issue and cite Woolf et al. (1990) in emphasizing the need to assess the potential for harm from any intervention. This study has provided valuable insight in this respect as negative participant response to the feedback element of the intervention was not anticipated. A 'trial' screening session prior to commencing the intervention could have highlighted difficulties and allowed for a judgement to be made about potential for harm versus potential for benefit at this stage. In practice it is likely that the potential for harm may only become apparent over a period of time and action taken in response to this. Nevertheless, this aspect should be given explicit consideration in a future study and a screening procedure added to the recruitment process.

The negative response to feedback demonstrated by several participants also highlights the importance of the performance feedback element of the intervention and consideration of the ability of the nasal speaker to accept and respond to the feedback. Hayhow (2011) discusses the development of self-evaluation in young children and the importance of the nature of the 'praise' or feedback given, concluding that this should be sufficient, specific, reliable and genuine. The role of performance feedback as a component of VFTh therefore warrants further investigation and elucidation. This is particularly important for a future larger study where more than one SLT would be delivering the intervention, introducing the possibility of varying styles of performance feedback. A first step could be to develop a script of core

feedback phrases ensuring a level of consistency and specificity of performance feedback.

6.2.3.5 Implications for current clinical practice.

Whilst the results of this small study are inconclusive at this stage in relation to current clinical practice it appears that there is scope for exploring the use of nasometry as a visual biofeedback tool, beyond its current routine assessment role and outwith the research context. The biofeedback features of this tool have been largely neglected due to the lack of support for behavioural treatments for nasal speech. Although this study has only provided a preliminary indication that this approach could be beneficial it would be valuable to now share this experience with speech therapy colleagues. This process has already begun and the results of this study were presented to specialist speech and language therapy colleagues at a Special Interest Group (Cleft & Craniofacial SIG, Craniofacial Conference, April 2012). The aim was to facilitate greater understanding of the mechanisms underlying biofeedback and related performance feedback in the context of VFTh. The presentation was positively received and generated considerable discussion as well as subsequent email correspondence to the author enquiring about the potential use of VFTh in clinical practice. This is likely to be an important precursor to involving other cleft treatment centres in any future multicentre trial.

6.2.4 Summary of feasibility discussion

Despite some recruitment and retention difficulties this study has demonstrated the feasibility of recruiting patients with nasal speech in a routine clinical setting. The study also illustrates the feasibility of this behavioural intervention with children as young as 7 years of age, as well as with older children and potentially with adults. All participants were able to respond to some degree to the tailored performance feedback and visual biofeedback but required different levels of support, repetition, rehearsal and reinforcement, as would be expected in a cohort of varying ages. The

findings therefore provide further information regarding visual biofeedback as well as preliminary evidence regarding VFTh as a therapeutic strategy.

Although this study has provided valuable information about many aspects of feasibility there are questions remaining concerning participant eligibility and effective screening as well as optimum frequency and intensity of the intervention. These issues will require consideration in order for further evaluation to take place and recommendations for addressing them are made in the following chapter.

6.3 Study design

The study has a paired design and so uses paired analysis. There are limitations to this pre post-test design but pairing in this way does exploit the fact that the measures are used with the same individual before and after the intervention. This allows for comparison of the participant with themselves to counterbalance within-group variation, and increase the likelihood of detecting an effect of the intervention. It is acknowledged that the small sample size and developmental nature of the study means that results must be interpreted with caution. The small sample limits the potential of the study to show significant change and it could be argued that the use of group data is not appropriate; for this reason the emphasis is on presenting individual results before and after the intervention (see Chapter Five). In fact, the use of any inferential analysis may be considered inappropriate given the feasibility focus of the study. However, the use of non-parametric analysis means that differences in measurements can be evaluated whilst making no assumptions about the distribution of the data or generalisability of the results at this stage.

6.3.1 Outcome measures

The study design is strengthened by the selection of outcome measures that include a direct physiological measurement of speech (nasometry) and a quality of life measure,

as well as blinded perceptual assessment. This is in contrast to the majority of previous studies in this field (see pages 46-9).

The selection of multiple measures in this study was designed to capture both subjective and objective outcomes and was successful in this respect. The perceptual measure, CAPS-A, provided the listener's view of the participants' speech and the expert listeners were blinded in their evaluation of pre and post-intervention speech recordings. The instrumental measure, nasometry, complemented the perceptual assessment by providing an objective physiological measurement of speech whilst the quality of life measures aimed to record the effect on the individual of having nasal speech.

6.3.1.1 Perceptual outcome measure: CAPS-A

Perceptual assessment of nasal speech, as used in this study, represents the 'gold standard' of speech outcome measurement, as it is accepted that the primary determinant of change is how speech sounds to the listener (Kuehn & Moller, 2000; Bhuta et al., 2004; Sell, 2005). This is essentially a subjective judgement, but CAPS-A provides a validated framework for this judgement.

CAPS-A was the measure used at all assessment points in the study. At pre-recruitment routine assessment, when eligibility for study participation was considered, the specialist SLT used a different perceptual measure; the Great Ormond Street Speech Assessment (GOS.SP.ASS). This raises the possibility that the variability observed between pre-recruitment and pre-intervention assessments were related to the different assessment tools used. The GOSS.SP.ASS could have been selected as the perceptual measure for the study instead of CAPS-A, but whilst it was developed first in the UK as a standardised approach to perceptual assessment, it was considered to be too detailed for use in routine audit. This prompted the development of CAPS-A as a specific audit tool, which is closely aligned to GOSS.SP.ASS and shares a common speech sample, and which has been evaluated for validity and reliability (John et al.,

2006; Sell et al., 2009) making it more suitable for use in a research context. The two measures can therefore be used independently but the results are comparable (Sell et al., 1999), suggesting that this is unlikely to account for the variability seen between recruitment and pre-intervention assessment.

The CAPS-A perceptual ratings were made by consensus listening involving five speech and language therapists (expert listeners), all specialists in speech disorders related to cleft palate and VPD and so experienced in evaluating nasal speech. It could be argued that consensus rating was not the best approach for analysis of the speech recordings. The idea is that areas of disagreement are resolved through discussion, and if necessary, review of the disputed segment (Shriberg et al., 1984). Although this approach has been advocated by the authors of CAPS-A (Sell et al., 2009) there is a risk that aspects of the expert listener assessment could be lost in the consensus process depending on the dynamics of the group as well as duration of the process, where listening fatigue could be also be a factor.

In the current study the expert listeners were provided with reference non-nasal speech samples as an 'external anchor' (Yiu et al., 2007) or 'modulus' (Chapman et al., 2008) to supplement their internal representations and increase reliability. This would be an important inclusion of a future study if consensus listening is used again.

Whether the evaluation should be carried out by expert or lay listeners is also open to debate and both approaches have been used in previous speech studies. Sell (2005) cautions that whilst the use of lay listeners may add validity to outcomes the methodology has not been adequately developed. In contrast, Sell argues that the expert listener has a framework for evaluating nasal speech developed through experience and training; although it could also be argued that the lay listener gives a better view of the day to day reality of how 'normal' speech sounds. Of course it would be possible to include both perspectives in a future study, as previous studies have

reported (Köster et al., 2007; Whitehill, 2002b), and a comparison between the ratings of the two groups could be illuminating.

In addition the question of what constitutes a clinically worthwhile benefit requires further examination as the perspective of this may differ between patient and clinician. This highlights the need for a self-rating measure as an additional component of any future research. This differs from a quality of life measure as it relates directly to the individual's own perception of their speech. It could be used in a complementary way, perhaps as the addition of a specific question about perception of degree of nasal speech quality or satisfaction with speech, presented in a developmentally appropriate format such as a simple rating scale. This type of measure would require preliminary development and testing and it is important to note that any self-report measure can only assess the individual's beliefs about their speech, which may differ from the judgement of another person. However the validity of any such measure can be strengthened by comparing it with the other measures used in a future study.

6.3.1.1.1 Intra-rater reliability

Intra-rater reliability for the expert listeners was reported in the form of weighted kappa and percentage agreement, based on results from previous CAPS-A training (see Chapter Five). Reliability results were best for rating of hypernasality; ranging from good to very good. Rating of audible nasal emission ranged from moderate to very good and nasal turbulence had the lowest level of reliability, ranging from moderate to good. Uebersax (2010) discusses the usefulness of kappa statistics to assess rater agreement and comments that categorising ranges of kappa as 'good', 'fair', 'poor' etc. is inappropriate and also not comparable across studies. This is supported by Sim and Wright (2005) who describe these type of kappa standards as 'arbitrary'. Despite these criticisms of labelling, kappa is widely used and reported in health research in effort to demonstrate the validity of observed outcomes (McGinn et al., 2004). The consensus in the literature appears to be that the kappa statistic does

have a place in assessing agreement beyond that expected by chance but with consideration given to influencing factors such as prevalence, bias and non-independence (Petrie & Sabin, 2000). Prevalence is an issue that can affect kappa ratings as greater emphasis is attached to large differences between ratings, reflecting the fact that some disagreements are more serious than others. In the present study weighted kappa was reported which is appropriate for the ordinal nature of the perceptual data as it incorporates a notion of distance between rating categories (Fleiss et al., 2004).

The kappa results of the expert listeners were supplemented by percentage agreement results, again from previous CAPS-A training. This describes the level of agreement between speech ratings carried out on a first and second occasion (Sell et al., 2009). Three of the five expert raters achieved percentage agreement of 80% or higher for all speech parameters The remaining two raters had over 80% agreement for two out of three speech parameters.

Percentage or percent agreement has been widely used as a measure of reliability but has its limitations, namely that it is over-liberal and does not take account of chance agreement (Lombard et al., 2006). However when used alongside another measure such as kappa or Intra-class coefficient (ICC) it does provide useful information about the reliability of judgements that is not affected by prevalence issues. This was the approach taken by Sell et al (2009) in their study of 32 speech and language therapists undertaking a standardised CAPS-A training programme and has been replicated here. Whilst cautioning against the use of percent agreement, Lombard and colleagues do agree that where it is used, this is alongside another more conservative measure.

The question remains whether the reliability results, kappa and percent agreement from previous training are applicable to the current study. The results do give an indication of the expert listeners' reliability as measured during CAPS-A training but there are a number of factors that could affect this. First of all, the length of time

since CAPS-A training varied between the expert listeners from seven years to two years. However, all five listeners had attended regular and ongoing consensus listening practice following training as well as having the opportunity to administer the CAPS-A assessment for routine clinical audit. It is possible that an element of observer bias was present in the current study that could threaten the validity of reliability results obtained in training. This is because all of the expert listeners were members of the same team, working with the chief investigator, although none were involved in the clinical care of any of the participants during the study. For this reason, blinding and random ordering of the speech samples presented to the expert listeners was used to maximise independence of the ratings, particularly in the context of a consensus listening approach.

On balance, the reporting of reliability for the expert listeners in this study is a positive feature. In 1996 Wyatt et al noted the inadequacies of previous speech research in the field of cleft palate, with little attention paid to questions of reliability or bias. Similarly, in a later review of fifty-seven studies of speech intelligibility Whitehill (2002b) states that half of the studies did not report reliability. This study presents both kappa and percentage agreement results in order to give a balanced view of the intra-rater reliability of the expert listeners. If kappa or ICC was to be used in a future study it may be appropriate to calculate this based on the study speech samples rather than results from a different sample as prevalence does not apply to the whole population. Finally, the use of the percentage agreement measure does provide additional information to allow for the weakness of kappa in relation to prevalence but nonetheless caution should be taken in applying reliability results across studies.

6.3.1.2 Instrumental outcome measure: Nasometry

The instrumental measure of nasal speech used in this study has previously been reported to have good test-retest reliability and a strong relationship with perceptual ratings of nasal speech (Sweeney & Sell, 2008). As an outcome measure it therefore

provides valuable objective information to supplement the subjective perceptual measure. In addition, when there is discordance between perceptual and instrumental outcomes, as in this study, Sweeney (2011) emphasises the importance of asking why this should be so, rather than assuming one measure is incorrect or inadequate.

The nasometer was used as both an outcome measure and an intervention tool in this study. Whilst the dual function of the nasometer might be considered problematic this was not found to be so as the assessment and intervention procedures differed. Both involved wearing the headset during speech production but the assessment procedure did not require the participant to view the nasometer feedback screen during recording of the standard speech sample. In contrast the feedback screen was the main focus during the intervention sessions when different speech stimuli were used to those in the nasometry assessment speech sample.

A further positive feature was that the portable nasometer was easy to set up and use with a laptop and allowed for the equipment to be taken to a location more accessible to the participants, including homes, schools and health centres.

Finally, nasometry norms (Sweeney et al., 2004) have been included in the results for reference but were not used as a comparison in the study as the aim was not to normalise nasalance levels but rather to explore the effect of VFTh on nasalance. This is discussed further in relation to the study results later in this chapter.

6.3.1.3 Quality of life outcome measure

The subjective measure of nasal speech used in this study was a quality of life (QoL) questionnaire. Two questionnaires were used with the child participants, one generic (Peds QL4.0) and one condition specific (speech, language, communication needs, PedSal QoL). One condition specific questionnaire was used with the adult participant (VoiSS). None of the measures used specifically asked participants 'do you feel your

nasal speech is better?' however the PedSalQoL and the VoiSS do aim to capture a perception that speech has improved in some way.

As well as the question of feasibility there is also an ethical issue to consider in relation to the QoL outcome measures used in the study. Children were required to complete two QoL questionnaires as the condition specific PedSaLQoL had not been identified at the time of the ethics application. The inclusion of this questionnaire was subsequently approved as a substantial amendment by the ethics committee as it was designed specifically for children with speech and language difficulties. It was decided to keep the original questionnaire (PedsQL 4.0) as this was part of the ethics submission. In practice the completion of two questionnaires proved challenging for a number of the children, irrespective of age, and two children were not able to complete both. It was important in the study to include a measure designed to capture the child's thoughts and feelings, alongside the perceptual and instrumental speech measures as perceptions of speech and impact do not necessarily correlate. However future research should carefully consider the burden such a measure places on a child and how best to select and deliver it within the study protocol. The experience of this study suggests that one questionnaire is preferable and that this needs to be valid and reliable for the target population, so that a condition specific measure is likely to be preferable , although further evidence of sensitivity and specificity would be beneficial.

It could be argued that the QoL measures used in this study only partially addressed the need to evaluate the intervention from the perspective of the participants. The design of all three questionnaires used (PedSaL QoL, PedsQL4.0 and VoiSS) aims to capture the functional impact of speech and language difficulties, including how the individual feels about their speech, but does not necessarily capture their feelings or views about the intervention offered and therefore did not specifically address the issue of acceptability. According to Kazdin (1980, page 259) acceptability refers to 'judgments about the treatment procedures by non-professionals, lay persons, clients, and other potential consumers of treatment.'

Despite this limitation the data presented regarding recruitment and completion provides some information about acceptability as well as feasibility, suggesting that the intervention was broadly perceived as acceptable. In particular, observations of participants' responses indicated that the use of the nasometer, which involved wearing a headset, was well tolerated. One child described the benefit of being able to 'see what I'm trying to do', although as previously discussed the visual feedback was not well received by all participants. This child's comment is one of a small number of ad hoc examples in this thesis of participants' verbalising what they thought or felt about the intervention. A systematic approach to eliciting and evaluating such comments would be beneficial in a future study, potentially providing a rich source of information relating to feasibility and acceptability from the perspective of the individual receiving the intervention. This is particularly important for behaviourally based interventions like VFTh as compliance is likely to be greater if a treatment is seen as acceptable by users (Reimers et al., 1992).

There are a number of ways that the user perspective could be captured using qualitative methodology that would address the desire to understand the 'social meanings and social processes' of the intervention (Needleman & Needleman, 1996). Nastasi and Schensul (2005) discuss the ongoing debate regarding the relative value of qualitative and quantitative approaches in school-based intervention research and report a growing recognition of the added value of mixed methods. This a view is supported by Yanchar, 2006, amongst others, but qualified by the added comment that primarily methods must match the research question and the match be subject to critical reflection and sound rationale. In the case of VFTh, qualitative methods could add value to a future study in providing both formative and evaluative data. A variety of approaches are worthy of consideration, as outlined by Conrad (2001) and listed below;

- Observation – to capture comments made by participants about the intervention during VFTh sessions.

- Interviewing or 'guided conversation' – in relation to the experience of having nasal speech and or receiving the VFTh intervention.
- Focus groups– to capture views and feelings about nasal speech and or the VFTh intervention through discussion and interaction between individuals.
- Documents – capturing comments about the intervention from the homework diary or electronic record where the participant is specifically asked for a comment on each session.

Selection of the most suitable approach/es would depend on a number of factors including burden on participants and time available for subsequent data analysis. The methods available for analysis will not be discussed in detail here but would be an iterative process from describing, to classifying, to connecting concepts and through to the overall account, as described by Dey (1993).

6.4 VFTh: Effect

Although the primary focus of this study was assessment of feasibility statistical analysis was also carried out to make a preliminary examination of the data. Shanyinde et al. (2011) comment that many pilot and feasibility studies actually focus on reporting efficacy rather than addressing methodological and planning issues. However measurement of effect can be viewed here as an essential aspect of the feasibility testing of the intervention, as some preliminary evidence of effect is needed to justify further research.

This feasibility study cannot differentiate spontaneous improvement from a specific benefit of the intervention (Carey & Boden, 2003). In addition the inherent variability of the nasal speech symptoms studied makes it particularly challenging to evaluate the effect of the intervention. The following research question has been addressed;

Does VFTh improve nasal speech and quality of life as measured by a range of perceptual, instrumental and patient-reported outcome measures?

Within this heterogeneous group of participants, individuals showed the capacity to respond positively to VFTh. Analysis of CAPS-A ratings from baseline assessment (combined A1+A2) to follow-up (A4) revealed that eight out of twelve participants showed improvement in one or more nasal speech symptom following the VFTh intervention. As the changes coincided with the intervention period it is possible to tentatively interpret a treatment effect. This finding is also consistent with previous studies using biofeedback approaches where individuals responded positively to nasendoscopic biofeedback in particular.

A note of caution is required at this point however, as whilst the following data provide preliminary evidence that some speakers can modify nasal speech symptoms in response to visual feedback, this does not at this stage indicate evidence of efficacy of the intervention.

6.4.1 Perceptual outcomes: Hypernasality

As reported in Chapter Five, a comparison of combined baseline (A1+A2) with follow-up (A4) showed that seven participants had reduced hypernasality ratings, three had not changed and two were increased. Non-parametric analysis of the reduction in hypernasality ratings indicated a less than 5% likelihood that the findings could have occurred by chance ($p = 0.040$). It is acknowledged that statistical significance does not necessarily equate to clinical significance and the intra-rater reliability of the expert listeners ranged from moderate to very good, so could account for some of the differences observed in this sample.

Another possible explanation for the changes observed in hypernasality is that the participant altered the position of their tongue to modify resonance. This type of compensatory elevation and retraction of the tongue was identified in an early paper

by Shelton (1969) and also commented on by Roll (1973). This feature, also described as tongue humping, can be seen on lateral videofluoroscopy (palate X-ray) and is often associated with retracted articulation generally (Grunwell, 1998; Wyatt, 1996). This means that whilst it may assist with soft palate closure and reduce hypernasality it can also result in abnormal articulatory patterns and reduced clarity of speech. None of the participants were identified with this compensatory feature at routine videofluoroscopy prior to the intervention, although this does not necessarily preclude its development in response to VFTh. Post-intervention videofluoroscopy would have been necessary to absolutely exclude its occurrence and this is not routinely carried out due to the ethical considerations around the use of radiation, particularly in children. Nevertheless, the CI was alert to the possibility of participants developing negative compensatory speech behaviours such as retracted articulation or misuse of the voice and direct advice was given to manage any evidence of these features.

6.4.2 Perceptual outcomes: Audible nasal emission and nasal turbulence

Audible nasal emission showed a reduction in ratings from combined baseline (A1+A2) to follow-up (A4), but this was not significant ($p=0.258$). However the small number of participants presenting with this speech symptom and the use of imputed data means that firm conclusions cannot be drawn from these results.

Ratings for nasal turbulence changed for some participants and not others after VFTh. Comparison of ratings at combined baseline (A1+A2) and follow-up (A4) showed that four participants had decreased severity of nasal turbulence, four did not change and four had increased severity, but again this was not statistically significant ($p=0.334$).

One reason for the greater variability in results for audible nasal emission and nasal turbulence as compared to hypernasality could relate to the nature of the speech symptoms. They are not interdependent and can occur in isolation or together (Sell & Sweeney, 2001). Therefore a reduction in one symptom would not necessarily mean a

reduction in the others. Hypernasality is perceived on vowel sounds, whilst audible nasal emission and nasal turbulence describe nasal airflow errors i.e. the nasal escape of air accompanying consonant sounds. The perception of nasal airflow errors can therefore be influenced by other distortions of consonant sound production, such as substitution or imprecise articulation. In particular, the presence of active nasal fricatives, as previously discussed, might have influenced rating of nasal airflow errors for some participants. The expert listeners in the study had previously demonstrated satisfactory intra-rater reliability for all three speech parameters but did report nasal airflow errors as the most problematic areas for reaching consensus agreement. This may reduce the validity of the results for these parameters.

6.4.3 Instrumental outcomes: Nasalance

Eight out of twelve participants showed a reduction in nasalance (the acoustic correlate of resonance) after VFTh, (follow-up, A4), $p=0.073$). There is debate about the degree of variation in nasometry score that can be interpreted as clinically significant. Sapir et al. (2007) propose that a 10% change in nasometry from pre-intervention is needed for clinical significance whilst Watterson et al. (2005) suggest a 5% threshold. Watterson et al. (2005) based this level on a comparison of the reliability of two previous models of the Nasometer™ where they found subject performance variability of up to five percentage points in repeated measures. In this study three participants achieved reductions of more than 10% and three participants more than 5% but less than 10%. The remaining six participants either had increased scores of more than 5% (two), less than 5% (two) or minimal decreases (<5%, two).

It could be argued that any reduction in nasalance is not meaningful unless the post-intervention level falls close to or below a normal threshold for a comparable population. From Sweeney's study of English-speaking Irish children the norm for the standard speech sample was between 26% and 35% nasalance. In the current study, after VFTh two of the seven participants who showed a reduction in nasalance had a

score below 26%, using the same speech sample as in the Sweeney study. One of these participants had a nasometry score of 40% at the second baseline assessment (A2) and achieved scores of 25% and 24% respectively, immediately after VFTh (A3) and at three month follow up (A4). The second participant who scored below the 26% threshold improved by 19 percentage points to 24%, whilst the remaining five participants showed a wide range of positive changes in nasalance after VFTh, from 3% to 34% reduction, but with a post-intervention level above 26%.

The reason for the wide variation in changes in nasalance after VFTh is unclear. As previously discussed in relation to perceived nasal speech symptoms, it could be explained as inherent inconsistency of the recruited participants' speech. Analysis of the difference between the nasometry scores for each individual's two baseline assessments (A1 and A2) indicates that the differences were not statistically significant ($p=0.645$) but this should be interpreted with caution as non-significance does not equate to equivalence. The fact that an improvement in nasometry score coincided with the VFTh intervention for some participants lends some support to the existence of a treatment effect. However, it is noteworthy that those participants who had decreased (improved) nasometry scores after VFTh did not necessarily have decreased perceptual severity rating even though nasalance is the acoustic correlate of perceived resonance. Of the seven participants with decreased nasalance, four also had decreased perceptual severity rating but three participants did not. A larger sample size would more clearly demonstrate whether there was an association between these two outcomes.

6.4.4 Quality of life outcomes

The two paediatric quality of life (QoL) measures produced mixed results. A higher score on both measures is associated with increased quality of life. Some participants had an increased score on one measure but a decrease on the other, only three out of nine participants who completed both measures had increased scores on both.

Analysis of the PedSaLQoL, the condition specific questionnaire, showed a statistically significant positive increase in both Total Score and score for the Communication and Feelings subsection. In contrast, the generic PedsQL4.0 did not show any significant change from pre to post intervention but five participants had a decreased score, suggesting reduced quality of life. This result is difficult to explain but one possibility is that the differing results for the two measures relates to order of administration. The PedSaLQoL was always administered first so there may have been a fatigue factor in completion of the second measure PedsQL4.0. It should also be noted that there were results for all eleven children on the PedSaL QL but for only nine children for the PedsQL4.0. This was a clinical decision based on the reluctance of two of the children to complete two questionnaires, which were time consuming, contained some similar questions and required careful listening and concentration.

This difference in results could also be explained by the fact that the PedsQL4.0 is a generic quality of life measure, not designed specifically for children with speech and language difficulties and so was not sensitive to the characteristics of interest in the study. However this does not necessarily explain why some children had decreased scores post-intervention and it is most likely that fatigue was the influential factor

Despite positive results for the PedSaLQoL, three participants did have decreased Total QoL scores on this measure after the intervention. A possible explanation for this is that after the period of contact with the speech therapist they became more aware of their speech symptoms. This could be viewed both positively and negatively; either that the individual was more able to honestly express negative feelings after a period of therapy or that in fact the individual was made to feel more self-conscious about their speech as a result of intervention. This is a well-recognised aspect of any speech therapy intervention where the clinician is aims to achieve a balance between drawing attention to speech 'deficits' whilst at the same time providing therapeutic intervention

There was an interesting difference in result between the School sections on the two questionnaire measures. The decrease in score on PedSaLQoL might be expected given that VFTh was not delivered through school staff. This does not account for the increase seen on the PedsQL4.0 but despite a similar heading to the two sections the questions are different in content (see Appendix F). PedsQL4.0 has five questions relating to school attendance and paying attention in class. PedSaLQoL also has five questions but which focus on support at school, with questions such as 'how often do you feel you get enough help at school?' which may account for the difference in result between the two sections. The response of the child participants to two of the questions in the PedSaLQoL section was also interesting. These questions ask how often the child talks to their teacher about how they are doing at school and participants often seemed confused by this line of questioning; with one parent commenting, 'Well, they don't really do that at this age do they, it's more us, the parents.'

The results of the adult QoL measure (VoiSS) are interesting at an individual level, but cannot be compared as there was only one adult in the study. Scores were improved in all sections of the questionnaire, and the total score halved from pre to post intervention, indicating improved quality of life. In particular the Impairment (IMP) score showed improvement reflecting the report of the participant that speaking was less effortful after intervention.

6.4.5 Perceptual and instrumental outcomes: Inconsistency or change in nasal speech symptoms?

Differences in participant profiles from recruitment to pre-intervention were highlighted earlier in this chapter and possible reasons for this discussed. It is therefore also possible that the differences observed in the perceptual and instrumental results of this study relate to factors other than the intervention and subject variability in particular. However, such variability is more likely to be seen at a

within-conversation level i.e. at sentence level versus word level, rather than between assessment points. The use of a standard speech sample at all assessment points, consisting of sentence repetition and conversational speech, means that the participant's speech is directly comparable before and after the intervention and increases the likelihood of detecting an intervention effect. Nevertheless in this small study the variability between assessments (including between pre-recruitment and pre-intervention, as previously discussed) coupled with missing perceptual and instrumental data are problematic in terms of evaluating the effect of the intervention.

6.4.6 Management of pre and post-intervention assessment data

Two pre-intervention assessments (perceptual and instrumental), A1 and A2, were carried out two weeks apart in this study in an attempt to evaluate the stability of nasal speech symptoms for comparison with post-intervention assessments. As reported in Chapter Five there was variability evident between A1 and A2 but statistical analysis showed a strong association between the two sets of results. Therefore it was judged that an average of A1 and A2 combined provided the more robust representation of pre-intervention status. It is acknowledged that the perceptual (CAPS-A) data is ordinal and would not normally be treated as continuous or numerical data in this way. However this approach was taken in order that baseline data from the perceptual and instrumental measures was treated consistently.

Frison and Pocock (1992) describe the use of simple summary statistics for analysing repeated measures, using a mean pre-treatment level, arguing for the value of such approaches in clinical trials. In this study the approach has been to use a mean of baseline (pre-intervention) measures and to compare this with post-intervention measures (A3 and A4). The post-intervention measures were not combined as they served different purposes; A3 to detect change immediately post-intervention and A4 as the final follow-up assessment after three months of no intervention.

6.4.7 Missing data

As reported in the previous chapter, despite the small number of participants in the study there was missing data (see Chapter Five, page145). In determining an appropriate strategy for managing this issue it is important that the nature of 'missingness' is identified (Carpenter & Kenward, 2008). This depends on whether the missing data is random or non-informative, or conversely dependent on subject characteristics. In most clinical studies missing data relates to drop-outs, therefore is informative or dependent and in some cases could be seen as a failure of the treatment of interest. The two participants who withdrew from the study arguably could have been included in the analysis as their withdrawal was related to the nature of the intervention. A decision was taken not to do so as the majority of the missing data related to technical failures in transferring speech recordings from one device to another and so can be described as random (missing completely at random, MCAR).

The strategy adopted in the study was a simple ad hoc imputation approach; last observation carried forward (LOCF) as described in Chapter Five. This has been widely used but is criticised as not being underpinned by statistical theory by Carpenter and Kenward (2008) who recommend avoiding this type of simple, ad hoc approach. Hamer and Simpson (2009) also acknowledge that LOCF makes assumptions about the relationship between repeated measures on the same subject, namely assuming that they remain constant. As Kenward and Molenberghs (2009) point out this increases the risk of bias, with the direction and degree of this dependent on true but unknown treatment effects. This is also problematic in this study where a result at the second baseline assessment, pre-intervention (A2) is carried forward to the first post-intervention assessment (A3) as this immediately assumes that the intervention has had no effect.

Given that the data in this study was predominantly MCAR an alternative approach would have been to carry out only complete case (CC) analysis. However this would

have reduced the analysis by a third to eight participants, which is not ideal in a small study. The small sample size was one of the reasons for choosing the LOCF approach as well as the fact that all participants did have first baseline (A1) and follow-up (A4) results.

It is acknowledged that the approach to missing data in this study has been simple and ad hoc in order to maximise the data available for analysis and that this could have led to an under or overestimation of the results. Soon (2009) advises that reducing missing data is the best way to address this issue and adjustments to the study protocol to increase the efficiency of speech sample recording are recommended. In addition a future study would include a plan for statistical analysis strategies, to include the analysis of participants who drop-out (De Souza et al., 2009).

6.4.8 Hawthorne effect

The possibility that participants responded to therapeutic attention rather than the intervention itself cannot be ruled out by this study (Carey & Boden, 2003). This is known as the 'Hawthorne effect', where change is seen in an experimental group as an effect of being treated differently (Bowling 2002). The influence of the clinician on therapy effectiveness is acknowledged (Carding et al., 1998) and is consequently difficult to separate from the intervention itself.

The repeated measures design allows participants' performance to be directly compared before and after the VFTh intervention but a number of questions remain in relation to the lack of comparison or control group. First of all the effect of history, how do we know that an external influence did not influence the outcomes? It is possible to state that no other speech therapy treatment took place for the duration of the study and this makes it more likely that the outcomes are related to the intervention. Secondly how do we know that participants would not have changed spontaneously irrespective of the study intervention or any other intervention? In this

respect, all participants presented to the clinical service with a history of ongoing nasal speech i.e. this had been present for at least three months prior to the start of the study and usually much longer than this. Finally how do we know that participants did not just improve on the outcome measures because they had been tested before? It is possible that there was a practice effect in that participants might use their 'best' speech at assessment after the intervention. However as the aim of VFTh was to examine whether participants could voluntarily modify their speech this could be viewed as a positive effect of the intervention. In addition not all participants showed improvement suggesting that factors other than familiarity were at work.

6.4.9 Potential for improvement

This study explored one of the assumptions put forward by previous research and underpinned by learning theory that potential for improvement in speech needs to be evident for a behavioural intervention such as VFTh to have any effect (Kummer, 2004). The documented individual response of participants at each intervention session lends support to this view. As previously discussed, all participants had been identified at routine assessment, prior to recruitment, as demonstrating inconsistency in nasal speech symptoms and potential for less nasal speech. All went on to show some ability to use visual feedback to modify their speech during the course of the intervention sessions, as evidenced in the individual case studies, although this was not necessarily reflected in the study outcomes. Hayhow (2011) emphasises the value of this approach to analysing the details of participants progression through treatment in an attempt to recognise patterns and profiles and to 'dismantle' or 'unpick' the components of the treatment.

6.5 Summary

The results of this feasibility study make the case for further testing of the VFTh intervention and have highlighted two key questions for consideration in planning a future study. First of all, is the intervention well enough developed to permit further

evaluation and secondly, is there preliminary evidence that the intervention is likely to be beneficial? (Sibbald & Roland, 1998). The following chapter considers these questions by summarising the conclusions that can be drawn from this study. It then recommends the most appropriate direction for further evaluation of VFTh, informed by the findings of the current study.

Chapter Seven: Conclusion

7.1 Potential value of this research

At the present time speech therapy is not routinely offered for nasal speech associated with velopharyngeal dysfunction and the current emphasis in treatment is often on surgery. Evidence that speech therapy could reduce nasal speech symptoms would highlight an alternative less costly and less invasive treatment option for some individuals with nasal speech.

This thesis describes a study designed to explore a newly developed speech therapy intervention for nasal speech (VFTh) and is the first study to examine the feasibility and effect of such an intervention. It provides a comprehensive initial evaluation of VFTh, in twelve participants with mild to moderate nasal speech associated with velopharyngeal dysfunction. The main purpose of this feasibility study was to identify potential limitations in preparation for a future larger trial. The question is whether the results from this study provide sufficient evidence, given the variability in individual outcomes, to warrant further evaluation of VFTh?

This study does suggest that VFTh, based on a sound theoretical model, may be a feasible intervention, and has provided valuable information to inform further research. The next stage will aim to make future evaluation more feasible (to increase recruitment and retention) and as robust as possible in terms of detecting any benefit. This will involve refining the intervention; determining optimal duration and intensity and primary outcome measures. Specific recommendations for addressing these aspects are given in this chapter.

7.2 Limitations and future research

This study has fulfilled the feasibility aims discussed in Chapter Six and has identified limitations which require consideration relating to the intervention and its evaluation. These are summarised here, along with recommendations for refinements and modifications to address these issues in a future study.

7.2.1 Disruptions to intervention schedule

Disruptions to the intervention schedule were an unplanned variable in the current study, as discussed in Chapter Six. This means that questions remain about whether a different 'dose' of the intervention (frequency/duration) would have resulted in speech gains for those participants who did not improve. It is also possible that more practice, including documented home practice, might have result in greater improvements for those participants who did show reductions in nasal speech symptoms.

In planning a future study there is little empirical evidence to draw upon regarding optimal frequency and intensity of speech therapy treatments. The pragmatic approach, based on general theories of learning, is to allow long enough for an effect to be seen i.e. weeks rather than days, whilst maximising practice and minimising disruptions. The current study has shown that some individuals were able to respond over the course of eight sessions, although these were not delivered at the same intervals. It is therefore proposed that eight sessions is the minimum for a future study and that scheduling is planned to allow for a maximum two week break at any time. This will ensure a more consistent approach to delivery of the intervention but may result in withdrawal of participants if unforeseen circumstances lead to a break of more than two weeks. In this situation it may be possible for a participant to re-enter the study after an appropriate 'wash-out' period, although the learning element of the intervention may mean that this is not possible.

7.2.2 Potential for adverse effect

This study has highlighted the need for caution in relation to the intervention as a negative reaction to or inability to effectively use the feedback could be potentially harmful; affecting self-esteem and motivation in an individual who is already aware of having speech that sounds different to others. This potential for adverse effect was not anticipated and indicates the need for a screening process whereby potential participants undergo a suitability assessment, in the form of a 'trial' session, to determine whether they are comfortable with and can make use of the visual biofeedback and performance feedback. Specific advice/training would also be required for clinicians delivering the intervention to alert them to the risk of an adverse response and identify appropriate and agreed management strategies, including robust procedures for early withdrawal where necessary.

7.2.3 Outcome measures

The conflicting individual results obtained within and between the different outcome measures do make it difficult to ascertain the true effect of the intervention and to determine which outcome is most important. However the use of multiple outcome measures does constitutes a strength of the current study due to the multidimensional nature of the measures, reflecting the complex nature of the intervention. Obtaining a balance in outcome measures between sensitivity to change and meaningful change for the individual is essential but challenging, as demonstrated by the current study. In order to determine the optimal approach for a future study consideration of the following is needed:

7.2.3.1 Perceptual outcome

Perceptual assessment, as the gold standard in speech analysis, is a minimum requirement of a future study. This would be the primary outcome measure. As previously discussed, consensus rating of speech samples using the perceptual measure CAPS-A, as used in this study, is already well established in routine speech

audit in the UK and would be appropriate and straightforward to repeat in future research. The use of an extra 'external' listener i.e. a specialist speech & language therapist from a different cleft centre is recommended to reduce the risk of bias.

The inclusion of a specific self-rating perceptual measure i.e. the participant's view of their speech, lacking in the current study, would be another important addition, as discussed in Chapter Six. This measure of would require preliminary development and testing of validity.

7.2.3.2 Instrumental outcome

The instrumental measure, nasometry, provided an objective result to complement the perceptual outcomes. The results of the perceptual and instrumental measures in this study do not always correlate but in a study of this size it is difficult to draw conclusions about this. As nasometry is also used a biofeedback tool in the study it could be argued that it would be better not to also use it as an outcome measure, but to concentrate on the perceptual and QoL outcomes instead. However this decision should not be made on the basis on conflicting results from CAPS-A and nasometry but rather based on consideration of the best outcome measures for the research question and study design. In this respect nasometry can be seen as a valuable secondary outcome measure as it provides an objective physiological measurement of the acoustic correlate of nasal speech. Both perceptual and instrumental measures are therefore essential in future research.

7.2.3.3 Quality of Life outcome

Two quality of life questionnaire measures were used with the children in the study, one generic and one specific to speech and language difficulties which was introduced after initial ethical approval. It was clear that two questionnaires presented an unnecessary burden for the child participants and this may have affected their responses. The PedSaLQoL is currently the most appropriate tool available to measure the functional and emotional aspects of speech behaviour in children. The current study has demonstrated that the tool was broadly acceptable to the child participants

and showed some evidence of sensitivity to the intervention so it would be appropriate to use in a future study.

In addition to a QoL measure consideration should be given to the addition of a qualitative element to the study, as discussed in Chapter Six. This could take several forms, for example audio-recording of VFTh sessions to capture participant comments; a focus group of participants to discuss their experience of nasal speech and receiving VFTh and an electronic record of home practice to include a comment from the participant on each session. Appropriate methods of data collection and recording would be required with particular attention given to any additional burden placed on participants.

7.2.4 Missing data

Missing data was a factor that limits the scope of meaningful analysis in this study and would require a clear strategy for a future study, both practically and for analysis. The use of video-recording (with high quality microphone) as well as the voice recorder would have provided a back-up recording and is a recommendation for a future study to minimise technical loss of data. There was also missing data for the quality of life measures as two children were unable to complete both of the quality of life questionnaires. For this and other reasons (as previously discussed in section 7.2.3.3 above) in a future study the use of a single questionnaire measure is recommended.

7.2.5 Sample size

As a feasibility study this study was not designed to demonstrate statically significant differences but to look for indications of possible positive effect. Nevertheless the small sample does increase the risk of a Type II error, even in the context of a feasibility study and inevitably limits the conclusions that can be drawn. Bowling (2002) highlights the potential for both Type I (false positive) and Type II (false negative) error in health research and it is possible that the results of this study could

suggest an effect that in reality does not exist or that the study is too small to detect an effect that does exist.

This study is not designed to test a hypothesis and therefore the tolerance of Type I error can also be higher at this preliminary stage to allow any therapeutic effect to emerge (Robey, 2004). The first step in planning a future study would therefore be to estimate the sample size required to provide adequate power with a Type I error rate of 0.05 and a power of 80%, Type II error rate of 1 in 5.

7.2.6 Lack of control group

Whilst the pre-test post -test design allows for individuals to effectively act as their own 'control', this study is limited by the lack of control group in terms of the conclusions that can be drawn from the results. A non-randomised, non-controlled trial ultimately cannot address the question of efficacy, or exclude the possibility of a placebo effect of the intervention. In order to do so in a subsequent study it would be necessary to include a control intervention or non-intervention group as a comparison.

The question is what type of control intervention or non-intervention group would be needed as a comparison? There are several possible research designs that could contribute to testing the findings of this study. Arguably there is insufficient evidence of effect at this stage to consider an active comparator study, for example a waiting list control group and it is suggested that further preliminary evaluation is needed.

7.2.7 Future study design

In order to design a future controlled study therefore a number of interim steps are required and the use of a larger series of case studies is suggested as the most appropriate next step. This would provide more data about individual response to the VFTh intervention, building on the approach taken in this study. At a later stage, ideally the design for a pilot study, and in due course a main RCT, would involve

randomisation to intervention or control groups as this would give greater confidence in detecting a real treatment effect. The most likely control group would be non-treatment as there is no 'standard' treatment alternative for comparison. As there appears to be some evidence that VFTh will benefit some individuals a randomised waiting list study could be used, as described by Campbell et al. (2000), whereby all participants ultimately receive the intervention. Alternatively, it is possible a non-randomised control group could be selected from a cleft treatment centre where nasometry is not available or not used as a biofeedback tool, although this would be less desirable.

7.3 Treatment effect or variability?

An important finding of the current study relates to the variability in responses for individuals across the three nasal speech symptoms and in self-reported quality of life. Some participants showed improvement in speech symptoms on the perceptual measure but not on the instrumental measure. Some showed decreased severity of one symptom but an increase in another. Some participants showed improved quality of life scores and others did not. This does makes it difficult to draw conclusions about the changes observed between pre and post-intervention points and emphasises the importance of the feasibility stage of evaluation employed in the current study.

All of the participants were identified as having inconsistent speech symptoms before starting VFTh and unsurprisingly this was also identified between symptoms measured at the two pre-intervention assessments. However it is not clear from the current study whether the variability between measures and between assessment points is a true finding or a measurement issue. There was an attempt to evaluate stability of nasal speech symptoms pre-intervention to allow for comparison with post-intervention results. A future study would be strengthened by the inclusion of three baseline measurements, as commonly used, providing a mean average. Alternatively, given the ordinal nature of the perceptual rating scale, a median could be used. It

should be noted however that the validity of baseline data results calculated in either way is reliant on minimising missing data.

7.4 Summary of recommendations for future research

This study suggests that individuals with nasal speech may be able to modify habitual nasal voice settings in response to VFTh. There is a need for further preliminary evaluation before testing of efficacy can be considered, in order to provide confirmatory evidence of potential effect and acceptability. A case series design is therefore recommended in preparation for a subsequent pilot study and RCT, if appropriate, to include the following key aspects:

- A series of single case studies: to provide further data on individual response to VFTh, in individuals with nasal speech.
- Recruitment at additional cleft treatment centre sites to increase sample size and to include both children and adults.
- Addition of a 'trial' VFTh session to the recruitment protocol to screen for adverse response to the intervention.
- Primary outcome measure to be perceptual (CAPS-A), plus development of an additional self-rating measure , as well as instrumental (nasometry) and quality of life (Ped SaLQoL) measures.
- Consideration of the addition of a qualitative element to the study to evaluate the subject experience and acceptability of the intervention.
- Intervention to include both visual biofeedback and performance feedback as in the current study but with modifications i.e. use of the nasometer as the sole biofeedback tool and scripting of performance feedback phrases for consistency.
- Disruptions to intervention schedule to be kept to a minimum by scheduling eight sessions with a maximum of two week break.
- Home practice to be emphasised as a requirement of the study and include use of electronic devices (e.g. smartphone, tablet, laptop) to increase compliance with completing and recording home practice, where possible.

- Follow up: to include more than one follow-up over a longer time period to assess durability of effect (e.g. 1 month and 6 months post-intervention).

7.5 Concluding remarks

This study makes an important step forward in understanding how Visual Feedback Therapy might work and how it can be evaluated. The model proposed for VFTh integrates theory from psychology, linguistics and speech acoustics to explain how new settings for less nasal speech can be elicited, monitored and reinforced. In addition the model provides a clear framework for the role of the individual with nasal speech and the speech therapist in generating and using feedback in a therapeutic intervention.

Alongside the modelling of the intervention, the results of the study lend tentative support to previous research findings that some individuals with nasal speech can respond to behavioural speech therapy treatment using biofeedback techniques, if they demonstrate potential to change habitual nasal speech settings. Taking these aspects together, the study makes an important contribution to understanding how an intervention like VFTh might work and how it could be delivered in clinical practice.

Most importantly it suggests that further evaluation is warranted and has provided important insights into how this should be conducted. In this respect the study fulfills the recent criteria recommended by Law et al. (2012) in relation to the evaluation of speech and language therapy interventions; first of all it demonstrates sound theoretical underpinning and secondly it has undergone formal evaluation, in this case feasibility. The results have provided what Law et al. describe as an 'indicative' level of evidence, i.e. good face validity but limited research evidence, at this stage.

These findings add to the body of knowledge in the area of non-surgical treatments for nasal speech and will provide the impetus for further research; initially in the form

of refining of the intervention and evaluation methods. If positive findings are seen in a future larger study this would raise important questions about the nature and management of persisting nasal speech associated with VPD; presenting the possibility of a lower risk, lower cost and more acceptable alternative to surgery for some individuals.

LIST OF APPENDICES

APPENDIX A: NHMRC Evidence Hierarchy

Table 3 NHMRC Evidence Hierarchy: designations of 'levels of evidence' according to type of research question (including explanatory notes)

Level	Intervention [1]	Diagnostic accuracy [2]	Prognosis	Aetiology [3]	Screening Intervention
I [4]	A systematic review of level II studies	A systematic review of level II studies	A systematic review of level II studies	A systematic review of level II studies	A systematic review of level II studies
II	A randomised controlled trial	A study of test accuracy with: an independent, blinded comparison with a valid reference standard,[5] among consecutive persons with a defined clinical presentation[6]	A prospective cohort study[7]	A prospective cohort study	A randomised controlled trial
III-1	A pseudorandomised controlled trial (i.e. alternate allocation or some other method)	A study of test accuracy with: an independent, blinded comparison with a valid reference standard,[5] among non-consecutive persons with a defined clinical presentation[6]	All or none[8]	All or none[8]	A pseudorandomised controlled trial (i.e. alternate allocation or some other method)
III-2	A comparative study with concurrent controls: • Non-randomised, experimental trial[9] • Cohort study • Case-control study • Interrupted time series with a control group	A comparison with reference standard that does not meet the criteria required for Level II and III-1 evidence	Analysis of prognostic factors amongst persons in a single arm of a randomised controlled trial	A retrospective cohort study	A comparative study with concurrent controls: • Non-randomised, experimental trial • Cohort study • Case-control study
III-3	A comparative study without concurrent controls: • Historical control study • Two or more single arm study[10] • Interrupted time series without a parallel control group	Diagnostic case-control study[6]	A retrospective cohort study	A case-control study	A comparative study without concurrent controls: • Historical control study • Two or more single arm study
IV	Case series with either post-test or pre-test/post-test outcomes	Study of diagnostic yield (no reference standard)[11]	Case series, or cohort study of persons at different stages of disease	A cross-sectional study or case series	Case series

NHMRC levels of evidence and grades for recommendations
December 2009

APPENDIX B: CONSORT STATEMENT CHECKLIST

CONSORT 2010 checklist of information to include when reporting a randomised trial*

Section/Topic	Item No	Checklist item	Reported on page No
Title and abstract			
	1a	Identification as a randomised trial in the title	
	1b	Structured summary of trial design, methods, results, and conclusions (for specific guidance see CONSORT for abstracts)	
Introduction			
Background and objectives	2a	Scientific background and explanation of rationale	
	2b	Specific objectives or hypotheses	
Methods			
Trial design	3a	Description of trial design (such as parallel, factorial) including allocation ratio	
	3b	Important changes to methods after trial commencement (such as eligibility criteria), with reasons	
Participants	4a	Eligibility criteria for participants	
	4b	Settings and locations where the data were collected	
Interventions	5	The interventions for each group with sufficient details to allow replication, including how and when they were actually administered	
Outcomes	6a	Completely defined pre-specified primary and secondary outcome measures, including how and when they were assessed	
	6b	Any changes to trial outcomes after the trial commenced, with reasons	

Sample size	7a	How sample size was determined	
	7b	When applicable, explanation of any interim analyses and stopping guidelines	
Randomisation:			
Sequence generation	8a	Method used to generate the random allocation sequence	
	8b	Type of randomisation; details of any restriction (such as blocking and block size)	
Allocation concealment mechanism	9	Mechanism used to implement the random allocation sequence (such as sequentially numbered containers), describing any steps taken to conceal the sequence until interventions were assigned	
Implementation	10	Who generated the random allocation sequence, who enrolled participants, and who assigned participants to interventions	
Blinding	11a	If done, who was blinded after assignment to interventions (for example, participants, care providers, those assessing outcomes) and how	
	11b	If relevant, description of the similarity of interventions	
Statistical methods	12a	Statistical methods used to compare groups for primary and secondary outcomes	
	12b	Methods for additional analyses, such as subgroup analyses and adjusted analyses	
Results			
Participant flow (a diagram is strongly recommended)	13a	For each group, the numbers of participants who were randomly assigned, received intended treatment, and were analysed for the primary outcome	
	13b	For each group, losses and exclusions after randomisation, together with reasons	
Recruitment	14a	Dates defining the periods of recruitment and follow-up	

	14b	Why the trial ended or was stopped	
Baseline data	15	A table showing baseline demographic and clinical characteristics for each group	
Numbers analysed	16	For each group, number of participants (denominator) included in each analysis and whether the analysis was by original assigned groups	
Outcomes and estimation	17a	For each primary and secondary outcome, results for each group, and the estimated effect size and its precision (such as 95% confidence interval)	
	17b	For binary outcomes, presentation of both absolute and relative effect sizes is recommended	
Ancillary analyses	18	Results of any other analyses performed, including subgroup analyses and adjusted analyses, distinguishing pre-specified from exploratory	
Harms	19	All important harms or unintended effects in each group (for specific guidance see CONSORT for harms)	
Discussion			
Limitations	20	Trial limitations, addressing sources of potential bias, imprecision, and, if relevant, multiplicity of analyses	
Generalisability	21	Generalisability (external validity, applicability) of the trial findings	
Interpretation	22	Interpretation consistent with results, balancing benefits and harms, and considering other relevant evidence	
Other information			
Registration	23	Registration number and name of trial registry	
Protocol	24	Where the full trial protocol can be accessed, if available	
Funding	25	Sources of funding and other support (such as supply of drugs), role of funders	

*We strongly recommend reading this statement in conjunction with the CONSORT 2010 Explanation and Elaboration for important clarifications on all the items. If relevant, we also recommend reading CONSORT extensions for cluster randomised trials, non-inferiority and equivalence trials, non-pharmacological treatments, herbal interventions, and pragmatic trials. Additional extensions are forthcoming: for those and for up to date references relevant to this checklist, see www.consort-statement.org

APPENDIX C: SLT Cochrane database results 2010

Systematic Review	Protocol
1. Speech and language therapy for aphasia following stroke 2010 2. Speech and language therapy for dysarthria due to non-progressive brain damage* 2005 3. Speech and language therapy for dysarthria in Parkinson's disease* 2001 4. Speech and language therapy versus placebo or no intervention in Parkinson's disease* 2001 5. Speech and language therapy interventions for children with primary speech and language delay or disorder* 2003 6. Speech and language therapy to improve the communication skills of children with cerebral palsy* 2003 7. Interventions for childhood apraxia of speech 2008 8. Interventions for apraxia of speech following stroke 2009 9. Electropalatography for articulation disorders associated with cleft palate 2009 10. Speech therapy for children with dysarthria acquired before the age of 3 years 2009 11. Interventions for dysarthria associated with acquired brain injury in children and adolescents 2008 12. Pharmocolgical treatments for aphasia* 2001 13. Interventions for treating functional dysphonia in adults 2007 14. Interventions for preventing voice disorders in adults 2007 15. Surgical versus non-surgical interventions (voice therapy, medical treatment) for the resolution of vocal cord nodules 2001	1. Altered auditory feedback treatment for stuttering in childhood and adolescence 2004
	Registered Title
	1) Treatment for speech disorder in Friedreich ataxia 2009

APPENDIX D: CAPS-A

Cleft Audit Protocol for Speech - Augmented

Please rate all items. Omissions are ambiguous. Circle number '8' only if it is not possible to make a judgement.

Name:		Date:	Audit No:
Date of Birth:	Age:	Centre:	
Male/Female	First Language:	Therapist:	
Type of Cleft/Structure:			
Background information (e.g. current URTI, Voice disorder)			

Insufficient speech sample to audit the case:		8
1 Intelligibility/Distinctiveness of speech		
Rating	Description	
0	Normal.	8
1	Different from other children's speech, but not enough to cause comment.	
2	Different enough to provoke comment, but possible to understand most speech.	
3	Only just intelligible to strangers.	
4	Impossible to understand.	
2 Voice		
Rating	Voice Characteristics	
0	Absent	8
1	Distinctive or abnormal voice quality	
3 Resonance		
3a Hypernasality		
Rating	Description	
0	Absent	8
1	Borderline - minimal	
2	Mild - evident on close vowels e.g. zoo, three, six / ũ, ĩ, ɪ̃ /	
3	Moderate - evident on open and close vowels	
4	Severe - evident on vowels and voiced consonants	
3b Hyponasality		
Rating	Description	
0	Absent	8
1	Mild – partial denasalization of nasal consonants and adjacent vowels	
2	Marked - denasalization of nasal consonants / m̃, ñ, ŋ̃ / and adjacent vowels	
4 Nasal Airflow		
4a Audible Nasal Emission		
Rating	Description	
0	Absent on pressure consonants	8
1	Occasional: pressure consonants affected <10% of the sample	
2	Frequent: pressure consonants affected >10% of the sample (judged as highly pervasive or highly distinctive)	
4b Nasal Turbulence		
Rating	Description	
0	Absent on pressure consonants	8
1	Occasional: pressure consonants affected <10% of the sample	
2	Frequent: pressure consonants affected >10% of the sample (judged as highly pervasive or highly distinctive)	
5 Grimace		
Rating	Grimace	
0	Absent	8
1	Grimace behaviour – sufficient to distract the listener	

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6 Consonant Production																					
6a Consonant Production																					
Realization	Labial			Dental			Alveolar						Post Alveolar			Velar			Gtl	/s/Clstr	
Initial Realization																					
Correct Target	m	p	b	f	v	ð	n	l	t	d	s	z	ʃ	ʧ	ʤ	ŋ	k	g	h	st	skr sl
Final Realization																					
6b Observations of spontaneous speech: note any distinctive characteristics in speech/sound imitation																					

7 Cleft Type Characteristics (CTCs) Summary				
7a Cleft Type Characteristics (CTCs) Rate individual CTCs		Absent 0	1 or 2 consonants affected	3 or more consonants affected
Anterior Oral CTCs		NB. Transcribe consonants affected		
1	Dentalization/ inter-dentalization e.g. [t̪] [t̼]			
2	Lateralization / lateral e.g. [ls] [ɬ] or [sˡ tˡ]			
3	Palatalization / palatal e.g. [tʲ] [ç ɟ]			
Posterior Oral CTCs				
4	Double articulation e.g. [t͡k] [p͡k]			
5	Backed to velar/uvular e.g. /t d s n l/ => [k g x ŋ] [q G χ N]			
Non-Oral CTCs				
6	Pharyngeal articulation e.g. [ħ ʕ]			
7	Glottal articulation e.g. [ʔ], [h ɦ]			
8	Active nasal fricatives e.g. [m̰̃] [ñ̰] [ŋ̰̃]			
9	Double articulation e.g. [b͡ʔ] [d͡ʔ]			
Passive CTCs				
10	Weak and or nasalized consonants e.g. [b̞, b̃, d̞, d̃]			
11	Nasal realization of plosives e.g. b=>[m], d =>[n], and/or suspected passive nasal fricative e.g. f => [(f)m̥], [(f)m̩] or s / ʃ => [(s)n̥ (ʃ)ŋ̥]			
12	Gliding of fricatives/ affricates / f v / => [w], /s z ʃ / => [j]			

7b Non-cleft speech immaturities / errors	Absent	Present	Describe/ Transcribe examples
Rating	0	1	e.g. fronting, stopping, gliding, cluster reduction

7c Evidence of influencing factors: general comments on child's speech and language, hearing etc.

8 Perceived Need		
Speech and Language Therapy required for cleft speech problems at some point	Yes	No

Procedure for Expert Listening

An exploratory study of visual feedback therapy for mild to moderate nasal speech associated with velopharyngeal dysfunction

Ginette Phippen

Expert rating

Method

After all participant audio speech recordings have been collected and copied to CD they will be listened to by a group of specialist speech and language therapists. The SLTs will rate the nasal speech symptoms using a nationally adopted scale (CAPS-A). A consensus rating will be used. Listening conditions will be standardized through the use of a standard location and equipment. The speech recordings will be randomly presented so that the expert listeners are not aware of the order of their collection. A sample of non-nasal speech will be inserted between participant recordings to minimise direct comparison between consecutive recordings. This sample will be matched for age and gender.

Number of participants = 12
Male = 6
Female = 6
Age range = 7 – 44 years

CAPS-A

The standard CAPS-A listening protocol (see below) will be used to rate the parameters of interest i.e. Resonance, Audible Nasal Emission and Nasal Turbulence.

The range of ratings should not exceed 0-3 as study inclusion criteria states that hypernasality is mild to moderate only.

Speech sample	Action
Spontaneous speech	Listen only (orientation)
Counting 1-20, 60-70, Sentences	Rate Resonance and Nasal Airflow
Sentences	Review Nasal airflow rating (%)
Counting 1-20, 60-70, Sentences	Repeat listen if required
	Agree consensus ratings

Additional notes

Hypernasality

If hypernasality is inconsistent, the most prevalent nasality in the speech sample is rated. It is noted that perceived resonance could be markedly elevated by nasal substitutions of vowels or approximants, in addition to plosives. Nasal realizations of vowels should be noted and taken into account when making a resonance rating.

Hyponasality

Under hyponasality, if perceptually there is partial denasalisation then rate as mild but if the nasals /m n ng / are perceived as completely denasalised and sound like [b d g] on more than one occasion, then hyponasality should be rated as marked. If cul de sac resonance is perceived, then hypernasality and hyponasality would be rated as absent. ‘Cul de sac resonance’ should be noted in the comments section.

Nasal Airflow

It should be emphasized that when nasal emission and nasal turbulence replace consonant targets this should be noted as nasal fricatives, with an absent rating for nasal emission and nasal turbulence

APPENDIX E: Nasometry Stimulus Sentences

1. Paul likes apple pie.
2. Gary's got a bag of Lego.
3. Vicky's got a very heavy bag.
4. The zebra lives at the zoo.
5. The shoe shop was shut.
6. We were away all year.
7. Will you wear a lily?
8. Ben is a baby boy.
9. Tim had a tart for tea.
10. Daddy mended the door.
11. Kevin's looking at the book.
12. The phone fell off the shelf.
13. I saw Sam sitting on a bus.
14. John jumped off a bridge.
15. The children were watching a football match.
16. Mum came home early.

APPENDIX F: Quality of life questionnaires

Q. No:

Please read the accompanying directions before starting. Please complete in black or blue ink and answer all the questions so that we get complete information. Thank you.

Background details	
Please enter today's date (DD/MM/YYYY):	
Please describe your relationship with the young person (e.g. parent, teacher) here:	
Please describe in a few words the type of speech and language difficulties the young person has:	
Please describe any other medical or learning difficulties the young person may have (e.g. Down Syndrome, Cerebral Palsy, Dyslexia):	
Type of education (please circle):	Special / Mainstream / Support Unit / Not in Education / Other, please specify:
Does the young person have a Statement of Special Educational Needs? (please circle):	Under consideration / Yes / No / Don't Know

Please introduce the scale to the young person with the following statement:

"I want to find out how you feel about things in your life. Please listen carefully to the questions I am going to ask you and then help me to choose an answer for you. Remember, there are no right or wrong answers and nothing you say will get you into any trouble. You can talk to me about your answers first and ask me anything you want to."

0 never / 1 almost never / 2 sometimes/ 3 often /4 almost always/ 5 always

"OK, now let's talk about how you get on with people and some of your feelings"

	"Communication and feelings"	No.
9.	*How often **do** people understand you at home?*	
10.	*How often do you get angry, if people **don't** understand you at home?*	
11.	*How often **do you** understand your friends?*	
12.	*How often **do you** understand what people say at home?*	
13.	*How often is it hard to understand other children when you are talking to them?*	
14.	*How often do you get angry, when people **don't** understand you at school?*	
15.	*How often **don't you** understand your teachers?*	
16.	*How often are other children nasty to you?*	
17.	*How often **do** people understand you at school?*	
18.	*How often is it hard for you to get on with other children (e.g. people at school)?*	
19.	*How often do you feel sad?*	
20.	*How often do you get cross, if people **don't** understand you?*	

"This is all about school"

	"Independence and Participation at school"	No.
21.	*How often **do** you know what you should be doing at school?*	
22.	*How often do you need help in class?*	
23.	*How often do you have trouble keeping up with your work in class?*	
24.	*How often do you have trouble keeping up with your homework (e.g. reading, topic work)?*	
25.	*How often can you manage to do your work on your own?*	
26.	*How often do you forget things you've been asked to do at school?*	

27.	*How often is it hard to listen to your teacher/s?*	
28.	*How often do you try to answer questions in class?*	
29.	*How often do you feel you're doing well with your school work?*	
30.	*How often do you feel that you don't need any help?*	

0 never / 1 almost never / 2 sometimes/ 3 often /4 almost always/ 5 always

"Now let's talk all about the help you get at school"

	"Support at school"	No.
31.	*How often are you able to talk to your teacher about what you need to learn at school?*	
32.	*How often do you talk to your teacher about how you're doing at school?*	
33.	*How often do you ask for help when you need it in class?*	
34.	*How often do you feel you get enough help at school?*	
35.	*How often do you ask teachers to explain things to you?*	

"Ok, almost done. I'm just interested in your spare time"

	Activities	Choose answer from: Daily Every few days Weekly Monthly Yearly Never N/A / NR / DKN
36	How often do you go to a club or group?	
37	How often have you been able to join in with clubs or activities that are **not** at school?	

"Nearly finished"

You might never feel sad, but is there something that makes you sad?_____

Tell me about something that makes you happy _____

When we do this with other children, do you have any ideas for us?_____

That's all. Before we finish, do you want to say anything else or have a chat about anything we just talked about?

_____

Thank you very much for your time

ID# _____

Date:_____

PedsQL™

Paediatric Quality of Life Inventory

Version 4.0 – UK English

CHILD REPORT (ages 8-12)

DIRECTIONS

On the following page is a list of things that might be a problem for you.

Please tell us **how much of a problem** each one has been for you

during the **PAST MONTH** by circling:

0 if it is **never** a problem

1 if it is **almost never** a problem

2 if it is **sometimes** a problem

3 if it is **often** a problem

4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the **PAST MONTH**, how much of a **problem** has this been for you ...*

1.6.1.1 About My Health and Activities *(PROBLEMS WITH...)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

1.6.1.2 About My Feelings *(PROBLEMS WITH...)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

1.6.1.3 How I Get On with Others *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting on with other children	0	1	2	3	4
2. Other children do not want to be my friend	0	1	2	3	4
3. Other children tease me	0	1	2	3	4
4. I cannot do things that other children my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other children	0	1	2	3	4

1.6.1.4 About School *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

PedsQL™

ID# _____

Date:_____

Paediatric Quality of Life Inventory

Version 4.0 – UK English

TEENAGER REPORT (ages 13-18)

DIRECTIONS

On the following page is a list of things that might be a problem for you.

Please tell us **how much of a problem** each one has been for you

during the **PAST MONTH** by circling:

0 if it is **never** a problem

1 if it is **almost never** a problem

2 if it is **sometimes** a problem

3 if it is **often** a problem

4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the **PAST MONTH**, how much of a **problem** has this been for you*

1.6.1.5 About My Health and Activities *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than a couple of streets	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

1.6.1.6 About My Feelings *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

1.6.1.7 How I Get On with Others *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting on with other teenagers	0	1	2	3	4

2. Other teenagers do not want to be my friend	0	1	2	3	4
3. Other teenagers tease me	0	1	2	3	4
4. I cannot do things that other teenagers my age can do	0	1	2	3	4
5. It is hard to keep up with other teenagers my age	0	1	2	3	4
1.6.1.8 About School / College *(PROBLEMS WITH…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my school / college	0	1	2	3	4
4. I miss school / college because of not feeling well	0	1	2	3	4
5. I miss school / college to go to the doctor or hospital	0	1	2	3	4

VoiSS

The VoiSS- Voice Symptoms Scale

Your Name……..

Your Date of Birth……..

Today's Date……/……/…….

Please circle one answer for each item
Please do not leave any blank items

1.	Do you have difficulty attracting attention?	Never	Occasionally	Some of the time	Most of the time	Always
2.	Do you have problems singing?	Never	Occasionally	Some of the time	Most of the time	Always
3.	Is your throat sore?	Never	Occasionally	Some of the time	Most of the time	Always
4.	Is your voice hoarse?	Never	Occasionally	Some of the time	Most of the time	Always
5.	When talking in company do people fail to hear you?	Never	Occasionally	Some of the time	Most of the time	Always
6.	Do you lose your voice?	Never	Occasionally	Some of the time	Most of the time	Always
7.	Do you cough or clear your throat?	Never	Occasionally	Some of the time	Most of the time	Always
8.	Do you have a weak voice?	Never	Occasionally	Some of the time	Most of the time	Always
9.	Do you have problems talking on the telephone?	Never	Occasionally	Some of the time	Most of the time	Always
10.	Do you feel miserable or depressed because of your voice problem?	Never	Occasionally	Some of the time	Most of the time	Always
11.	Does it feel as if there is something stuck in your throat?	Never	Occasionally	Some of the time	Most of the time	Always
12.	Do you have swollen glands?	Never	Occasionally	Some of the time	Most of the time	Always
13.	Are you embarrassed by your voice problem?	Never	Occasionally	Some of the time	Most of the time	Always
14.	Do you find the effort of speaking tiring?	Never	Occasionally	Some of the time	Most of the time	Always
15.	Does your voice problem make you feel stressed and nervous?	Never	Occasionally	Some of the time	Most of the time	Always
16.	Do you have difficulty competing against background noise?	Never	Occasionally	Some of the time	Most of the time	Always

Please Turn Over ⇒

VoiSS

Please circle the correct answer for each item
Please do not leave any blank items

17.	Are you unable to shout or raise your voice?	Never	Occasionally	Some of the time	Most of the time	Always
18.	Does your voice problem put a strain on your family and friends?	Never	Occasionally	Some of the time	Most of the time	Always
19.	Do you have a lot of phlegm in your throat?	Never	Occasionally	Some of the time	Most of the time	Always
20.	Does the sound of your voice vary throughout the day?	Never	Occasionally	Some of the time	Most of the time	Always
21.	Do people seem irritated by your voice?	Never	Occasionally	Some of the time	Most of the time	Always
22.	Do you have a blocked nose?	Never	Occasionally	Some of the time	Most of the time	Always
23.	Do people ask what is wrong with your voice?	Never	Occasionally	Some of the time	Most of the time	Always
24.	Does your voice sound creaky and dry?	Never	Occasionally	Some of the time	Most of the time	Always
25.	Do you feel you have to strain to produce voice?	Never	Occasionally	Some of the time	Most of the time	Always
26.	How often do you get throat infections?	Never	Occasionally	Some of the time	Most of the time	Always
27.	Does your voice 'give out' in the middle of speaking?	Never	Occasionally	Some of the time	Most of the time	Always
28.	Does your voice make you feel incompetent?	Never	Occasionally	Some of the time	Most of the time	Always
29.	Are you ashamed of your voice problem?	Never	Occasionally	Some of the time	Most of the time	Always
30.	Do you feel lonely because of your voice problem?	Never	Occasionally	Some of the time	Most of the time	Always

Thank you for completing this questionnaire
Have you remembered to circle one response for each item?

For Office use:

Total VoiSS= ……

Impairment: 1, 2, 4, 5, 6, 8, 9, 14, 16, 17, 20, 23, 24, 25, 27 (max 60) = ……..

Emotional: 10, 13, 15, 18, 21, 28, 29, 30 (max 32) = ……..

Physical: 3, 7, 11, 12, 19, 22, 26 (max 28) = ……..

APPENDIX G: Research protocol and NRES approval

RESEARCH PROTOCOL

Title:

A study to examine the feasibility and effect of a behavioural speech therapy intervention on mild to moderate nasal speech symptoms in patients with velopharyngeal dysfunction.

Background:

Nasal speech is a common side effect of repaired cleft palate affecting up to 30% of individuals, even after the original cleft palate has been repaired. This is caused by poor function of the soft part at the back of the roof of the mouth (soft palate) which has to close against the back of the throat during speech. When this closure does not happen air escapes down the nose and causes speech to have a nasal quality. Velopharyngeal dysfunction is the term used to describe the lack of closure of the soft palate against the back of the throat.

Current treatment options:

Surgery is the appropriate treatment where there is a clear structural basis for the velopharyngeal dysfunction i.e. the palate is too short or does not move well. In less clear cut cases behavioural speech therapy may be offered or no treatment at all.

Clinical experience:

A significant number of children and adults present with persisting nasal speech following primary cleft palate repair in infancy. The effect on speech is to make it different to other people and difficult to understand. This can lead to lowered levels of confidence and self-esteem and a negative impact on quality of life.

Current evidence:

There is a lack of evidence for the use of behavioural speech therapy as an alternative treatment to surgery or no treatment, in patients who do not have a clear structural defect of

the palate. However behavioural speech therapy is widely used in speech and language therapy across a range of client groups and disorders and previous studies have suggested that some individuals could benefit. These studies have generally been small with a variety of approaches and outcome measures.

Research Question:

What is the feasibility of a trial of a behavioural speech therapy intervention for nasal speech in individuals with velopharyngeal dysfunction?

Clinical Context:

[redacted] twin site Cleft Centre -- [redacted]

Participants:

Minimum age of 7 years to allow understanding and cooperation with behavioural intervention. No upper age limit. Participants will be recruited as they present to the clinical service i.e. are referred with nasal speech and meet the inclusion criteria for the study.

Design:

Pre and post intervention design using within subject comparison at four assessment points over a period of six months.

Methods:

Intervention

Participants will receive the intervention once a week for 45 minutes over eight consecutive weeks from the Chief Investigator (a specialist speech and language therapist). This will take place in a hospital or community clinic speech and language therapy department, depending on the patient's geographical location. Participants will also be requested to carry out home practice for 15 minutes/ day during the study period and to keep a home practice diary.

The speech therapy intervention uses speech tasks and the participant progresses in a graded way through the tasks from practice of single speech sounds to sound combinations, single words, word combinations and finally sentences. The behavioural element of the

intervention involves the Chief Investigator providing feedback about the presence and degree of nasal speech symptoms verbally and visually using See-Scape™ and Nasometry equipment. The See-Scape™ provides the participant with visual feedback about the amount of air escaping down the nose during speech. Similarly the Nasometer provides visual feedback about the degree of nasal speech, in picture and graph form, via a computer.

Speech Recordings

Participant will provide a speech recording at each of the four assessment points. This will comprise of a standard speech sample in the clinic setting using a Philips Digital Voice Recorder and transferred to a computer in the speech therapy department. A participant number only will be used to identify the recordings. The recordings will then be copied to audio CD for expert rating.

Expert rating

After all speech sample recordings have been collected and copied to CD they will be listened to by a group of four specialist speech and language therapists (SLTs). The SLTs will rate the nasal speech symptoms using a nationally adopted scale. A consensus rating will be used. Listening conditions will be standardized through the use of a standard location and equipment. The speech recordings will be randomly presented so that the expert listeners are not aware of the order of their collection. A sample of non-nasal speech will be also be inserted between participant recordings to minimise direct comparison between consecutive recordings. This sample will be matched for age and gender and will be recruited from hospital colleagues and their family members.

Data Analysis

The within-subjects design allows each participant's performance to be directly compared over time, in relation to the intervention. Although a causal relationship between the intervention and the nasal speech symptoms cannot be inferred, it may be possible to hypothesise that a relationship exists and to suggest the likely direction of this relationship.

This study is looking for evidence of feasibility and a potential benefit to participants from the speech therapy intervention. Appropriate parametric and non-parametric statistical techniques will be used to analyse any differences between participant's speech before and after the intervention.

Outcome measures:

1 Perceptual assessment – analysis of speech samples by 4 expert listeners (speech and language therapists) using CAPS-A, a nationally adopted and validated speech outcome tool

2 Instrumental assessment – analysis of ratio of oral and nasal airflow during speech using routine Nasometry. This is expressed as a percentage nasometry score.

3 Quality of life assessment -using a validated quality of life questionnaire..

Ethical issues:

This study is subject to NHS Research Governance and independent ethical approval. It is being undertaken as part of a Clinical Doctorate programme at the University of Southampton and is therefore also subject to the University's research governance process.

Children

This study will include children and young people from the age of 7 years. This will require that that both parental consent and assent or consent from the child is gained. Young people aged 16 or 17 years will be presumed competent to give consent for themselves. Clear and appropriate verbal and written explanations of the study aims and procedures will be provided) and consent/assent documented .As cooperation with treatment is required it is essential that all participants are willing to comply. Clinical experience indicates that there can be a conflict between the motivation of the child and the parent/s to undergo therapy. A child will only be accepted as a participant if both he/she and the parent/s are in agreement.

Expected contribution of research to practice:

The value of this study lies in the attempt to investigate whether a behavioural speech therapy intervention has a positive effect for some patients with nasal speech, whose

characteristics are carefully described. In addition there is an opportunity to explore the theoretical rationale for any changes observed as well as to discuss how best to identify other patients who might benefit.

Speech and Language Therapists (SLTs) have a well-defined role in the multidisciplinary assessment of VPD. However their role in the treatment of nasal speech associated with VPD is less clear and there may be more that specialist SLTs can offer as an effective and acceptable alternative to surgical treatment or no treatment. A well designed exploratory study will contribute to the body of knowledge and debate regarding the effect of non-surgical interventions for nasal speech and could form the basis for a future randomised controlled trial.

Ginette Phippen

Speech and Language Therapist

Chief Investigator

NOTICE OF SUBSTANTIAL AMENDMENT

For use in the case of all research other than clinical trials of investigational medicinal products (CTIMPs). For substantial amendments to CTIMPs, please use the EU-approved notice of amendment form (Annex 2 to ENTR/CT1) at *http://eudract.emea.eu.int/document.html#guidance*.

To be completed in typescript by the Chief Investigator in language comprehensible to a lay person and submitted to the Research Ethics Committee that gave a favourable opinion of the research ("the main REC"). In the case of multi-site studies, there is no need to send copies to other RECs unless specifically required by the main REC.

Further guidance is available at *http://www.nres.npsa.nhs.uk/applicants/review/after/amendments.htm*.

Details of Chief Investigator

1.6.1.8.1.1.1 **Name**:
Ginette Phippen

Address: Speech and Language

Therapy Department,

Telephone:

Email:

1.6.1.8.1.2 Fax:

Full title of study:	An exploratory study of the efficacy of behavioural speech therapy for mild to moderate nasal speech symptoms in patients with velopharyngeal dysfunction.
Name of main REC:	Southampton and South West Hampshire REC (A)
REC reference number:	09/H0502/19
Date study commenced:	01/10/2009
Protocol reference *(if applicable),* current version and date	n/a
Amendment number and date:	Amendment 1: 30/10/2009

Type of amendment (indicate all that apply in bold)

(a) Amendment to information previously given on the NRES Application Form

~~*Yes*~~ *No*

If yes, please refer to relevant sections of the REC application in the "summary of changes" below.

(b) Amendment to the protocol

~~Yes~~ No

If yes, please submit <u>either</u> the revised protocol with a new version number and date, highlighting changes in bold, <u>or</u> a document listing the changes and giving both the previous and revised text.

(c) Amendment to the information sheet(s) and consent form(s) for participants, or to any other supporting documentation for the study

Yes *No*

If yes, please submit all revised documents with new version numbers and

dates, highlighting new text in bold.

Is this a modified version of an amendment previously notified to the REC and given an unfavourable opinion?

~~*Yes*~~ *No*

Summary of changes

The approved application contained details to measure quality of life as part of the trial protocol. The measure proposed to do this with children was Peds QL, a generic measure of quality of life. However, since the favourable opinion, a more valid measure of QoL for children with nasal speech has become available. The measure has been developed and validated in populations of children and young people with speech, language and communication difficulties and consequently offers a much more internally and externally valid evaluation of quality of life in the population we propose to recruit. We would like to include the paediatric Speech and Language Quality of Life scale in our study (Markham, 2008). The scale takes approximately 10 minutes to complete and would be administered by a parent / qualified speech and language therapist. A copy of this measure is enclosed for your information.

Markham, C (2008) The development and validation of a quality of life scale for children with speech, language and communication needs. Unpublished, PhD thesis.

Any other relevant information

1.6.1.9 **Applicants may indicate any specific ethical issues relating to the amendment, on which the opinion of the REC is sought.**

This amendment does not raise any new ethical dilemmas

List of enclosed documents

Document	*Version*	*Date*
Ped SaL QoL		

Declaration

- I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.

- I consider that it would be reasonable for the proposed amendment to be implemented.

Signature of Chief Investigator: ……

Print name: ……

Date of submission: ……

23.11.2009

Mr Edward Carter

Chair

Southampton and South West Hampshire

Research Ethics Committee (A)

1st Floor, Regents Park Surgery

Park Street

Shirley

Southampton

SO16 4RJ

Dear Mr Carter

REC reference number: 09/H0502/19

Please find enclosed a Notice of Substantial Amendment form in relation to the above approved study. The amendment relates to the addition of a paediatric quality of life questionnaire which I also enclose.

Thank you for your attention.

Yours sincerely

Ginette Phippen

cc. Dr Martina Prude, Head of Research Governance

31.10.2008

Mr Stephen Robinson

Chair

CLAPA Wessex

Dear Steve

Request for donation for speech therapy research

Title of research: Behavioural speech therapy for nasal speech.

I would be most grateful if CLAPA Wessex would consider making a donation towards the above research project. The research will be looking at whether speech therapy can help children and adults with cleft palate who have nasal speech.

At the moment some patients can benefit from further surgery to improve their speech. However this is not the right thing for everyone and others may be left with nasal speech that makes them difficult to understand and has a significant effect on their quality of life.

It is generally thought that speech therapy cannot help with nasal speech but we do know that some people can benefit from some approaches. This research will add to our understanding about whether speech therapy can help and if so, who it can help.

As part of the research I will be using a piece of equipment called a See-Scape™. This helps a person to see if there is air coming down their nose when they speak. I would like to buy additional See-Scapes for use in the research so that I can give them to participants to take home and practise. If CLAPA Wessex are willing to make a donation I would use this to buy ten See-Scapes at a cost of £45 each (total £450). After the research has finished the See-Scapes can be used to give on loan to patients having speech therapy from The Spires Cleft Centre.

The results of the research will be disseminated locally and nationally via presentations, journals and websites.

Thank you for considering this request, please let me know if you require any more information.

Kind regards

Ginette Phippen

Speech and Language Therapist/Chief Investigator

[redacted] Cleft Centre

Speech and Language Therapy

03.04.2009

Mr Edward Carter

Chair

Southampton and South West Hampshire

Research Ethics Committee (A)

1st Floor, Regents Park Surgery

Park Street

Shirley

Southampton

SO16 4RJ

Dear Mr Carter

REC reference number: 09/H0502/19

Thank you for your letter dated February 17th 2009, in respect of the above application. I am delighted that the Committee are content to give a favourable ethical opinion of the research, subject to clarification of the Participant Information Sheets and Consent Forms.

I enclose the revised documentation as follows

1. Participant Information Sheet: Children 16+, Version 2, 24 March 2009.

(previously Participant Information Sheet: Children 16+, Version 1, 30 December 2008)

Participant Information Sheet: Parent, Version 2, 24 March 2009.

(previously Participant Information Sheet: Parent, Version 1, 30 December 2008)

2. Parental Consent Form Version 2, 24 March 2009.

(previously Participant Consent form: Children 7-15 years Version 1, 30 December 2008)

3. Assent Form for children age 7 -15 years, Version 2, 24 March 2009.
(previously Participant Consent form: Children 7-15 years Version 1, 30 December 2008)

I can also confirm that all participant information and forms have been checked for typographical errors.

Yours sincerely
Ginette Phippen

cc. Dr Martina Prude, Head of Research Governance

APPENDIX H: Participant information sheets

Child [Version 2]

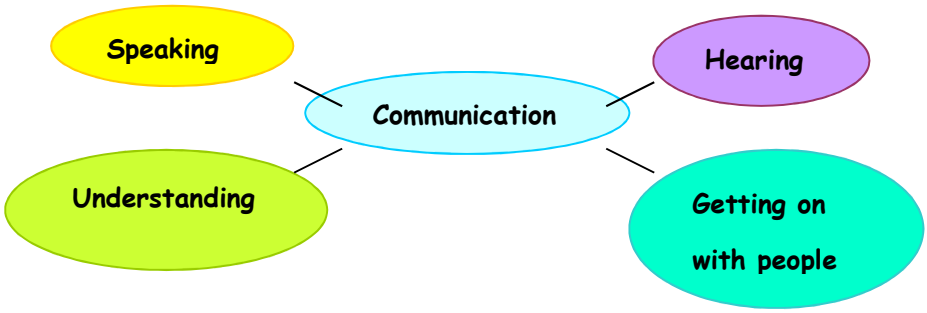
Hospital Headed Paper

Speech Therapy Research

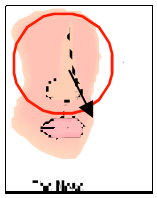
I would like to ask you to take part some research about Speech Therapy. It is important that you understand what this means, so please read this information and talk to your Mum or Dad or another grown up about it.

Research means finding out about things and looking for evidence

Speech Therapy is helping people who have problems with communication



You have been chosen to take part in this research because you had a cleft palate. This makes your speech sometimes sound like it is coming down your nose and not very clear. We call this **nasal speech.**



Air coming down the nose when you speak

If you take part you will be asked to come and have some Speech Therapy to see if this makes your speech better. You would have to come to all of the testing and Speech Therapy appointments, which will be 12 appointments altogether. The therapy will be practising words and sentences. I will use games and a computer to make this fun.

What might be hard?

☒ It might be difficult for you to come to therapy appointments at the hospital. You might have to miss time at school. I will try to fit it in so that you don't miss your favourite subjects. Also the speech therapy might not change your speech so that it might still sound the same as it did before the therapy.

What might be good?

☑ The speech therapy might make your speech sound clearer and not so nasal. Also what I find out from this research will help me to decide whether to use speech therapy with other children like you who have nasal speech.

I will keep all your details private during the research and no-one will know you are taking part unless you tell them.

What happens at the end?

At the end of the research I will tell you and other people about the **results.** The results are what I find out from doing the research with lots of people. No-one will know which results are yours except me.
We will talk about how it has gone and whether it would help you to have any more Speech Therapy. Even though the research has finished you will still be able to have more therapy if you need it.

You are in charge!

It is up to you whether you decide to take part. If you do you can still decide to pull out at any time and you do not need to tell me why. If you decide you do not want to take part or you want to pull out this is fine.

Thank you very much for reading this. If you decide you would like to take part in this research please fill in the **Consent form** with your parents and send it back to me at the hospital.

Ginette Phippen, Speech and Language Therapist

Participant information sheet

Adult [Version 1]

Participant Information Sheet

I would like to ask you to take part in some research about speech therapy for nasal speech. Before you decide, it is important that you understand why the research is being done and what it will involve. Please read this information sheet carefully and discuss it with other people if you wish. Please ask me if there is anything that is not clear or if you need more information. Take your time to decide whether or not you wish to take part.

1. What is the purpose of the research?

This research aims to find out whether speech therapy can change your speech so that it sounds less as if you are speaking through your nose (nasal), and whether this change lasts over time.

When we speak, we sound 'nasal' if too much air comes through the nose instead of out of the mouth. This happens when the soft part at the back of the roof of the mouth (soft palate) does not work properly. Sometimes this is because the soft palate is too short or does not move well, even though an operation may have already been done for a cleft palate. Sometimes the only treatment for this is another operation to make the palate longer or the muscles work better. But sometimes the problem is that the soft palate only moves well some of the time. In this case speech

therapy might help. However we are not sure who would benefit most from therapy or what the best therapy is.

2. What am I being asked to do?

You are being asked to have speech therapy You will be asked to come to the Speech & Language Therapy department two weeks before the therapy starts to record your speech. You will then have eight therapy sessions (one session each week). Each session will last 45 minutes. In between each session you will be asked to practise at home for 15 minutes each day and to keep a diary of your home practice.

After the last therapy session you will come back for your speech to be recorded again, two months later. The total number of visits is 12.

3) Why have I been asked?

You have been asked because you have been referred to [redacted] with nasal speech.

4) What are the possible disadvantages of taking part?

It might be difficult for you to come to therapy sessions at the cleft centre. I will arrange these at times to suit you if I can. I can also arrange to pay your travel expenses (at public transport rate) for visits to the hospital.

The speech therapy may not make your speech sound less nasal and it may stay the same as at the start of the research.

5) What are the possible benefits of taking part?

The aim of the therapy is to make your speech sound less nasal. The information I get from this research will help to treat other patients with nasal speech in the future.

6) Will my details be kept private during this research?

All information gathered about you will be kept private and any information that leaves the hospital will not have your name and address on it. If you agree, I will let your GP know if you take part in the research.

7) Do I have to take part?

It is up to you to decide whether or not to take part.

If you do decide to take part please keep hold of this information sheet and sign the agreement (consent) form attached. If you take part you can still decide to pull out at any time and without giving a reason.

A decision not to take part or to pull out will not affect any future care you receive.

8) What happens when the research stops?

Your care will continue with [redacted], if you need it. We can talk about this at the last research therapy session.

9) What if something goes wrong?

If you have a concern or a complaint about this study you should contact Susan Rogers, Head of Research & Enterprise Services, at the School of Health Sciences (Address: University of Southampton, Building 67, Highfield, Southampton, SO17 1BJ; Tel: +44 (0)23 8059 7942; Email: S.J.S.Rogers@soton.ac.uk). If you remain unhappy and wish to complain formally Susan Rogers can provide you with details of the

University of Southampton Complaints Procedure.

You can also use the National Health Service complaints procedures

The following contact may be helpful:

NHS Patient Advice and Liaison Service (PALS)

10) What will happen to the results of the research?

The research study will be part of my Doctorate in Clinical Practice at the University of Southampton. I aim to have the results of the research published in the Cleft Palate–Craniofacial medical journal and on the Cleft Lip and Palate Association (CLAPA) website. Your details will not be included in any report or publication.

11)Who is organising and funding the research?

The research is being organised through [redacted]. I will be applying to CLAPA and the hospital Trust Fund for a small amount of money to buy extra therapy equipment and pay travel expenses.

12)Who has checked the research?

The research has been checked by a Research Ethics Committee and the Hospital Research and Development department.

13)Contact for further information

If you need any more information about this research please contact either me or [redacted], Clinical Director, at the address given at the top of this information sheet.

Thank you

Thank you for taking the time to read this information sheet.

If you decide you would like to take part in the research please complete the consent form attached and send it to me in the enclosed stamp addressed envelope.

Best wishes,

Ginette Phippen, Speech and Language Therapist

Hospital Headed Paper

Participant information sheet

Parent Information Sheet [Version 2]

I would like to ask your child to take part in some research about speech therapy for nasal speech. Before you decide, it is important that you and your child understand why the research is being done and what it will involve. Please read this information sheet carefully and discuss it with your child and other people if you wish. Please ask me if there is anything that is not clear or if you need more information. Take your time to decide whether or not you want your child to take part.

1. What is the purpose of the research?

This research aims to find out whether speech therapy can change your child's speech so that it sounds less as if he/she is speaking through their nose (nasal), and whether this change lasts over time.

When we speak, we sound 'nasal' if too much air comes through the nose instead of out of the mouth. This happens when the soft part at the back of the roof of the mouth (soft palate) does not work properly. Sometimes this is because the soft palate is too short or does not move well, even though an operation may have already been done for a cleft palate. Sometimes the only treatment for this is another operation to make the palate longer or the muscles work better. But sometimes the problem is that the soft palate only moves well some of the time. In this case speech

therapy might help. However we are not sure who would benefit most from therapy or what the best therapy is.

2. What is my child being asked to do?

Your child is being asked to have speech therapy, which is practising saying sounds, words and sentences clearly and without sounding nasal. The Speech Therapist will help your child to work out how to make his/her speech sound less nasal using feedback from her and from a computer, so that they can hear and see the difference in how their speech sounds.

He/she will be asked to come to the Speech & Language Therapy department twice weeks before the therapy starts to record their speech. Your child will then have 8 therapy sessions (one session each week). Each session will last 45 minutes. In between each session your child will be asked to practise at home for 15 minutes each day and to keep a diary of your child's home practice.

After the last therapy session your child will come back for their speech to be recorded again, two weeks and then two months later. The total number of visits is 12.

3) Why have I been asked?

Your child has been asked because he/she have been referred to [redacted] with nasal speech.

4) What are the possible disadvantages of taking part?

It might be difficult for you to bring your child to therapy sessions at the cleft centre. I will arrange these at times to suit you if I can. I can also

arrange to pay your travel expenses (at public transport rate) for visits to the hospital.

The speech therapy may not make your child's speech sound less nasal and it may be the same as at the start of the research.

5) What are the possible benefits of taking part?

The aim of the therapy is make your child's speech sound less nasal. The information I get from this research will help to treat other patients with nasal speech in the future.

6) Will my details be kept private during this research?

All information gathered about your child will be kept private and any information that leaves the hospital will not have his/her name and address on it. If you agree, I will let your child's GP know if he/she takes part in the research.

7) Do I have to take part?

It is up to you and your child to decide whether or not to take part.

If you do decide to take part please keep hold of this information sheet and sign the agreement (consent) form attached. If your child takes part he/she can still decide to pull out at any time and without giving a reason.

A decision not to take part or to pull out will not affect any future care your child receives.

8) What happens when the research stops?

Your child's speech therapy care will continue with [redacted], if your child needs it. We can talk about this at the last research therapy session.

9) What if something goes wrong?

If you have a concern or a complaint about this study you should contact Susan Rogers, Head of Research & Enterprise Services, at the School of Health Sciences (Address: University of Southampton, Building 67, Highfield, Southampton, SO17 1BJ; Tel: +44 (0)23 8059 7942; Email: S.J.S.Rogers@soton.ac.uk). If you remain unhappy and wish to complain formally Susan Rogers can provide you with details of the University of Southampton Complaints Procedure.

You can also use the National Health Service complaints procedures

The following contact may be helpful:

NHS Patient Advice and Liaison Service (PALS)

10)What will happen to the results of the research?

The research study will be part of my Doctorate in Clinical Practice at the University of Southampton. I aim to have the results of the research published in the Cleft Palate–Craniofacial medical journal and on the Cleft Lip and Palate Association (CLAPA) website. Your child's details will not be included in any report or publication.

11)Who is organising and funding the research?

The research is being organised through [redacted]. I will be applying to CLAPA and the hospital Trust Fund for a small amount of money to buy extra therapy equipment and pay travel expenses.

12)Who has checked the research?

The research has been checked by a Research Ethics Committee and the Hospital Research and Development department.

13)Contact for further information

If you need any more information about this research please contact either me or [redacted], Clinical Director, at the address given at the top of this information sheet.

Thank you

Thank you for taking the time to read this information sheet.

If you decide that you would like your child to take part in the research please complete the consent form attached and send it to me in the enclosed stamp addressed envelope.

Best wishes,

Ginette Phippen

Speech and Language Therapist

APPENDIX I: Participant details form for recruiters

Study: Behavioural Speech Therapy for Nasal Speech

Chief Investigator: Ginette Phippen, Lead Speech &Language Therapist

Participant details:

Name:

Address:

Telephone number:

NHS number:

Affix label if available

Hospital number:

GP name and address:

Please tick one box:

☐ This individual would like to participate (Consent/Assent forms attached)

☐ This individual would like more time to think about taking part and is happy to be contacted by phone in one weeks time.

SLT signature:_____

Date:_____

APPENDIX J: CONSENT FORMS

CONSENT FORM

Title of Research: Speech therapy for nasal speech

Name of Researcher: Ginette Phippen, Speech and Language Therapist

Please initial box

1	I confirm that I have read and understood the Information Sheet dated March 2009 inviting me to take part in the above research and that I have had the opportunity to ask questions.	
2	I understand that my participation is voluntary and I am free to withdraw at any time, without giving any reason and without my medical care or legal rights being affected.	
3	I agree to audio recordings being made of my speech to be listened to anonymously by a group of four specialist speech and language therapists. If any other use is required, I understand that my agreement will be specifically requested.	
4	I agree to take part in this study.	

_____ _____ _____

Name of Patient Date Signature

_____ _____ _____

Researcher Date Signature

Cc. for patient; researcher; hospital notes

PARENTAL CONSENT FORM

(for children age 7 to 15 years)

Title of Research: Speech therapy for nasal speech

Name of Researcher: Ginette Phippen, Speech and Language Therapist

Please initial box

1	I confirm that I have read and understood the Information Sheet dated December 2008 inviting my child to take part in the above research and that I have had the opportunity to ask questions.	
2	I understand that the participation of my child is voluntary and I am free to withdraw them at any time, without giving any reason and without their medical care or legal rights being affected.	
3	I agree to audio recordings being made of my child's speech to be listened to anonymously by a group of four specialist speech and language therapists. If any other use is required, I understand that my agreement will be specifically requested.	
4	I agree to my child taking part in this study.	

_____ _____

Name of Child Date of birth

_____

Name of Parent Date Signature

_____ _____

Researcher Date Signature

Cc. for patient/parent; researcher; hospital note

302

ASSENT FORM for children age 7 to 15 years

Title of Research: Efficacy of behavioural speech therapy for nasal speech

Name of Researcher: Ginette Phippen, Speech and Language Therapist

Please initial box

1	I confirm that I have read and understood the Information Sheet dated March 2009 which invites me to take part in the above research and that I have been able to ask questions.	
2	I understand that taking part is voluntary and I am free to pull out at any time, without giving any reason and without my medical care or legal rights being affected.	
3	I agree to audio recordings being made of my speech, to be listened to by a group of four speech therapists, without them knowing who I am. If any other use is required, I understand that I will be asked again for my permission to use the recordings.	
4	I agree to take part in this study.	

_____ _____ _____

Name of Child Date Signature

_____ _____ _____

Name of Parent Date Signature

_____ _____ _____

Researcher Date Signature

Cc. for patient/parent; researcher; hospital notes

APPENDIX K: Statistical tests used in analysis

Test	Rationale
Wilcoxon Signed Ranks	**Non-parametric test to evaluate differences between paired data**
Mann Whitney U	**Non-parametric test to evaluate differences between unpaired data**
Chi Square	**Test of association**
Kruskal Wallis	**Non-parametric test to evaluate differences between groups**
Spearmans Rho	**Test to evaluate degree of association between variables**

Appendix L: Additional results & analysis

Participants

Table L-1: Nasal speech symptoms observed in each participant prior to recruitment

ID	Hypernasality	Audible nasal emission	Nasal turbulence
*A	✓	✓	x
*B	✓	✓	x
C	✓	✓	x
D	✓	x	x
*E	✓	✓	✓
*F	✓	✓	x
*G	✓	x	x
*H	✓	x	✓
I	✓	x	x
J	✓	x	x
K	✓	✓	x
L	✓	x	x

*Active nasal fricative (ANF) as inconsistent feature noted pre-recruitment

Perceptual results

Table L-2: Group results (%) of CAPS-A ratings for pre & post assessment points

	A1	A2*	A3**	A4
CAPS-A rating	**Pre-intervention**	**Pre-intervention**	**Post-intervention**	**Post-intervention**
HYPER				
0 absent	33% (4)	17% (2)	25% (3)	33% (4)
1 borderline	17% (2)	0%	25% (3)	58% (7)
2 mild	33% (4)	25% (3)	25% (3)	8% (1)
3 moderate	17% (2)	25% (3)	0%	0%
ANE				
0 absent	75% (9)	67% (8)	75% (9)	92% (11)
1 occasional	25% (3)	0	0	8% (1)
2 frequent	0%	0	0	0%
NT				
0 absent	67% (8)	42 % (5)	50 % (6)	58% (7)
1 occasional	33% (4)	8% (1)	17% (2)	25% (3)
2 frequent	0%	17% (2)	8% (1)	17% (2)

* missing data for 4 participants ** missing data for 3 participants

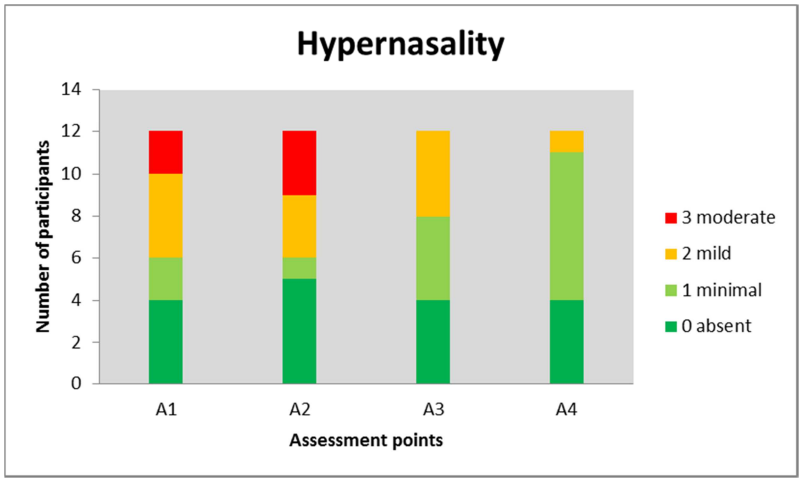


Figure L-1: Group results of CAPS-A hypernasality ratings

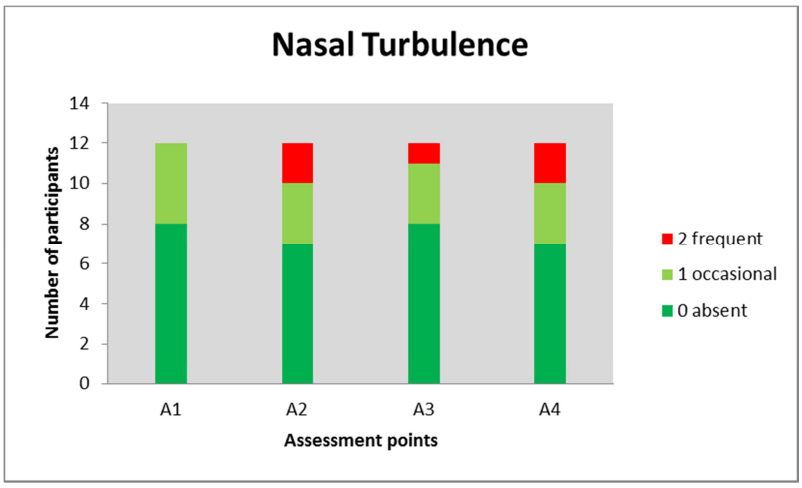


Figure L-2: Group results of CAPS-A nasal turbulence ratings

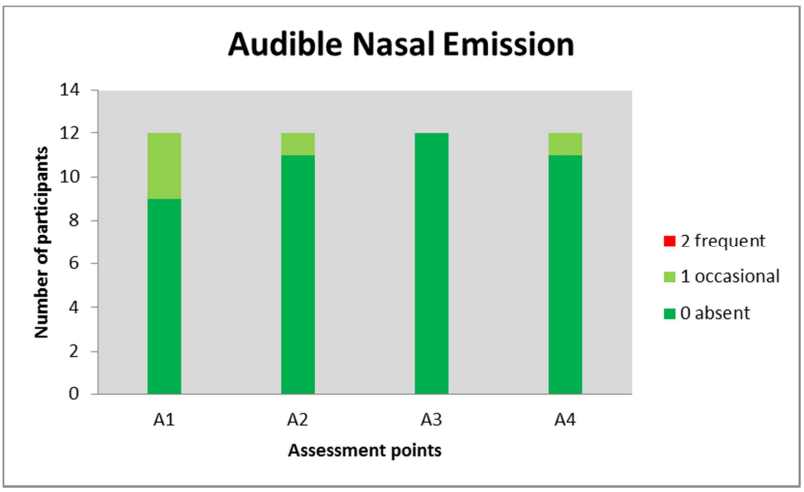


Figure L-3: Group results of CAPS-A audible nasal emission ratings

Table L-3: Difference between pre-intervention CAPS-A ratings (A1 and A2)

Test statistic	HYPER A2 minus HYPER A1	ANE A2 minus ANE A1	NT A2 minus NT A1
Z	.000	-1.414	-1.134
Asymp. Sig. (2-tailed)	1.000*	.157*	.257*

*p =>0.05

Table L-4: Analysis of differences in CAPS-A hypernasality ratings between pre and post intervention assessment points

Test statistic	HYPER A3 minus HYPER A1+A2	**HYPER A4 minus HYPER A1+A2**	HYPER A4 minus HYPER A3
Z	-1.382	**-1.750**	-1.000
†P-value	0.085	**0.040**	0.317*

†Wilcoxon Signed Ranks p=<0.05 * Two sided test

Table L-5: Analysis of differences in CAPS-A audible nasal emission ratings between pre and post intervention assessment points

Test statistic	ANE A3 minus ANE A1+A2	ANE A4 minus ANE A1+A2	ANE A4 minus ANE A3
Z	-1.342	-.557	-1.000
†P-value	0.090	0.258	0.317*

†Wilcoxon Signed Ranks p=<0.05

* Two sided test

Table L-6: Analysis of differences in CAPS-A nasal turbulence ratings between pre and post intervention assessment points

Test statistic	NT A3 minus NTA1+A2	NT A4 minus NT A1+A2	NT A4 minus NT A3
Z	.000	-.427	.000
†P-value	0.50	0.334	1.000*

†Wilcoxon Signed Ranks p=<0.05

* Two sided test

Instrumental results

Table L-7: Difference between pre-intervention nasometry scores (A1 & A2)

Test statistic	Nasometry A2 minus A1
Z	-.461[a]
Asymp. Sig. (2-tailed)	.645

Table L-8: Analysis of differences in nasometry scores between assessment points

Test statistic	A3 minus A1 +A2	A4 minus A1 +A2	A4 minus A3
Z	-.593	-1.452	-.140
†P-value	.271	.073	.888*

†Wilcoxon Signed Ranks

* two sided test

Instrumental (nasometry) results: comparison to norms

In Figure L-4 the black lines indicate the published literature for the cut-off range for nasal speech. This is the range of nasometry scores that would be considered 'normal' in an English speaking population, i.e. between 26% and 32% (Sweeney 2011). Above

the upper black line (>32%) would be considered abnormal and would be characterised by increased nasality (hypernasality). Below the lower black line (<26%) would also be abnormal and would be characterised by reduced nasality (hyponasality).

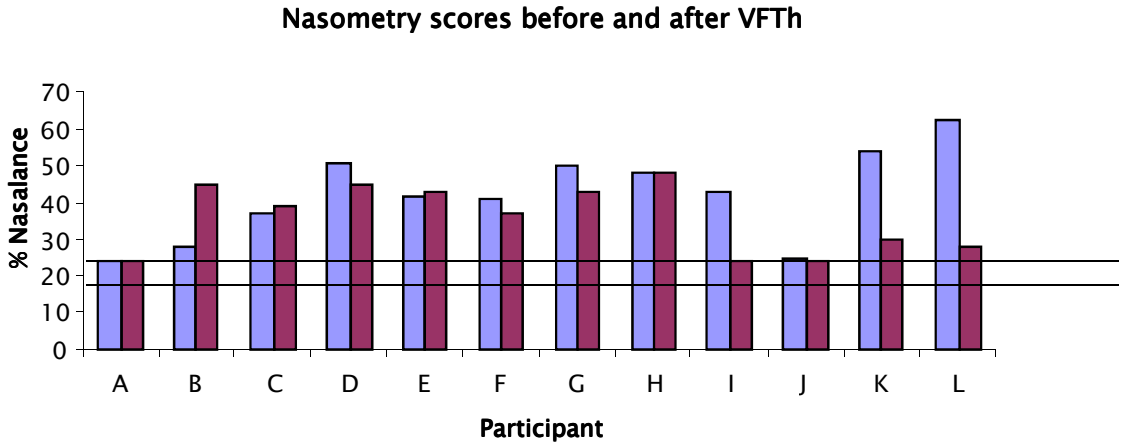


Figure L-4: Individual nasometry scores pre and post-intervention with norms comparison

Table L-9: Group mean nasalance scores across assessment points (%)

Assessment	N	Mean	Std. Deviation
A1	12	42.1	11.9
A2	12	41.2	10.4
A3	9	37.8	11.8
A4	12	35.8	9.3

Quality of Life results

Table L-10: Analysis of differences in PedSalQoL scores pre and post-intervention

Test statistic	SATIS A4 minus A1	**COMM & FEEL A4 minusA1**	IND & PAR A4 minus A1	SUPP SCH A4 minus A1	**TOTAL SCORE A4 minusA1**
Z	-1.024	**-2.274**	-.939	-.140	**-1.958**
†P-value	0.153	**0.012**	0.175	0.444	**0.025**

†Wilcoxon Signed Ranks

Table L-11: Analysis of differences in PedsQL4.0 scores pre and post-intervention

Test statistic	Hact A4 minus A1	Feel A4 minus A1	Geton A4 minus A1	Sch A4 minus A1	Phys A4 minus A1	Total A4 minus A1
Z	-.135	-.105	-1.289	-1.429	-.059	-.652
†P-value	.448	.451	.100	.076	.475	.260

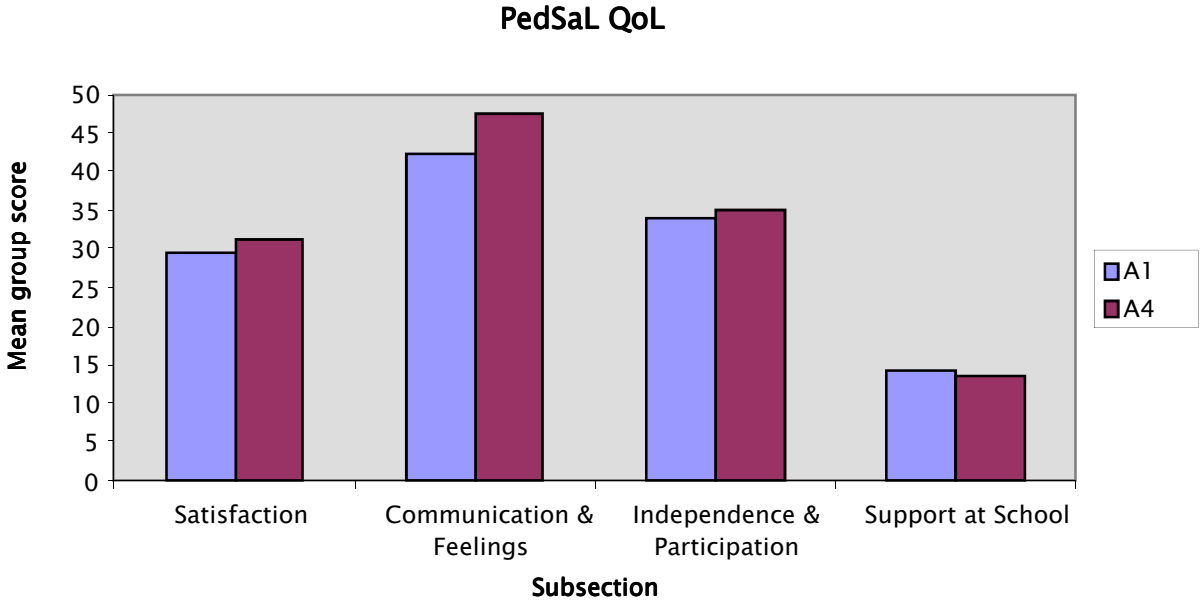
†Wilcoxon Signed Ranks

PedSaLQoL: group results

Table L-12: Summary of PedSaLQoL group mean scores pre and post-intervention (*n*=11)

PedSaL QoL	A1		A4	
Subsection	Mean	Std Dev	Mean	Std Dev
Satisfaction	29.5	4.0	31.1	3.2
Communication & Feelings	42.3	8.6	47.5	6.1
Independence & Participation	33.8	5.5	34.9	5.6
Support at school	14.2	4.5	13.6	4.7
Total score	119.7	15.9	127.2	13.6

A1 pre intervention, A4 post intervention



Figures L-5 & L-6: PedSaL QoL mean group subsection & Total score pre and post-intervention

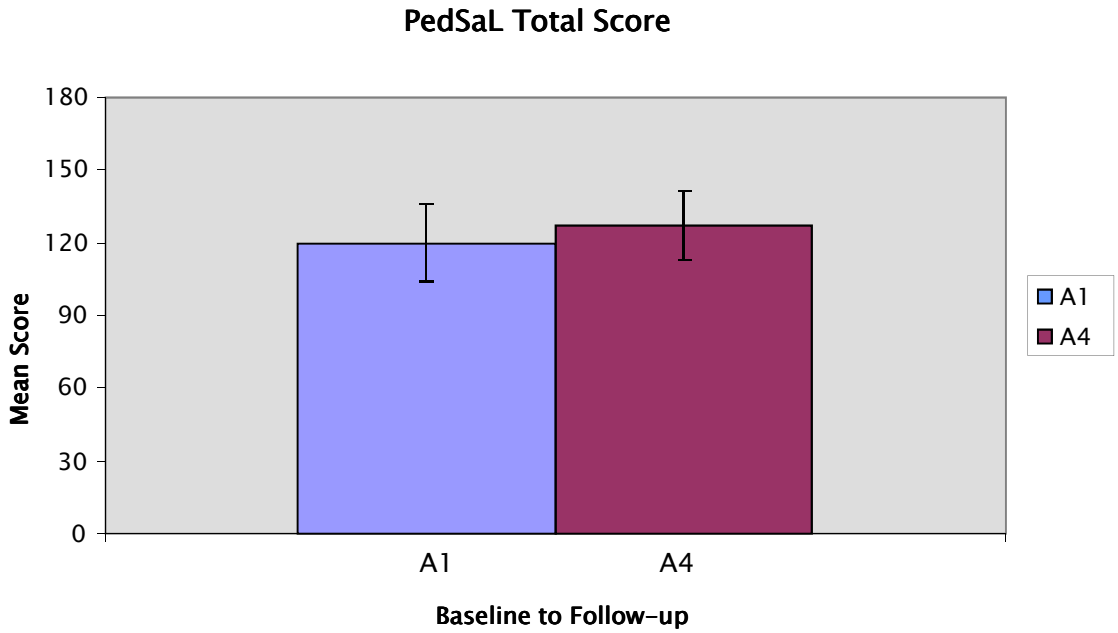
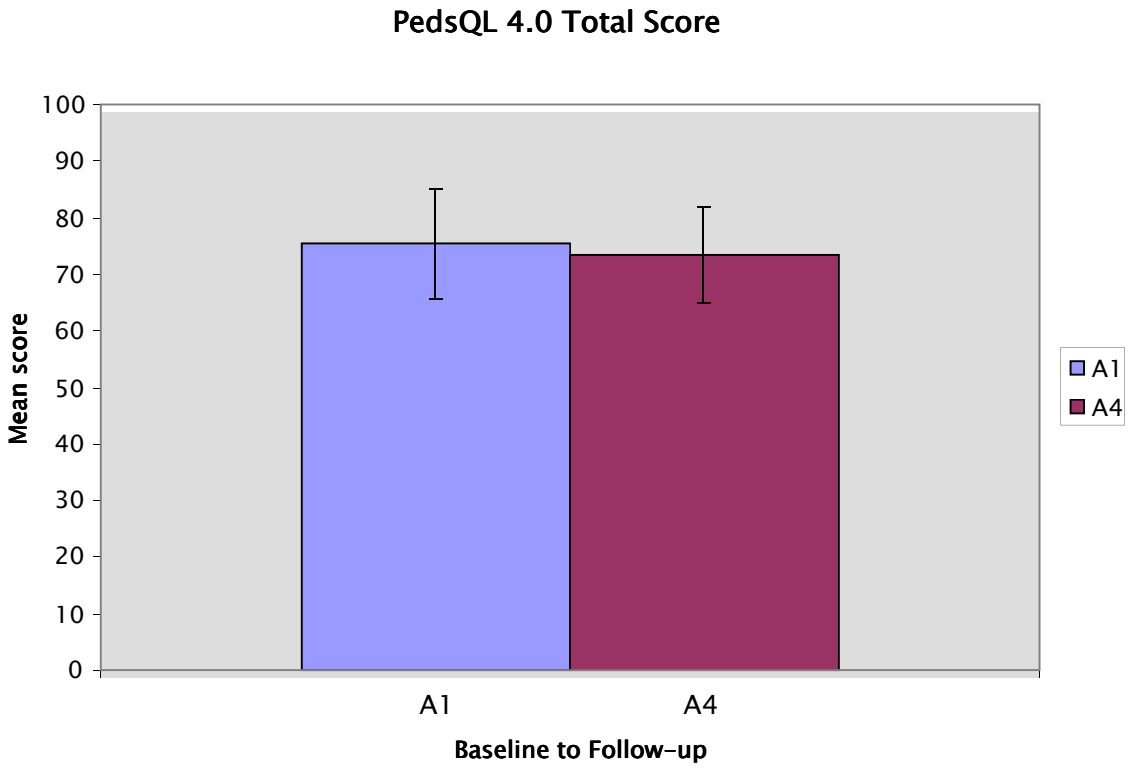
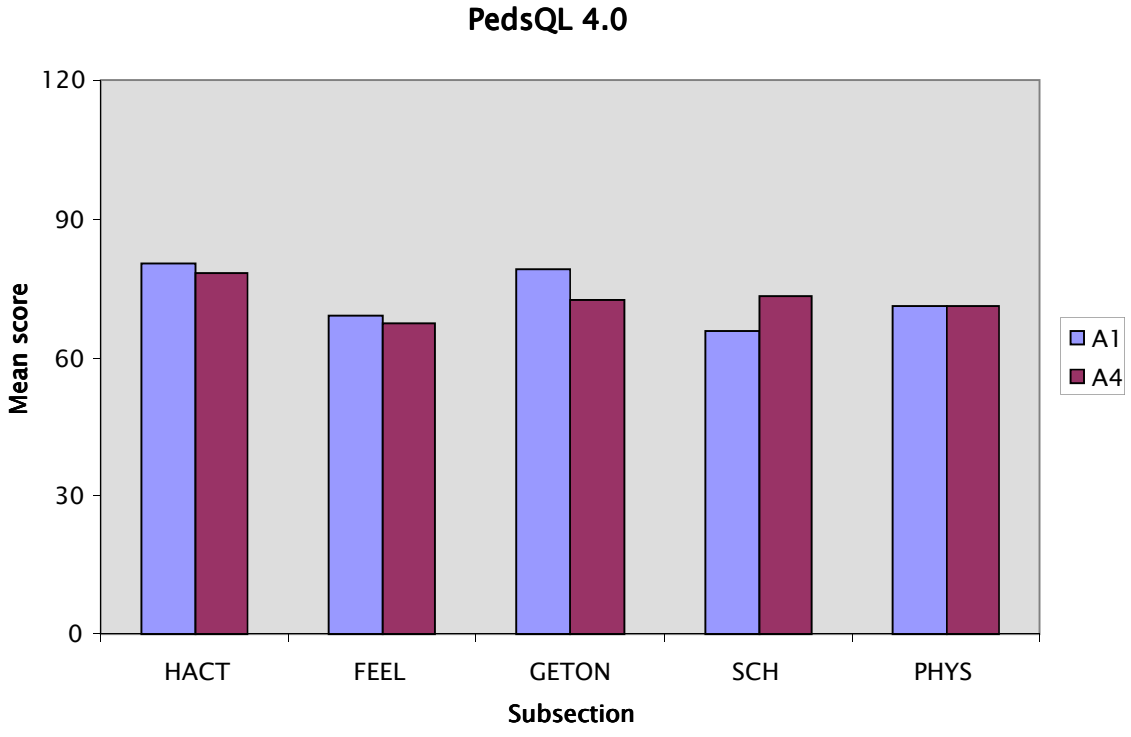


Table L-13: Summary of PedsQL4.0 group mean scores pre and post-intervention (*n*=9)

PedsQL 4.0	A1		A4	
Subsection	**Mean**	**Std Dev**	**Mean**	**Std Dev**
Health & Activities (Hact)	80.2	10.5	78.1	11.6
Feelings (Feel)	68.9	13.9	67.2	6.2
Get on with others (Geton)	78.9	12.9	72.2	20.1
School (Sch)	65.6	14	73.3	14.6
Physical (Phys)	71.1	8.7	70.9	8.2
Total score	75.3	9.8	73.4	8.6





Figures L-7 & L-8: PedsQL 4.0 mean group subsection score and Total score pre and post-intervention

List of references

Adams, C., Lockton, E., Freed, J., Gaile, J., Earl, G., McBean, K., Nash, M., Green, J., Vail, A. and Law, J. (2012), The Social Communication Intervention Project: a randomized controlled trial of the effectiveness of speech and language therapy for school-age children who have pragmatic and social communication problems with or without autism spectrum disorder. *International Journal of Language & Communication Disorders*, 47: 233–244

Adams, S.G. and Page, A.D. (2000), Effects of selected practice and feedback variables on speech motor learning. *Journal of Medical Speech-Language Pathology,* 8: 215–220.

Adult Communication Coalition England (ACCE) (2009), Available at http://www.rcslt.org/news/docs. [Accessed 08 August 2012].

Albery, E. and Russell, J., 1990, Cleft palate and orofacial abnormalities. In P. Grunwell (Ed.) *Developmental Speech Disorders* (London: Whurr), Chapter 4, pp. 63–82.

Albery, E. and Russell, J. (2005), *Practical Intervention for Cleft Palate Speech*. Milton Keynes: Speechmark Pub Ltd.

Allen, K.D., Bernstein, B. and Chait, D.H. (1991), EMG biofeedback treatment of pediatric hyperfunctional dysphonia. *Journal of behavior therapy and experimental psychiatry*, 22(2): 97–101.

Altman, D.G. (1991), *Practical Statistics for Medical Research.* London: Chapman & Hall. 403–409.

Altmann, G.T.M. (2001), The language machine: Psycholinguistics in review. *British Journal of Psychology,* 92: 129–170.

American Speech & Hearing Association (ASHA) (2010) Motor Speech Disorders Evaluation. [online] Available at http://www.asha.org/uploadedFiles/slp/healthcare/AATMotorSpeech.pdf [Accessed 11 October 2010].

American Speech & Hearing Association (ASHA) (2008) Available at http://www.ASHA.org/research/reports/children. [Accessed 08 August 2012].

Annett, J. (1969), *Feedback and human behaviour: The effects of knowledge of results, incentives and reinforcement on learning and performance.* Oxford, England: Penguin Books.

Arain, M., Campbell, M. J., Cooper, C.L. and Lancaster G.A. (2010), What is a pilot or feasibility study? : A review of current practice and editorial policy. *BMC Medical Research Methodology* ,10:67. Available at http://www.biomedcentral.com/1471-2288/10/67. [Accessed 12 May 2012].

Arvedson, J., Clark, H., lazarus, C., Schooling, T. and Frymark, T, (2010), The effects of oral-motor exercises on swallowing in children: an evidence-based systematic review. *Developmental Medicine & Child Neurology,* 52(11): 1000-1013.

Atherton, J.S. (2010), *Learning and Teaching; Piaget's developmental theory*. [online] Available at: http://www.learningandteaching.info/learning/piaget.htm#Key Accessed 04 October 2010.

Bacsfalvi, P., Bernhardt, B.M. and Gick, B. (2007), Electropalatography and ultrasound in vowel remediation for adolescents with hearing impairment. *International Journal of Speech-Language Pathology*, 9(1): 36-45.

Baker, D.F. and Buckley, M.R. (1996), A historical perspective of the impact of feedback on behaviour, *Journal of Management History* (Archive), 2(4): 21-33.

Baker, L. and Cantwell, D. (1987), A Prospective Psychiatric Follow-up of Children with Speech/Language Disorders. *Journal of the American Academy of Child and Adolescent Psychiatry,* 26(4): 546-553.

Barr, L., Thibeault, S.L., Muntz, H. and de Serres, L. (2007), Quality of Life in Children With Velopharyngeal Insufficiency. *Archives of Otolaryngology- Head & Neck Surgery,* 133(3): 224-229.

Bercow, J. (2008), *The Bercow Report: A Review of Services for Children andYoung People (0-19) with Speech, Language and Communication Needs*. Department of Child & Family Services: London.

Bernhardt, B., Gick, B., Bacsfalvi, P. and Adler-Bockl, M. (2005), Ultrasound in speech therapy with adolescents and adults. *Clinical Linguistics & Phonetics,* 19(6-7): 605-17.

Bessell, A., Sell, D., Whiting, P., Roulstone, S., Albery, L., Persson, M., Verhoeven, A., Burke, M., Ness, A.R. (2013), Speech and Language Therapy Interventions for Children With Cleft Palate: A Systematic Review. *The Cleft Palate-Craniofacial Journal*: January 2013, Vol. 50, No. 1, pp. e1-e17.

Bhuta. T., Patrick L. and Garnett J.D. (2004), Perceptual evaluation of voice quality and its correlation with acoustic measurements. *Journal of Voice,* September, 18(33): 299-304.

Bivati, M.J. (2002), Velopharyngeal Insufficiency. Available at: http://www.emedicine.medscape.com/article/873018. Accessed on 19 Sept 2005.

Blood, G.W. (1994), Efficacy of a computer-assisted voice treatment protocol. *American Journal of Speech-Language Pathology*, 3(1): 57.

Borden, G.J. (1980), Use of feedback in established and developing speech IN: *Speech and language: advances in basic research and practice*. Volume 3. Academic Press 223-242.

Boseley, M. E., & Hartnick, C. J. (2004). Assessing the outcome of surgery to correct velopharyngeal insufficiency with the pediatric voice outcomes survey. *International journal of pediatric otorhinolaryngology*, *68*(11), 1429-1433.

Bowen, C. (1998), *Children's speech sound disorders: Questions and answers.* [online] Available at: http://www.speech-language-therapy.com/phonol-and-artic.htm [Accessed 11 September 2010].

Bowen, C. (2005), What is the evidence for.....? Oral motor therapy. *ACQuiring Knowledge in speech, Language and Hearing* 7(1): 144-147.

Bowen, C. (2009), *Children's Speech Sound Disorders.* Wiley-Blackwell

Bowling, A. (2002), *Research methods in health: Investigating health and health services*. Berkshire: Open University Press

Bray, M., Ross, A. and Todd, C. (1999), *Speech and Language Clinical Process and Practice.* London: Whurr Publishers.

Bradbury, E. (2001), Growing up with a Cleft: the Impact on the Child. IN: Watson ACH, Sell DA and Grunwell P (eds). *Management of Cleft Lip and Palate*. London: Whurr Publishers.

Bressman, T., Klaiman, P. and Fischbach, S. (2006), Same noses, different nasalance scores: data from normal subjects and cleft palate speakers for three systems for nasalance analysis. *Clinical Linguistics & Phonetic,* 20 (2-3):163-170.

Brunner, M., Stellzig-Eisenhauer, A., Proschel, U., Verres, R. and Komposch, G. (2005), The effect of nasopharyngoscopic biofeedback in patients with cleft palate and velopharyngeal dysfunction. *Cleft Palate -Craniofacial Journal,* 42(6): 649-57.

Bunton, K. (2008), Speech versus Nonspeech: Different tasks, Different Neural Organization. *Seminars in Speech and Language,* 29(4) 267-275.

Burzynski, C.M. and Starr, C.D. (1985), Effects of feedback filtering on nasalization and self perception of nasality. *Journal of Speech & Hearing Research,* 28: 585-588.

Bzoch, K.R. (2004), Introduction to the study of communicative disorders in cleft palate and related craniofacial anomalies IN: K.R. Bzoch (ed.) *Communication Disorders related to Cleft Lip and Palate*. Austin, Texas: Pro-Ed.

Cahill, L.M., Turner, A.B., Stabler, P.A., Addis, P.E., Theodoros, D.E. and Murdoch, B.E. (2004), An evaluation of continuous positive airway pressure (CPAP) therapy in the treatment of hypernasality following traumatic brain injury: a report of 3 cases. *Journal of Head Trauma Rehabilitation,* 19(3): 241-53.

Campbell, M., Fitzpatrick, R., Haines, A., Kinmonth, A. L., Sandercock, P., Spiegelhalter, D., & Tyrer, P. (2000). Framework for design and evaluation of complex interventions to improve health. *BMJ: British Medical Journal*, *321*(7262), 694.

Canadian Interior Health Evidence hierachy, (2006) Available at: http://phprimer.afmc.ca [Accessed on 13 January 2012]

Carding, P.N., Horsley, I.A. and Docherty, G.J. (1998), The effectiveness of voice therapy for patients with non-organic dysphonia. *Clinical Otolaryngology & Allied Sciences*, 23(4): 310-318.

Carding, P.N. (2000), *Evaluating Voice Therapy*. London: Whurr Publishers.

Carding, P. and Hillman, R. (2001), More randomised controlled studies in speech and language therapy: complex behavioural interventions can be evaluated. *British Medical Journal,* 22:323 (7314): 645-646

Carey, T.S. and Boden, S.D. (2003), A critical guide to case series reports. *Spine,* 28(15):1631-1634.

Carpenter, J. R., & Kenward, M. G. (2008), Missing data in clinical trials—a practical guide. *National Institute for Health Research, Publication RM03/JH17/MK: Birmingham*.

Carter, P., & Edwards, S. (2004). EPG therapy for children with long-standing speech disorders: predictions and outcomes. *Clinical linguistics & phonetics*, 18(6-8), 359-372.

Caruso, A.J. and Strand, E.A. (1999), *Clinical management of motor speech disorders in children.* New York: Thieme.

Carver, C.S., Sheier, M., Higgins, T.E. and Sorrentino, R.M. (1990), Principles of self regulation: Action and emotion IN: *Handbook of motivation and cognition: Foundations of social behaviour,* (Vol 2) 3-52, NY, US: Guilford Press.

Casserly, E.D. and Pisoni, D.B. (2010), Speech Perception and Production. *WIREs: Cognitive Science,* 1(5): 623-773.

Cavalli, L. (2011), Voice assessment and Intervention IN: Howard S & Lohmander A (eds.) *Cleft Palate Speech: Assessment and Intervention*. Chichester, UK: Wiley-Blackwell, 181-195.

Chapey, R. (1994), Cognitive Intervention. IN: Chapey R (ed.) *Language intervention strategies in adult aphasia*. Baltimore: Williams & Wilkins, 220-245.

Chapman, K. L., Hardin-Jones, M. A., Goldstein, J. A., Halter, K. A., Havlik, R. J., & Schulte, J. (2008). Timing of palatal surgery and speech outcome. *The Cleft Palate-Craniofacial Journal*, *45*(3), 297-308.

Chen, M.Y. (1997), Acoustic correlates of English and French nasalized vowels. *Journal of Acoustical Society of America*, 102: 2360-2370.

Chernobelsky, S. (2002), The use of electroglottography in the treatment of deaf adolescents with puberphonia. *Logopedics Phonatrics Vocology*, 27(2): 63-65.

Cleft Lip and Palate Association (CLAPA) (2009), *Understanding Cleft Lip and Palate* .[online] Available at: http://www.clapa.com/medical/cleft_lip_article/107/. [Accessed 16 August 2010].

Clark, H.M. (2003), Neuromuscular treatments for speech and swallowing: a tutorial. *American Journal of Speech and Language Pathology,* 12(4): 400-15.

Cohen, A. and Gureje, O. (2007), Making sense of evidence. *International Review of Psychiatry,* 19: 583-91.

Cole, R.M. (1979), Direct muscle training for improvement of velopharyngeal activity. IN: Bzoch K (ed.) (2nd edition) *Communicative Disorders Related to Cleft Lip and Palate.* Texas: Pro-Ed.

Conrad, P. (2001). Health research, qualitative. *International encyclopedia of the social & behavioral sciences*, *10*, 6608-12

Consent: a guide for children and young people (2001) Department of Health publication 24473, Crown Copyright.

CONSORT checklist (2010) available at: www.consort-statement.org [Accessed on 18 November 2011].

Cooper, J., Moodley, M. and Reynell, J. (1978), *Helping language development: A developmental program for children with early language handicaps*. New York: St. Martin's Press.

CRANE. CRANE Database Annual Report (2009). London: Clinical Effectiveness Unit, The Royal College of Surgeons of England.

Cremeens, J., Eiser, C. and Blades, M. (2006), Factors influencing agreement between child self-report and parent proxy-reports on the Pediatric Quality of Life Inventory™ 4.0 (PedsQLTM) generic core scales. *Health and Quality of Life Outcomes* 4(58): [online] Available at: http://www.halo.com/content/4/1/58. [Accessed 15 December 2011].

Cruice, M. (2008), The contribution and impact of the International Classification of Functioning, Disability and Health on quality of life in communication disorders. *International Journal of Speech-Language Pathology* , 10 (1-2): 38-49.

Daly, D.A. and Johnson, H.P. (1974), Instrumental modification of hypernasal voice quality in retarded children: case reports. *Journal of Speech and Hearing Disorders,* 39(4): 500-507.

Dalston, R.D., Warren, D.W. and Dalston, E.T. (1991), Use of nasometry as a diagnostic tool for identifying patients with velopharyngeal impairment. *The Cleft Palate-Craniofacial Journal*, 28(2): 184-189.

Damico, J.S., Simmons-Mackie, N.N. and Schweitzer, L.A. (1995), Addressing the Third Law of Gardening: Methodological Alternatives in Aphasiology. *Clinical Aphasiology*, 23: 83-93.

Deane, K., Whurr, R., Playford, E.D., Ben-Shlomo, Y. and Clarke, C.E. (2009), Speech and language therapy for dysarthria in Parkinson's disease: a comparison of techniques (Review). *The Cochrane Library,* 2009 (1).

Deary, I.J., Wilson, J.A., Carding, P.N. and MacKenzie, K. (2003), VoiSS: a patient derived voice symptom scale. *Journal of Psychosomatic Research,* 54(5): 483-489.

DeSouza, C. M., Legedza, A. T., & Sankoh, A. J. (2009), An overview of practical approaches for handling missing data in clinical trials. *Journal of Biopharmaceutical Statistics*, 19(6), 1055-1073.

Dey, I. (1993). *Qualitative data analysis: A user-friendly guide for social scientists*. Routledge.

Dixon-Wood, V.L. (2004), Correcting Retracted Articulation Error Patterns: Recent Techniques. IN: Bzoch KR (ed.) *Communicative Disorders Related to Cleft Lip and Palate*. (5th edition). Austin, Texas: Pro-Ed.

Dromey, C., Ramig, L.O. and Johnson, A.B. (1995), Phonatory and Articulatory Changes Associated With Increased Vocal Intensity in Parkinson Disease: A Case Study *Journal of Speech & Hearing Research,* 38: 751-764.

Dworkin, J.P., Marunick, M.T. and Krouse, J.H. (2004), Velopharyngeal Dysfunction: Speech Characteristics, variable Etiologies, Evaluation techniques, and Differential Treatments. *Language, Speech & Hearing Services in Schools,* 35(4): 333-352.

Earles, J., Kerr, B. and Kellar, M. (2003), Psychophysiologic treatment of vocal cord dysfunction. *Annals of Allergy Asthma & Immunology,* 90(6): 669-71.

El Dib, R.P. and Atallah, A.N. (2006), Evidence-based speech, language and hearing therapy and the Cochrane Library's systematic reviews. *Sao Paulo Medical Journal,* 124(2): 51-4.

Estrem, T. & Broen, P. (1989), Early speech production of children with cleft palate. *Journal of Speech, Language, and Hearing Research* Vol 32, pp.12-23.

Evans, L. (2002), *Reflective practice in educational research: Developing advanced skills*. Continuum International Publishing Group.

Evarts, E. V. (1971). Central control of movement. V. Feedback and corollary discharge: a merging of the concepts. Neurosciences Research Program Bulletin, 9(1), 86.

Fang, T.J., Li, H.Y., Gliklich, R.E., Chen, Y.H., Wang, P.C. and Chuang, H.F. (2008), Quality of Life Measures and Predictors for Adults With Unilateral Vocal Cord Paralysis. *The Laryngoscope*, 118: 1837-1841

Fant, G. (1981), The Source Filter concept in voice production. *STL-QPSR* 22(1): 22-37

Fathalla, M.F. & Fathalla, M.M.F. (2004), *A practical guide for health researchers*. WHO Regional Publications, Eastern Mediterranean series; 30.

Fisher, H.R. (2004), Preliminary Studies on Efficacy of Prolonged Nasal Cul-De-Sac with High Pressure Speech Acts (P.i.N.C.H.) on Hypernasality. *The Internet Journal of Allied Health Sciences and Practice,* January 2(1).

Fleck, S.J. and Kraemer, W.J. (2004), *Designing resistance training programs*. Human Kinetics Publishers.

Fleiss, J. L., Levin, B., & Paik, M. C. (2004), The measurement of interrater agreement. *Statistical Methods for Rates and Proportions, Third Edition*, 598-626.

Fletcher, S.G. (1972), Contingencies for Bioelectronic Modification of Nasality*. Journal of Speech & Hearing Disorders,* 37: 329-346.

Fletcher, S.G. and Higgins, J.M. (1980), Performance of Children with Severe to Profound Auditory Impairment in Instrumentally Guided Reduction of Nasal Resonance*. Journal of Speech & Hearing Disorder*s, 45: 181-194.

Frattali, C. (1998), *Measuring outcomes in speech-language pathology*. New York: Theime.

Frison, L., & Pocock, S. J. (2007). Repeated measures in clinical trials: analysis using mean summary statistics and its implications for design. *Statistics in medicine*, *11*(13), 1685-1704.

Fujiki, M., Brinton, B., Isaacson, T. and Summers, C. (2001), Social Behaviors of Children With Language Impairment on the Playground: A Pilot Study. *Language Speech & Hearing Services in Schools,* 32(2): 101-113.

Fujiwara, Y. (2007), Electropalatography home training using a portable training unit for Japanese children with cleft palate. *International Journal of Speech-Language Pathology*, 9(1): 65-72.

Garber, S.R. and Moller, K.T. (1979), The Effects of Feedback Filtering on Nasalization in Normal and Hypernasal Speakers*. Journal of Speech & Hearing Research*, 22(2): 321-333.

Garrett, Z. and Thomas, J. (2006), Systematic reviews and their application to research in speech and language therapy: a response to T.R. Pring's 'Ask a silly question:

two decades of troublesome trials.' (2004). *International Journal of Language and Communication Disorders,* 41: 95-105.

Gibbon, F. E., McNeill, A.M., Woods, S. and Watson, J.M. (2003a), Changes in linguapalatal contact patterns during therapy for velar fronting in a 10-year-old with Down's syndrome. *International Journal of Language & Communication Disorders,* 38(1): 47-64.

Gibbon, F.E. and Wood, S.E. (2003b), Using electropalatography (EPG) to diagnose and treat articulation disorders associated with mild cerebral palsy: a case study. *Clinical linguistics & phonetics*, 17(4-5): 365-374.

Glassman, W.E. and Hadad, M. (2008), *Approaches to psychology.* Fifth edition. : Maidenhead, UK Open University Press/McGraw-Hill Education.

Golding-Kushner, K.J. (2001), *Therapy techniques for cleft palate and related disorders*. San Diego: Singular.

Golding-Kushner, K.J. (2009) IN: Bowen C (ed.). *Children's speech sound disorders*. Wiley. Q13: 85

Goldrick, M. (2007), Connectionist principles in theories of speech production. IN: M.G. Gaskell (Ed.) The Oxford handbook of psycholinguistics. Oxford: Oxford University Press. pp. 515-530.

Greene, M.C.L. and Mathieson, L. (1989), *The Voice and its Disorders*. London: Whurr Publishers.

Grimes, D.A., & Schulz, K.F., (2002). Bias and causal relationships in observational research, The Lancet, Vol 359 (19), pp. 248-252.

Grunwell, P., 1993. *Assessing Cleft Palate Speech*, London, Whurr Publishers.

Guideline for Good Clinical Practice E6 (R1) (1996) The international conference on harmonisation of technical requirements for registration of pharmaceuticals for human use (ICH) (1996) Available from http://www.ich.org/LOB/media/MEDIA482.pdf Accessed 08 October 2010.

Gulian, E., Fallside, F., Hinds, P. and Keiller, C. (1983), Acquisition of frication by severely hearing-impaired children. *British Journal of Audiology*, 17(4): 219-231.

Hagg, M. and Larsson, B. (2004), Effects of motor and sensory stimulation in stroke patients with long-lasting dysphagia. *Dysphagia*, 19(4): 219-30.

Hall, R. (1998), *Within-Subjects Designs*. Available at: http://web.mst.edu/psyworld/within_subjects.htm. [Accessed 14 February 2012]

Hamer, R. M. and Simpson, P. M. (2009), Last Observation Carried Forward Versus Mixed Models in the Analysis of Psychiatric Clinical Trials *American Journal of Psychiatry, 1*66:639-641.

Harding, A. and Grunwell, P. (1996), Characteristics of cleft palate speech. *European Journal of Disorders of Communication,* 31(4): 331-57.

Harland, K. (1996) *An extended audit of complete bony unilateral cleft lip and palate patients aged 5-6 and 8-11: speech outcomes*. Presented at the annual meeting of the Craniofacial Society of Great Britain and Ireland; 1996; Egham, Surrey, U.K.

Hartley, J. (1998), *Learning and Studying: A research perspective*. London: Routledge.

Havstaam, C. and Lohmander, A. (2011), Communicative Participation. IN: *Cleft Palate Speech: assessment and intervention*, Howard S & Lohmander A (eds.) 305-312.

Hayhow, R., Cray, A.M. and Enderby. P., (2002), Stammering and therapy views of people who stammer*. Journal of Fluency Disorders,* 27(1): 1-17.

Hayhow, R. (2011), Does it work? Why does it work? Reconciling difficult questions. *Journal of Language & Communication Disorders,* 46(2): 155-168.

Henriksson, T.G., Hakelius, M., Andlin-Sobocka, A., Svanholm, H., Low, A. and Skoog, V. (2005), Intravelar veloplasty reinforced with palatopharyngeal muscle: a review of a 10-year consecutive series. *Scandinavian journal of plastic and reconstructive surgery and hand surgery*, 39(5): 277-282.

Hogikyan, N.D. and Sethuraman, G. (1999), Validation of an instrument to measure voice-related quality of life (V-RQOL). *Journal of Voice*, 13(4): 557-569.

Holmberg, E.B., Hillman, R.E., Hammarberg, B., Sodersten, M. and Doyle, P. (2001), Efficacy of a behaviorally based voice therapy protocol for vocal nodules. *Journal of Voice,* 15(3): 395-412.

Hombert, J.M. (1986), The development of nasalized vowels in the Teke language group (Bantu). *The phonological representation of suprasegmentals. Dordrecht: Foris*: 359-379.

Hooper, C.R. (2004), Treatment of voice disorders in children. *Language Speech & Hearing Services in Schools,* 35(4): 320-6.

Horneman, G., Folkesson, P., Sintonen, H., von Wendt, L. and Emanuelson, I. (2005), Health-related quality of life of adolescents and young adults 10 years after serious traumatic brain injury. *International Journal of Rehabilitation Research*, 28(3): 245-249.

Horton, S. (2006), A framework for description and analysis of therapy for language impairment in aphasia. *Aphasiology,* 20(6): 528-564.

Huang, H., Wolf, S.L. and He, J. (2006), Recent developments in biofeedback for neuromotor rehabilitation. *Journal of Neuroengineering Rehabilitation,* 3:11.

I CAN, Talk (2006) Speech, Language and Communication Needs and Literacy Difficulties Issue 1; available at: www.ican.org.uk [Accessed on 6 November 2011].

Jacobson, B.H., Johnson, A., Grywalski, C., Silbergleit, A., Jacobson, G., Benninger, M.S. and Newman, C.W. (1997), The voice handicap index (VHI): development and validation. *American Journal of Speech-Language Pathology*, 6(3): 66.

John, A., Sell, D., Sweeney, T., Harding-Bell, A. and Williams, A. (2006), The cleft audit protocol for speech-augmented: A validated and reliable measure for auditing cleft speech. *Cleft Palate Craniofacial Journal,* 43(3): 272-88.

Johns, D.F., Rohrich, R.J. and Awada, M. (2003), Velopharyngeal incompetence: a guide for clinical evaluation. *Plastic & Reconstructive Surgery,* 112(7): 1890-7.

Jones, D.L., Morris, H.L. and Vandemark, D.R. (2004), A Comparison of Oral-Nasal Balance Patterns in Speakers Who Are Categorized as Almost but Not Quite and Sometimes but Not Always. *The Cleft Palate-Craniofacial Journal* ,41(5): 526-534.

Jones, D.L. (2006), Patterns of Oral-nasal Balance in Normal Speakers with and without Cleft Palate. *Folia Phoniatrica Logopaedica,* 58: 383-391.

Kamhi, A.G. (1989), Language Disorders in Children. IN: Leahy, M (ed.) *Disorders of Communication: The Science of Intervention*. London: Taylor & Francis.

Kamm, K., Thelen, E. and Jensen, J.L. (1990), A dynamical systems approach to motor development. *Physical therapy*, 70(12): 763-775.

Katz, W.F., Carter, G.C. and Levitt, J.S. (2007), treating buccofacial apraxia using augmented kinematic feedback. *Aphasiology,* 21(12): 1230-1247.

Kazdin, A.E. (1980), Acceptability of alternative treatments for deviant child behaviour. *Journal of applied Behaviour Analysis*, 135(2): 259-273.

Keuning, K.H.D., Wieneke, G.H. and Dejonckere, P.H. (1999), The intrajudge reliability of the perceptual rating of cleft palate speech before and after pharyngeal flap surgery: The effect of judges and speech samples. *The Cleft Palate - Craniofacial Journal*, 36(4): 328.

Kent, R.D. and Read, C. (2002), *Acoustic analysis of speech.* Second edition. NY, USA: Delmar, Cengage Learning.

Kenward, M. G., & Molenberghs, G. (2009). Last observation carried forward: a crystal ball?. *Journal of biopharmaceutical statistics*, *19*(5), 872-888.

Kleim, J.A. and Jones, T.A. (2008), Principles of Experience-Dependent Neural Plasticity: Implications for Rehabilitation after Brain Damage. *Journal of Speech Language & Hearing Research,* 51(1): S225-239.

Klintö, K., Salameh, E.K., Svensson, H. and Lohmander, A. (2011), The impact of speech material on speech judgement in children with and without cleft palate. *International Journal of Language & Communication Disorders*, 46(3): 348-360.

Knox, E. and Conti-Ramsden, G. (2003), Bullying risks of 11-year-old children with specific language impairment (SLI): does school placement matter? *International Journal of Language & Communication Disorders,* 38(1): 1-12.

Köster, O., Jessen, M., Khairi, F., & Eckert, H. (2007, August). Auditory-perceptual identification of voice quality by expert and non-expert listeners. In *Proceedings of the 16th International Congress of Phonetic Sciences, Saarbrücken, Gertmany* (pp. 6-10).

Krischke, S., Weigelt, S., Hoppe, U., Kollner, V., Klotz, M., Eysholdt, U. and Rosanowski, F. (2005), Quality of Life in Dysphonic Patients. *Journal of Voice,* 19(1):132-137.

Kuehn, D.P. and Moller, K.T. (2000), Speech and Language Issues in the Cleft Palate Population: The State of the Art. *The Cleft Palate -Craniofacial Journal,* 37(4): 348-350.

Kuehn, D.P., Imrey, P.B., Tomes, L., Jones, D.L., O'Gara, M.M., Seaver, E.J., Smith, B.S., Van Demark, D.R. and Watchel, J.M. (2002), Efficacy of continuous positive airway pressure for treatment of hypernasality. *The Cleft Palate-Craniofacial Journal*, 39(3): 267-276

Kummer, A.W. (2004), Velopharyngeal dysfunction: Current thinking on the cause, effect, assessment and treatment. *Otolaryngology & Head and Neck Surgery,* 10(6): 455-459.

Kummer, A.W. & Lee, L. (1996), Evaluation and Treatment of Resonance Disorders. *Language Speech & Hearing Services in Schools*, 27(3): 271-281.

Kunzel, H.J. (1982), First Applications of a Biofeedback Device for the Therapy of Velopharyngeal Incompetence. *Folia Phoniatrica et Logopaedica,* 34(2): 92-100.

Lallh, A.K. and Rochet, A.P. (2000), The effect of information on listeners' attitudes toward speakers with voice or resonance disorders. *Journal of Speech Language & Hearing Research,* 43(3): 782-95.

Lancaster, G.A., Dodd, S. and Williamson, P.R, (2004), Design and analysis of pilot studies:recommendations for good practice. *Journal of Evaluation in Clinical Practice,* 10(2): 307-312.

Landis, J. and Koch, G. (1977), The measurement of observer agreement for categorical data. *Biometrics,* 33: 159-174

Laukkanen, A.M., Syrja, T., Laitala, M. and Leino, T. (2004), Effects of two-month vocal exercising with and without spectral biofeedback on student actors' speaking voice*. Logopedics Phoniatrics Vocology,* 29(2): 66-76.

Laures, J. and Shisler, R. (2004), Complementary and alternative medical approaches to treating adult neurogenic communication disorders: a review. *Disability & Rehabilitation,* 26(6): 315-25.

Law, J., Boyle, J., Harris, F. and Harkness, A. (1998), Screening for Primary Speech and Language Delay: A Systematic Review of the Literature. *International Journal of Language & Communication Disorders,* 33(S1): 21-23.

Law, J., Garrett, Z. and Nye, C. (2004), The efficacy of treatment for children with developmental speech and language delay/disorder. *Journal of Speech, Language, and Hearing Research*, 47:924-943.

Law, J. and Forsyth, K. (2006), Plugging the evidence base gap. RCSLT conference presentation, Edinburgh.

Law, J., Lee, W., Roulstone, S., Wren, Y., Zeng, B., & Lindsay, G. (2012), 'What Works': interventions for children and young people with speech, language and communication needs. Department for Education research report DFE-RR247-BCRP10.

Lead Cleft SLT Forum UK (2010) VPD referral data (unpublished).

Leahy, M. and Stemple, J.C. (1989), Aspects of the anatomy and physiology of speech. IN: *Disorders of Communication: The Science of Intervention*. Leahy M (ed.) London: Taylor & Francis.

Lee, A.S.Y., Ciocca, V. and Whitehill, T.L. (2003), Acoustic correlates of hypernasality. *Clinical Linguistics & Phonetics,* 17(4-5): 259-264.

Lee, A.S.Y., Law, J. and Gibbon, F.E. (2009), Electropalatography for articulation disorders associated with cleft palate. *Cochrane Database Systematic Reviews,* (3): CD006854.

Levelt, W.J. (1993), Lexical access in speech production. *Knowledge and language: From Orwell's problem to Plato's problem*: 241.

Lewis, B.A., Freebairn, L.A., Hansen, A. J., Iyengar, S.K. and Taylor, H.G. (2004), School-age follow-up of children with childhood apraxia of speech. *Language Speech & Hearing Services in Schools,* 35(2):122-40.

Lewis, B.A. and Freebairn, L. (1992), Residual Effects of Preschool Phonology Disorders in Grade School, Adolescence, and Adulthood. *Journal of Speech & Hearing Research,* 35(4): 819-831.

Lewison, G. and Carding, P. (2003), Evaluating UK research in speech and language therapy. *International Journal of Language & Communication Disorders*, 38(1): 65-84.

Locker, D., Jokovic, A. and Tompson, B. (2005), Health-related quality of life of children aged 11 to 14 years with orofacial conditions. *Cleft Palate Craniofacial Journal*, 42(3): 260-6.

Lohmander, A. & Olsson, M. (2004), Methodology for perceptual assessment of speech in patients with cleft palate: a critical review of the literature. *The Cleft Palate - Craniofacial Journal*, 41(1): 64-70.

Lombard, M., Snyder - Duch, J., & Bracken, C. C. (2006), Content analysis in mass communication: Assessment and reporting of intercoder reliability. *Human communication research*, *28*(4), 587-604.

Louw, P. (1996), *Coping with Stuttering*. Johannesburg: Delta Books.

Lubker, J.F. (1975), Normal velopharyngeal function in speech. *Clinics in Plastic Surgery* , 2: 249-59

Lubit, E.C. (1969), The Lubit Palatal Exerciser: a preliminary report. *The Cleft Palate Journal*, 6: 120-133.

Lubit, E.C. and Larsen, R.E. (1971), A Speech Aid for Velopharyngeal Incompetency. *Journal of Speech & Hearing Disorders,* 36(1): 61-70.

Ludlow, C. L., Hoit, J., Kent, R., Ramig, L. O., Shrivastav, R., Strand, E., Yorkston, K. & Sapienza, C. M. (2008). Translating Principles of Neural Plasticity Into Research on Speech Motor Control Recovery and Rehabilitation. *Journal of Speech, Language, and Hearing Research*, *51*(1), S240.

Maas, E., Robin, D.A., Austermann Hula S.N., Freedman, S.E., Wulf, G., Ballard, K.J. and Schmidt, R.A. (2008), Principles of motor learning in treatment of motor speech disorders. *American Journal of Speech-Language Pathology,* 17(3): 277-298.

Mackenzie, C. and Lowit, A. (2007), Behavioural intervention effects in dysarthria following stroke: communication effectiveness, intelligibility and dysarthria impact. *International Journal of Language & Communication Disorders,* 42(2): 131-153.

Main, A., Kelly, S. and Manley, G. (1999), Instrumental assessment and treatment of hypernasality, following maxillofacial surgery, using SNORS: a single case study. *International Journal of Language & Communication Disorders,* 34(2): 223-38.

Marchant, J., McAuliffe, M.J. and Huckabee, M. (2008), Treatment of articulatory impairment in a child with spastic dysarthria associated with spastic cerebral palsy. *Developmental Neurorehabilitation,* 11(1): 81-90.

Markham, C. (2008), The development and validation of a quality of life scale for children with speech, language and communication needs. Unpublished, PhD.

Markham, C. and Dean, T. (2006), Parents' and professionals' perceptions of Quality of Life in children with speech and language difficulty. *International Journal of Language & Communication Disorders,* 41(2): 189-212.

Markham, C, van Laar D., Gibbard, D. and Dean, T. (2009), Children with speech, language and communication needs: their perceptions of their quality of life. *International Journal of Language and Communication Disorders,* 44(5): 748-768.

Mason, C., Nickels, L., McDonald, B., Moses, M., Makin, K., & Taylor, C. (2011), Treatment of word retrieval impairments in aphasia: Evaluation of a self-administered home programme using personally chosen words. *Aphasiology*, *25*(2), 245-268.

Massengill, R.J., Quinn, G.W., Pickrell, K.L. and Levinson, C. (1968),Therapeutic exercise and velopharyngeal gap. *Cleft Palate Journal,* January (5): 44-7.

Massengill, R.J. & Quinn, G. (1974), Adenoidal atrophy, velopharyngeal incompetence and sucking exercises: a two year follow-up case report. *Cleft Palate Journal,* April (11): 196-9.

Mathieson, L. (2001), IN: Greene MCL and Mathieson L (eds.) *The Voice and its Disorders*. 6th Edition London: Whurr Publishers.

McAuliffe, M.J. and Cornwell, P.L. (2008), Intervention for lateral /s/ using electropalatography (EPG) biofeedback and an intensive motor learning approach: a case report. *International Journal of Language & Communication Disorders,* 43(2): 219-229.

McCormack, J., McLeod, S., McAllister, L. and Harrison, L.J. (2009), A systematic review of the association between childhood speech impairment and participation across the lifespan. *International Journal of Speech-Language Pathology,* 11(2): 155-170.

McGinn, T., Wyer, P.C., Newman, T.B., Keitz, S. and Leipzig, R. (2004), Tips for learners of evidence-based medicine: 3. Measures of observer variability (kappa statistic). *Canadian Medical Association Journal*, 171(11): 1369-1373.

McLeod, S. (2004), Speech pathologists' application of the ICF to children with speech impairment. *Advances in Speech-Language Pathology,* 6(1): 75-81.

McLeod, S. & Bleile, K. (2004), The ICF: a framework for setting goals for children with speech impairment. *Teaching and Therapy,* 20(3): 199-219.

McLeod, S. & McKinnon, D.H. (2007), Prevalence of communication disorders compared with other learning needs in 14500 primary and secondary school students, *International Journal of Language & Communication Disorders,* 42(s1): 37-59.

McLeod, N.M.H., Arana-Urioste, M.L., & Saeed, N.R. (2004). Birth prevalence of cleft lip and palate in Sucre, Bolivia. Cleft Palate-Craniofacial Journal, 41(2), 195-198

Medical Research Council (MRC) (2000) *A framework for development and evaluation of RCTs for complex interventions to improve health.*

Medical Research Council (MRC) (2008) *Developing and evaluating complex interventions: new guidance.* Available at: http://www.mrc.ac.uk/complexinterventionsguidance

Mercer, N.S.G. and Pigott, R.W. (2001), Assessment and Surgical Management of Velopharyngeal Dysfunction. IN: Watson ACH, Sell DA and Grunwell P (eds.) *Management of Cleft Lip and Palate*. London: Whurr Publishers.

Michie, S., Johnston, M., Francis, J., Hardeman, W. and Eccles, M. (2008), From theory to intervention;mapping theoretically deived behavioural determinants to behaviour change techniques. *Applied Psychology* , 57(4) October :660-680.

Moher, D., Pham, B., Jones, A., Cook, D. J., Jadad, A. R., Moher, M., ... & Klassen, T. P. (1998). Does quality of reports of randomised trials affect estimates of intervention efficacy reported in meta-analyses?. *The Lancet*, *352*(9128), 609-613.

Moller, K.T., Path, M., Werth, L.J. and Christiansen, R.L. (1973), The Modification of Velar Movement. *Journal of Speech & Hearing Disorders*, 38(3): 323-334.

Moon JB & Kuehn DP (2004) Anatomy and physiology of normal and disordered velopharyngeal function for speech. IN: Bzoch K R (ed.) *Communicative Disorders Related to Cleft Lip and Palate.* Austin, Texas: Pro-Ed.

Morley, M.E. (1966), Speech and speech therapy in cleft palate. *Modern Trends in Plastic Surgery,* 2: 255-67.

Morley, M.E. (1967), *Cleft palate and speech*. London: E. & S. Livingstone Ltd.

Mossey, P. A., Little, J., Munger, R.G., Dixon, M.J. and Shaw, W.C. (2009), Cleft lip and palate. *The Lancet,* 374(9703): 1773-1785.

Muntz, H. (1992), Navigation of the nose with flexible fiberoptic endoscopy. *Cleft Palate Craniofacial Journal,* 29(6): 507-10.

Murdoch, B.E., Pitt, G., Theodoros, D.G. and Ward, E.C. (1999), Real-time continuous visual biofeedback in the treatment of speech breathing disorders following childhood traumatic brain injury: report of one case. *Pediatric Rehabilitation,* 3(1): 5-20.

Murray, J. (2006), Cybernetic circularity in teaching and learning. *International Journal of Teaching and Learning in Higher Education*, 18(3): 215-221.

Nastasi, B. K., & Schensul, S. L. (2005). Contributions of qualitative research to the validity of intervention research. *Journal of School Psychology*, *43*(3), 177-195.

Needleman, C., & Needleman, M. L. (1996). Qualitative methods for intervention research. *American Journal of Industrial Medicine*, *29*(4), 329-337

NHMRC Evidence Hierarchy (2009) from: Merlin T, Weston A and Tooher R (2009) Extending an evidence hierarchy to include topics other than treatment: revising the Australian'levels of evidence'. *BMC Medical Research Methodology*, *9(*1): 34.

NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC) [2008], Available at: http://www.netscc.ac.uk/glossary/#glos6. [Accessed on 14 July 2011]

Nohara. K., Tachimura, T. and Wada, T. (2006), Levator veli palatini muscle fatigue during phonation in speakers with cleft palate and borderline velopharyngeal incompetence. *The Cleft Palate Craniofacial Journal* ,43: 103-7.

Nordberg, A., Carlsson, G. and Lohmander, A. (2011), Electropalatography in the description and treatment of speech disorders in five children with cerebral palsy. *Clinical linguistics & phonetics*, 25(10): 831-852.

Oxford Centre for Evidence based Medicine (OCEBM) [2011], The Oxford 2011 levels of evidence. Available at: http://www. cebm.net/index.aspx?o=5653. [Accessed on 14 July 2011]

Palmer, R., Enderby, P. and Hawley, M. (2007), Addressing the needs of speakers with longstanding dysarthria: computerized and traditional therapy compared. *International Journal of Language & Communication Disorder* , 42(Suppl 1): 61-79.

Pamplona, M.C., Ysunza, A., González, M., Ramírez, E., & Patiño, C. (2000), Linguistic development in cleft palate patients with and without compensatory articulation disorder. *International journal of pediatric otorhinolaryngology*, 54(2-3): 81.

Pantelemidou, V., Herman, R. and Thomas, J. (2003), Efficacy of speech intervention using electropalatography with a cochlear implant user. *Clinical linguistics & phonetics*, 17(4-5): 383-392.

Paradise, J.L. (1996), Tonsillectomy and adenoidectomy. *Pediatric otolaryngology*, 2: 1054-1065.

Parahoo, K. (2006), *Nursing Research: principles, process and issues.* Second edition. Hampshire, UK: Palgrave Macmillan.

Peat, B., Albery, E.H. and Pigott, R.W, (1994), Tailoring velopharyngeal surgery: The influence of etiology and type of operation. *Plastic and Reconstructive Surgery,* 93(5): 901-1116.

Pennington, L., Miller, N. and Robson, S. (2010), Speech therapy for children with dysarthria acquiored before three years of age. *Cochrane Library* 2010 (1),

Peterson, S. (1974), Electrical Stimulation of the Soft Palate. *Cleft Palate Journal*, January, 11(1): 72-86.

Peterson, G.E. and Shoup, J.E. (1966), A physiological theory of phonetics, *Journal of Speech, Language, and Hearing Research,* 9: 5-67.

Peterson-Falzone, S.J., Trost-Cardamone, J.E., Karnell, M.P. and Hardin-Jones, M.A. (2006), *The Clinicians Guide: Treating Cleft Palate Speech,* St. Louis, MO: Mosby Elsevier.

Petrie, A. and Sabin, C. (2000), *Medical Statistics at a Glance.* Oxford, UK: Blackwell Science Ltd.

Plint, A.C., Moher, D., Morrison, A., Schulz, K., Altman, D.G. and Gaboury, I. (2006), Does the CONSORT checklist improve the quality of reports of randomised controlled trials? a systematic review. *The Medical Journal of Australia*, September, 4 ,185 (5): 263-267.

Powers, G.L. and Starr, C.D. (1974), The effects of muscle exercises on velopharyngeal gap and nasality. *Cleft Palate Journal,* January 11(1): 28-35.

Pratt, D. (1978), Cybernetic Principles in the Design of Instruction. Paper presented at the Annual Meeting of the American Educational Research Association (Toronto, Canada, March 1978). Available at: http://www.eric.ed.gov.

Pring, T (2004), Ask a silly question: two decades of troublesome trials. *International Journal of Language & Communication Disorders,* 39(3): 285-302.

Pring, T. (2005), *Research methods in communication disorders*. UK: Wiley.

Rattenbury, H.J., Carding, P.N. and Finn, P.L. (2004), Evaluating the effectiveness and efficiency of voice therapy using transnasal flexible laryngoscopy: a randomized controlled trial. *Journal of Voice,* 18(4): 522-33.

RCSLT (2005), *Royal College of Speech and Language Therapists: Clinical Guidelines*. Milton Keynes: Speechmark Publishers Ltd.

Ramig, L.O. and Verdolini, K. (1998), Treatment efficacy: voice disorders. *Journal of Speech Language & Hearing Research*, 41(1): 101-16.

Reilly, S. (2004), The challenges in making speech pathology practice evidence based. *International Journal of Speech-Language Pathology*, 6(2): 113-124.

Reimers, T., Wacker, D.P., Cooper, L.J. and deRaad, A.O. (1992), Clinical evaluation of the variables associated with treatment acceptability and their relation to compliance. *Behavioural Disorders,* 18(1): 66-76.

Ren, Y.F., Isberg, A. and Henningsson, G. (1995), Velopharyngeal incompetence and persistent hypernasality after adenoidectomy in children without palatal defect, *The Cleft Palate-Craniofacial Journal,* 32(6): 476-482.

Resnick, H.E., Fries, B.E. and Verbrugge, L.M. (1997), Windows to their World: The Effect of Sensory Impairments on Social Engagement and Activity Time in Nursing Home Residents. *Journal of Gerontology: Social Sciences,* 52B(3): S135-S144.

Riski, J.E. (2004), Secondary Surgical Procedures to Correct Postoperative Velopharyngeal Incompetencies Found After Primary Palatoplasties. IN: Bzoch

KR (ed.) *Communicative Disorders Related to Cleft Lip and Palate*. 5th edition. Austin, Texas: Pro-Ed.

Riski, J.E. and DeLong, E. (1984), Articulation development in children with cleft lip/palate, *Cleft Palate Journal,* 21(2): 57-64.

Robey, R.R. (2004), A five-phase model for clinical-outcome research. *Journal of Communication Disorders,* 37(5): 401-11.

Robey, R.R. and Shultz, M.C. (1998), A model for conducting clinical-outcome research: an adaptation of the standard protocol for use in aphasiology. *Aphasiology,* 12: 787-810.

Robbins, J., Butler, S.G., Daniels, S.K., Diez Gross, R., Langmore, S., Lazarus, C.L. and Rosenbek, J. (2008), Swallowing and dysphagia rehabilitation: translating principles of neural plasticity into clinically oriented evidence. *Journal of Speech Language & Hearing Research,* 51(1): S276-300.

Rogers, C. (1951), *Client centered therapy: its current practice, and theory*. Oxford. Houghton Mifflin.

Rogers, C. R. (1985), Toward a more human science of the person. *Journal of Humanistic Psychology*, *25*(4), 7-24.

Rohrich, R.J., Rowsell, A.R., Johns, D.F., Drury, M.A., Grieg, G., Watson, D.J. and Poole, M. D. (1996), Timing of hard palatal closure: a critical long-term analysis. *Plastic and reconstructive surgery*, 98(2): 236.

Rohrich. R.J., Love, E.J., Byrd, H.S. and Johns, D.F. (2000), Optimal timing of cleft palate closure. *Plastic and reconstructive surgery*, 106(2): 413-422.

Roll, D.L. (1973), Modification of nasal resonance in cleft-palate children by informative feedback. *Journal of Applied Behavioural Analysis,* 6(3): 397-403.

Rosenbek, J.C. (1985), Treating apraxia of speech IN: Johns DF (ed.) *Clinical management of neurogenic communication disorders,* Boston: Little Brown 267-312.

Ross, K. and Wertz, R. (2003), Quality of life with and without aphasia. *Aphasiology*, 17(4): 355-364.

Rothman, K. J. (1976). Causes. *American Journal of Epidemiology*, *104*(6), 587-592

Roulstone, S., Glogowska, M., Enderby, P. and Peters, T.J. (1999), Issues to consider in the evaluation of speech and language therapy for preschool children. *Child: care, health and development*, 25(2): 141-155.

Roy, N. and Weinrich, B. (2003), Three treatments for teachers with voice disorders: a randomized clinical trial. *Journal of Speech Language & Hearing Research,* 46(3): 670-88.

Ruscello, D.M. (1982), A selected review of palatal training procedures. *Cleft Palate Journal,* 19: 181-93

Ruscello, D.M. (2006), *Treatment of velopharyngeal closure for speech: discussion and implications for management*, [online} Available at: http://www.thefreelibrary.com/Treatment of velopharyngeal closure for speech. [Accessed 20 September 2010].

Ruscello, D.M. (2008), An examination of nonspeech oral motor exercises for children with velopharyngeal inadequacy. *Seminars in Speech & Language,* 29(4): 294-303.

Ruscello, D.M., Shuster, L.I. and Sandwisch, A. (1991), Modification of Context Specific Nasal Emission. *Journal of Speech and Hearing Research,* February 34: 27-32.

Russell, B. (2006), *Treatment of Hypernasality in Children with Down's Syndrome Post Adenoidectomy,* [online] Available at: http://www/speechpathology.com/articles/index.asp. [Accessed 20 September 2010]

Russell J, and Grunwell P. Speech development in children with cleft lip and palate. In Grunwell P, ed. *Analysing cleft palate speech*. London: Whurr, 1993

Russell, J. and Harding, A. (2001), Speech Development and Early Intervention, in *Management of Cleft Lip and Palate* IN: Watson ACH, Sell DA and Grunwell P (eds.) London: Whurr Publishers

Sapir, S., Spielman, J.L., Ramig, L.O., Story, B.H. and Fox, C. (2007), Effects of intensive voice treatment (the Lee Silverman Voice Treatment [LSVT]) on vowel

articulation in dysarthric individuals with idiopathic Parkinson disease: acoustic and perceptual findings. *Journal of Speech, Language and Hearing Research*, 50(4): 899.

Saunders, N.C., Hartley, B.E.J., Sell, D. and Sommerlad, B. (2004), Velopharyngeal in, sufficiency following adenoidectomy. *Clinical Otolaryngology,* 29(6): 686-688.

Schmidt, R. A. (1975), A schema theory of discrete motor skill learning. *Psychological review*, *82*(4), 225.

Schmidt, R.A. and Lee, T.S. (2005), *Motor control and learning: A behavioural emphasis*. 4th edition. Champaign, IL: Human Kinetics.

Scobbie, J.M., Wood, S.E. and Wrench, A.A. (2004), Advances in EPG for treatment and research: an illustrative case study. *Clinical linguistics & phonetics*, 18(6-8): 373-389.

Seaver, E.J., Dalston, R.M., Leeper, H.A. and Adams, L.E. (1991), A study of nasometric values for normal nasal resonance. *Journal of Speech and Hearing Research,* 34 (August): 715-721.

Sell, D.A., Harding, A. and Grunwell, P. (1999), GOS.SP.ASS.'98: an assessment for speech disorders associated with cleft palate and/or velopharyngeal dysfunction (revised). *International Journal of Language & Communication Disorders,* 34(1): 17-33.

Sell, D.A. and Grunwell, P. (2001), Speech Assessment and Therapy. IN: Watson ACH, Sell DA and Grunwell P (eds.) *Management of Cleft Lip and Palate.* London: Whurr Publishers.

Sell, D. (2003), Management of VPD. Paper presented at Postgraduate Cleft Training Course. Homerton College, Cambridge, UK

Sell, D. (2005), Issues in perceptual speech analysis in cleft palate and related disorders: a review. *International Journal of Language & Communication Disorders,* 40(2): 103-21.

Sell, D.A., Mars, M. and Worrell, E. (2006), Process and outcome study of multidisciplinary prosthetic treatment for velopharyngeal dysfunction. *International Journal of Language & Communication Disorders,* 41(5): 495-511.

Sell, D., Britton, L., Hayden, C., Phippen, G. and Russell, J. (2008), Speech and Language Therapy and Nasendoscopy for Patients with Velopharyngeal Dysfunction Position Paper, Royal College of Speech and Language Therapists.

Sell, D., John, A., Harding-Bell, A., Sweeney, T., Hegarty, F. and Freeman, J. (2009) Cleft Audit Protocol for Speech (CAPS-A): a comprehensive training package for speech analysis. *International Journal of Language & Communication Disorders,* 44(4): 529-548.

Serlin, R.C., Mendoza, T.R., Nakamura, Y., Edwards, K.R. and Cleeland, C.S. (1995), When is cancer pain mild, moderate or severe? Grading pain severity by its interference with function. *Pain,* 61(2): 277-284

Shanyinde, M., Pickering, R.M. and Weatherall, M. (2011), Questions asked and answered in pilot and feasibility randomized controlled trials. *BMC Medical Research Methodology,* 11:117. Available at: http://www.biomedcentral.com/1471-2288/11/117. [Accessed on 18 August 2012]

Shelton, R.L., Chisum, L., Youngstrom, K.A., Arndt, W.B. and Elbert. M. (1969) Effect of articulation therapy on palatopharyngeal closure, movement of the pharyngeal wall, and tongue posture. *Cleft Palate Journal*, October 16: 440-448.

Shelton, R.L., Beaumont, K., Trier, W.C. and Furr, M.L. (1978), Videoendoscopic Feedback in Training Velopharyngeal Closure. *Cleft Palate Journal,* 15(1): 6-12.

Sherman, S.A., Eisen, S., Burwinkle, T.M. and Varni, J.W. (2006), The PedsQL Present Functioning Visual Analogue Scales: preliminary reliability and validity. *Health Quality of Life Outcomes,* 4: 75.

Shimokawa, T., Yi, S. and Tanaka, S. (2005), Nerve supply to the soft palate muscles with special reference to the distribution of the lesser palatine nerve. *Cleft Palate Craniofacial Journal,* 42(5): 495-500.

Shprintzen, R.J., Sher, A.E. and Croft, C.B. (1987), Hypemasal speech caused by tonsillar hypertrophy. *International journal of pediatric otorhinolaryngology*, 14(1): 45-56.

Shprintzen, R.J. and Marrinan, E. (2009), Velopharyngeal insufficiency: diagnosis and management. *Current Opinion in Otolaryngololy & Head and Neck Surgery,* 17(4): 302-7.

Shprintzen, R.J., McCall, G.N. and Skolnick, M.L. (1975), A new therapeutic technique for the treatment of velopharyngeal incompetence. *Journal of Speech & Hearing Disorders,* 40(1): 69-83.

Shriberg, L. D., Kwiatkowski, J., & Hoffmann, K. (1984). A procedure for phonetic transcription by consensus. *Journal of Speech, Language and Hearing Research*, *27*(3), 456.

Sibbald, B., & Roland, M. (1998). Understanding controlled trials. Why are randomised controlled trials important?. *BMJ: British Medical Journal*, *316*(7126), 201.

Siegel-Sadewitz, V.L. and Shprintzen, R.J. (1982), Nasopharyngoscopy of the normal velopharyngeal sphincter: an experiment of biofeedback. C*left Palate Journal,* 19(3): 194-200.

Sim, J. and Wright, C. C. (2005), The kappa statistic in reliability studies: use, interpretation, and sample size requirements. *Physical therapy*, 85(3): 257-268.

Sime, W.E. and Healey, E.C. (1993), An interdisciplinary approach to the treatment of a hyperfunctional voice disorder. *Applied Psychophysiology and Biofeedback*, 18(4): 281-287.

Skolnick, M. L., Shprintzen, R. J., McCall, G. N., & Rakoff, S. (1975), Patterns of velopharyngeal closure in subjects with repaired cleft palate and normal speech: A multi-view videofluoroscopic analysis. *Cleft Palate Journal*, *12*(4), 369-375.

SLTRU (2011)Bristol Speech and Language Therapy Research Unit at http://www.speech-therapy.org.uk/research

Sommerlad, B. C. (1981), Nasendoscopy IN *Recent Advances in Plastic Surgery 2* (Ed: I T Jackson). Churchill Livingstone, Edinburgh

Sommerlad, B.C. (2003), A Technique for Cleft Palate Repair. *Plastic and Reconstructive Surgery,* 112(6): 1542-1548.

Soon, G. (2009): Editorial: Missing Data—Prevention and Analysis, *Journal of Biopharmaceutical Statistics*, 19:6, 941-944.

Specialised Services National Definitions Set, No.15, Cleft Lip and Palate (all ages) (3rd edition) available from www.specialisedcommissioning.nhs.uk

Spriestersbach, D. C., Dickson, D. R., Fraser, F. C., Horowitz, S. L., McWilliams, B. J., Paradise, J. L., & Randall, P. (1973). Clinical research in cleft lip and cleft palate: the state of the art. *The Cleft palate journal*, *10*, 113.

Stackhouse. J., Wells, B. and Snowling, M.J. (1997), *Children's speech and literacy difficulties: A psycholinguistic framework*. London: Whurr Publishers.

Stackhouse, J., Wells, B., Pascoe, M. and Rees, R. (2002), From phonological therapy to phonological awareness. In *Seminars in Speech and Language,* 23(1): 23-34.

Stal, P.S. and Lindman, R. (2000), Characterisation of human soft palate muscles with respect to fibre types, myosins and capillary supply. *Journal of Anatomy,* 197: 275-290.

Stanier, P. and Moore, G.E. (2004), Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. *Human Molecular Genetics,* 13(s1): R73-R81.

Stevens, K.N. (1972), The quantal nature of speech: Evidence from articulatory-acoustic data. IN: David, E.E. and Denes, P.B (eds.) *Human communication: A unified view* , pp. 51-66. McGraw-Hill, New York.

Stewart, K.J., Ahmad, T., Watson, A.C.H. and Razzell, R. (2002), Altered speech following adenoidectomy: a 20 year experience. *British Journal of Plastic Surgery,* 55(6): 469-473.

Stewart, T. and Brosh, H. (1997), The use of drawings in the management of adults who stammer. *Journal of Fluency Disorders,* 22(1): 35-50.

Strand, E.A. and Skinder, A. (1999), Treatment of developmental apraxia of speech: Integral stimulation methods. *Clinical management of motor speech disorders in children*, 109-148.

Straus, S.E. (2007), Evidence-based health care: Challenges and limitations. *Evidence-Based Communication Assessment and Intervention*, 1(1): 48-51.

Sweeney, T., Sell, D. and O'Regan, M. (2004) Nasalance scores for normal-speaking Irish children. *Cleft Palate-Craniofacial Journal,* 41: 168-174

Sweeney, T. and Sell, D. (2008), Relationship between perceptual ratings of nasality and nasometry in children/adolescents with cleft palate and/or velopharyngeal dysfunction. *International Journal of Language & Communication Disorders,* 43(3): 265-282.

Sweeney, T. (2011), Nasality-Assessment and Intervention IN: Howard S & Lohmander A (eds.) *Cleft Palate Speech: Assessment and Intervention*, Chichester, UK: Wiley-Blackwell.

Sweeney, T. and Sel,l D. (2011), Relationship between perceptual ratings of nasality and nasometry in children/adolescents with cleft palate and/or velopharyngeal dysfunction. *International Journal of Language & Communication Disorders*, *43*(3): 265-282.

Tachimura, T., Nohara, K., Hara, H. and Wada, T. (1999), Effect of placement of a speech appliance on levator veli palatini muscle activity during blowing. *The Cleft Palate-Craniofacial Journal,* 36(3): 224-32.

Tash, E.L., Shelton, R.L., Knox, A.W. and Michel, J.F. (1971), Training voluntary pharyngeal wall movements in children with normal and inadequate velopharyngeal closure. *Cleft Palate Journal,* 8: 277-90.

Teager, H.M. and Teager, S.M. (1990), Evidence for nonlinear sound production mechanisms in the vocal tract. *Speech production and speech modelling*, 55: 241-261.

Thabane, L., Ma, J., Chu, R., Cheng, J., Ismaila, A., Rios, L.P., Robson, R., Thabane, M., Giangregorio, L. and Goldsmith, C.H. (2010), A tutorial on pilot studies: the

what, why and how. *BMC Medical Research Methodology,* 10:1, Available at http://www.biomedcentral.com/1471-2288/10/1 [Accessed on 8 June 2012]

The Communication Trust, *Explaining Speech, Language and Communication needs (SLCN)*. 2010: London.

Thelen, E. (1991), Motor aspects of emergent speech: a dynamic approach. IN: Krasnegor NA, Rumbaugh DM, Schiefelbusch RL and Studdert-Kennedy (eds.) *Biological and behavioural determinants of language development,* New Jersey: Lawrence Erlbaum 339-362.

Timing of Palate Surgery (TOPS) Clinical Trial, Available at: http://clinical trials.gov/ct2/show/NCT00993551

Titze, I., Riede, T. and Popolo, P. (2008), Nonlinear source-filter coupling in phonation: Vocal exercises. *The Journal of the Acoustical Society of America*, 123(4): 1902.

Threats, T.T. and Worrall, L. (2004), Classifying communication disability using the ICF. *International Journal of Speech-Language Pathology*, 6(1): 53-62.

Treiman, R., Clifton, C. Jr., Meyer, A.S. and Wurm, L.H. (2003), Language comprehension and production IN: *Comprehensive handbook of pyschology, Vol 4; Experimental Psychology.* New York: John Wiley & Sons, Inc. 527-548.

Trost, J. E. (1981). Articulatory additions to the classical description of the speech of persons with cleft palate. *Cleft Palate J*, *18*(3), 193-203.

Trost-Cardamone, J.E. (1989), Coming to terms with VPI: a response to Loney and Bloem. *Cleft Palate Journal*, 26(1): 68-70.

Tudor, C. and Selley, W.G. (1974), A palatal training appliance and a visual aid for use in the treatment of hypernasal speech. A preliminary report. *The British Journal of Disorders of Communication,* 9(2): 117-22.

Tugade, M.M., Fredrickson, B.L. and Feldman Barrett, L. (2004), Psychological Resilience and Positive Emotional Granularity: Examining the Benefits of Positive Emotions on Coping and Health. *Journal of Personality,* 72(6): 1161-1190.

Turner, H.M., Nye, C., Ortiz, M., Liu, E. and Eisenstein, E.L. (2007), Producing systematic reviews of interventions in speech-language pathology: A framework

for sustainability. *Evidence-Based Communication Assessment and Intervention*, *1*(4), 201-212.

Uebersax, J. (2010), Kappa coefficients. Available at: http://www.john-uebersax.com/stat/kappa.htm [Accessed September 2012].

Vallino-Napoli, L.D. (2011), Chapter 18 IN: Howard S & Lohmander A (eds.) *Cleft Palate Speech: Assessment and Intervention*, Chichester, UK: Wiley-Blackwell,

Varni, J.W., Limbers, C.A. and Burwinkle, T.M. (2007), How young can children reliably and validly self-report their health-related quality of life?: an analysis of 8,591 children across age subgroups with the PedsQL 4.0 Generic Core Scales. *Health Quality of Life Outcomes,* 5: 1.

Vogel, A.P., Ibrahim, H.M., Reilly, S. and Kilpatrick, N. (2009), A Comparative Study of Two Acoustic Measures of Hypernasality. *Journal of Speech Language & Hearing Research,* 52(6): 1640-1651.

Yanchar, S. C. (2006). Beyond the qualitative-quantitative divide: How to proceed? *New Ideas in Psychology*, *24*(3), 275-281

Watson, A.C.H. (2001), Embryology, Aetiology and Incidence. IN: Watson ACH, Sell DA and Grunwell, P. (eds.) *Management of Cleft Lip and Palate.* London: Whurr Publishers.

Watterson, T., Lewis, K.E. and Deutsch, C. (1998), Nasalance and nasality in low pressure and high pressure speech. *The Cleft Palate - Craniofacial Journal,* 35(4): 293-298

Watterson, T., Lewis, K.E. and Brancamp, T. (2005), Comparison of Nasalance Scores Obtained With the Nasometer 6200 and the Nasometer 6400.*The Cleft Palate - Craniofacial Journal,* 42(5): 574

Weber, J. Jr., Jobe, R.P. and Chase, R.A. (1970), Evaluation of muscle stimulation in the rehabilitation of patients with hypernasal speech. *Plastic & Reconstructive Surgery,* 46(2): 173-4.

Weiss CE (1971) Success of an Obturator Programme. *Cleft Palate Journal* July 8(3): 291-7.

Weismer, G. (2006), Philosophy of research in motor speech disorders. *Clinical Linguistics & Phonetics,* 20(5): 315-49.

Wenke, R.J., Theodoros, D. and Cornwell, P. (2010), Effectiveness of Lee Silverman Voice Treatment (LSVT) on hypernasality in non-progressive dysarthria: the need for further research. *International Journal of Language & Communication Disorders,* 45(1): 31-46.

West, R., Kennedy, L. and Carr, A. (1937), *The rehabilitation of speech: A textbook of diagnostic and corrective procedures.* New York: Harper & Bros.

Whitehill, T.L. (2002a), *Behavioural Treatment of Resonance and Nasal Emission*. [online] Available at: http://www.smiletrainchina.org/update/20100202202810565.pdf. [Accessed 20 September 2010].

Whitehill, T.L., (2002b), Assessing Intelligibility in Speakers With Cleft Palate: A Critical Review of the Literature. *The Cleft Palate-Craniofacial Journal*, January 2002, Vol. 39, No. 1, pp. 50-58.

Williams, P., Stephens, H. and Connery, V. (2006), What's the evidence for oral motor therapy: A response to Bowen 2005. *Acquiring Knowledge in speech, Language and Hearing,* 8(2): 89-90.

Witt, P.D., O'Daniel, T.G., Marsh, J.L., Grames, L.M., Muntz, H.R. and Pilgram, T.K.F. (1997), Surgical management of velopharyngeal dysfunction: outcome analysis of autogenous posterior pharyngeal wall augmentation. *Plastic & Reconstructive Surgery,* 99(5): 1287-96; discussion: 1297-300.

Witt, P.D., Rozelle, A.A., Marsh, J.L., Marty-Grames, L., Muntz, H., Gay, W.D. and Pilgram, T.K. (1995), Do palatal lift prostheses stimulate velopharyngeal neuromuscular activity? *The Cleft Palate -Craniofacial Journal,* 32(6): 469-75.

Witzel, M.A., Tobe, J. and Salyer, K. (1988), The use of nasopharyngoscopy biofeedback therapy in the correction of inconsistent velopharyngeal closure. *International Journal of Pediatric Otorhinolaryngology,* 15(2): 137-42.

Witzel, M.A., Tobe, J. and Salyer, K.E. (1989), The use of videonasopharyngoscopy for biofeedback therapy in adults after pharyngeal flap surgery. *Cleft Palate Journa*l, 26(2): 129-34; discussion 135.

Wolfaardt, J.F., Wilson, F.B., Rochet, A. and McPhee, L. (1993), An appliance based approach to the management of palatopharyngeal incompetency: a clinical pilot project. *The Journal of Prosthetic Dentistry,* 69: 186-95.

World Health Organization. (2001), International Classification of Functioning, Disability and Health. Geneva. *World Health Organization*, 1-303.

World Health Organization. (2003), Skills for health: skills-based health education including life skills: an important component of a child-friendly/health-promoting school. Geneva. *World Health Organization*, 1-85.

Wyatt, R., Sell, D., Russell, J., Harding, A., Harland, K. and Albery, E. (1996), Cleft palate speech dissected: a review of current knowledge and analysis. *British Journal of Plastic Surgery,* 49(3): 143-9.

Yamaoka, M., Matsuya, T., Miyazaki, T., Nishio, J. and Ibuki, K. (1983), Visual training for velopharyngeal closure in cleft palate patients; a fibrescopic procedure (preliminary report). *Journal of Maxillofacial Surgery,* 11(4): 191-3.

Yiu, E.M.L., Chan, K. M., and Mok, R.S.M. (2007), Reliability and confidence in using a paired comparison paradigm in perceptual voice quality evaluation. *Clinical Linguistics & Phonetics*, *21*(2), 129-145.

Young, A. R., Beitchman, J. H., Johnson, C., Douglas, L., Atkinson, L., Escobar, M., & Wilson, B. (2002) Young adult academic outcomes in a longitudinal sample of early identified language impaired and control children. *Journal of Child Psychology and Psychiatry,* 43(5): 635-645.

Ysunza, A., Pamplona, M., Femat, T., Mayer, I. and Velasco-Garcia, M. (1997), Videonasopharyngoscopy as an instrument for visual biofeedback during speech in cleft palate patients. *International Journal of Pediatric Otorhinolaryngology,* 41(3): 291-8.

Yules, R.B. and Chase, R.A. (1969), A training method for reduction of hypernasality in speech. *Plastic & Reconstructive Surgery,* 43(2): 180-5.