VOICE QUALITY OF CHILDREN AND YOUNG PEOPLE WITH DOWN'S SYNDROME AND ITS IMPACT ON LISTENER JUDGEMENT

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For my dad, who wanted to be here to share this achievement with me

ABSTRACT

Background: Voice quality in Down's syndrome (DS) is accepted as unusual, often perceived as harsh and whispery with lower pitch and altered nasal resonance. Less is known about the resulting impact, particularly in relation to how children and young people with DS are accepted by their peers.

Method: This is a quantitative study of the voice quality of children and young people with DS compared to age-matched typically-developing (TD) controls. Expert raters use the Vocal Profile Analysis Scheme to perceptually rate voice, which is compared to instrumental analysis of fundamental frequency, perturbation measures and spectral tilt. The impact of typical and atypical voice quality is evaluated in a study of listener judgments of character, ability, age, gender and social desirability using a specially designed semantic-differential questionnaire completed by special-needs and mainstream education staff and TD peers based on audio-recordings.

Results: Perceptually, a number of features, including lip, tongue and jaw settings, pharyngeal constriction and respiratory support were found to be atypical compared to controls, whilst other features, notably phonation type and nasality, echoed typical patterns but were more severe in presentation in the speakers with DS. Contrary to hypotheses only spectral tilt differed significantly in instrumental analysis. All groups of raters judged the speakers with DS significantly more negatively than controls across all questionnaire parameters. TD peers showed a strong preference for the company of TD children over those with DS.

Conclusions: Perceptual differences are evident in the voices of children with DS, but these are not always supported by instrumental findings, perhaps indicating that the constellation of differences give rise to more negative perception. Close agreement between education staff groups suggests that children with DS are no more disadvantaged by the perception of teachers in mainstream than in special-schools; however particular difficulties are highlighted for the development of friendships with TD peers.

Keywords: voice quality, Down's syndrome, social inclusion

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1
INTRODUCTION

1.1 SUMMARY OF RESEARCH

This PhD thesis is an investigation of the voice characteristics of children and young people with Down's syndrome (DS) and the impact that those features might have on social inclusion.

DS is a genetic disorder, most typically involving the presence of an additional chromosome 21, which occurs with an incidence of 1/732 live births (Canfield, 2006). It is associated with a range of physical, health and sensory impairments and most commonly with some degree of intellectual disability (ID). Speech and language deficits are well-documented, being predominantly expressive in nature and affecting the acquisition of grammatical structures more than vocabulary (Chapman & Hesketh, 2001). However, less is known about the vocal characteristics of people with DS, despite early observations of voice disorder (Strazzulla, 1953; Schlanger, 1962; Tredgold & Soddy, 1963; Blanchard, 1964; Benda, 1965; Novak, Sedlackova, Klajman & Betlycwski, 1967).

The term 'voice' in this study is used to describe the habitual quality which occurs as a consequence of the small adjustments of the articulators of speech (including the lungs, larynx, pharynx, velum, tongue and lips) which produce the distinctive patterns that make individual speakers easily identifiable. Thus voice is dependent on both the anatomical structure of the vocal tract and the speaker's unique vocal configuration (Mathieson, 2001).

Three distinct studies were completed to gain a comprehensive picture of the voice features associated with DS:

- Study 1 is a questionnaire-based investigation of listener judgements about the voices of children and young people with DS and a control group of age and gender matched typically-developing (TD) peers
- Study 2 is an instrumental analysis of a range of parameters associated with voice
- Study 3 is an expert perceptual analysis of voice using the Vocal Profile Analysis Scheme (VPAS; Laver, Wirz, Mackenzie & Hiller, 1991).

In this thesis the abbreviated terms 'DS speakers' and 'TD speakers' are used to represent the groups of children and young people with DS and the control group of children and young people without developmental disorder respectively. It is acknowledged that it is more appropriate to use the fuller phrases to avoid children being defined by their diagnoses; however due to the considerable amount of repetition of these terms abbreviated forms were felt to be necessary to maintain the clarity and flow of the writing.

1.2 MOTIVATION FOR RESEARCH

This study is driven by the lack of data concerning the voice features of young people with DS and their impact on social acceptance. It is recognised that poor intelligibility (encompassing deficits across language, phonology and articulation) contributes towards negative perceptions of speakers, however much less is known about the role of voice in social judgements. Previous research studies have tackled the acoustic and perceptual features of voice and its social impact separately, meaning that fundamental differences in their individual methodologies make it difficult to identify relationships between findings. This Ph.D. is the first study to have attempted to define the acoustic and perceptual features of voice in DS and their impact on listener judgements as well as looking at possible interactions between those three variables via a series of statistical correlations, thus offering a significant new insight into voice quality in DS.

Changes in social policy over recent decades mean that people with DS, along with those with generalised ID, are now enjoying a much higher profile within society. Most notably, within education the policy of increased inclusion has seen, when appropriate, a swing towards the education of children with special educational needs within mainstream schools rather than special schools. Much consideration has been given to meeting the curricular needs of such children, but less is known about the opportunities this inclusive model affords for the development of meaningful friendships. This factor was a significant motivation in making education the focus of study 1 (questionnaire-based listener judgements of voice). By evaluating the perception of special-needs and mainstream education staff it is hoped to establish whether differences in perception, perhaps leading to differences in expectation, exist between staff in different models of education. In addition, inclusion of a group of TD peer raters is hoped to gain insight into how voice affects the development of social relationships within schools.

Vast improvements in healthcare have seen a rise in the life expectancy of people with DS, which together with increased understanding of this disorder and its associated conditions means that numbers of people with this syndrome are expected to rise (Fonseca, Amaral, Ribeiro, Beserra & Guimaraes, 2005); this is despite the decrease in births as a consequence of more prenatal screening for this condition (Sadovnick & Baird, 1992). This population increase means that more research needs to be done to determine typical features and their consequent impact in the various domains affected by DS to be able to provide effective support for individuals and their families.

On a personal note, as a person who has worked with people with ID for a number of years in a residential care and social and educational support capacity I am aware that these individuals are very often disadvantaged in society and perceived negatively. Moreover, as a speech and language therapist I recognise the increasing demands made on the service to meet the needs of people with multiple and complex needs and their families via both direct and indirect therapeutic interventions. It is hoped that this research will go some way to inform those involved in the care, therapy and education of people with DS of the characteristic vocal profile of this disorder, its associated causes, as well as the possible judgements of ability and character which they engender. Perhaps this knowledge will help to make people more aware of the potential for making negative judgements based on vocal qualities which in DS are likely to be constrained by the structure and function of the vocal tract.

1.3 AIMS AND HYPOTHESES

In summary, this research aims:

- To identify the judgements of character, ability, age, gender and social desirability made by listeners about the voices of children and young people with DS and a group of age and gender-matched TD peers
- To investigate the voice quality of children with DS in comparison to TD age-matched peers, using both instrumental and perceptual measures
- To identify whether specific instrumental and perceptual ratings of voice quality correlate with specific listener judgements

Several key hypotheses have been made about the outcome of this research:

Study 1: Listeners will judge the abilities and characteristics of the children with DS more negatively than those of age and gender-matched TD peers. Judgements of gender will be less accurate for the DS group and their age judged to be younger than their actual age and that of the TD peer group.

Study 2: Instrumental analysis of a range of voice-related parameters will reveal significant differences between the children with DS and age-matched TD controls. These include:

- Increased jitter values
- Increased shimmer values
- Decreased harmonic-to-noise-ratio (HNR) values
- Increased spectral tilt (SPT) values
- F0 will not differ significantly

Study 3: Perceptual analysis of voice will reveal significant differences between the children with DS and age-matched TD controls across the various areas of the VPAS:

- Labial settings
- Mandibular settings
- Lingual settings
- Pharyngeal settings
- Velopharyngeal settings
- Larynx height settings
- Muscular tension
- Phonation features
- Prosodic features
- Temporal organization
- Other features

More detailed hypotheses for each of the three studies are described within the research method (study 1, 3.3.9; study 2, 3.4.6; study 3, 3.5.7) and a rationale given to support each based on previous findings within the literature. Where no evidence could be found to suggest the outcome of an analysis no a priori hypothesis was made.

2
LITERATURE REVIEW: BACKGROUND INFORMATION

2.1 INTRODUCTION

This chapter will give an overview of DS, including the genetic causes and some of the medical, sensory and psychological conditions which are associated with this group. It will also provide a summary of general speech and language development in DS, social communication skills and how they affect inclusion, and finally a description of typical voice features and their social impact.

Early observations of individuals with Down's syndrome (DS) suggest that people with this genetic condition have been present within society for many years. The first description of what was likely to be a child with DS is suggested to be that of Esquirol in 1838, although at this early point in history there was not yet recognition of its existence as a distinct medical syndrome. The term "furfuraceous idiocy" was used by Seguin in 1846 to describe a child with similar features to those noted by Esquirol, and shortly after in 1866 Duncan wrote of a girl who had a small round head, Chinese looking eyes, a large protruding tongue and a severely limited vocabulary (Pueschel, 1992). However, it was the British physician John Langdon Down (1866) who recognised the common pattern of physical characteristics and cognitive impairment present in these children, giving the condition a name and acknowledgement as a syndrome in its own right.

DS is now acknowledged as the oldest and most common recognised genetic syndrome causing cognitive impairment (Pennington, Moon, Edgin, Stedron & Nadel, 2003) affecting 1/732 live births (Canfield, 2006). A number of physical characteristics occur with high frequency in DS and as such are considered to be classical signs. These include: a small skull with a shortened anterior-posterior diameter; small oral cavity causing protruded tongue position; small ears; small nose with flattened bridge; upward-slanting eyes with marked epicanthic folds; a wide gap between the first and second toes; and reduced muscle tone (Pueschel, 1992). Whilst these are typical features of DS, individual presentation will vary considerably and “none of these features can be considered pathognomonic for this chromosome disorder” (Pueschel, 1992, p. 1). For this reason the only reliable method of diagnosis is genetic karyotyping, which enables identification of the distinct variant of DS present.

2.2 The genetics of DS

Despite DS being recognised for well over a century it was only in 1959 that LeJeune, Gautier and Turpin identified its genetic basis: the presence of an additional chromosome 21.

Our genetic make-up is stored in genes which are grouped together in long, thread-like structures called chromosomes. All body cells (with the exception of the sex cells) contain 46 chromosomes, grouped in 23 pairs. One chromosome from each of these pairs comes from the mother, and the other from the father. The pairs are numbered from 1 to 23 according to size, with chromosome 1 being the largest, and each has a long and a short arm. The 23rd pair differ in that they are the sex chromosomes, termed either X or Y and contain only a single set of 23 chromosomes. Thus when an egg cell is fertilised by a sperm cell the balance of 46 chromosomes is restored; the genetic material from this initial cell then divides to form all the cells required to produce human life (Kessling & Sawtell, 2002).

In DS there are 3 types of chromosomal abnormality:

- Trisomy 21
- Translocation
- Mosaicism

Trisomy 21 is the most common type of DS, occurring in approximately 94% of cases (Pennington, Moon, Edgin, Stedron & Nadel, 2003). It is also known as Regular, Standard or Free Trisomy 21. This occurs when an egg or a sperm cell contains an additional chromosome 21, which results in the presence of an extra chromosome 21 in every subsequent cell created (i.e. 47 chromosomes rather than the typical 46). The production of an extra chromosome in the egg or sperm cell can occur from uneven division of the chromosomes during either of the two phases of meiosis (cell division); the reason for this is as yet unknown (Kessling & Sawtell, 2002).

Translocation accounts for approximately 4% of cases of DS (Kessling & Sawtell, 2002). In such cases individuals do not have the whole of the additional chromosome 21, rather only a part of it which has attached itself (or translocated) to another chromosome. The chromosomes which can be involved are 13, 14, 15, 21 and 22 (Kessling & Sawtell, 2002) although only translocation of chromosome 21 produces DS. As in Trisomy 21, this genetic material will be replicated during cell division, causing all cells to be affected.

Mosaicism accounts for the remaining few cases of DS. It is characterised by the presence of the additional chromosome 21 in only some of the cells, resulting in a mixture of trisomic and normal cells. The extent of this mix can vary greatly and will have considerable impact on the severity and type of DS features experienced. Mosaicism arises from an aberration in cell division, where a chromosome goes astray, producing a single cell with the additional chromosome 21 (Kessling & Sawtell, 2002), this cell then continues to divide alongside the unaffected cells.

Chromosome 21 is believed to contain around 1% of the body's genes (Kessling & Sawtell, 2002). A recent advance has seen the mapping of this chromosome (Hattori, Fujiyama, Taylor, Watanabe, Yada & Park, 2000) and from this it appears that the number of genes present is approximately 225; a majority of these 225 genes are in the DS region on the long arm of chromosome 21 (Pennington et al., 2003). At present it is unclear which of the genes on chromosome 21 are over-expressed and with what specific results, however it has been speculated that trisomy causes developmental instability in a general way that does not allow for the specific phenotypic features to be traced back to extra doses of specific genes (Reeves, Baxter & Richtsmeir, 2001); although this is not a universally accepted proposition.

The number of genes involved in DS means that its genetic aetiology is more complicated than that of other genetic conditions such as Fragile X syndrome or William's syndrome (Pennington et al., 2003).

Unlike genetic hereditary syndromes which are passed on by a parent who carries the affected gene, only a very small percentage of cases of DS (1%) can be traced through family lines; in these cases all the individuals with DS have a rare form of translocation (Kessling & Sawtell, 2002). In such cases a parent who is a carrier has the standard two whole 21 chromosomes, but one of them is translocated onto another chromosome. Parents who are carriers are at an increased risk of having a second child with DS at any age. In contrast, the birth of a child with the Trisomy 21 type of DS is more common in older women (although as more women have children when they are younger there are naturally more births of children with DS to these younger women). The reason for the increased incidence of DS in older women is not known for certain, although it has been speculated that all women store some eggs with the extra 21 chromosome and that these eggs may be fertilised only towards the end of the woman's natural reproductive lifespan. A second theory is that older women have the same rate of trisomic conceptions as younger women, but are at a reduced risk of miscarriage (Kessling & Sawtell, 2002).

2.3 Intellectual disability

ID is defined as an IQ of less than 70, with further approximate bands to indicate the extent of impairment. According to Ring, Zia, Lindeman and Himlok (2007) the bands routinely employed in clinical practice in the UK are: 'mild' ID, between 50 and 70 IQ; moderate ID, between 35 and 50; severe ID between 20 and 35; and profound ID, <20. Although these bands provide a guide to intellectual function, severity should be considered alongside any compensatory or adversarial factors, such as a supportive environment which enables the child or adult to make fuller use of his or her potential (Abudarham, 2002).

Although a few individuals are reported to possess an IQ within the normal range (Fishler, 1975; Epstein, 1989) most adults with DS fall within the moderate to severe ID range (Gibson, 1978) and DS is recognised as the most common identified cause of intellectual disability (ID) in young children (Wishart, 1993), accounting for almost 40% of cases of moderate to profound ID (Pennington et al., 2003).

At birth, the brains of infants with DS have been found to be little different from those of infants without DS (Schmidt-Sidor, Wisniewski, Shepard & Sersen, 1990). However, differences begin to emerge within the first few months of life. Over the course of the first few years the phenotypic characteristics of the adult DS brain develop, including an overall reduced volume (microcephaly), with particular diminishment of the cerebellum, hippocampus, frontal lobes and prefrontal cortex (Pennington et al., 2003); although this is at odds with Pueschel (1992) who reports that true microcephaly is rarely observed in DS. By adulthood, brain abnormalities are invariably present "because virtually all adults with DS have developed some of the neuropathological features of Alzheimer's disease by around the age of 35" (Pennington et al., 2003).

2.4 MENTAL HEALTH

Individuals with generalised ID have been found to experience a greater incidence of psychiatric problems than those without ID. Factors such as higher levels of educational failure and family disruption are thought to be likely to cause increased stress leading to more frequent occurrences of personality disorders and neurosis (Dobson, 2002); although the contribution of biological factors has not been ruled out. Despite their higher prevalence, mental health problems can often remain undiagnosed as the resulting behavioural disturbances can be similar to those caused by the underlying condition or syndrome causing ID (Leudar & Fraser, 1985); which might suggest that the true extent of these problems is even greater than is presently reported.

Pueschel (1998) states that there is a high prevalence of psychiatric disorder in persons with DS and that it has been noted that there appears to be no neurobiological basis to suggest that, for example, major depressive disorder and DS are mutually exclusive (Khan, Osinowo & Pary, 2002). Compared to adults with ID from other aetiologies, adults with DS have been found to have a higher rate of depressive illness (Collacott, Cooper & McGrother, 1992). However, in contrast, children with DS have been reported to be at lower risk of psychopathology than children with generalised ID (Dykens, Shah, Sagun, Beck & King, 2002) and similarly adults with ID not caused by DS were reported to be six times more likely to experience psychiatric illness than adults with DS (Haveman, Maaskant, van Schrojenstein, Urlings & Kessels, 1994); although the latter study did exclude dementia in its description of psychiatric illness which may have affected findings.

There is considerable evidence of a significantly increased risk of dementia of the Alzheimer's type in DS, and of onset at an earlier age (Rowe, Lavender & Turk, 2006). A prevalence rate of 9.4% has been found in the fourth decade, with an increase to 54.5% in the sixth decade (Prasher, 1995) and it has been noted that the characteristic amyloid plaques and neurofibrillary tangles are found in almost all individuals with DS over the age of 40 (Mann, 1988). Features of dementia include impairments in short and long-term memory, abstract thinking, judgement, and other higher cortical functions, as well as changes in personality (Myers, 1992).

2.5 NEUROMUSCULAR CONTROL AND MOTOR FUNCTION

There is a general acceptance of the presence of motor impairment in DS and that this deficit is likely to result in the delayed attainment of general motor milestones such as walking, reaching and grasping (Hayley, 1986; Palisano, Walter, Russell, Rosenbaum, Gemus & Galuppi, 2001) and of vocal motor milestones, such as babbling (Cobo-Lewis, Oller & Lynch, 1996). Motor-developmental outcomes in DS have been found to be influenced by both the individual's genetic make-up and their learning environment (Flint, 1999, Nokelainen & Flint, 2004) making it difficult to identify a single causative factor; although such delays are often attributed to the presence of low muscle tone, also referred to as 'hypotonia' (Harris, 1985).

Strong evidence of hypotonia has been a finding of even the earliest studies of people with DS. McIntire and Dutch (1964) found great variation in the general presentation of eighty-six infants with DS but found that hypotonia was extremely common, being exhibited in all the major muscle groups in eighty-four of the infants (97.7%); a finding subsequently corroborated by Dummer (1978) and Griffiths (1976). Later research by Morris, Vaughan, and Vaccaro (1982) also found evidence of hypotonia in children with DS, and moreover identified a slight increase in muscular tone with increasing age indicating that hypotonia decreases as the child matures, echoing the much earlier findings of Penrose and Smith (1966). A positive correlation between muscular strength and muscle tone was also observed in both the TD children and children with DS; improvements in both being suggested to be related to increased physical activity as the children matured.

More recently (1994) used electromyography to evaluate the electrical activity which occurs when a nerve impulse triggers the contraction of an individual group of fibres in a muscle. It was found that individuals with DS require considerably greater effort to initiate the musculature of the larynx (a mean of 131.57 microvolts of energy in the DS group compared to a mean of 72.52 in the TD control group). Pryce notes that although this does not directly prove the presence of hypotonia, the greater energy required to set in motion the vibration of the vocal folds for the purpose of phonation is consistent with the greater level of energy required to initiate a more flaccid (hypotonic) mechanism.

2.6 GROWTH

The endocrine system is instrumental in regulating mood, growth and development, tissue function, metabolism, sexual function and reproductive processes. Historically, DS was suggested to be associated with a generalized endocrine disturbance, due to the high level of pathologic findings in almost every endocrine gland of an individual with DS during postmortem evaluation (Benda, 1949). However, more recent investigations suggest that most individuals with DS in fact do not have significant endocrine disturbances (Pueschel & Blaymore Bier, 1992).

Pueschel (1993) notes one of the main characteristics of children with DS as deficient linear growth. A longitudinal investigation of ninety children with DS identified that by 36 months the majority fell below the third percentile, with only 10% having a normal growth velocity (Cronk, 1978). Growth has been found to be most reduced between the ages of 6 months and 3 years, becoming in line with norms after this period (Cronk, Crocker, Pueschel, Shea, Zachai, Pickens and Reed, 1988; Sara, Gustavson, Annerén, Hall and Wetterberg, 1983). Studies have identified that there is no obvious deficiency in the growth hormone serum itself (Annerén, Sara, Hall and Tuvemo, 1986) and it has been suggested that the excess genetic material resulting from trisomy of chromosome 21 may cause primary and/or secondary effects which interfere with the normal pattern of growth (Pueschel, 1993).

2.7 THYROID DISORDER

Thyroid dysfunction is a well examined aspect of DS; the most common finding being hypothyroidism which is decreased thyroid hormone production (Criscuolo, Perrone, Sinisi, Bellastella & Faggiano, 1986; Cutler, Benezra-Obeiter & Brink, 1986; Dinani & Carpenter, 1990). The symptoms include tiredness, weight gain, mental slowing, feeling cold and aches and pains. In infants low levels of thyroid hormones can result in decreased growth, delayed development, low muscle tone, dry skin, constipation and an enlarged tongue; all of which are commonly found in infants with DS (Leshin, 2005). Similarly, as individuals with DS mature they tend to be shorter than average height, slower and less active (thus having a tendency to gain weight) and have coarser skin and rougher voice quality (Dinani & Carpenter, 1990). This overlap in the typical clinical presentation of DS and hypothyroidism makes early and accurate diagnosis of thyroid disorder difficult; for this reason regular assessment of thyroid function is recommended (Dinani & Carpenter, 1990; Pueschel, Jackson, Giesswein, Dean & Pezzullo, 1991).

More recently a contrasting opinion has been proposed by Prasher and Haque (2005) which suggests that healthy individuals with DS are being diagnosed with thyroid disorder as a consequence of inappropriate comparison with normal reference ranges for the general population. The authors argue that the present TD diagnostic standards fail to take into account factors such as the possible effects on thyroid function of the premature aging which is seen in adults with DS and that given comparison to an appropriate DS reference group these individuals would demonstrate ‘normal’ thyroid function. Evidence for their proposal comes from their study of 110 healthy adults with DS, believed to be representative of the total DS population. Over a quarter of those who displayed no clinical symptomology of thyroid disorder and who generally appeared to be fit and healthy were found to have abnormal plasma thyroid hormone levels (Prasher and Haque, 2005).

Treatment of hypothyroidism consists of the replacement of the natural thyroid hormone with synthetic thyroxine; this treatment is usually life-long (Leshin, 2005). Leshin also notes that the treatment can result in a child who was previously overly calm and sedate due to the influence of hypothyroidism being perceived as ‘hyperactive’, when in fact he or she has merely returned to his or her natural behavioural state.

2.8 CARDIOVASCULAR FEATURES

The frequency of congenital heart disease in DS is purported to be very high (Marino, 1992). The Atlanta Down Syndrome Project (Freeman, Taft, Dooley, Allran, Sherman, Hassold, Khoury & Saker, 1998) report congenital heart defects in 44% of 227 infants observed in a population-based study of DS, whilst a slighter higher rate of 50% is reported by Marin, Rosenbaum and Sardegna (1989); these figures compare to only 0.3% in children without chromosomal abnormalities (Ferencz, Rubin, McCarter, Brenner, Neill, Perry, Hepner & Downing, 1985). A higher incidence has also been found in individuals with DS compared to those with an intellectual disability of other aetiology (Chaney, Eyman & Miller, 1985). It is thought that a locus on chromosome 21 is involved in the development of cardiac defects in DS, although environmental factors and other genes are likely also to play a role (Vis, Duffels, Winter, Weijerman, Cobben, Huisman & Mulder, 2009). Interestingly, adults with DS have been found to not be significantly more at risk of acquired cardiac disease than those with ID of other aetiologies or the general population (van den Akker, Maaskant & van der Meijden, 2006) possibly due to relatively low levels of atherosclerosis (Marino, 1992).

2.9 VISUAL IMPAIRMENT

Visual impairment is common in DS and prior to the introduction of karyotyping ocular findings were of great significance in making a positive diagnosis (Catalano, 1992). There is a higher risk of both congenital cataracts and early developing cataracts and people with DS are also more likely to have long or short sight, blepharitis (inflammation of the margins of the eyelids) sticky eyes and conjunctivitis (Woodhouse, 2002). Additionally, keratoconus, which is a distortion of the cornea (Cullen, 1963), strabismus (Jaeger, 1980; Shapiro & France, 1985) a squint caused by the axes of the eyes not being parallel (Lindsay & Bone, 2004) and nystagmus (ocular ataxia) characterised by an involuntary 'to and fro' eye movements (Shapiro

& France, 1985; Wagner, Caputo & Reynolds, 1988) have all been found to be prevalent.

At birth, many TD babies are long or short sighted but over the first few years their sight gradually adjusts to within normal limits. Woodhouse (2002) notes that babies with DS tend to be born with a similar spread of refractive errors but that unlike TD infants in some these deficits do not fade over time; consequently by school-age approximately 40-45% of children with DS will require glasses to correct their vision.

2.10 HEARING IMPAIRMENT

Hearing loss has been found to be more common in individuals with DS than in both those without ID (Brooks, Wooley & Kanjilal, 1972) and those with ID not resulting from DS (Dahle & McCollister, 1986; van Schrojenstein Lantman-de Valk, Haveman & Crebolder, 1996). It is the most common of the ENT pathologies associated with DS (Venail, Gardiner & Mondain, 2004) and although estimates of prevalence vary, it has been reported that as many as 78% of people with DS have some degree of loss (Balkany, Downs, Jafek & Krajicek, 1979).

Of the 107 children and adults with DS evaluated by Balkany et al. (1979) the identified hearing loss was predominantly conductive in nature (83%), falling in the range of mild to moderate loss, echoing the findings of other early otologic studies (Schwartz & Schwartz, 1978; Keiser, Montague, Wold, Maune & Pattison, 1981). Conductive hearing loss is consistent with middle ear abnormalities. Children with DS have been frequently found to have impacted wax due to the particularly narrow ear canal, retracted tympanic membranes and middle ear effusion (Dahle & McCollister, 1986). Venail, Gardiner and Mondain (2004) report that 90% of cases of conductive loss are caused by otitis media with effusion (OME) also known as glue ear. OME is known to be more common in DS and it is speculated that this is due to anatomical and functional differences such as poor immune defence mechanisms and abnormalities of the eustachian tube resulting in poor drainage (Clarke, 2005). The eustachian tube runs from behind the eardrum, draining down into the nasal cavity. It normally opens into the nasopharynx only when muscles in the palate contract (e.g. during swallowing and yawning). People with DS are suggested to have what is termed a 'patulous' eustachian tube, whereby it is constantly in an open position, thus increasing the risk of chronic ear infections (Loury, 2006). Hypotonicity is proposed to be a factor in the poor function of the eustachian tube (Dahle & McCollister, 1986) affecting ability to achieve and maintain adequate closure.

The treatment of OME involves the insertion of ventilation tubes called grommets, which aid drainage of fluid. These require regular replacement as long as OME is present, however this increases the risk of perforation of the tympanic membrane; 'T' tubes can be used as a longer term option, reducing the risk of this complication (Venail, Gardiner & Mondain, 2004). Untreated or undertreated OME results in mastoiditis (inflammation of the mastoid air cells) which in acute states can lead to sclerosis of these cells; a finding observed by Roizen, Martich, Ben-Ami, Shalowitz and Yousefzadeh (1994) in 42% of 53 children with DS. Balkany et al. (1979) observed ossicular abnormalities arising from inflammation in 60% of the children evaluated (including malformation or erosion of the malleus, incus and stapes) but noted that congenital abnormalities of the stapes were also found in DS.

Although conductive hearing loss is most common, sensorineural hearing loss, or a mixed loss, also occur. There appears to be a correlation between age and sensorineural loss: of thirty-eight individuals with DS below the age of 21 a sensorineural element to their loss was found in 21% (compared to 15% of TD controls), whereas a much higher incidence (55%) was found in forty individuals with DS over the age of 21 (compared to only 10% of controls). This suggests that rather than being of a congenital aetiology sensorineural loss develops over time, possibly as a result of long-standing middle ear disease (Brooks, Wooley & Kanjilal, 1972). This concurs with more recent findings of mild sensorineural loss mostly being owed to early presbycusis, which is hearing loss as a consequence of the natural aging process (Nespoli, Burgio, Ugazio & Maccario, 1993). As in the TD population, sensorineural loss is treated with hearing aids, cochlear implants and speech and language therapy (Venail, Gardiner & Mondain, 2004).

Studies of hearing loss in otherwise TD children have identified that even a mild loss has been found to affect all aspects of development. This includes delay in language (Holm & Kunze, 1969), emotional and educational development (Menyuk, 1977) and interpersonal skills (Needleman, 1977); clearly, the effects of a hearing loss on children who are already disadvantaged by ID will be even more profound. “As rehabilitation of hearing can bring about dramatic improvements in the quality of life of both child and family” (Clarke, 2005), and as children with DS are accepted as being at a high risk of hearing impairment, regular otological evaluation is crucial.

2.11 SPEECH AND LANGUAGE PROFILE

A considerable amount of research has been done to investigate the development of speech and language in DS. The following is a summary of some of the features found in key stages of development as well as some of the contributing factors.

For typically developing (TD) infants, the first 18 months of life are considered to be the prelinguistic stage of development where the basic underlying principles of communication begin to be explored. According to Jenkins and Ramruttun (1998, p.53) prelinguistic communication “refers to the rich interpersonal, intentional, meaningful and essentially non-linguistic communication that characterises early face to face interactions between infants and their care givers”. In Down’s syndrome (DS) this prelinguistic stage is extended with evidence showing that children with DS may rely on prelinguistic strategies for the first 2-3 years of their lives (Rondal, 2003). It is within this stage that children begin to develop more mature cognitive and sensorimotor processes based on human interaction and the physical exploration of their environment. The delayed motor development of children with DS, which is most commonly associated with low muscle tone (hypotonia), together with intellectual disability (ID), degrades the ability of these infants to access more than their immediate physical environment. This reduced developmental experience is believed to have a significant impact on cognitive development (Rondal, 2003) which has implications for the rate and quality of future linguistic development.

Despite slower developmental progression, research has shown that infants with DS are primed to attend to human voices in the same way as their peers, which according to Glenn, Cunningham and Joyce (1981) is an important factor in the development of receptive language. Similarly, the early babbling sounds used by infants with DS, which are recognised as a good predictor of later language (Stoel-Gammon, 2001), although perhaps being slightly behind that of their peers (Lynch, Oller, Steffens & Buder, 1997) are recognised to follow more-or-less the same patterns as TD infants (Dodd, 1972; Smith & Oller, 1981) suggesting that babbling may not be a learned behaviour and therefore not determined by cognitive ability.

Linguistic imitation is also a skill which contributes towards the early development of language, helping the child to develop new vocabulary (Rodgon & Kurdek, 1977) and grammatical structures (Clark, 1977). Furthermore linguistic imitation enables the child to interact with another person in a manner which reflects the natural turn-taking of more mature conversation, at a stage before their language skills allow for this. Children with DS have been shown to imitate the speech sounds of their parent less than their peers (Rondal, 1980; Sokolov, 1992) which Sokolov suggests reflects fundamental differences in the interactions between young children with DS and their carers. Parents of children with DS have been found to use more directive language than parents of TD children (Cardoso-Martins & Mervis, 1985) and the use of less linguistic models which are appropriate for imitation can thus reduce the opportunity and motivation for children with DS to imitate (Snow, 1989).

Children with DS have also been found to have a heavier reliance on gestures than their TD peers (Greenwald & Leonard, 1979; Legerstee, Varghese & van Beek, 2002) and to retain gesture for longer, even increasing the amount and complexity at a time when TD peers are favouring more verbal strategies (Caselli, Vicari, Longobardi, Lami, Pizzoli Stella, 1998). However, this nonverbal specialisation is contended by other research which finds no reliable differences between the gestures used by children with DS and their peers and that children with DS actually have smaller gestural repertoires than peers (Iverson, Longobardi & Caselli, 2003). Regardless, it has been observed that children with DS find gesture to be a particularly helpful strategy in supporting understanding of spoken language (Caselli et al., 1998) as well as helping them to express their wants and needs well before they begin to use formal verbal communication (Kumin, 1996).

Despite some inconsistencies in research findings it appears that children with DS may not be significantly more disadvantaged than their TD peers prior to the development of speech. However, with the onset of verbal communication children with DS begin to show marked differences from their TD peers, with the extent of language difficulty being greater than would be indicated by cognitive level (Wisniewski & Miezejeski, 1988). Language comprehension has been found to correlate with nonverbal mental age, and to be less impaired than expressive language (Chapman & Hesketh, 2001); verbal expressive difficulties being recognised to cause considerable frustration for children who struggle to be understood (Kumin, 1996). Marked deficits are apparent in syntax (Caselli et al., 1998) especially in the area of morphology (Chapman, Schwartz & Bird, 1998) whilst semantics has been shown to be a relative strength (Grela, 2002). People with DS have been observed to continue to develop lexical and syntactic skills through adolescence, suggesting that there is not a critical period of language acquisition (Chapman, Schwartz & Bird, 1998); a finding which has marked implications for continued therapeutic and educational input into adulthood.

The presence of ID will have a significant effect on language development, however other factors are also considered to be contributory. Reduced opportunities for language learning are proposed to occur as a consequence of infants with DS being less able to explore their environment due to differences in motor development (Rondal, 2003) as well as from receiving fewer appropriate language models by parents who tend to use more directive speech than with TD children (Chapman, Schwartz & Bird, 1998). Hearing loss is believed to account for some of the difficulties observed (Chapman, Schwartz & Bird, 1998) and is further compromised by poor levels of attention (Landry & Chapieski, 1990). Finally, auditory short-term memory has been found to correlate with expressive language deficits and children with DS shown to score less highly than peers in tests of this type (Hick, Botting & Conti-Ramsden, 2005).

Language deficits are compounded by difficulties in phonological acquisition and articulation which occur frequently in DS. Children with DS are suggested to use a similar range of phonological processes as TD children but to retain them for much longer (Shriberg & Widder, 1990) which according to (Stoel-Gammon, 2001) can cause unintelligibility to persist into adulthood. There is also some evidence of differences in ability to discriminate between phonological contrasts (Eilers & Kimbrough Oller, 1980; Keller-Bell & Fox, 2007) indicating that errors are not only expressive in nature. The pattern of phonological simplification strategies observed in DS has prompted the proposal that development follows a typical path, however Dodd and Thompson (2001) contend that the greater variability in speech productions by people with DS indicates disordered rather than delayed phonological acquisition. Articulation errors are also frequent in DS (Stoel-Gammon, 1980) and more severe than would be predicted by cognitive ability (Kumin, 1996). Errors most commonly affect fricatives, affricates and liquids (Bleile & Schwarz, 1984) and tend to be eliminated at a slower rate than is seen in TD children.

Anatomical and functional differences, such as a small oral cavity and relatively large tongue, and muscle weakness as a consequence of hypotonia are proposed to be significant factors in articulation errors (Stoel-Gammon, 2001) although more recently apraxia of speech, which affects motor-planning and motor-coordination, has been suggested as another possible factor (Kumin, 2006). Hearing impairment and auditory short-term memory deficits may reduce the quantity and quality of phonological representations (Hulme & Mackenzie, 1992) whilst a failure of adults to correct variable productions may not help children with DS to realise that consistency is crucial for intelligibility (Dodd & Thompson, 2001). "The speech difficulties associated with Down syndrome are thought to result from impairments in almost all of the systems required for successful speech production…this makes it extremely difficult to design interventions for improving speech in persons with Down syndrome" (Timmins, Cleland, Rodger, Wishart, Wood & Hardcastle, 2009, p. 20).

2.12 SOCIAL INTERACTION AND SOCIAL ACCEPTANCE

Children with DS are proposed to demonstrate a profile of more linguistic than cognitive impairment, and to have relative strengths in social domains (Beeghly, Weiss-Perry & Cicchetti, 1990). The ability to optimise this social competence is crucial as these skills influence considerably the extent to which individuals with DS are able to adjust to vocational or living settings as adults (Sigman & Ruskin, 1999) as well as influencing their ability to access education and interact with their peers through their younger years.

2.12.1 Pragmatic ability in DS

Pragmatics is the study of the social use of language, including awareness of social rules such as the ability to turn-take in conversation, maintain appropriate eye-contact and physical proximity, as well as the ability to interpret the intentions or beliefs of others. Therefore pragmatics can be said to encompass both the use and understanding of verbal and non-verbal language within a social context. Increasingly, more emphasis has been placed upon the investigation of these skills as it is believed that communicative functioning significantly predicts later social and adaptive competence (Spiker, 1990).

The term 'speech acts' is used to encompass a range of functional communicative behaviours, such as obtaining information, making requests and social contracts, and sharing beliefs, which can be expressed both linguistically and non-verbally within interactions (Abbeduto & Keller-Bell, 2003). When matched with both children with similar mental age (MA) and children with similar grammatical ability as measured by mean length of utterance (MLU), children with DS were found to produce a greater variety of speech acts than the children matched for MLU, but not to differ significantly from the MA matched group, thus demonstrating asynchronous pragmatic and syntactical development (Beeghly, Weiss-Perry and Cicchetti, 1990). In this study Beeghly and colleagues also noted a specific and significant difference in the 'requesting behaviour' of the children with DS who made fewer requests than MA matched controls, performing more in line with those matched for MLU. There have been conflicting findings on the ability of children with DS to make verbal and nonverbal requests: evidence of a specific requesting deficit in DS was proposed by Mundy, Kasari, Sigman and Ruskin (1995), while more recent research suggests that requesting emerges at the expected time (Moore & Oates, 2000).

Young children with DS have been found to be able to make use of contextual information to make inferences about speech acts in order to understand what is expected of them in any given interaction (Leifer & Lewis, 1984) but without a familiar context or sensitive communication partner the ability to recognise speech acts may be significantly compromised (Abbeduto & Keller-Bell, 2003). It has been observed that children with DS have demonstrated less mature communicative behaviour in less structured settings or settings with peers than in structured settings with adults (Beeghly, Weiss-Perry & Cicchetti, 1990). For example, within the mainstream school environment, children with DS have been shown to fail to observe established conversational codes, such as maintaining mutual gaze and responding to initiations by peers (Sinson & Wetherick, 1982); underlying difficulties in visual attention, arousal level and information processing being potential contributing factors to social deficits of this type (Wagner, Ganiban & Cicchetti, 1990).

The ability to utilise situational and linguistic information is crucial for the listener to establish the referent (or focus) of the speaker's utterance and contributes significantly to the successful completion of the desired speech act. Similarly, the speaker must be aware of the need for the listener to share a joint understanding of what is being spoken about; for example, a communication breakdown is likely to occur should the speaker fail to declare the intended referent (e.g. '*my mum', 'the dog'*) before utilising pronouns such as '*she'* and '*it'*. This ability to make referents clear to others is a well documented area of deficit in DS and in other adults with intellectual disabilities (ID), with ability being poorer than would be expected from non-linguistic function (Abbeduto, Weissman & Short-Meyerson, 1999).

Closely linked to these skills is the ability to recognise, maintain and advance the conversation topic. This requires collaboration between the speaker and listener to ensure that utterances are appropriate both to the general theme and, more specifically, to the sense of the preceding utterance (Abbeduto & Keller-Bell, 2003). Early studies of discourse skills identified that children with DS interacted in significantly longer sequences of topic-appropriate turn-taking than MLU matched controls, not differing significantly from MA matched controls (Beeghly, Weiss-Perry & Cicchetti, 1990). Although more recently, children with DS were found to have reduced conversational repertoires (Berglund, 2001) and to be less likely to introduce new topics during conversation with their mothers (Tannock, 1988). However, Abbeduto and Hesketh (1997) question whether increased parental control during conversation may be a factor in this finding. It has also been proposed that lower motivation in people with DS (Abbeduto & Keller-Bell, 2003), the degree of ID and the potential for reduced quantity and quality of opportunities to develop these skills (Abudarham, 2002) may contribute towards the identified deficits in social behaviour.

2.12.2 Comparison of social abilities between DS and TD children and children with other developmental disorders

The behavioural effects of genetic disorders have increasingly become a focus of research in recent years, seeking to investigate whether there are uniform effects relating to specific syndromes.

In comparison with children with Prader-Willi syndrome (PWS) and Williams's syndrome (WS) Rosner, Hodapp, Fidler, Sagun and Dykens (2004) found that children with DS had the lowest ratings in skill and time spent in activities, possibly reflecting the lower cognitive function of this group. However, they also demonstrated high sociability scores, showed good interaction and low levels of maladaptive behaviours; supporting earlier similar findings in comparison to adults with generalised ID (Collacott, Cooper, Branford & McGrother, 1998). Moreover, higher social competence and activity competence scores were moderately associated with increasing chronological age, which might suggest that individuals with DS continue to learn skills into adulthood; a finding which has implications for continued clinical intervention into adulthood.

In relation to specific language impairment (SLI), WS and TD peers, young people with DS were found to demonstrate relative strengths in social communication, with pragmatic composite scores being above the level indicative of pragmatic language disorder (Laws & Bishop, 2004). Specific difficulties were identified in the coherence of narratives, although it was noted that this could be associated with the speech production and grammatical deficits observed in DS. These individuals with DS were not reported to have unusual or restricted range of interests, and significantly, unlike the WS and SLI groups, were judged as having social relationships at a level equivalent to the TD controls; although a preference for interaction with adults rather than peers was noted.

In contrast, research by Sigman and Ruskin (1999) comparing the social interaction skills of young children with DS, autism and generalised ID found that despite the stereotype of overt sociability in DS the children with generalised ID engaged in more high level play (e.g. organised games/turn-taking activities) as opposed to low level parallel play than the children with DS and that no differences were found between those two groups for initiating interactions (the group with autism scoring lowest in both measures). The three groups did not differ in ability to maintain those interactions.

Unsurprisingly, social deficits were also found in later research comparing the social abilities of children with DS to their TD peers. During free play children with DS were noted to make less initiations, to play in a more solitary manner, to have a preference for adult company (Virji-Babul, Hovorka & Jobling, 2006) to spend more time in passive behaviour (Linn, Goodman & Lender, 2000) to be less likely to adapt their level of interaction in an appropriate way to different activities and to show less interest in co-operative play (Hamilton, 2005).

People with DS have been shown to be significantly better than those with autism at matching emotions to facial expressions Celani, Battacchi and Arcidiacono (1999) and to perform better than those with autism when matching expressions to the corresponding voice (Loveland, Tunali-Kotoski, Chen, Brelsford, Ortegon & Pearson, 1995). More recently Williams, Wishart, Pitcairn and Willis (2005) evaluated emotion recognition ability in comparison to TD children and children with non-specific ID. Results identified that children with DS were significantly poorer than both the other groups in their ability to match photographs of unfamiliar adults on the basis of displayed emotion. Williams et al. (2005) suggest that deficits in emotion-recognition are a consequence of the neurological profile of DS and question the proposal of uniform strengths in social skills. Emotion recognition is a skill which begins early in life with TD infants of less than one year being able to discriminate between happy and sad faces (Haviland & Lelwica, 1987), and by approximately 4 years of age being able to recognise and label a wider range of emotions (Denham & Couchoud, 1990; Serrano, Iglesias & Loeches, 1995). Children with DS have been found to recognise emotion at a level similar to that of MA matched TD children of approximately 3 years of age, however, by the age of 4 years the TD and ID children have been shown to outperform children with DS (Kasari, Freeman & Hughes, 2001) which indicates that despite increases in MA a comparable growth in emotion recognition does not occur. Kasari et al. propose that this indicates both aetiological and developmental differences in the emotion recognition skills of children with DS. Recognition of how emotions are represented in facial expressions is a key skill in the ability to form social relationships, with indices of children's emotional knowledge being closely correlated to peer and teacher ratings of likeability and friendliness (Denham, 1998).

Despite specific deficits in emotion recognition individuals with DS are proposed to demonstrate communicative strengths utilising the visual channel. Research suggests that visual memory is more effective than auditory memory, and consequently children with DS may find it easier to communicate through gestures than spoken words (Buckley, 1993). Children with DS with a MA of between 11 and 17 months, who have had both sign and spoken input, have been found to have larger combined vocabularies than MA matched TD children, thus illustrating that there is an advantage in early vocabulary development associated with the use of sign (Clibbens, Powell & Atkinson, 2002). Similarly, children with DS aged 3 years who received systematic sign input were found to have vocabularies comparable to children with DS, aged 4 years, who had had no sign input (Launonen, 1996). Moreover, significant gains in social communication, speech and language, self-help skills and cognitive and motor development were reported subsequently for the sign group, implying wider benefits from sign input. Therefore it is unsurprising that a 'total communication' approach, involving the use of alternative or augmentative communication systems has proved effective in aiding expressive and receptive communication for individuals with DS (Kumin, 1994). For children who go on to develop an adequate system of spoken language these transitional techniques diminish in favour of verbal communication (Kumin, 1996) but even for those with relatively good speech, signing can still contribute towards increased intelligibility into adulthood (Powell & Clibbens, 1994). Due to the considerable heterogeneity of this population, some individuals with DS will always require AAC support (Kumin, 1996) making their primary mode of communication different to that of their TD peers and, in an environment where not all children or adults may be familiar with AAC, increasing the likelihood of social isolation.

2.12.3 Social interaction in school and peer perception of children with DS

Increased educational inclusion has resulted in more individuals with DS being taught in mainstream schools rather than schools specifically for children with special educational needs. As a result, much consideration has been given to meeting their educational and curricular needs, with however, relatively little focus on social needs, despite this being the area about which parents and carers are often most concerned (Cuckle, 1997).

It has been suggested that children with developmental delays interact with peers more frequently in integrated rather than specialised schools (Guralnick, Connor, Hammond, Gottman & Kinnish, 1996) and develop more positive social exchanges (Guralnick & Groom, 1987) perhaps due to exposure to increased models of appropriate social behaviours which promote the acquisition of effective social skills (Odom & Diamond, 1998). However, as research has identified that children with DS between 8 and 11 years, despite being selected often as work-partners at school are seldom invited home or considered as 'best friends' (Laws, Taylor, Bennie and Buckley, 1996) and more recently that TD children, if presented with the opportunity, would choose to interact with another TD child rather than a child with DS (Hamilton, 2005) then there is a clear risk of social exclusion. In a review of the existing literature Odom and Diamond (1998) confirm this divide and note that children with ID are often involved in more solitary and less co-operative play, and may fail to take account of contextual cues to interpret when it is, or is not, appropriate to interact with peers. Taken together, these factors put children with developmental disabilities at risk of being segregated within what are supposedly integrated or inclusive education environments. Moreover, it is notable that according to a study of teenagers during the 1980s and 1990s, that the single variable relating to interpersonal relationships in which children in special schools possessed an advantage over their ID peers in mainstream is that special schools allowed for real, reciprocated and supportive relationships based on shared interests and abilities (Buckley & Bird, 2000).

The transition to secondary school, where traditionally children begin to assert increasing independence and develop important social relationships can be a particularly sensitive time for individuals with developmental disabilities. Young people with DS are likely to have had less experience of being independent from their parents and carers, often relying on them for opportunities to interact socially (Sloper, Turner, Knussen & Cunningham, 1990), and may appear socially, physically and developmentally less mature than their peers (Cuckle & Wilson 2002). Additionally, the dynamics of the secondary school are likely to be very different from that of primary education, and may involve a new peer group, as well as changes to the social and educational support network offered by the school.

According to research by Cuckle and Wilson (2002) young people with DS have been shown to have strong and appropriate notions of what constitutes friendships, although the emotional immaturity of some of those youngsters was indicated in talk about social activities which were more appropriate to much younger children (such as playing with dolls). Friends were perceived as important people in their lives, and tellingly, most said they would like to have more friends. This research also identified that the people with DS in mainstreams schools with special provisions for individuals with ID (including identified bases to register at and join mainstream lessons from, specialist support from learning support assistants during lessons and integrated lunchtimes) tended to have more friends than those attending less well-resourced mainstream schools; although, support workers identified that those friends also tended to have special needs, and this was confirmed by the individuals with DS themselves. In both types of schools adult direction was often required to include the person with DS in the classroom or at lunchtimes and individuals with DS rarely chose TD study-partners or included themselves in groups, demonstrating social passivity.

Social relationships are particularly important during the teenage years. The findings of the Cuckle and Wilson study further confirm that there is a genuine danger of social isolation for adolescents with DS within mainstream schools, particularly if strategies are not in place to support integration.

2.12.4 Parental and professional perception of ability and disability in DS

In contrast to other syndromes and developmental disorders causing ID, parents of children of children with DS have been found to feel less stress (Hodapp, Ricci, Ly & Fidler, 2003) and more reward (Hodapp, Ly, Fidler & Ricci, 2001) whilst their siblings exhibit fewer adjustment problems (Rodrigue, Geffken & Morgan, 1993). Furthermore, children with DS are often described in positive terms, using adjectives such as affectionate, cheerful and readily pleased (Fidler, 2003).

With regard to physical features, young people and adults with DS have been found to retain certain immature craniofacial characteristics and it has been suggested that these may also contribute to perception of character. Fidler (2003) notes that the typical DS craniofacial findings reported by Allanson, O'Hara, Farkas and Nair (1993) bear a strong resemblance to those which Zebrowitz (1997) describes in a craniofacial phenomenon termed 'babyfaceness' which is typified by infant-like features including a small nose, larger forehead with a sunken bridge, fuller cheeks and a rounder chin. Zebrowitz observed that people associated these features with positive attributes such as being affectionate, warm and honest, but also with more negative traits such as weakness naivety and gullibility' (which may reflect perception of immaturity). Further to this, Fidler (2003) went on to identify that the speech patterns of parents with DS, when speaking to their children, tended to reflect those used by parents of younger TD children, thus also suggesting the perception of immaturity. Perceived immaturity is likely to lower expectations (Fidler, 2003) which may result in children with DS not attaining their full developmental potential. Moreover, Fidler argues that if this is the case, parents may not actively discourage immature social behaviours, such as displays of over-affection, which may negatively affect the emotional and social growth of children with DS, having negative consequences for self-esteem and mental health in the future.

Parents of children with DS have been found to be no different from parents of children with generalised developmental delay with regard to their attitudes towards the disabilities of their children, preferences for support and satisfaction with services provided (Reiter, Tirosh, Bar-Tikvah & Adam, 1992). The vast heterogeneity of people with ID has seen considerable variation in parental preference within a range of services; most notably educational provision. It has been found that parents of children with developmental delays are unlikely to hold similar views on inclusive education based merely on the underlying aetiology of the developmental delay, rather parents tend to consider specific needs and benefits to the individual child (Palmer, Borthwick-Duffy, Widman & Best, 1998). This suggests that service providers should primarily be focussed on the individual wants and needs of the family, rather than constructing care packages which are tailored to specific aetiologies (Reiter, Tirosh, Bar-Tikvah & Adam, 1992). Differences in preference for inclusive or specialised services can be influenced by many variables, including the degree of ID, behavioural problems or physical or medical needs. Similarly, parents may weight academic success and social integration differently (Palmer, Borthwick-Duffy, Widman & Best, 1998). Some may feel that their child may benefit most from a specifically adapted curriculum and more intensive teaching support in special education, whilst others may perceive the greatest benefits to their child in mixing with TD children who may serve as good social role models and friends. In a study of the attitudes of parents with autism and DS to inclusive education the parents of children with DS were found to be significantly more likely to support full-time inclusive placements for their children than the parents of children with autism, who preferred the option of part-time inclusive education (Kasari, Freeman, Bauminger & Alkin, 1999). Furthermore, the existing educational placement and age of the child were found to influence preferences; those parents with children in special education were less positive about inclusive practices than those already accessing mainstream schools, and parents of younger children were more likely to endorse inclusion than those with older children.

Parents of children with DS tend to recognise and accept their child's academic limitations and consequently are often reported to be satisfied with academic progress at school, whether in mainstream or special education; however it has been identified that they may continue to have very real concerns over social development and the formation of friendships (Cuckle & Wilson, 2002). Parents can often face a very difficult dilemma, desiring to encourage social development and independence, yet fearing inappropriate friendships and relationships borne out of naivety and social immaturity (Cuckle & Wilson, 2002) which may limit the opportunities open to young people with DS to create friendships and mature socially alongside their TD peers.

It has been reported that parents sometimes perceive professionals as overly negative and at times patronising and unable to perceive the true impact of disability on the family. This issue was examined by Bhattacharya and Sidebotham (2000) in a questionnaire-based study of the levels of disability of children with DS. They found significant discrepancies between parental and professional judgements in a number of areas. Parents rated their child as having more disability in the areas of vision, behaviour, health and growth and personal care but less disability in terms of speech and language. Familiarity with their child's individual communication strategies is likely to account for the parental perception of relative communication strengths. With regard to behaviour, doctors identified behavioural difficulties in 55% of children with DS, while parents identified difficulties in 81%, which seems to indicate that professionals underestimate the difficulties experienced by parents. However, according to Coe, Matson, Russell, Slifer, Capone, Baglio and Stallings (1999) more agreement has been found between parents and teachers, who in their research both rated children with DS to present behavioural problems at a level exceeding that of the TD children rated by a margin of 3:1. It is possible that children may not demonstrate behavioural problems during more brief assessments and therefore some professionals may not observe an accurate reflection of the child's habitual behaviour, thus there is a clear need for the right questions to be asked to determine the real difficulties (Bhattacharya & Sidebotham, 2000).

Both public and professional perceptions of disability have changed in recent years. Hutchison (1995) proposes that differences in professional perspectives may influence perception of ID, with those professionals who have a background rooted in the 'medical model' focussing on deficits, whilst parents and professionals from educational and social welfare backgrounds may be more likely to look beyond impairment and consider the child's future potential. Bhattacharya and Sidebotham (2000) state that in their view a combination of the medical and social models is the most desirable course as this enables both identification of disability and implementation of therapeutic and educational measures which will help to minimise the impact of disability. Thus the most effective form of assessment will utilise formal and informal testing by trained professionals as well as parental reports to build up an accurate picture of individual strengths and weaknesses.

2.13 VOICE CHARACTERISTICS

2.13.1 Introduction

This section will give an overview of the process of voice production and the role of voice in communication, summarising from the literature the particular qualities which are associated with the voices of typically developing individuals and individuals with DS, the possible reasons for those differences and the subsequent impact of atypical vocal features.

2.13.2 What is voice?

Voice quality is a result of a speaker's individual muscular adjustments to the whole of the vocal apparatus, including the lungs, larynx, pharynx, tongue, jaw and lips. These adjustments, or 'voice settings', are learnt over time and are often consciously or subconsciously influenced by the social norms which are representative of the speaker's environment. The result is distinctive, habitual patterns which contribute to the recognisable features that identify speakers from each other. The degree to which a speaker can manipulate their voice quality is constrained by the physical properties of the vocal apparatus (Laver, 1980). For example, a relatively small oral cavity may cause the tongue to sit in a more anterior position, and it may be difficult for the speaker to actively change the resulting voice quality. Thus, it can be summarised that an individual's distinctive vocal characteristics are dependent both on the anatomical structure of the vocal tract and the speaker's unique vocal settings (Mathieson, 2001).

2.13.3 Anatomical properties of the vocal tract

The human vocal tract consists of the upper and lower respiratory tracts. The upper tract includes the larynx, pharynx and the oral and nasal cavities, whilst the lower portion contains the trachea, bronchi and lungs. The structures within these two aspects of the vocal tract are generally referred to according to their relative positions from the vocal folds; those above the vocal folds are termed 'supraglottic' and those below, 'subglottic'. The configuration of the vocal tract, and the interaction between individual structures, has a considerable bearing on the make-up of vocal characteristics.

In the following sections the contribution of each structure to the process of voice production will be examined individually, using the anatomical labels favoured by Mathieson (2001).

2.13.3.1 <u>The subglottic tract</u>

The **trachea** is the portion of the airway to and from the lungs. It comprises of open rings of cartilage which are supported by fibroelastic membranes. It begins below the cricoid cartilage of the larynx and divides, left and right, into the lungs via the bronchi.

The **lungs** are situated in the chest cavity or thorax, receiving protection from the surrounding bone-structure. They are attached to, and supported within the rib cage by a series of muscles which allow for the expansion and contraction which correspond to the inspiration and expiration sequences of the respiratory process. Inspiration is enabled by the active contraction of the inspiratory muscles which overcomes the resistance of the lung and chest wall, enabling the diaphragm to lower to allow increased thoracic volume. The upward movement of the diaphragm in expiration is passive; rather it is the expiratory muscles which pull the ribs down, diminishing the volume of the thoracic cavity. The expiratory muscles are less strong than the inspiratory muscles, and only predominate as the inspiratory muscles relax (Mathieson, 2001).

Active expiration is enabled by involvement of the abdominal muscles which contract, forcing the diaphragm upwards, compressing the lungs and thus forcefully expelling air through the respiratory tract. Breathing during speech differs from typical respiratory patterns in a number of ways, which are influenced by the manner of the utterance (e.g. volume, length, word stress). It involves a shorter inspiration phase, which is oral rather than nasal (to speed up the process) thus minimising any disruption to continuous speech. Wyke (1983) notes that the vocal folds are abducted and the larynx is lowered during the inspiration immediately before phonation and terms this the ‘pre-phonatory inspiratory phase’. Adduction of the vocal folds during phonation causes a high resistance within the vocal tract, generating subglottic air pressure. Once this pressure is sufficient to overcome their resistance, at the point termed the ‘phonation threshold pressure’ (Farley & Barlow, 1994) the vocal folds will be pushed apart, beginning the cycle of vibratory movements required for phonation (Mathieson, 2001). Mathieson notes that the process of speech breathing should not be an effortful one, as once the diaphragm lowers and the thoracic cavity expands, the interplay of external and inter-thoracic air pressures cause exterior air to rush in to equalise the negative thoracic pressure.

2.13.3.2 The supraglottic tract

The **larynx** is a continuation of the airway to and from the lungs, positioned from the base of the tongue to immediately above the trachea. It is suspended from the hyoid bone, allowing vertical movement which modifies the corresponding length of the pharynx above. Laver (1980, p. 25-26) notes that “the hyoid bone is unique in being the only bone in the body which is not articulated with any other bone, and its muscular suspension from the larynx, pharynx, tongue and jaw, with the muscular tensions of the different sling systems having to be appropriately balanced for the accurate production of almost every single act of the vocal apparatus, makes the hyoid complex the prime example of mutually-influencing interaction of different muscular systems in speech". The larynx comprises of a series of cartilage, supported by a framework of ligaments, muscles and membranes. The lowest cartilage, positioned immediately above the tracheal rings, is the inflexible cricoid cartilage, which is frequently compared in shape to a signet ring with the narrow segment to the front. The thyroid cartilage sits superior to the cricoid; it is a large cartilage with a notch in the central point which is visible externally as the 'Adam's apple' or thyroid prominence. Above and attached to the thyroid is the epiglottis which extends upwards and backwards to the tongue base, while the arytenoid cartilages (a pair of pyramid-shaped cartilages which join the cricoid on either side of the midline by means of cricoarytenoid joints) allow the arytenoids to make a range of movements which correspondingly change the shape and alignment of the vocal folds (Mathieson, 2001).

The **vocal folds** are the vibratory mechanism within the larynx which contributes to the act of phonation. The two folds are attached to the anterior part of the thyroid cartilage at the 'anterior commissure', and run back to the 'vocal processes', the point at which the arytenoids extend into the vocal folds (Mathieson, 2001). The main body of the vocal folds is comprised of the lower part of the thyroarytenoid muscle (vocalis muscle) while the superior portion forms the false vocal folds (also termed the ventricular folds). Each true vocal fold is made up of five layers: the outermost layer consists of mainly ciliated columnar epithelium with a 'reinforced' layer of stratified squamous epithelium to protect against forcible adduction during phonation. Three layers of connective tissue lie under the epithelium, forming the lamina propria. The first layer of the lamina propria, also known as Reinke's space, is formed of a gelatinous-like substance which enables it to vibrate freely. The second layer (the intermediate layer) is a flexible layer which is made up of elastic fibres. The final layer of the lamina propria is made up of collagenous fibres, and is known as the ‘deep layer’. Together these properties enable the vocal folds to produce a smooth, undulating wave during phonation.

A series of muscles (the intrinsic laryngeal muscles) allow movement of the various laryngeal cartilages to which they are attached, causing the vocal folds to adduct and abduct, and lengthen and shorten, altering the thickness of the folds; thus changing their tension and vibratory characteristics according to the needs of speech (Mathieson, 2001). The majority of the intrinsic laryngeal muscles function in pairs, thereby enabling symmetrical vocal fold adjustments. Key muscles are the thyroarytenoid muscle which enables adduction of the true and false vocal folds, causing the folds to shorten and thicken; the posterior cricoarytenoid muscles, which elongate and abduct the vocal folds by pulling them upwards and backwards; the lateral cricoarytenoid muscles which have the opposite effect, rotating the arytenoids slightly forward and inwards to enable adduction of the vocal folds; and the cricothyroid muscles, which cause the cricoid cartilage to pivot, elevating it at the front and lowering it at the back, thus elongating and thinning the vocal folds (Mathieson, 2001). Stretching of the vocal folds is associated with increased frequency (which correlates with the listener’s perception of increased pitch) therefore the cricothyroid muscles are particularly important for this prosodic function. With the exception of the sternothyroid muscle, all the extrinsic laryngeal muscles are attached to the hyoid bone and enable movement of the larynx within the neck. They are made up of two groups: the suprahyoid muscles, which function as laryngeal elevators, and the infrahyoid muscles, which depress the larynx (Mathieson, 2001).

The **pharynx** is a muscular tube of approximately 12cm in length with a roughly 4cm opening at the top and a narrower opening at the bottom of around 2cm (Zemlin, 1964). It continues the airway immediately above the larynx anteriorly and the oesophagus posteriorly, running up to the nasal and oral cavities. Atkinson and McHanwell (2002) identify three divisions to the pharynx: the lowest, the laryngopharynx, runs from the entrance of the larynx/oesophagus up to the level of the tip of the epiglottis; above this is the oropharynx, this is the area at the back of the oral cavity and the pharyngeal section (the posterior third) of the tongue; and lastly, the nasopharynx, which is the uppermost portion of the pharynx where the pharynx enters the nasal cavity (the nasopharynx being separated from the oropharynx at the point where the elevated velum makes contact with the pharyngeal wall). The pharynx is able to constrict and relax according to the innervation of the inferior, middle and superior pharyngeal constrictor muscles, which correspond to the nasopharynx, oropharynx and laryngopharynx respectively (Mathieson, 2001). Constriction and relaxation within the pharynx is required for the process of swallowing, but also has considerable impact on the properties of voice; this is particularly evident when the speaker has muscular tension associated with being upset or tearful (Mathieson, 2001).

The **faucal arches** are two sets of muscular arches which sit one behind the other at the junction between the oral cavity and the pharynx. These pairs of muscles act in various ways to alter the physical property of the vocal tract during speech. The principle action of the arches is to pull the velum into a downward position, however when the velum is ‘fixed’ contraction of the palatoglossus muscles will cause elevation of the tongue, while contraction of the palatopharyngeal muscles will cause the larynx to elevate (thus shortening the length of the pharynx); similarly, when both the velum and larynx are fixed, contraction of both sets of muscles causes the pairs to move together, narrowing the vocal tract at the site of the arches (Laver, 1980).

The **nasal cavity** consists of two chambers (divided by the septum) into which the paranasal sinuses drain. It is separated from the oral cavity below by the hard palate and the soft palate or velum, which is able to lower and elevate to open and close the airway to and from the nose. Velic closure is not a simple case of elevation of the velum, it also includes (to a lesser degree) the back wall of the nasopharynx moving slightly forward (Luchsinger & Arnold, 1965). The mechanism for lowering the velum involves contraction of the paired palatoglossus and palatopharyngeus muscles (Laver, 1980).

By opening up the nasal cavity, sound energy can pass into the nasal tract to produce the nasal segments associated with English speech (e.g. /m/, /n/). It is quite typical for the velum to be slightly open during speech without causing the overt perception of nasality (Van Riper & Irwin, 1958) however, where there is insufficient closure oral sounds may be produced in a nasalised manner and there may be audible nasal escape. Conversely, reduced nasal airflow in nasal segments will contribute to the perception of denasality.

The **oral cavity** extends forward from the oropharynx, housing a number of structures whose complex interactions are key to speech production. At its posterior point are the faucal arches, whilst anteriorly the oral cavity is enclosed by the jaw and lips. The roof of the cavity consists of the hard palate, and more posteriorly, the soft palate or velum, which elevates to close off the nasal cavity above. On the floor of the oral cavity, the tongue is anchored to the hyoid bone posteriorly and the mandible anteriorly. The jaws are able to lower and rise, by means of the temporomandibular joint, changing the dimensions of the oral cavity and thus altering its resonant qualities. There is a muscular linkage between the jaw, tongue and hyoid bone which is reflected in sympathetic movements between the three (Laver, 1980). The lips can be rounded, protruded or spread by means of various sphincter and dilator muscles – principally the orbicularis oris (Laver, 1980), whilst the tongue utilises a series of intrinsic muscles (including the genioglossus, geniohyoid and mylohyoid) to make the complex movements required of individual phonetic segments of speech. Specifically the styloglossus contracts to pull the tongue up and back; the palatoglossus pulls the body of the tongue upwards when the velum is fixed; the hyoglossus exerts a pull on the tongue body downwards and backwards; whilst the genioglossus (which is the bulk of the tongue body) pulls the tongue body forward when the jaw is in a fixed position (Laver, 1980). As with the jaw, a lowered, elevated, fronted or backed tongue will change the shape of the oral cavity, thus altering its acoustic properties.

2.13.4 The function of voice

It is widely recognised that the human voice is able to convey a range of information from speaker to listener. Habitual voice patterns function as a conscious mechanism for the speaker to present him or herself in a particular manner or to indicate membership of a particular social community. This kind of intentional communication through voice quality is termed the communicative function of voice. Paralinguistic features of voice are those which alter these habitual characteristics, most obviously through changes in emotional state such as anger, grief or excitement (Mathieson, 2001) or perhaps through intended alterations such as such as reducing volume or using whispered phonation, to indicate confidentiality or intimacy. Much of the information exchange between speaker and listener is intended, yet voice quality can also signal factors over which the speaker has no control, but which are still interpreted by the listener; for example, changes in emotional state (e.g. depression) can bring about physical changes, such as increased tension in the vocal tract or reduced jaw and lip movements, which will alter the perceptual features of voice. Additionally, listeners have been found to make extralinguistic judgements about the age, sex or physical build of speakers, or even to make assumptions concerning the speaker's social background or level of intelligence, from voice features alone. This kind of unintentional communication is termed the informative function of voice.

Aside from providing information about the speaker and his or her state of mind, voice quality also has a linguistic function (Mathieson, 2001). This can be further broken down to two levels: segmental and non-segmental phonology. The first refers to the ability to make a distinction between phonemes which have the same place and manner of production but differ only in their voiced versus voiceless contrast (Mathieson, 2001). For example, the words /bad/ and /bat/, which have individual meaning, would be undistinguishable without the ability to contrast the voiced alveolar plosive /d/ with its voiceless counterpart /t/. Non-segmental or suprasegmental phonology is concerned with the prosodic features of stress, pitch, volume and rhythm, which together make the meaning of utterances clear, minimising or removing possible ambiguities (Crystal, 1981). It involves vocal practices such as rising intonation to signal a question or falling intonation to signal a statement or finality, thus indicating to the listener that he or she can take a turn to speak.

2.13.5 Variation in vocal characteristics – typical and disordered

2.13.5.1 Development of habitual vocal features

An individual's vocal features are greatly constrained by the anatomy of the vocal tract. Thus for example the pitch level in children will be considerably higher than in adult voices as a direct consequence of smaller larynx size and generally reduced stature. However, within the typical course of maturation it is normal for speakers to adapt vocal characteristics to enable them to fit in with the people and environment in which they live and these features gradually become more concrete with increasing age. This process begins in early childhood through exposure to the norms of the family, friends and wider social group and can be likened to the way that individuals alter language or dress-code between home, work or school. For example, it is accepted that adolescent boys often lower their pitch before there is evidence of actual physical change, which Holmes (2001) identifies as social and cultural influence. Similarly, in the past it was frowned upon for children to use regional dialect within the classroom, requiring a 'code-shift' into the more formal Standard English. Mathieson (2001) notes that although individuals routinely adapt their language, behaviour or appearance, voice quality tends to be adapted less as the majority of speakers are relatively unaware of their habitual vocal features.

2.13.5.2 Regional variations

Although phonological differences, such as the replacement of /ð/ with /f/, which often occurs in the south-east of England, or language-based differences which reflect local dialect, are overt indicators of geographical origin, voice quality differences may also be perceived, allowing speakers from different regions and countries to be identified from their particular vocal profile. Similarly, these voice settings can help to identify the perceived social class of the speaker; a practice which is noted to be particularly prevalent in the UK where significant weight is attributed to the way a speaker sounds (Mathieson, 2001). Differences in vocal settings may be generalised across countries as well as within different regions or cities; for example it has been observed that American males may utilise a lower pitch range than British males (Giles & Powesland, 1975) and a higher degree of nasality is common in American and Australian speakers than is associated with British speakers, with the possible exception of speakers of Received Pronunciation (RP) in England (Laver, 1980). A backed and lowered tongue setting may also be associated with RP speakers, and this accent is often perceived as a marker of higher social status in the UK (Kramerac, 1982). In speakers from Edinburgh, increased use of creaky phonation has been associated by listeners with a similar high status, whilst whispery and harsh phonation type have suggested a lower social standing (Esling, 1978). Velarised voice with denasality is prominent in parts of Lancashire (Abercrombie, 1967) whilst in Liverpool, a backed and elevated tongue setting (causing constriction in the oropharynx) together with a raised laryngeal setting and close jaw gives rise to the stereotypical ‘adenoidal’ quality associated with ‘Scouse’ speakers (Knowles, 1978).

2.13.5.3 Effects of aging

Changes in vocal characteristics occur naturally as a consequence of the aging process. Within elderly populations diminishing function caused by muscular atrophy, skeletal ossification, slower nerve conduction, reduced cardiovascular and respiratory function together with hormonal differences all compromise the voice production system, which can result in phonation that is perceived as rough or breathy and decreased in amplitude (Mathieson, 2001). Acoustically, fundamental frequency decreases markedly, the degree of change being much larger in females than in males (Nishio & Niimi, 2008) levels of jitter and shimmer are increased and the noise-harmonic ratio reduced (Mathieson, 2001).

2.13.5.4 Effects of vocal misuse and abuse

Muscle tension dysphonia is a relatively common example of hyperfunctional voice disorder which is often a consequence of the vocal misuse associated with professions which demand a high-level of voice use, such as call centre workers and teachers. The excessive muscular tension typically results in a harsh tone, increased noise, pitch breaks and vocal instability due to fatigue from increased effort (Mathieson, 2001). Similarly, elderly people may further damage their vocal apparatus by attempting to force their voice in order to combat the natural effects of aging (Close & Woodson, 1989). Vocal abuse, such as smoking is strongly linked to conditions like Reinke's oedema, where the superficial layer of the vocal folds (the lamina propria) becomes filled with fluid, thus impairing effective vocal fold movement (Zeitals, Hillman, Bunting & Vaughn, 1997). Reinke's oedema is associated with reduced pitch and volume, as well as increased jitter, shimmer and noise in the acoustic signal (Mathieson, 2001).

2.13.5.5 Effects of ill health

Degenerative conditions, such as Parkinson's disease are also associated with voice changes. Parkinson's disease results in a range of difficulties associated with dysarthria (muscle weakness) which affects all the muscles of the speech/voice system. Thus impaired respiratory function, vocal fold movement and velum control contribute to weak, breathy and rough phonation, raised but monotonous pitch, increased nasality and reduced loudness (Mathieson, 2001). Motor Neurone Disease is a progressive disorder in which mixed neurological signs are present, as a result of which vocal characteristics can vary greatly from patient to patient (Strand, Buder, Yorkston & Ramig, 1994); in those with bulbar involvement (as opposed to pseudobulbar) one of the earliest symptoms is voice deterioration (Robert, Pouget, Giovanni, Azulay & Triglia, 1999). A mixed presentation is also associated with the various brain lesions which occur in Multiple Sclerosis (MS), although there is a general agreement of the occurrence of deviations in fundamental frequency, noise, and jitter, which have been found to affect women with MS less than men (Feijó, Parente, Behlau, Haussen, De Veccino & de Faria Martignago, 2004).

As well as having consequences for speech and language, CVA or stroke can affect voice through partial or full paralysis of the vocal folds, poor neuromuscular, oropharyngeal and laryngeal control and the presence of secretions (Altman, Maronian, Lundy, Moore, Heman-Ackah & Schaefer, 2005). Reduced control may take the form of muscle weakness or in irregularities in the timing of the motor movements required for speech (apraxia), which will have a significant impact on voice production also. Symptoms will vary tremendously amongst individuals dependent on the site of lesions and extent of damage.

Oral, laryngeal and lung cancers can all cause disruption to voice. In the case of lung cancer this can be due to reduced respiratory volume and the potential for pressure on the recurrent laryngeal nerve which can impair vocal fold control (Lee, Carding & Fletcher, 2008). Tumours further up the vocal tract present a physical impedance to the structure and function of the voice system (for example a tumour on the vocal folds will impair the smooth undulating movement of the folds, resulting in perturbation, which is associated with harsh phonation). Surgery involving the removal of the tumour may also necessitate the removal of an area of healthy tissue around the malignancy, or even removal of whole structures, such as the larynx.

2.13.5.6 Effects of congenital disorders and syndromes

Congenital conditions can be the cause of physiological or neurological differences which can result in dysphonia. In the case of cleft palate voice quality has been described from early on as 'breathy' and 'hoarse' (McDonald & Baker, 1951), ‘harsh’ (Berry & Eisenson, 1956), ‘rough’ (Leder & Lerman, 1985) ‘strangled’ (McWilliams, Lavorato & Bluestone, 1973) and ‘strained’ (D’Antonio, Muntz, Province & Marsh, 1988). Pitch findings have been variable; D’Antonio et al. (1988) finding excessive high or low habitual pitch level and range as well as reduced or excessive loudness, whilst Tarlow and Saxman (1970) found levels of fundamental frequency to be within normal limits. Alterations in laryngeal valving (McDonald & Baker, 1951), incomplete glottal closure (D’Antonio et al., 1988) and excessive pharyngeal and laryngeal tension (Berry & Eisenson, 1956) are proposed factors in these vocal differences.

Prader-Willi syndrome (PWS) is a congenital genetic disorder which is characterised by excessive appetite, immature physical development, hypotonia, emotional instability and some degree of intellectual disability (Prader-Willi Syndrome Association, UK, 2005). Research has shown irregularities in pitch, in some but not all subjects, with levels being both higher and lower than would be expected for age and gender which may be related to altered laryngeal growth (Lewis, Freebairn, Heeger & Cassidy, 2002). A mixed pattern of hypernasality and hyponasality have been identified, both of which are suggested to be a consequence of poor velopharyngeal function due to reduced muscle tone (Lewis et al., 2002). Inadequate vocal intensity, as well as harsh and hoarse voice quality has also been reported (Akefeldt, Akefeldt, & Gillberg, 1997; Downey & Knutson, 1995). Lewis et al. (2002) query whether the use of growth hormone in the PWS population might have gone some way to alter vocal characteristics.

Similarly, the dysarthria associated with athetoid cerebral palsy is recognised to have significant effects on control of the vocal articulators, particularly affecting respiratory support, tongue movements and vocal fold coordination (Cerebral Palsy Source, 2005) which can result in strained or strangled voice quality (Ratusnik, Wolfe, Penn & Schewitz, 1978).

DS is also a congenital genetic syndrome which is associated with unusual voice features. The remainder of this chapter will examine the findings of research which has sought to describe the specific vocal profile of this population, as well as the possible causes and the potential social and emotional impact.

2.13.6 Perceptual and acoustic voice research in children with DS

2.13.6.1 Pitch and quality differences in children with DS

Historically, many authors have noted a high incidence of voice disorder in DS (Strazzulla, 1953; Schlanger, 1962; Tredgold & Soddy, 1963; Blanchard, 1964; Benda, 1965; Novak, Sedlackova, Klajman & Betlycwski, 1967). More specifically West, Kennedy and Carr (1947) described the voice of this group as very hoarse, loud and with inflectionless phonation, whilst Penrose and Smith (1966) used the terms 'guttural and low-pitched'. From early on, there appears to be a general consensus that children with DS have particularly low-pitched voices (Benda, 1949; Strazzulla, 1953; Blanchard, 1964); in fact Benda (1949) went so far as to propose that the 'low and raucous' voices of children with DS was so typical that it often allowed diagnosis to be made without even seeing the child.

Early studies of voice in DS tended to be concerned with making description of vocal characteristics based on perceptual clinical judgements of parameters such as pitch level and range or phonation type. The development of technology which enabled empirical instrumental analysis of acoustic features associated with voice production added new depth to the debate. Researchers were then able to look for correlations between perceptual judgements of the voices of individuals with DS and acoustic analysis findings.

In one of the earliest instrumental studies, **Michel and Carney (1964)** investigated the speaking pitch characteristics of eight boys with DS (8; 6-10; 6 years) in comparison to three groups of TD boys; all of whom had adequate hearing across the speech frequencies. The study also collected data on the height and weight of each child.

Unlike the previous perceptual studies which had found the voices of children with DS to be lower in speaking pitch than TD children, the mean pitch level of these boys with DS was found to be essentially normal, at least with respect to age. Overall, the pitch of the DS boys tended to be closest to that of the TD boys aged 10 years, which approximately corresponds with the average age of the DS group (9; 9 years). Pitch range was also found to be essentially normal. Differences were found between boys with DS and their peers with respect to physical development, boys with DS tending to be between one-and-a-half to two years behind in terms of physical growth. The authors suggest that this may affect the perception of pitch, as listeners may perceive that the child's pitch is abnormally low with respect to immature physical appearance when in fact it is appropriate to chronological age. The authors also speculated that other aspects of speech, such as unusual voice quality or intonation patterns rather than low pitch, may actually be responsible for the listener judgements of abnormal vocal characteristics in DS.

The Michel and Carney study is important as it is the first of its kind to challenge the accepted view that the pitch level of individuals with DS is abnormal. By providing empirical evidence that pitch is in fact appropriate to chronological age (at least in boys with DS) it was able to question the accuracy of perceptual judgements of pitch. The study however is not without flaw. Firstly, it does not use age-matched controls, making direct comparisons against the typically developing boys problematic. Secondly, the speech samples of the boys with DS were selected from spontaneous speech whilst the voice samples from the control groups were obtained by reading from a passage. Although the authors judged this methodology not to affect the validity of comparisons, more recent research has argued that within reading tasks speakers tend to adopt a more formal style which can result in a more restricted frequency range than is found in more spontaneous conversational speech, which is considered to be more representative of a speaker's habitual patterns (Selby & Wilson, 1997).

A logical progression from the study of pitch in boys with DS was the analysis of the pitch level of girls with DS by **Hollien and Copeland (1965).** This study of 'speaking fundamental frequency' (F0) replicated that of Michel and Carney (1964) using the voice samples of nine girls with DS (ages 8; 4-11; 4) and three groups of typically developing girls (ages 7, 8 and 11 years); again all had adequate hearing across the speech frequencies. Unlike the previous study, speech samples for both the DS and TD groups were selected from spontaneous conversation.

Although the speaking F0 of the girls with DS was found to be a little lower than all of the other groups, it did not deviate significantly from any of the control groups (or that of boys with DS of the same age in the previous study). Pitch range also echoed that of the TD group. The girls with DS, who had an average age of approximately 10 years, were found to be physically comparable to the youngest TD group (aged 7 years) demonstrating a similar pattern of immature physical development to the previous study.

Interestingly, the authors reject the suggestion that pitch may be perceived as low in DS due to the small physical stature of these girls in relation to their chronological age, rather they propose that it is the interaction of other vocal quality features which combine to give the illusion of low pitch. They tentatively suggest that the 'whispered voice' associated with DS may be a particular factor, recommending further analysis of the acoustic correlates of apparent vocal abnormalities associated with DS.

The new-found consensus of 'normal' pitch level in children with DS was short-lived. In **1970, Weinberg and Zlatin** published the findings of their research into the speaking F0 characteristics of five and six year old children with DS in spontaneous speech. This study was motivated by the influence of hypotonia (low muscle tone) on the voice of children with DS. Generalised hypotonia has been identified as a frequent characteristic of these children; McIntire and Dutch (1964) finding evidence of hypotonia in 97.7% of eighty-six children below six years of age whom they studied. The severity of hypotonia was believed to diminish with increasing age

(Penrose & Smith, 1966) and from this, Weinberg and Zlatin hypothesised that hypotonia would have its maximal effect on laryngeal muscle function (having the effect of lowering F0) in very young children with DS. This study differed from the previous studies in a number of ways: it encompassed a more homogenous study group, consisting of only those children with DS with a positive diagnosis of Trisomy 21; included only children who came from non-institutionalised backgrounds; used the same method of collecting voice data for both the TD and DS groups; and matched the two groups for age and sex, thus ensuring valid comparison of results. Data on physical development (height and weight) was also collected.

The F0 of twenty-seven children with DS (aged 5; 1-6; 11) with adequate hearing for speech was compared to that of sixty-six TD peers. The outcomes were somewhat unexpected with the mean speaking F0 for the overall DS group being significantly higher than that of the TD group, refuting the proposed hypothesis.

Weinberg and Zlatin state that the smaller physical stature, and therefore smaller larynx size of these children compared to their peers would naturally result in higher F0; however this does not explain why school-age children with DS, who are also physically smaller than their peers, do not have significantly different F0 values, and nor does it address why hypotonia has not had the hypothesised effect on the larynx. Weinberg and Zlatin suggest that the differences in the findings of this study compared to those of school-aged children are explained by differences in methodology (the use of a more homogenous sample group and more consistent data collection between groups), and secondly, that typical F0 values may have altered naturally over time, as they identified lower F0 levels for their control subjects than those of controls reported in earlier studies.

The authors are naturally cautious about generalising their findings to all children with DS due to the heterogeneous nature of this syndrome. They suggest that as phenotypic characteristics and clinical presentation vary within individuals, it could be that future studies find other young children with DS to exhibit a lower speaking

F0 than peers. They argue that these individual differences may become useful in the future in the possible identification of subgroups of DS.

The speaking F0 characteristics of school-aged, institutionalised children with DS were further investigated by **Montague, Brown and Hollien (1974).** This study was similar to those of Michel and Carney (1964) and Hollien and Copeland (1965) but crucially it addressed the criticisms made about these studies regarding the irregularities of sampling and age-matching. The voices of twenty children with DS, ten male and ten female (ages 7; 8-13; 5 years) were compared to those of twenty age-matched TD controls (ages 8; 0-13; 3 years). Unlike Weinberg and Zlatin (1970) the authors did not deem it necessary to control for specific genetic subgroups of DS as even though they conceded that it undoubtedly created a more homogenous DS group, they argued that they were not aware of any research which supported the proposition of vocal differences within different genetic subgroups of DS.

As elevated auditory thresholds were considered to affect the ability of children to self-monitor, potentially contributing to deviant vocal characteristics, the children with DS were screened for hearing impairment. Overall, the DS group were found to have elevated thresholds compared to the TD group. It could be argued that it is not valid to compare the voices of children with an identified hearing impairment to those without hearing loss, however, as hearing impairment is so prevalent in DS (Balkany, Downs, Jafek and Krajicek (1979) observing that as many as 78% of individuals with DS have some degree of loss) then a study group including these individuals may actually be better representative of the typical DS vocal profile.

The results indicated that there was no overall difference between the speaking F0 of children with DS and the TD children (supporting the findings of Michel & Carney, 1964, and Hollien & Copeland, 1965). However three of the boys with DS had significantly higher speaking F0 than the TD controls (in keeping with the findings of Weinberg & Zlatin (1970) in younger children with DS). The authors suggest it may be the case that in DS F0 is naturally higher in pre-school children, with a decrease at around 5 to 6 years of age bringing them roughly in line with age-matched TD peers, however some children with DS may fail to 'normalise' F0 and remain above the levels of their TD or DS peers.

More specifically, there was very little variation of the mean F0 between the three words within each subject's sample evidencing of a fairly consistent F0 level and no significant difference in speaking F0 for either 'group' or 'sex'. There was however a highly significant interaction between 'group' and 'sex', boys with DS being found to have a significantly higher speaking F0 than girls with DS and TD boys. Montague, Brown and Hollien propose that a possible contributing factor in this is the "heavy weighting in the upper end of the male Down's syndrome cell" (p. 417). Finally no correlation was found between speaking F0 and IQ.

In a follow-up study **Montague, Hollien, Hollien and Wold (1978)** examined listener perception of pitch in relation to its acoustic counterpart, F0, again in institutionalised children with DS. In this study sixteen undergraduate college students were asked to evaluate whether the children's pitch level was too high, too low, or normal with respect to their perceived age and sex. The listeners were not told that any of the children in the study had DS. On the first pass students made gross judgements of high, low or normal pitch, and on the second rated those children with too high or too low pitch on a seven-point scale of severity to qualify judgements. As this study used the same DS voice data as the previous study by Montague, Brown and Hollien, (1974) which had been demonstrated to have similar F0 levels to controls, the authors therefore hypothesised that there would be no difference in the perceived pitch levels between the voices of children with DS and their TD peers.

The results indicated that overall the children with DS were rated as exhibiting significantly lower pitch than the TD controls (60.2% of the DS group compared to only 29.7% of the TD group). However, a number of the DS group were also rated as having excessively high pitch (24.8% of the DS group compared to 19.7% of the TD group); evidencing a much greater range of deviation than the TD subjects who tended to cluster around the norm (50.6%, compared to only 15% of the DS group).

Essentially, this DS profile demonstrates a bimodal distribution, with peaks at both low and high pitch, which is in contrast to the normal distribution curve seen in the TD children. Females of both groups were judged as having significantly lower pitch levels than male subjects.

This study provides further evidence that despite ‘normal’ levels of F0, listeners typically judge pitch in DS to be different (generally lower) to that of their TD peers. As a greater variance in F0 was also perceived, the authors suggest that it may be this variance which is be responsible for the perceptual rating of low pitch, rather than a deviance in the actual F0 level.

This study demonstrates good intra-rater reliability and reasonably good inter-rater reliability. However, there are a couple of criticisms which the authors concede. The first is in regard to the listeners themselves who are regarded as being relatively unsophisticated with no background in participation in ‘critical auditory processing tasks’; although Montague and Hollien (1973) found naïve listener judgements to be approximately equal to experienced listeners in roughly equivalent conditions to this study. Secondly, the voice samples are not naturalistic and were rather short in duration for the purpose of making voice judgements, although it is acknowledged that it can be extremely difficult to elicit long samples from populations with ID.

Many early studies of voice quality in children with DS concentrated solely on the issue of pitch and its relation to F0. In **1983, Pentz and Gilbert** widened the debate to examination of the relationship between a range of other acoustical parameters and perceptual ratings of voice quality. This study appears to have been partly motivated by the proposition that the F0 level cannot be solely responsible for the perception of low pitch in DS (Montague, Hollien, Hollien & Wold, 1978) and that pitch level alone is not responsible for the perception of deviant voice quality within this population (Hollien & Copeland, 1965). The authors noted that previous perceptual studies of voice in DS have identified other deviant vocal characteristics: children with DS have been described as husky and monotonous (West, Ansberry & Carr, 1957); raucous (Benda, 1969); and judged to exhibit more breathiness, roughness and nasality than non-ID controls (Montague & Hollien, 1973). Within the general population acoustic studies had already identified a relationship between increased frequency perturbation or 'jitter' and the perception of harshness (Moore & Thompson, 1965); a finding echoed by Kitajima (1977) in relation to increased amplitude perturbation or 'shimmer', and by Isshiki, Yanagihara and Morimoto (1966) in relation to increased spectral noise-to-harmonic ratio.

As a result of these findings Pentz and Gilbert proposed to investigate the influence of F0 level and range, frequency and amplitude perturbation (jitter and shimmer) and noise-to-harmonic ratio on the voice characteristics of non-institutionalised pre-adolescent children with DS, and to look for correlations between identified differences in acoustical parameters and listener ratings of voice quality

The study used audio-recordings of fourteen children with DS, six males and eight females (mean age 9.42 years) with no evidence of hearing impairment, and the voice samples of a similarly matched typically developing control group. The recordings consisted of a series of sustained vowels /u/, /i/ and /a/, and twenty mono- and poly-syllabic words containing these vowels, which were played backwards in order to minimise the effects of articulatory errors on voice judgements. To allow analysis of intra-rater reliability four additional samples from two DS subjects and two TD controls were included. No information about the identity of the subjects was given, except age. The seven judges (volunteer graduate students with no previous experience of rating voice quality) were first exposed to recorded examples of the voice parameters being evaluated.

The results indicated that perceptually, children with DS were rated to be significantly different from the TD group in terms of the overall severity of voice quality disorder. However, these children were not judged as significantly different to the TD group on any of the individual parameters being evaluated (pitch; airloss; tension; nasality; rate; intensity; and vocal range). This would seem to be an astonishing disparity and would suggest that the judges were skilled sufficiently to identify that the voices of children with DS were 'different' but not to be able to identify the source of those differences (however, the short nature of the recordings and the 'backwards' presentation of those words are acknowledged to have made their task particularly challenging).

In terms of instrumental measures of F0 level and F0 range no significant differences were found between the DS and TD groups, supporting the majority of previous studies. Differences however, were identified in the remaining acoustic measures, the DS group being found to have significantly higher jitter, shimmer and noise-to-harmonic ratios than the TD control group. It is worth pointing out here that differences in the terminology can create the potential for confusion; Pentz and Gilbert use the term 'noise-to-harmonic ratio' (higher levels indicating increased noise in the acoustic signal) whilst later research adopts the reversed term 'harmonic-to-noise ratio' (HNR) and therefore talks of reduced levels in DS. With regard to correlations between acoustic parameters and perceptual ratings, the significantly higher severity rating of the DS group was found to correlate with jitter factors (0.50), shimmer factors (0.53), and noise-to-harmonic ratios (0.61).

The authors of this study raise the interesting question of whether children from a non-institutionalised background are perceived differently from those from an institutionalised background. This study of non-institutionalised children, although identifying a significantly higher severity rating for children with DS compared to their TD peers failed to identify any differences in specific parameters of voice (pitch; airloss; tension; nasality; rate; intensity; and vocal range). This is in contrast to the findings of other studies of institutionalised children with DS, which have found perceptual ratings of increased breathiness, roughness and nasality (Montague & Hollien, 1973) and differences in pitch (Montague et al., 1978). Pentz and Gilbert suggest these differences may be evidence of a different vocal profile between institutionalised and non-institutionalised children with DS, on the basis that the non-institutionalised children may be exposed to more and often better communication models, which allow them to learn and maintain more appropriate speech and voice habits than their institutionalised counterparts. However, they also concede that "to infer that residence in a non-institutional setting as opposed to an institutional setting is sufficient in itself to have produced these differences is speculative at best" (Pentz & Gilbert, 1983, p. 209). Although differences in learning and experience are considered likely to impact on subsequent vocal quality to some degree, in this case the use of minimally trained listeners rather than experienced voice professionals must be considered as the most significant factor in the lack of specifically identified vocal differences.

From their findings, the authors recommended additional investigation of the effects of jitter, shimmer and noise-to-harmonic ratio in DS as their contribution to differences in voice quality perception remains unclear. They also suggest that other factors may interact with these three acoustic parameters, such as elevated levels of acoustic damping in the oral and nasal tracts and structural differences.

Within the non-ID population differences in noise-to-harmonic ratios had often been found to be accompanied by reductions in the relative formant amplitude levels of vowels (Isshiki, Yanagihara & Morimoto, 1966). Essentially, in speakers with high noise-to-harmonic ratios there is a reduction in the intensity of production of the high energy regions in the vowel spectra (those portions which help listeners to distinguish one vowel from another) whilst the normally lower energy regions between formants (which often represent aperiodicity or 'noise' in the vowel sound) are often increased.

Using the data of the Pentz and Gilbert (1983) study **Pentz (1987)** sought to clarify the vowel formant amplitude levels of speakers with DS in comparison to their peers. His findings revealed that there was a significant difference between groups with the DS group demonstrating significantly lower formant amplitude intensity levels than the control group. More specifically, the children with DS were found to have significantly lower second formant (F_2) amplitude levels which were less distinct than is typical from non-formant regions than those of the TD group across all the vowels studied (/u/, /i/ & /a/). This indicates that this formant is most profoundly affected by the differences in DS speech. The author notes that it would be expected that vowels which have a higher second F_2 (e.g. /a/) would be more vulnerable to this distortion than vowels which typically have a lower second formant (e.g. /i/). The impact of these differences is a reduction in the harmonic or periodic component of speech together with an increase in the aperiodic or noise component, these differences are thought to be significant factors in what the author describes as the 'hoarse', 'breathy' and 'nasal' perceptual voice quality which is characteristic of DS. Although these patterns may be borne from physiological differences Pentz also speculates that they may be exacerbated by poor vocal habits which may in fact constitute vocal abuse and thus further degrade an already compromised system. In terms of therapy he therefore recommends that children with DS be encouraged to achieve and habitually use their optimum vocal potential (taking into account physical limitations) by eliminating problem behaviours and encouraging better vocal conservation and vocal hygiene.

It was some twenty years later that further investigation of the formants of vowels in the speech of children with DS was completed. In a large-scale study of Portuguese speaking children, aged between 4 and 8 years of age, with a confirmed genetic diagnosis of Trisomy 21, **Moura, Cunha, Vilarinho, Cunha, Freitas, Palha, Pueschel and Pais-Clemente (2008)** found an interesting pattern in the production of formants for the five vowels studied (/a/, /e/, /i/, /o/ & /u/). The back, low vowels /a/ and /o/ demonstrated predominant variations in F_1, whilst the high vowels /i/, /e/ and /u/ were more typically characterised by variations in F_2. Overall the differences between F_1 and F_2 in the DS group were found to be smaller than those of an age-matched TD control group, and are believed to be a likely cause of reduced discrimination between vowel sounds. The authors attribute differences between the groups to differences in anatomy and physiology and difficulties in neuromuscular and aerodynamic control. Specifically, the decrease in F_2 seen in the mid-upper-front vowels /e/ and /i/ is suggested to be a consequence of decreased pharyngeal space caused by ineffective anterior movement of the tongue, whilst the large increase in F_2 in the upper-back /u/ may be related to limited range of tongue movement in this position. The variation observed in the DS group in F_2 between vowels /i/ and /u/ is proposed to be a significant factor in distinguishing the voices of children with DS from their TD peers.

In addition to formant analysis, Moura et al., (2008) investigated a range of other acoustic parameters including: F0; intensity (average and maximum); frequency perturbation; amplitude perturbation; harmonic-to-noise ratio; and spectral tilt.

Analysis of F0 (average, standard error, variability coefficient and highest and lowest level) for the five vowels was found to produce a very different pattern of F0 in these school-aged children with DS than found in previous English-speaking studies. When adjustments for age and gender were made, statistical analysis revealed highly significant differences between the two groups on all parameters with the exception of the average F0 for the vowel /u/. Across all 5 vowels, the DS group produced lower values than the TD group in all of the F0 measures, with the exception of the variability coefficient. This is a stark contrast to earlier findings of roughly equivalent F0 level and range between school-age children with DS and their TD peers (Michel & Carney, 1964; Hollien & Copeland, 1965; Montague, Brown & Hollien, 1974; Pentz & Gilbert, 1983) and may be indicative of language-specific differences. The authors suggest that the lower F0 together with increased variability of F0 may reflect the perceived vocal instability associated with this group. Increased variability is noted in neurological disorders, and according to Hirose, Imaizumi and Yamori (1995) alterations in vibration frequency can be a result of a lack of control of vocal fold tension. In the control group the standard error of F0 was found to decrease with age, but not in the DS group, whose standard error values were significantly higher than controls. This lack of improvement is significant as it signals a persistent disorder in DS.

Both frequency perturbation (jitter) and amplitude perturbation (shimmer) were significantly higher in the DS group. The authors speculate that low firing rate of motor units in the thyroarytenoid muscle related to general muscular hypotonia may be a factor; increased firing rate being likely to smooth the motion of the muscle and thus contribute towards more stable phonation.

The harmonic-to-noise ratio was lower in the DS group than in the control group across all five vowels; this reflects higher levels of noise in the acoustic signal (in common with the findings of Pentz & Gilbert, 1983). Similarly spectral tilt, which is concerned with the rate at which the amplitudes of harmonics decline, was also lower in comparison to controls, and is suggested to be a factor in the increased levels of perceived breathiness and forced voice.

To evaluate the impact of these acoustic findings, a perceptual analysis was completed by two speech and language therapists who were experienced in using a version of the GRBAS scale (Hirano, 1981) adapted to Portuguese. Parameters such as 'roughness', 'breathiness', asthenic speech', and 'strained speech' were rated. In line with previous perceptual assessments of voice in DS, all parameters were judged as statistically more severe in the DS group compared to controls, although the asthenic speech category was slightly less statistically relevant than the others.

2.13.6.2 <u>Nasal resonance differences in children with DS</u>

The only study to date which includes specific evaluation of the perception of nasality in children with DS is that of **Montague and Hollien (1973)**. They investigated the issue of voice disorder in DS, specifically examining listener perception of breathiness, roughness and hyper-nasality between children with DS and TD children.

Matched samples (single word utterances elicited from a picture naming task) from twenty children with DS (with an average age of 6.27 years) and an age and gender matched TD group were recorded and presented in a randomised format to listeners. In order that verbal components other than voice quality (e.g. articulatory difficulties) did not influence judgements, the speech samples were presented backwards. Four different judging groups were used to rate the recordings (G1 and G2 were naïve listeners; G3 were qualified speech and language therapists (SLTs) with post-graduate qualifications; whilst G4 consisted of SLTs educated to BSc level). All listeners completed a speech discrimination hearing test and those scoring less than 92% were excluded. Prior to the task, the examiners explained and provided examples of the three voice qualities to be analysed (roughness, breathiness and hyper-nasality). On the first occasion pass the raters were asked to make a judgement as to whether or not each voice sounded disordered, then on the second pass to rate severity on a eight-point scale for each parameter (0, representing no disorder and 8 a severe disorder). Listeners were not told that some of the children whom they were listening to had a diagnosis of DS.

All the listener groups evaluated the children with DS as exhibiting significantly more breathiness and roughness than controls, and all groups judged the female children with DS to have significantly more breathiness and roughness than males. Only G1 did not agree that the children with DS exhibited a greater degree of nasality than the control group (although as untrained listeners less confidence might be placed in their judgements of voice features). Interestingly, males were found to produce more nasality than females by two of the listener groups, and when all the listener group ratings were combined (however the authors note that caution should be exercised due to the possibility of type 1 error). In terms of intra-rater reliability, the judges showed a good degree of consistency in judgements, with a high level of reliability on measures of breathiness and roughness. Reliability for hyper-nasality was somewhat lower, and as such the authors suggest that nasality is possibly more difficult to evaluate than the voice conditions of breathiness and roughness. Between the four listening groups, a higher level of inter-rater reliability was found on the measure of nasality than breathiness or harshness (a reversal of the intra-rater findings).

The children with DS in this study were identified as having a significant bilateral hearing loss (the flat audiometric configuration suggesting a possible conductive impairment). The authors suggest this is likely to be a contributing factor in the judged voice characteristics due to interference in the process of self-monitoring of vocal output. They concede that the level of the hearing loss, in conjunction with the small group size, makes their findings difficult to generalise across the wider DS population, demonstrating a need for more analysis of this area.

2.13.7 Perceptual and acoustic voice research in adults with DS

2.13.7.1 Pitch and quality differences in adults with DS

The majority of studies of voice in DS have focussed on paediatric populations. It was this paucity of adult based research which motivated **Moran and Gilbert (1978)** to investigate the speaking fundamental frequency characteristics of institutionalised adults with DS.

The study used the data of sixteen adults (eight males, mean age 38.4 years & eight females, mean age 41.2 years) who had passed a hearing screen test, and sixteen gender-matched, TD controls (males, mean age 37.8 years & females, mean age 36 years). Control subjects reported no history of hearing impairment or laryngeal pathology. Matched voice samples for each subject were obtained via a counting task, prolonged vowels and repetition of a declarative sentence.

The results, in contrast to the studies of F0 in school-age children with DS, identified that adults with DS had a significantly higher speaking F0 than the TD group. Interestingly, this result is in agreement with the findings of elevated F0 levels in pre-school children with DS (Weinberg & Zlatin, 1970). Unlike the results of the paediatric studies of Montague, Brown and Hollien (1974) and Montague, Hollien, Hollien and Wold (1978) male DS subjects were found to have a significantly lower F0 than females; this being in keeping with the lower F0 found in TD adult males in comparison to adult females.

Significant differences were found between the tasks, counting being associated with a significantly higher mean F0 than the repetition of sentences. Other studies have noted that differences in F0 between tasks should be expected (Michel & Wendahl, 1971; Schultz-Coulon, 1975) indicating the importance of consistent methodologies between subject and control groups. Intriguingly, these particular findings are in contrast to those of Schultz-Coulon (1975) who found that counting and spontaneous speech tasks yielded lower F0 than other speech tasks.

The authors suggest some factors which may contribute to the disparity in speaking F0 between adults with DS and their TD peers. Firstly they consider the impact of the development of secondary sexual characteristics. Benda (1965) proposes that many females and some males with DS may be expected to reach full sexual development. Moran and Gilbert (1978) identify that this maturation has a direct impact on physical development and thus is likely also to affect the development of vocal characteristics. Seven out of eight males with DS had F0 levels considerably lower than values reported for the pre-adolescent males in the Michel and Carney (1964) and Montague, Brown and Hollien (1974) studies, providing evidence of a pubescent voice change. The most significant finding from this study is that the natural voice change in DS is not as great as that seen in TD populations. This may be related to the generally smaller stature in DS, thus evidencing less laryngeal growth during puberty than seen in TD populations. A direct correlation exists between pitch level and laryngeal size (Hollien, 1960) with a smaller larynx being associated with a higher level of pitch. The authors suggest the single male subject who did not exhibit significantly different F0 values from pre-pubescent males, may have failed to experience the voice change which is associated with the onset of puberty. The difference between pre-pubescent and adult females was less obvious. However two females with DS had a particularly high F0, which raised the mean of the whole adult female DS group. If these out-lying values were excluded then there was evidence of a possible pubescent voice change.

The authors propose a second theory related to hypotonia. The effects of hypotonia are proposed to diminish as individuals with DS mature (Penrose & Smith, 1966) resulting in increased laryngeal tone and thus higher pitch. Typically, this elevated pitch level would be ‘cancelled out’ by natural voice changes in puberty (where pitch levels tend to lower) however, in the absence of a pubescent voice change, the pitch level and corresponding F0 values would remain above typical adult values. This may have been the case for the two females with DS who demonstrated extremely high F0.

The finding of elevated speaking F0 in adults with DS was confirmed by **Beckman, Wold, and Montague (1983)** in a non-invasive acoustic study using computer-generated vocal-tract shapes and frequency perturbations. However, the authors stress that this was a study primarily of non-invasive methodology, which was based on only two speakers with DS (a female aged 22 years and a male aged 27 years), and as such the findings should not be interpreted as definitive data on DS voice quality. In addition to higher than typical F0, both individuals were found to exhibit larger than normal jitter ratios, and the female was found to be diplophonic; this is the phenomenon of having two fundamental frequencies.

In a follow-up to their 1978 study of F0 in adults with DS, **Moran and Gilbert (1982)** utilised the same data to evaluate the relationship between perceptual judgements and two other acoustic parameters: jitter and noise-to-harmonic ratio. The DS and TD voice samples were evaluated by seventeen 'Communication Disorders' graduate students for a range of voice parameters: breathiness; tension; pitch; and breathiness plus tension. The latter category (breathiness plus tension) was included as an additional perceptual category as the simultaneous presentation of breathiness and tension is characteristic of hoarseness (Darley, 1965; Morris & Spriestersbach, 1978).

The perceptual results indicated that breathiness (air loss) was the most commonly perceived characteristic (more than 60% of judges rated breathiness in eleven of the sixteen adults with DS). Laryngeal tension was perceived by more than 70% of judges in nine of the DS adults, and laryngeal tension plus breathiness was perceived by more than 60% of judges in six of the DS adults. Low pitch was perceived by more than 70% of judges in five of the adults with DS, whilst agreement on high pitch was significantly lower (not perceived in any single subject with DS by more than 58% of judges).

Acoustic analysis revealed that speaking F0 was lower in males with DS than females with DS, supporting the findings of the earlier adult study by Moran and Gilbert (1978). Jitter factors of greater than 6% were found in 3 of the DS subjects (2 males: 15% and 11% and 1 female: 12%). The noise-to-harmonic ratio revealed a mixed picture (shown in table 2.1).

NHR results in adults with DS (Moran & Gilbert, 1982)		
Category	**Description of category**	**Found in**
Category 1	no noise component	4 males and 3 females (43.75%)
Category 2	distinct harmonic component mixed with noise component	1 male and 1 female (12.5%)
Category 3	slight noise component from 3000-5000Hz and noise in second formant predominating over harmonic	1 male and 1 female (12.5%)
Category 4	noise only in the second formant, further intensified noise above 3000Hz	2 females and 3 males (31.25%)
Category 5	harmonic components hardly noticeable	not found in any subjects

Table 2.1: HNR results of adult DS speakers (Moran & Gilbert, 1982)

Correlations between perceptual ratings and acoustic characteristics for males and females revealed similarities and differences between the two. Four high-level correlations were identified for females with DS: breathiness significantly correlated with noise-to-harmonic ratio (r = 0.82); pitch correlated with F0 (r = 0.75); and breathiness plus tension correlated with F0 perturbation (r = 0.75) and with noise-to-harmonic ratio (r = 0.79). For males with DS there was only a single positive correlation: breathiness and noise-to-harmonic ratio (r = 0.73). The finding of a significant correlation between breathiness and noise-to-harmonic ratio in both male and female speakers with DS can be explained by the findings of Isshiki, Yanagihara and Morimoto (1966) who identified that turbulent airflow, caused by vocal fold insufficiency (as found in breathiness) contributes to the spectral noise component. The breathiness component may also be responsible for the significant correlation in the combined breathiness plus tension and noise-to-harmonic ratio categories in DS females; although it is not clear why this was not the case for the males. The finding of a significant correlation between F0 perturbation and combined perceptual rating of breathiness plus tension was deemed unsurprising as both F0 perturbation

(Lieberman, 1963; Isshiki et al., 1966) and combined breathiness plus tension (Darley, 1965; Morris & Spriestersbach, 1978) are characteristic of the same 'harsh' voice quality. Again, why this correlation was not identified in males with DS is unclear.

A high correlation was found between pitch and F0 for female subjects with DS ($r = 0.75$) while the same correlation for male DS subjects was extremely low ($r = 0.16$). The authors identify that the perception of pitch in male subjects in this study is in keeping with the findings of previous studies, where low pitch has been perceived, but F0 has been objectively identified as average or above average. They suggest that this may be the result of the interaction of several acoustic and perceptual factors (i.e. breathiness plus tension and high noise-to-harmonic component ratio may influence perceptual judgements of low pitch).

It was over twenty years until the next study investigating the adult DS voice was completed. **Lee, Thorpe and Verhoeven (2009)** examined parameters relating to intonation and phonation in young adults with DS (four males & five females aged between 17 and 29 years, mean age, 24.7) in comparison to an age and gender matched TD control group. The organic pitch range (OPR), i.e. the range of pitch available on the basis of laryngeal anatomy and physiology, was obtained by measuring ascending and descending pitch glides on a sustained vowel. Findings demonstrated that the range of the DS group was significantly lower than that of the control group; the DS range being limited to approximately one octave, with little difference between male and female DS subjects. In contrast the TD groups' OPR was almost 2.5 times greater; and in common with an earlier study within the general population (Henton, 1995) females were found to have a wider OPR than males. The authors note that the lack of difference between genders in the DS group is indicative of pitch range being constrained by physiological differences which are common to both males and females with DS. A reading task was used to measure characteristics of the pitch contour framework. Although linguistic pitch range (LPR), which is described as the pitch movements associated with phonological prosodic events such as pitch accents and syntactic boundaries, did not differ significantly between the groups, the DS subjects were found to have a reduced range; it is proposed that this may be indicative of different linguistic processing strategies as well as physiological differences. Findings of higher mean F0 were in keeping with those of Moran and Gilbert (1978) and Beckman, Wold, and Montague (1983). Interestingly, Lee, Thorpe and Verhoeven (2009) suggest that personality characteristics, such as submissiveness, may interact with differences in laryngeal physiology to bring about F0 differences. This, in conjunction with shallower declination (the general descending trend of pitch in utterances) caused the authors to state that the intonation of adults with DS is more monotonous than controls.

In terms of phonation, there were a number of surprises: unlike earlier findings of increased incidence of frequency perturbation (jitter) in DS (Pentz & Gilbert, 1983) this study found fewer frequency perturbations in the DS group. The authors suggest that the difference may be explained by differences in sampling; however it must also be considered that this study investigated an adult population, whilst the earlier study was of children with DS. A further explanation of differences proposed by the authors is that the adults with DS were all members of a theatre group, and thus may have been more aware of their voice production. Both jitter and shimmer were found to be lower in females than males in both groups, and no significant difference was found between ratings of shimmer between the groups. Measures of maximum phonation time (MPT) also threw up unexpected findings, with only a marginal difference being found between the DS and TD groups, suggesting effective vocal fold function without significant air leakage. This is in stark contrast to the findings of Pryce (1994) who found significantly reduced MPT, which is claimed to lead to increased respiratory effort. It is possible that the practise of using voice for performance may also have had a positive impact on the respiratory and laryngeal function of these subjects. Summing up their study, Lee, Thorpe and Verhoeven (2009) state that their results might suggest that the perceptual ratings of hoarse and breathy voice quality “may not reflect laryngeal parameters, but may be more affected by supralaryngeal factors whereby particular patterns of articulation affect the resonance characteristics of the vocal tract” (p. 86).

2.13.7.2 Nasal resonance differences in adults with DS

Earlier paediatric studies had provided some evidence for a higher prevalence of hypernasality in DS (Montague & Hollien, 1973). Physiological examination of three hundred and eighty-nine subjects with DS also revealed potential correlates with hypernasality: 4.63% with bifid uvula and 0.77% with submuccous cleft palate (Schendel & Gorlen, 1974). However, these percentages are relatively low, suggesting that other factors may be involved. **Rolfe, Montague, Tirman, and Vandergrift (1979)** sought to examine the velopharyngeal mechanism and its relation to objectively measured hyper- and hypo-nasality in adults with DS.

The study examined 6 non-institutionalised DS subjects (5 male, 1 female) between 26 and 30 years of age, who had been perceived as demonstrating hypernasality by two speech and language therapists. Five out of these six subjects were assessed as having a mild-moderate conductive hearing loss. Elicited single words were recorded backwards onto a tape (as in earlier studies in order to maintain the focus of judgements on resonance rather than mis-articulations). Two groups of judges: G1 consisting of six graduate-level SLT students trained in the evaluation of resonance disorders; and G2 consisting of twenty-one undergraduate students beginning a course in communication disorders, but with no experience of rating resonance. Before judging, both groups were given verbal instruction as to the differences between hypo- and hyper-nasality, and the tape was played once as an example. Judges were required to identify the presence of resonance disorder for each subject on the first pass, and on the second pass to identify whether that resonance disorder was hypo- or hyper-nasal, together with the degree of the disorder (mild, moderate or severe). The judges were not told that the subjects had DS or that an SLT had judged them as having hypernasality.

The results, which were rated on a scale of 1-7 (1 representing severe hyponasality; 4, ‘normal’ resonance; and 7, severe hypernasality) indicated no clear trend with a mean score for all six DS subjects of 4.23; indicating that the judges perceived the subjects to cluster around the norm. The single exception to this was subject number three, who was perceived as having a mean hypernasal rating of 5.58 (by G1) and 4.76 (by G2). Statistical analysis determined that the G1 rating for this subject (5.58) differed significantly from that of the remaining subjects with DS.

As subject three had been rated as having a significantly higher level of hypernasality than the remaining DS subjects he was subjected to further cinefluorographic evaluation. Production of sustained /s/ and /a/ and repeated /a, a, a/ in a rapid sequence allowed evaluation of his velopharyngeal competence. It was found that while the subject's soft palate did make contact with the nasopharynx (demonstrating some level of closure of the nasal cavity) there was relatively minimal tissue available to make the contact (80% of normal, in comparison to TD subjects). This, together with the identified reduced thickness of the soft palate, is proposed to have affected the quantity and quality of closure.

The authors also hypothesise that the perception of hypernasality may be related to the timing of the velum in making contact with the nasopharynx, as velopharyngeal valving requires milli-second neuromuscular control. They suggest that the presence of hypotonia and identified hearing loss may result in closure-timing errors. It may also be the case that more evidence of resonance disorder might have been found by evaluating voice samples from connected speech (as connected speech is associated with increased cognitive, linguistic and physiological demands compared to the production of isolated segments and words, which these subjects may have found relatively easy to produce; thus resulting in 'better' (less nasal) articulations). Both listener groups felt that the voice samples were too short to make adequate judgements about resonance. It might also be questioned whether the listener groups were sufficiently skilled in discriminating between hypo- and hyper-nasality, given the disparity between G1 and G2 and the pre-study SLT evaluation, identifying the presence of hypernasality.

The proposal of resonance differences in adults with DS was further examined by **Moran (1986)**. This study aimed to establish whether listeners can distinguish DS speakers from a control group of TD speakers with hoarse voice resulting from other pathologies, such as vocal nodules, vocal fold polyps and vocal fold paralysis. More specifically, if the above distinction was made, it hoped to determine whether resonatory differences might account for the listeners' ability to distinguish between the two groups.

Voice samples, prolonged vowels (/a/, /i/ and /u/) were collected from fourteen institutionalised adults with DS (eight males and six females, ages 20-43 years) and fourteen TD controls with hoarse voice, (eight males, six females, ages 19-54 years). Those with hearing impairment were excluded through a pure tone hearing screen prior to sampling as well as those with significant differences in F0 as it was felt this might distract the raters.

Two groups of listeners (G1, who as graduate students from the faculty of education were familiar with ID, and G2 as graduate students from the faculty of speech pathology were familiar with dysphonia) listened to each voice and judged whether it was a subject with DS or a TD adult with a voice disorder. Listeners were not told how many subjects were in each group. The results indicated that G1 correctly identified 65% of adults with DS and 55% of adults with dysphonia. As would be expected of SLT students, G2 demonstrated a higher level of accuracy, correctly identifying 74% of adults with DS and 76% of adults with dysphonia. At end of the listening task, the listeners were asked to explain what criteria they had used to identify subjects with DS; both groups described resonance differences ("sounds made through nose", "nasal emission") to distinguish the DS speakers. Some described hearing "*imprecise vowel production*" or made statements such as the subject's vowels "*did not sound exactly like the vowels of non-ID speakers*". Moran also considered the possibility that listeners may have associated shorter utterances with an ID population. To test this proposal the vowel samples of each group were matched for length, the results showed that a brief utterance was no more likely to result in the identification of DS than perceived hypernasality.

Crucially, further perceptual rating of the resonance of the vowel samples, by experienced listeners within the speech department, demonstrated low inter-judge reliability and failed to find significant differences between the two groups (although there was a trend towards higher nasality ratings in the DS group). Spectrographic analysis of the formant frequencies (F_1 & F_2) of the vowel samples also found no significant difference between the two groups.

This study failed to identify statistically significant differences in nasality ratings between adults with DS and TD dysphonic adults, which taken with the low nasality ratings of the previous study by Rolfe, Montague, Tirman, and Vandergrift (1979) suggests that resonance differences do not appear to be a particularly salient feature of the voices of adults with DS.

2.13.8 Possible factors in the voice quality of individuals with DS

Over the years, a range of factors have been proposed to explain the distinctive vocal quality associated with DS. Early investigations speculated that smaller physical stature relative to age caused the perception of low pitch in boys with DS (Michel & Carney, 1964) but this was subsequently rejected by Hollien and Copeland (1965) who attributed the same mis-perception in girls with DS to the interaction of a range of vocal features. This view of a combination of physiological and functional factors (Novak, 1972; Pentz & Gilbert, 1983; Moran, 1986; Pentz, 1987) is still considered to be the most valid explanation of vocal differences in DS. Yet even the most recent research into this syndrome notes that "the possible anatomical and physiological basis for the vocal phenotype remains largely undefined" (Moura et al., 2008, p. 35).

Three principle themes occur in the literature: structural differences within the vocal tract; hypotonia; and hearing impairment. Although additionally it could be argued that the ID associated with DS is likely to lead to poorer awareness of vocal differences and perhaps less inclination to adopt voice patterns which are typical in terms of gender, age, social group and geographical location. Poor vocal habits are also suggested to exacerbate difficulties (Pentz, 1987) and consequently children with DS need to be encouraged to achieve and habitually use their optimum vocal potential, taking into account physical limitations, by eliminating problem behaviours which may constitute vocal abuse and thus further degrade an already compromised vocal apparatus.

2.13.9 Structural differences associated with the DS vocal tract

Although a number of physiological differences frequently occur in DS it is not possible to generalise these differences to the whole of this population as presentation varies considerably between individuals in terms of the frequency and intensity of anomalies. Scientific advances mean that genetic diagnosis of DS is now the norm. This means that those individuals who presented with few clinical signs and previously escaped diagnosis or inclusion in research are now recognised, and serves to illustrate the diversity of this population (Mackenzie Beck, 1988).

2.13.9.1 Impact of endocrinological disorders on vocal structure

Growth deficiency is commonly associated with DS, with both males and females being of smaller stature than TD age-matched peers (Michel & Carney, 1964; Hollien & Copeland, 1965). It has been suggested that the significantly higher mean speaking F0 which has been identified in adults with DS may be a consequence of this growth deficiency (Moran & Gilbert, 1978) as there is a direct correlation between pitch level and laryngeal size (Hollien, 1960) with a smaller larynx producing a higher pitch. Similarly, Moran and Gilbert (1978) noted that despite F0 levels of school-age children with DS being roughly equivalent to those of TD peers, some adolescents with DS failed to demonstrate the lowering of pitch which is characteristic of typical development during puberty. They proposed that growth hormone deficiency may cause failure to develop secondary sexual characteristics, resulting in this failure to lower pitch. This finding has been echoed in recent studies of individuals with PWS, where it was identified that older subjects are more likely to have abnormally high pitch levels than younger subjects (Akefeldt, Akefeldt & Gilberg, 1997).

In TD populations alterations of voice in even mild cases of thyroid disorder are common indicating that thyroid hormone receptors are present in the larynx (Altman, Haines, Vakkalanka, Keni, Kopp & Radosevich, 2003). Early studies noted the presence of myxoedema in children with DS (Benda, 1969; Strazzulla, 1953). This condition is related to underactivity of the thyroid gland (hypothyroidism) and causes thickening of the submucosal tissues of the vocal folds resulting in a hoarse sounding voice (Mathieson, 2001). Strazulla (1953) proposed hypothyroidism as a reason for the low-pitched voice associated with this group as improvements in vocal quality were found after treatment with thyroxin.

Although a generalised growth deficit is likely to have implications for development of individual vocal tract features and their resulting quality, and hypothyroidism may impact on laryngeal function, it is not believed that endocrine disorder can completely account for the characteristic vocal features of DS. Novak (1972) concludes that due to the variability in endocrine disorders present in this population and the failure to observe evidence of thickening of the vocal folds as a consequence of these conditions within his research, that the harsh, rough voice cannot be explained solely by the presence of endocrinological differences. Additionally more recent investigation of endocrine disorder suggests that abnormal levels of plasma thyroid hormone do not necessarily result in symptoms of thyroid disorder in DS (Prasher & Haque, 2005). The authors therefore argue that it is not valid to compare hormone levels between DS and TD groups and that an appropriate reference scale for DS needs to be created to avoid the possibility of mass misdiagnosis of thyroid disorder and the resulting potential for unnecessary hormonal treatment.

2.13.9.2 The lungs

Respiratory infections in children with DS are common (Venail, Gardiner & Mondain, 2004) and abnormalities in the respiratory mucosa can exacerbate infection (Mackenzie Beck, 1988). Breathing is often described as shallow with a high incidence of abdominal breathing (Novak, 1972) which is explained by the presence of hypotonia which affects the intracostal muscles more expressly than the diaphragm muscle (McIntire, Menolascino & Wiley, 1965). Novak (1972) suggests that poor co-ordination between thoracic and abdominal breathing is the result of primitive control of breathing function, stemming from the ID associated with DS. Abdominal breathing is also common in those individuals who take no exercise and have excessive girth (Mathieson, 2001) as is common in DS, and these excessive fat deposits have been found to impair lung function (Cotes, 1979). The intercostal muscles are the most important for everyday speech as they control the volume of the rib cage and hence the volume of air required for speech (Mathieson, 2001) thus decreased strength of the costal musculature will see less thoracic air capacity and therefore reduced breath support in this population.

2.13.9.3 The larynx

Early small-scale studies suggest that in DS the larynx sits higher in the neck than in TD individuals (Benda, 1969) which is confirmed in a recent review of DS by Venail, Gardiner and Mondain (2004) who also note that the larynx is shorter than is typical.

The presence of thickened and fibrotic laryngeal mucosa was observed by Benda (1969) and suggested to be a consequence of disturbance of the pituitary gland causing hormonal disorder. However during laryngoscopic evaluation Novak (1972) found only light thickness of the mucosa of the vocal folds, with no evidence of vocal fold thickening. Further stroboscopic assessment revealed no expressive changes in the vibrations of the vocal cords in ten out of twelve of the DS subjects evaluated, causing Novak to conclude that the harsh, rough voice in DS could not be explained only by pathological findings on the vocal cords. This is supported by Pryce (1994) who comments that the early findings of Michael and Carney (1964) identifying no significant difference in F0 (the base note at which the larynx vibrates) indicates that the larynx in DS produces a normal vibration. The harsh, rough, pressed voice which Novak describes is proposed to be related to the presence of enormously high phonation pressure and the squeezing of both the true and false (ventricular) folds during phonation. However, Beckman, Wold and Montague (1983) suggested that the presence of hypotonia negated involvement of ventricular fold vibration observed in a single female DS subject with diplophonia (the phenomenon of having two fundamental frequencies). Rather they felt that the diplophonia could be related to large variations in the amount of time devoted to each phase of the vibratory cycle, which might in turn be linked to subglottic air pressure variations. It is speculated that "if diplophonia is widespread among persons having Down's syndrome, it might partially explain the large number of clinical reports suggesting a lower perceived pitch for the Down's subject" (Beckman, Wold & Montague, 1983, p. 313). However, the authors stress that their study is primarily an investigation of non-invasive acoustic analysis methodology and that it may be hazardous to generalise these findings across the wider DS population.

There is some suggestion of insufficiency of the vocal folds which might influence vocal quality. Novak (1972) notes what he terms as a 'wheezy admixture' in the voices of individuals with DS, caused by air escaping through the folds during phonation. Similarly, Pentz (1987) attributes the 'hoarse and breathy' voice quality in DS to increased 'noise' in the speech signal, due to insufficient closure of the vocal folds. Novak suggests this escape is related to increased phonation pressure; although hypotonia may also be a contributing factor causing weakness of the vocal folds. As in respiratory control, Novak proposes that ID results in primitive control of the glottis, causing irregular vibrations. Vocal fold irregularities (jitter and shimmer) have been found to correlate closely with harsh vocal quality (Wendahl, 1966) which is frequently associated with this population.

2.13.9.4 <u>The pharynx</u>

The pharynx in DS is generally considered to be constricted, causing a reduced airway (Jacobs, Gray & Todd, 1996). Excessive tonsillar mass was believed to be a cause of this constriction (Adran, Harker & Kemp 1972) however, surgical removal of these tissues revealed them to be similar or smaller than typical (Strome, 1981).

This disparity between visual appearance and actual size is suggested to be caused by the narrowing of the pharynx at the level of the faucial pillars which causes the tonsils to be more visible as they are unable to sit in their natural position behind the pillars (Mackenzie Beck, 1988). Further evidence of constriction within the pharynx comes from Beckman, Wold and Montague (1983) who suggest that pharyngeal constriction limits the movements of the posterior portion of the tongue, causing differences in the production of back vowels between DS and TD speakers; they also note open-mouth breathing and the fronted position of the tongue, particularly when swallowing in support of this constriction.

According to Novak (1972) the shape of the pharyngeal and oral cavities and their connections (the isthmus between the tongue and the hard or soft palate) influence the height and distance of the first and second formants of vowels. A narrow isthmus is characterised by a greater distance of resonating frequencies, whilst a broad isthmus (where the connection is loose) causes the frequencies to almost fuse. Using x-ray, Novak found the latter to be a clear distinguishing feature in children with DS. As well as making it more difficult to distinguish between vowels this loose connection was proposed to affect the timbre of voice itself. Imprecise vowel productions were also described by raters of adults with DS (Moran, 1986) however these vowels were not found to reflect the 'fused' pattern of formants described in children by Novak (1972). Moran suggests that the hypotonia seen in children with DS, which improves as the nervous system matures (Benda, 1960) may cause children, more than adults, to have a more flaccid protruding tongue which exacerbates the effects of the prognathic mandible and thus further affects vowel and voice production. Furthermore, the characteristics of the pharynx may be a factor in nasal resonance differences in DS. Pentz (1987) notes that many DS speakers have a vocal sound stream in which the vowel formants (especially the second ones) are depressed in amplitude and less distinct than is typical from non-formant regions; a pattern which can give rise to the perception of nasality (Pent, 1987). Pentz suggests that it is the constriction in the upper vocal tract that redirects too much sound energy into the nasal cavity rather than the oral cavity, thus causing increased acoustic damping which further reduces vowel formant amplitude levels.

Novak (1972) also notes the tendency of the mucosa in the relatively small, narrow pharynx in DS speakers to show signs of atrophy and a tendency to dry out. This may be related to the frequent open-mouthed posture which has a drying effect on the mucosal lining of the oral tract and the laryngeal areas (Pryce, 1994). Individuals with DS, and generalised ID, are also often reported to have reduced fluid intake (Pryce, 1994); these factors combined may impact on voice quality, as good hydration is associated with effective vocal hygiene (Mathieson, 2001).

2.13.9.5 The hard palate

There are many reports describing the dimensions of the hard palate in DS. Early studies observe it to be high and narrow (Oster, 1953) arched (Levinson et al., 1955) and possibly reduced in length (Engler, 1949); however, these descriptions are based on clinical observation rather than instrumental analysis. Shapiro et al. (1967) provided the first objective measurements of palatal dimensions in children and adults with DS, finding that contrary to earlier observational reports there was no significant difference in the height of the palate between DS and TD subjects, however, the DS group were found to have a significantly narrower and shorter palate. Similarly, in a more recent analysis it was found that the DS palatal measurements of width, length and average volume were significantly less than the corresponding values of TD controls, but in contrast, that the average height value of the palates of the children with DS was significantly greater than controls (Bhagyalakshmi, Renukarya & Rajangam, 2007).

The contour of the palate in DS has been described as 'steeple-shaped' (Shapiro et al., 1967); 'gothic' (Novak, 1972); 'vaulted' (Goodman & Gorlin, 198); and 'stair type' (Bhagyalakshmi, Renukarya & Rajangam, 2007). It is suggested by Benda (1969) that this shape is related to the underdevelopment of the bones connected to the nasal cavity. Palatal differences have been found to lessen with age-related changes in the growth of caraniofacial structures (Skrinjarić, Glavina & Jukić, 2004).

Implications include reduced oral cavity volume, impairing mastication (Goodman & Gorlin, 1983); poor palato-lingual contact resulting in defective articulation (Bhagyalakshmi, Renukarya & Rajangam, 2007) and in specific relation to voice, oral constriction, impacting on the relative position of the tongue, causing the perception of raised tongue body setting (Mackenzie Beck, 1988).

2.13.9.6 The tongue

Tongue fissuring and excessive growth of the papillae on the surface of the tongue (Benda, 1969; Novak, 1972; Temtamy, Aboul-Ezz, El-Hadidi, Soliman & Soliman, 1994; Bhagyalakshmi, Renukarya & Rajangam, 2007) and macroglossia (an enlarged tongue) are frequently described in DS (Venail, Gardiner & Mondain, 2004; Morgan, Friedman, Duncan & Sulek, 1996; Temtamy et al., 1994). Vogel, Mulliken and Kaban (1986) describe 'true macroglossia' as the presence of an enlarged tongue in correlation with histologic abnormalities, whilst in contrast the term 'relative macroglossia' is proposed to exist where histology does not provide a pathologic explanation, such as in DS where the tongue appears to be over-large in relation to the smaller oral cavity.

Pharyngeal constriction may displace the tongue into a more forward position, helping to maintain an adequate airway (Gisel, Lange & Ninman, 1984) which is exacerbated by the more forward position of the mandible, to which the tongue is anchored, in relation to the maxilla (Mackenzie Beck, 1988). In light of the already fronted tongue position, and the frequent open-mouthed posture (Pryce, 1994) hypotonia will further contribute towards a lax tongue which is often observed to loll forward out of the mouth.

In a French questionnaire study of the oral health of individuals with DS it was noted from parental responses that the prevalence of tongue protrusion reduced with age (Hennequin, Allison & Veyrune, 2000). This observation confirms that of Adran, Harker and Kemp (1972) and is believed to reflect a general realignment of the vocal tract as the child grows and the neuromuscular system matures (Moran, 1986) resulting in increased tone of the tongue and other orofacial muscles (Skrinjarić, Glavina & Jukić, 2004).

2.13.9.7 The lips

As an obvious external facial feature many comments have been made concerning the appearance of the lips of people with DS. The membranes of the lips have been reported to be thickened and white in appearance which results in fissuring as the lips grow (Butterworth, Leoni, Beeman, Wood & Stern, 1960) which is exacerbated by excessive flow of saliva due to the habitual open-mouth posture (Pueschel, 1992). Recently lip morphology was more formally assessed by Ferrario, Dellavia, Colombo and Sforza (2004) who compared children and adults with DS to gender and age-matched TD controls. They identified that DS subjects had significantly smaller mouth width and significantly smaller lower lips with regard to volume, area and vermilion height, whilst the upper lip was significantly larger in area and vermilion height.

Lip morphology is proposed to be typical at birth and to change with growth (Benda, 1969). It is not clear whether it is the habitual lip and facial postures which influence abnormal lip development or the physiological differences associated with DS which constrain the unusual lip posture (Mackenzie Beck, 1988) however a mix of the two is most probable.

Hypotonia has been found to affect the muscles of the lips of individuals with DS (Limbrock, Fischer-Brandies & Avalle, 1991; Mizuno & Ueda, 2001) although it is regarded as more common in the tongue than the lips (Kumin & Bahr, 1999). This low tone has been identified as impairing the sucking motion of babies with DS (Mizuno & Ueda, 2001). Reduced upper lip mobility together with decreased sensory awareness and decreased feedback have been observed in infants with DS, all of which contribute to feeding, eating, and drinking difficulties (Kumin & Bahr, 1999) as well as communication differences.

Accurate control of vocal-tract length is essential for the correct production of vowels (Riordan, 1977) with both the position of the larynx and lip posture acting to determine these features. Lip protrusion, which lengthens the vocal tract (Fitch, Tecumseh & Brown, 1995) will therefore influence vowel quality in DS whilst contributing towards alterations in voice characteristics, most notably pitch-related.

2.13.9.8 The nasal cavity and velum

Compared to age and gender-matched controls, the noses of children and adults with DS were found to be significantly smaller in volume and area and to be of a different shape (having a flatter slope and more acute nasal tip) whilst nasal bridge length, height of the nose and nasal tip protrusion measurements were reduced and the horizontal dimensions increased (Ferrario, Dellavia, Colombo & Sforza, 2004); all of which will have a direct effect on the resonating properties of the nasal cavity. There have been few direct studies of the dimensions of the nasopharynx in DS however a cinefluorographic case-study of an adult male revealed the depth of his nasopharynx to be significantly reduced from the norm for his age (Rolfe, Montague, Tirman & Vandergrift, 1979).

Bolfan-Stosic and Hedever (1999) attribute the differences in acoustical characteristics in children with DS to structural anomalies including an insufficient obstacle between the nasal and oral cavities. In a review of ENT and speech disorders in DS, Venail, Gardiner and Mondain (2004) report that the sinuses may be small and their ostia obstructed by hypertrophic mucosa, whilst Benda (1960) hypothesized that an absence of sinus formation in the skull may be in part responsible for the resonance differences in the voice of individuals with DS. However, this view is contentious as Proctor (1980) supports Benda's proposition, whilst Bunch (1982) asserts that the sinuses have little or no involvement in voice; current theory suggests that the latter holds true (Mathieson, 2001).

With regard to the velum, it has been suggested that resonance disorder is a perceived feature of DS, with both hypernasality and hyponasality being reported, although the former more frequently. In perceptual studies, Montague and Hollien (1973) report that children with DS exhibit a greater degree and severity of nasality than children without ID, these findings are suggested to be related to velopharyngeal insufficiency caused by hypotonia. In contrast, Moran (1986) failed to identify statistically significant differences in nasality ratings between adults with DS and non-ID dysphonic adults with hoarse voice quality, suggesting that nasality is not a distinguishing feature of the adult DS voice. It is possible that increasing muscular control due to diminishing hypotonia positively affected velopharyngeal function in the adult population. Fluoroscopic evaluation of an adult male with DS revealed that although there was contact between the subject's soft palate and nasopharynx during production of /s/ and /a/, that there was relatively minimal tissue (80% of typical) available to make that contact (Rolfe, Montague, Tirman & Vandergrift, 1979). The authors propose that this together with reduced thickness of the soft palate may have affected the quantity and quality of that closure. Furthermore, they speculate that hypotonia in conjunction with errors in the timing of velic closure may have added to the perception of hypernasality in this individual. However, as this was a single case study, it would be inappropriate to generalise these findings to other adults with DS. Perhaps more significantly, Novak (1972) found the movement of the soft palate to be normal in all of the twenty children with DS that he evaluated by laryngoscope.

2.13.9.9 Jaw and dentition

The dimensions of the mandible of individuals with DS appear to be close to typical norms, however, the maxilla is suggested to be underdeveloped (Mackenzie Beck, 1988). An evaluation of the craniofacial measurements of children and adults with DS in comparison to age and gender-matched controls revealed that with increasing age maxillary growth was reduced in relation to mandibular growth (Allanson, O'Hara, Farkas & Nair, 1993). The effect of these anomalies is what Mackenzie Beck (1988) describes as pseudo-prognathism, where the protruding mandible appears over-large in relation to the maxilla.

In terms of jaw function, the muscles which control jaw movement act more strongly to close the jaw rather than open it (Van Riper & Irwin, 1958); thus there is a tendency towards an open-mouth posture due to low muscle tone. Kumin and Bahr (1999) identified that although young children with DS were generally found to have symmetrical patterns in jaw movements, jaw instability contributed to feeding, eating, and drinking difficulties; this instability would also be likely to have a significant impact on articulation and thus speech intelligibility. Added to this the underdeveloped nature of the maxilla contributes towards the pharyngeal congestion which is typical in DS, and is therefore a factor in the resulting vocal differences.

The atypical jaw development has implications for the dentition of individuals with DS, which is described as malformed and misaligned (Bhagyalakshmi, Renukarya & Rajangam, 2007) with delayed eruption of the primary teeth (Temtamy et al., 1994). Angle’s class III malocclusion, where the mandibular dental arch is anterior to the maxilliary arch, has been described in several early studies (Brown & Cunningham, 1961; Kisling, 1966; Cohen et al., 1970). This is supported by more recent research into the frequency and type of anomalies in tooth alignment of children with DS compared to TD children and children with generalised ID by Ondarza, Jara, Bertonati and Blanco (1995) which found that the DS group showed a higher frequency of malalignments in both the deciduous and permanent dentitions, with the frequency of malalignments being higher in the permanent teeth than in the deciduous.

2.13.10 Functional differences associated with the DS vocal tract caused by hypotonia

In a study of children with DS under 6 years of age McIntire and Dutch (1964) identified hypotonia in all major muscle groups in 97.7% of the eighty-six children they examined, and subsequent studies have confirmed this high prevalence of hypotonia in this population (Griffiths, 1976; Dummer 1978; Morris, Vaughan & Vaccaro, 1982). Low muscle tone has been found to be related to low muscle strength (Morris, Vaughan & Vaccaro, 1982) and to decrease as the child with DS matures (Penrose & Smith, 1966; Morris, Vaughan & Vaccaro, 1982); thus its impact is felt most strongly in young children with DS. It would be expected then that low muscle tone would have the greatest effect on the vocal folds of very young children with DS, causing then to be particularly flaccid, which consequently would have the effect of producing lower F0. However, as shown by the findings of higher F0 in pre-school children with DS in relation to their TD peers (Weinberg & Zlatin, 1974) in comparison to the absence of differences between school-aged children with DS and their peers, then this is not the case.

Reduced muscle tone can have a significant effect on all the musculature of the vocal tract and hence affect voice production in a number of ways. In DS it has been found that abdominal breathing is prevalent due to the negative effects of hypotonia on the intracostal muscles (McIntire, Menolascino & Wiley, 1965) which impairs breath support. Low tone causes the extrinsic laryngeal muscles, which hold the larynx in position in the neck, and the muscles of the pharyngeal wall, which act as resonating areas, to be flaccid causing the harmonics which result in the 'gruff' voice described by Pryce (1994). Rolfe, Montague, Tirman and Vandergrift (1979) attribute the presence of hypernasality to inadequacy of the velopharyngeal mechanism caused by low tone in the muscles controlling velic closure. Similarly, the lips, and particularly the tongue, can be affected considerably by hypotonia (Kumin & Bahr, 1999) causing mis-articulation and therefore poor intelligibility.

PWS is also characterised by hypotonia and offers further evidence of the impact of low muscle tone on the vocal apparatus. According to Kleppe, Katayama, Shipley and Foushee (1990) the voice quality in PWS is comparable to that of patients with flaccid dysarthria, where hypernasality is a characteristic feature. Nasal emission and hypernasality in PWS are proposed to occur as a result of velopharyngeal incompetence (Branson, 1981; Munson-Davis, 1988; Lewis, Freebairn, Heeger & Cassidy, 2002) caused by hypotonia of the velum. As in DS, weak speaking volume, perceived pitch differences and breathy and/or hoarse voice quality (Munson-Davis, 1988) have also been identified and are believed to be related to hypotonia as well as hormonal differences and obesity (Defloor, Van Borsel, Curfs & De Bodt, 2001). Interestingly, it is proposed that high pitch in PWS may be the result of the compensatory stretching of the laryngeal muscles which are not typically used in speech, to combat the effects of hypotonia (Akefeldt, Akefeldt & Gilberg, 1997).

There is some suggestion that specially planned physical education programmes can bring about improvements in motor development (Morris, Vaughan & Vaccaro, 1982); although this proposal has been questioned by more recent studies, which suggest that long-term benefits are not likely (Mahoney, Robinson & Perales, 2004; Spiker & Hopmann, 1997). In terms of communication, non-speech motor exercises have not been found to be helpful in producing long term improvements in articulation (Forrest, 2002).

The vocal initiation levels of people with DS have been found to require almost twice the amount of energy to set in motion the vocal cord vibrations of speech compared to controls (Pryce, 1994). In that study Pryce found that dysphonic adults and adults with generalised ID also had to expend significantly more energy than control subjects, but not to the same degree as the individuals with DS. Although Pryce cannot conclusively attribute this difference to hypotonia, it is noted that greater energy would be required to initiate a more flaccid mechanism suggesting that low tone is a contributing factor. Pryce notes that should individuals with DS find it more effortful to initiate voice, then individuals may opt to communicate orally less frequently, moreover, as it may take longer for the individual with DS to reach the required initiation level the opportunity to contribute may be lost, particularly when the individual is competing with other speakers during rapidly-moving conversation Pryce argues that both these scenarios might contribute towards a culture of learned passivity in DS. Pryce also notes a possible link between initiation levels and communication style, suggesting that the telegraphic speech used by many individuals with DS may partly be due to the inability to sustain voice long enough to complete a grammatically complete sentence, although delayed language development must also be considered as a causative factor. The use of electromyography biofeedback is suggested by Pryce to be of potential benefit in providing individuals with DS with increased awareness of their muscle tension and therefore helping them to self-monitor their vocal output more effectively.

2.13.11 Impact of hearing impairment on DS voice

In DS some degree of hearing impairment is common, being identified in as much as 78% of the population (Balkany, Downs, Jafek & Krajicek, 1979). Conductive loss, as a result of middle ear abnormalities, occurs more frequently than sensorineural loss which involves cochlear deficits. Particularly prevalent is fluctuating hearing loss as a result of otitis media with effusion (OME), also known as ‘glue ear’, which is reported to account for 90% of cases of conductive loss (Venail, Gardiner & Mondain, 2004).

It has been suggested that hearing loss causes insufficient self-monitoring of vocal output which may be a possible contributory factor in the aberrant voice quality of children with DS (Montague & Hollien, 1973; Montague, 1976). Wold and Montague (1979) report that in their study of perceived voice deviations and hearing disorders in adults with DS that there was an indication that the subjects with the greatest degree of hearing impairment had the greatest degree of voice disorder. They, like earlier researchers, propose that hearing loss disrupts the feedback loop of the speech chain and thus impairs the ability to self-monitor the vocal parameters of quality, intensity and pitch. Similarly, Rolfe, Montague, Tirman, and Vandergrift

(1979) suggest that this loss may impair the ability of individuals with DS to make the fine motor timing and closure patterns necessary for appropriate velopharyngeal valving thus contributing towards hypernasality.

Evidence connecting hearing loss and voice deviations also comes from studies within the congenitally deaf population. By focussing on individuals without other co-existing conditions such as ID, hypotonia, or structural differences of the vocal tract more direct conclusions can be drawn about the impact of hearing loss. Such early studies have identified that deaf children have been found to have deviant voices which are most likely a direct consequence of their hearing loss (Boone, 1966). More recently, in a Vocal Profile Analysis Scheme (VPAS; Laver, Wirz, Mackenzie & Hiller, 1981) evaluation of forty deaf adult speakers with profound loss compared to forty hearing adults, Wirz (1986) describes deaf speakers as having highly significant differences in pitch and loudness, pharyngeal constriction, laryngeal tension and harsh voice quality. Mathieson (2001) notes that the laryngeal and pharyngeal tension associated with the deaf population can contribute to perceived hypernasality.

As well as contributing to differences in voice quality, hearing loss has been found to have a significant effect on other aspects of development. Even with the use of hearing aids infants and young children with bilateral profound hearing loss have been found to show substantial delays in vocal development (Ertmer, Young & Nathani, 2007) including late and less frequent canonical babbling (Oller & Eilers, 1988) and reduced phonetic and syllable shape inventories (Stoel-Gammon, 1988) which can disturb the acquisition of phonological awareness (Ertmer & Stark, 1995) and impair intelligibility. Expressive and receptive language acquisition (Holme & Kunze, 1969) emotional and educational development (Menyuk, 1977) and interpersonal skills (Needleman, 1977) have also all been found to be negatively impacted by hearing impairment. These wide-spread consequences of hearing loss indicate the need for regular screening and appropriate aural rehabilitation including the use of aids, sound field amplification, grommets and cochlear implants; this is particularly relevant for infants and young children with DS who are a high risk group for hearing loss.

2.13.12 Judgements made about speakers based on vocal characteristics

It is well established that physical appearance can have a significant effect on the judgements which are made about the personality and abilities of both children and adults. Studies specifically of individuals with DS have shown that the characteristic facial features can generate the perception of immaturity (Fidler & Hodapp, 1999) whilst the generally smaller physical stature associated with this syndrome is likely to further enhance this perception. Added to this presence of ID can lead to inappropriate social behaviour and language deficits both of which often reflect younger developmental patterns and may further encourage a less mature perception and thus foster low expectations (Fidler, 2003).

Less consideration has been given to the specific role of voice quality in listener judgements. Early perceptual studies have established that within TD populations, voice quality does impact on the way that speakers are perceived (Barbara, 1958; Murphy, 1964; Starkweather, 1961). Esling (1978) found that listeners judged whisper and harsh phonation types in Edinburgh speakers to be associated with low social status, whilst creaky phonation correlated with higher prestige; as both whisper and harsh phonation types are those which are most often associated with DS speakers this suggests further potential for negative social judgements about this syndrome also. More recently, Saxton (2006) identified that adult and adolescent listeners find lower pitch level more attractive when judging male voices, whilst child listeners preferred voices which had higher pitch; as adults have a lower habitual pitch level than children this would suggest that listeners demonstrate a preference for vocal features which reflect their own qualities.

More investigation of voice quality has been directed at acquired disorders such as progressive neurological conditions. Damage to specific neural subsystems is reported to underlie the type of dysarthria present and the particular perceptual characteristics of voice that result. Speakers with Parkinson's disease (PD) have been found to have increased 'huskiness', deeper pitch level with a more monotone quality and reduced volume, which can be attributable to acoustic changes in the level and variability of fundamental frequency, increased noise and lower sound intensity (Miller, Noble, Jones & Burn, 2006). In an investigation of the impact of communication changes on the lives of individuals with PD, patients reported that "listeners lacked an appreciation of difficulties they were facing in talking, talked over them, talked for them, did not wait for an answer, ignored them or assumed they were stupid" (Miller et al., 2006; p. 237). Anecdotal reports from patients with degenerative conditions that their communicative difficulties have been wrongly presumed to be the consequence of inebriation are also not uncommon; the embarrassment caused by such situations can cause patients to become isolated reducing their desire to communicate and lowering self esteem (Miller et al., 2006). Voice disorders which are associated with dysarthria often occur along with articulatory, resonance and respiratory impairments (Kent, Vorperian, Kent & Duffy, 2003) all of which are typical of speakers with DS also, making it difficult to isolate the specific contribution of voice to listener perception.

Perhaps a more focussed analysis of the effect of voice quality on judgements can be made within the field of laryngeal cancers, where specific phonation differences can be isolated without the conflicting presence of language disorder or cognitive impairment. These cancers can involve laryngectomy (partial or complete surgical removal of the larynx) which has obvious repercussions on phonation and may necessitate the implementation of alternative voice output strategies, such as oesophageal voice or the use of an electrolarynx. Recent research by Turcotte, Wilson, Harris, Seikaly and Rieger (2009) assessed listeners' ratings of speakers after treatment for laryngeal cancer in comparison to a control group with no history of voice disorder. The patients treated for laryngeal cancer were grouped into three groups: those having received a total laryngectomy and subsequently using a tracheoesophageal voice prosthesis; partial laryngectomy; and radiation therapy. The results showed that the latter non-surgical group and the control group were rated more positively than those who underwent surgery across the 7-point Likert scales examining how ‘attractive’, ‘clever’, ‘trustworthy’ and ‘sophisticated’ listeners perceived the speakers to be, whilst the two surgical groups received more negative ratings for the parameters of ‘scary’, ‘annoying’, and ‘intimidating’. Their findings led Turcotte et al. to conclude that those individuals who undergo surgery as a consequence of laryngeal cancer may be at an increased risk of experiencing social stigmatization. Laryngectomy patients have been found to frequently complain of adverse listener reactions to their voices, including being shouted at as if they were deaf, avoidance of conversation and eye contact, failure to indicate that the speaker has not been understood for fear of embarrassment of continued misunderstanding, being spoken to through a companion rather than directly, and angry or mocking responses to attempts to communicate via the telephone (Mathieson, 2001). Like individuals with progressive conditions these negative experiences can have a significant impact on the confidence of recovering patients and thus affect their ability and desire to return to work or social circles.

Degenerative disorders and laryngectomy are generally adult conditions, usually occurring after a considerable period of typical voice development, and as such the progression and impact can be very different to the experience of individuals with developmental differences, such as in DS. Therefore, perhaps more can be determined about how people with DS are perceived by looking at studies of perception of voice within other developmental conditions which also have associated voice disorders.

In such populations, at the most fundamental level, the mere presence of phonation has been found to be a desirable feature. In a study by Lilienfeld and Alant (2002) children were found to display a more positive response to a child with cerebral palsy (CP) when he used a voice output device than when he communicated without functional speech. Clear differences in the way adolescents perceive speakers with CP in comparison to their TD peers were identified by Lass, Ruscello, Harkins and Blankenship (1993) who found that the CP speakers were rated significantly more negatively than their peers across a set of twenty-two parameters evaluating personality and appearance traits. In common with adolescents, adult and child raters were also found to rate the same group of children with CP more negatively than controls (Lass, Ruscello & Lakawicz, 1988; Ruscello, Lass, Hansen & Blankenship, 1992).

Similarly, in children who have had voice disorder clinically diagnosed by speech and language therapists but where dysarthria is not a complicating variable, children (Lass, Ruscello, Stout & Hoffman, 1991) adolescents (Lass, Ruscello, Bradshaw & Blankenship, 1991) and adults (Ruscello, Lass & Podbesek, 1988) have again rated those children more negatively than TD control subjects across a similar set of variables; the adults being found to rate more negatively than both child and adolescent judges (Lass, Ruscello, Harkins & Blankenship, 1993).

Some children with autism have been found to display significant impairments in vocal quality (Sheinkopf, Mundy, Oller & Steffens, 2000). The impact of these features was investigated in a study of prosody and voice by Paul, Shriberg, McSweeny, Cicchetti, Klin and Volkmar (2005); findings revealed that inappropriate stress and hypernasality were factors behind children with autism being rated lower than TD peers in communication and sociability scales.

Individuals with PWS have been found to share a number of vocal features in common with DS including harsh/hoarse voice quality, flat intonation patterns, abnormal pitch contours (Lewis, Freebairn, Heeger & Cassidy, 2002) weak speaking volume, nasal emission and hypernasality (Munson-Davis, 1988). Although these individuals are often reported as being perceived negatively by their peers (Dykens & Rosner, 1999; Dykens, Hodapp & Finucane, 2000) due to the frequent occurrence of challenging behaviour associated with PWS it is difficult to speculate to what extent unusual vocal quality plays in negative social perception.

The studies above demonstrate the significant impact of aberrant voice features in the generation of negative judgements by listeners, and also serve to illustrate the considerable difficulties researchers face in evaluating the specific role of voice quality in how listeners perceive speakers within complex disorders which affect a variety of speech and language and social interaction mechanisms over and above voice disorder.

2.13.13 Judgements made about speakers with DS based on vocal characteristics

Very little is known about the impact of vocal features on judgements made by listeners about speakers with DS. To date a single study has looked at the effect of voice quality on adult perception of the personality and capabilities of children with this developmental disorder. Moran, Labarge and Haynes (1988) recognised that many earlier perceptual and instrumental studies had identified the presence of abnormal voice features in DS, and that within populations without ID that aberrant features had been found to influence listener perceptions; they therefore sought to determine whether this negative perception would also apply to children with DS.

The voices of ten children with DS (five males and five females, aged between 3 and 13 years) were compared to those of ten TD age and gender matched children. In order that the children with DS were able and willing to complete the task and so that low language level did not influence judgements, the voice sample was limited to the production of three isolated, prolonged vowels (/i/, /a/, /u/). The listeners (twenty-four university students in education related courses, none of whom had any experience of rating voice quality, or any training in voice disorders, and little direct experience of DS or ID) were asked to rate the speakers across twelve parameters (table 2.2). The parameters were presented in the form of a semantic differential scale where subjects could be rated at one end of the scale as having a strong association with the positive adjective (scoring 1), and at the other a strong association with the negative adjective (scoring 7), a neutral response was indicated by a mid-point rating of 4.

Parameters investigated by Moran, Labarge and Haynes (1988)	
Semantic differential parameters	Lazy-ambitious
	Unfriendly-friendly
	Distractible-attentive
	Excitable-calm
	Unsociable-sociable
	Not likeable-likeable
	Timid-confident
	Unpleasant-pleasant
	Uncooperative-cooperative
	Impulsive-controlled
	Helpless-capable
	Unreliable-reliable

Table 2.2: Parameters investigated by Moran, Labarge and Haynes (1988) within a 7-point semantic differential scale in their evaluation of the impact of voice quality on the judgements made by adult listeners about child speakers with DS

During the course of the task, listeners were randomly shown a photographic slide of each of the children with DS matched with their own voice recording of the three vowels, and also matched with the voice recording of an age and gender matched TD peer. Thus the researchers were able to contrast the ratings of the recordings in the two different conditions to determine if perception of voice was influenced by the accompanying image.

The results, as predicted, demonstrated that overall, listeners did rate the voices of children with DS significantly more negatively than the voices of their TD peers. In particular ratings of 'confidence' and 'capability' produced the greatest negative ratings. No significant difference was identified in two parameters: 'calm-excitable', and 'cooperative-uncooperative'. The authors suggest that 'calm' may actually have been interpreted by listeners as 'lethargic', thus representing a negative judgement, whilst they felt that the rating of cooperation may have been particularly difficult for the listeners to judge without direct interaction with the children. This study identifies that listeners do indeed use voice quality to make judgements about individuals with DS, and that this voice quality results in more negative perceptions of character and personality traits than attributed to TD peers. It might be argued however, that the use of prolonged vowels, although effectively ensuring that language is not a confounding factor in perception, does not adequately reflect the diversity of speech found in conversation or narrative samples and furthermore may not be of sufficient length for listeners to make accurate judgements.

The only other perceptual study to date for this population explored the impact of voice on the perceived age and gender characteristics of voices of institutionalised children with DS (Montague, 1976). In conjunction with unusual voice quality, children with DS are described as being less well developed in terms of their physical and sexual maturation. Montague proposed that these differences might result in the perception of immaturity and less concrete judgements about gender compared to TD children.

The groups studied consisted of ten boys and ten girls with DS from institutionalised backgrounds, with a mean age of 10.42 years and a control group of age and gender matched TD peers. The majority of the DS subjects were identified as having significantly elevated hearing thresholds, indicative of a mild conductive loss, whilst a further six children with DS could not be evaluated for hearing impairment.

For each subject three recorded words were selected from a list of words which were presented by the researcher and then repeated by each subject. In order to control for possible bias based on articulatory errors the samples were played backwards to the sixteen undergraduate students who rated the voices of the children. The listeners, who were unaware of the diagnosis of any of the children, were asked to judge only the gender of each sample on the first pass. The second playback involved judgements of age, where the listeners were given eight scaled choices ranging between 6 and 13 years.

The results indicated that listeners found it very difficult to accurately judge the gender of children with DS from their voice alone; the author suggests that this may in part be due to the physical and sexual underdevelopment of these children in relation to their chronological age. With regard to judgements of age, unlike the TD controls, the children with DS were perceived as being significantly younger than their actual age (the TD mean of 10.41 years was perceived as 10.45 years, whilst the DS mean of 10.42 years was judged as only 8.17 years). Montague notes that previous studies have found that five vocal characteristics are strong predictors of age in adult males (aged 40-80 years), these being: voice tremor; laryngeal tension; air loss; imprecise consonants; and slow rate of articulation (Ryan & Burke, 1974). As the first three relate directly to laryngeal physiology, Montague suggests that deviant physiological parameters may be related to age-judgements about children with DS also.

As in the previous study, there are some methodological factors which should be considered when interpreting the results. Firstly in relation to the recordings used, the duration of the voice samples is short, and again by playing the samples backwards it is likely that the natural features of voice are distorted, which may affect subsequent voice judgements. Additionally, rather than spontaneous speech, voice samples are of words which are first modelled by the investigator which may have resulted in recordings which don't necessarily reflect typical speaker stress or pitch patterns. The second issues concern the DS subjects used. The majority have evidence of some degree of hearing loss; a factor which is likely to have affected habitual voice quality. Although it is conceded that the presence of hearing impairment is so common in DS that these subjects may be an appropriate representation of the typical voice quality of this population, it may have been helpful to have excluded outright those who could not comply with audiological testing who may have had significantly elevated thresholds. Finally, as the children with DS all came from an institutionalised background (a constraint of the model of care for people with ID at the time of this study) it may be unwise to relate the results directly to current day populations of DS where large impersonal institutions are no longer prevalent.

From both these perceptual studies it can be seen that the atypical voice quality associated with speakers with DS does have a negative impact on how listeners judge their personality and abilities as well as making judgements of age and gender problematic.

2.14 SUMMARY

A range of medical and psychological conditions occur with a high frequency in DS. These include the presence of hearing loss (Balkany, Downs, Jafek & Krajicek, 1979) and visual impairments (Woodhouse, 2002), ID (Pennington, Moon, Edgin, Stedron & Nadel, 2003), mental health problems (Pueschel, 1998), growth deficits (Pueschel, 1993) hormone disturbances (Criscuolo, Perrone, Sinisi, Bellastella & Faggiano, 1986; Cutler, Benezra-Obeiter & Brink, 1986; Dinani & Carpenter, 1990) and congenital heart defects (Marino, 1992). Although the severity of these features may vary considerably within individuals, and indeed not all features may be present in any one individual, the constellation of potential difficulties reveal a complex pattern of disability which will have significant impact on both quality and quantity of life if not managed appropriately.

A wide range of speech and language difficulties are also associated with this population, with a tendency for deficits to be most marked with the onset of oral communication. These difficulties have been found to be more significant than would be expected from cognitive levels (Wisniewski & Miezejeski, 1988). Expressive language tends to be more affected than comprehension (Chapman & Hesketh, 2001) and grammatical ability more than vocabulary skills (Caselli et al., 1998). As with the phenotypic physical characteristics of DS these speech and language difficulties will affect individuals differently, necessitating personalised learning goals which take into account individual strengths and weaknesses and likes and dislikes.

Social communication skills, despite often being regarded as an area of strength, can be seen to vary considerably according to factors such as the degree of ID and opportunities to develop social competence. Specific social deficits such as fewer instances of requesting behaviours and inconsistent turn-taking skills, (Beeghly, Weiss-Perry & Cicchetti, 1990) reduced conversational repertoires (Berglund, 2001) less frequent initiation of and response to social interactions than peers (Sinson & Wetherick, 1982) and poorer ability to establish shared referents (Abbeduto, Weissman & Short-Meyerson, 1999) are all observed to varying degrees. Children with DS tend to play in a more solitary and less co-operative way (Hamilton, 2005) and have poorer attention and motivation (Abbeduto & Keller-Bell, 2003) compared to TD peers. Despite a good understanding of what friendship is and generally low levels of maladaptive behaviours, immature interests and reduced independence are likely to negatively impact on social inclusion and the development of meaningful friendships with TD peers both in and out of school (Cuckle & Wilson, 2002).

Differences in the perception of ability and behaviour have been reported between parents and some professionals (Bhattacharya & Sidebotham, 2000). In order that the optimum service can be provided both perspectives need to be considered. The vast heterogeneity of this population also requires the creation of individual programmes to develop skills as opposed to syndrome specific interventions (Reiter, Tirosh, Bar-Tikvah & Adam, 1992). Many young people with DS are now going on to further education at local colleges and are generally enjoying a higher profile within society. Given that there is evidence that social communication skills can continue to increase with age (Rosner, Hodapp, Fidler, Sagun & Dykens, 2004) such educational and therapeutic interventions may be appropriate into adulthood.

At the present time the voice quality of people with DS has not received the level of investigation attributed to other speech and language domains, despite the early observation of a high incidence of voice disorder (Strazzulla, 1953; Schlanger, 1962; Tredgold & Soddy, 1963; Blanchard, 1964; Benda, 1965; Novak, Sedlackova, Klajman & Betlycwski, 1967). From the very early days of voice research a range of aberrant vocal features have been described within the literature. The terms 'hoarse' (West, Kennedy & Carr, 1947) 'guttural' (Penrose & Smith, 1966) and 'raucous' (Benda, 1949) have all been attributed to speakers with DS, whilst perceptually pitch has been perceived to be lower than that of TD peers. Despite this, scientific advances revealed that F0 did not in fact differ from TD levels in school-aged

English-speaking children with DS (Michel & Carney, 1964; Hollien & Copeland, 1965; Montague, Brown & Hollien, 1974; Pentz & Gilbert, 1983) although it was found to be lower in young Portuguese-speaking children with DS (Moura et al., 2008) and significantly higher in English-speaking pre-school children (Weinberg & Zlatin, 1970) and also in adults with DS (Moran & Gilbert, 1978).

Other objective findings from acoustic analysis have identified an increase in the vocal fold irregularities of frequency and intensity perturbation and increased noise in the acoustic signal (Moran & Gilbert, 1982; Pentz & Gilbert, 1983; Moura et al., 2008) as well as lower than typical levels of spectral tilt (Moura et al., 2008). Although hypernasality has been identified by experienced raters of voice, these findings are not always replicated by studies using inexperienced listeners (Montague & Hollien, 1973; Rolfe, Montague, Tirman & Vandergrift, 1979; Pentz & Gilbert, 1983) suggesting that resonance disorder is less salient perceptually than phonation type, which is judged more consistently (Pentz & Gilbert, 1983).

There is a general consensus that it is the interaction of several variables rather than a single factor which results in the distinctive vocal profile of people with DS (Novak, 1972; Pentz & Gilbert, 1983; Moran, 1986; Pentz, 1987). Structural abnormalities of the vocal tract combine with functional differences brought about by hormonal factors affecting the laryngeal and vocal tract mucosa (Benda, 1969; Strazzulla, 1953) and low muscle tone causing the vocal tract musculature to be flaccid, contributing to vocal fold and velopharyngeal insufficiency and acoustic damping (Rolfe, Montague, Tirman & Vandergrift, 1979). Hearing impairment is proposed to affect the ability to self-monitor quality, intensity and pitch (Montague & Hollien, 1973; Montague, 1976) whilst poor vocal behaviours (Pentz, 1987), inadequate vocal hygiene and individual personality differences (Lee, Thorpe and Verhoeven, 2009) may further impair an already compromised system.

The vocal differences associated with DS, have been found to have an impact on the way in which these individuals are perceived by others. Children with DS have been judged more negatively than their TD peers on measures of personality and ability

(Moran, LaBarge and Haynes, 1988) and perceived as being younger than their chronological age, whilst judgements of gender have been found to be ambiguous (Montague, 1976). Perceived immaturity is likely to result in lower expectations of these individuals (Fidler, 2003) which may impact negatively on their self-esteem and mental health. The correct management of communication difficulties and voice disorder are noted to have a positive impact on the social integration and emotional wellbeing of children with DS (Venail, Gardiner & Mondain, 2004).

Advances in knowledge and understanding of DS and its associated conditions, together with more effective medical and therapeutic interventions have increased the life expectancy of people with DS over the last few decades (Fonseca, Amaral, Ribeiro, Beserra & Guimaraes, 2005) and thus it can be expected that the numbers of people with DS will increase, even in the presence of decreased births due to more prenatal screening (Sadovnick & Baird, 1992). In light of this, the medical, educational and therapeutic professions must work together to plan and implement strategies for long-term support for individuals with DS and their families.

3
METHOD

3.1 INTRODUCTION

This research project can be divided into three distinct studies:

- Questionnaire-based analysis of listener judgements of voice (study 1)
- Acoustic analysis of voice (study2)
- Perceptual analysis of voice (study 3)

Correlations between the results of the three studies are then analysed.

3.1.1 Study 1 is a questionnaire study, examining the judgements that listeners (special-needs education staff, SNES; mainstream education staff, MES; & typically-developing peers, PEERS) make about the personal characteristics and abilities of a group of eight children with Down's syndrome (DS) compared to an age- and gender-matched group of eight typically developing (TD) children, based on audio-recordings alone.

3.1.2 Study 2 is an objective analysis of a range of acoustic correlates of voice quality, including measures of fundamental frequency (F0), perturbation (jitter & shimmer), harmonic-to-noise ratio (HNR) and spectral tilt. It is based on a wider sample of DS and TD recordings (22 DS & 52 TD), but also includes a sub-analyses of DS and TD children by gender, and analysis of the sixteen (8 DS & 8 TD) voices presented in study 1.

3.1.3 Study 3 is a perceptual analysis of voice quality, based on a recognised voice rating scale, completed by expert raters of voice quality. This study is based on the same samples as study 2.

The research aims to determine whether there are significant differences in the above studies between and within groups, and furthermore to establish whether there are interactions between the findings from all three studies. It is anticipated that the findings will contribute significantly to the existing knowledge base of DS and TD voice characteristics, and help to form the basis of a framework which will increase understanding of the interaction between specific perceptual and acoustic voice features and listener judgements.

3.2 RECRUITMENT OF PARTICIPANTS (studies 1, 2 & 3)

3.2.1 Children with DS

The children and young people with DS taking part in this study of voice quality were recruited with the assistance of an MRC-funded research project running within the Speech Science Research Centre at Queen Margaret University, Edinburgh entitled: "Assessment and treatment of impaired speech motor control in children with Down's syndrome" (MRC grant number G0401388); henceforth this will be referred to as the 'MRC speech motor control' study.

With permission, information packs were posted or passed on in person to all parents whose children with DS were involved in the MRC speech motor control study, requesting that their child complete a single, short recording at one of their scheduled MRC study articulation therapy or assessment sessions at the university, in order to support the voice research. Packs contained information sheets, describing the nature of the study, what would be required of the child, any anticipated risks of taking part and contact details for the PhD researcher and a second member of research staff who was independent of the study, should further questions or concerns be raised (appendix I). For the children with DS, who all had some degree of intellectual disability, a simplified version of the information sheet utilising symbols and photographic images was used to gain informed consent directly from each child (appendix II). Where possible both a parent/guardian and the child were asked to sign a consent form agreeing to take part (appendix III). It was stressed that participation was voluntary, could be withdrawn at any time without explanation, and that participation would not reduce any allotted MRC study articulation therapy time nor would opting out of the voice study affect in any way the child's involvement in that research. Parents were also reassured that data would be held securely within the university, and that all recordings would be coded in order to protect the anonymity of their child. As all of the children with DS had already received an incentive (gift token) from the MRC speech motor control study, and as the recordings were completed during the articulation sessions which were part of that research, no further incentive was offered to the children to take part in this study.

Written consent was received from twenty-five parents/children approached (one participant, whose recording quality was poor was later excluded, as were a further two, who failed to produce a suitable narrative on the day of recording). All lived within the central belt of Scotland and were in full-time education at either a mainstream or special-needs school, or in the case of the older people with DS, attending a local further-education college.

All the children/young people with DS had already been subject to a strict exclusion criteria for the MRC speech motor control study, as such no further criteria were set for exclusion from the voice study.

Children were excluded from the MRC speech motor control study and the voice study if:

1. English was not the child's first language or the principle language used within the home
2. There was evidence of a severe hearing loss (designated as an aided threshold of >40dB)
3. The child had a dual diagnosis of autism
4. The child's cognitive age-equivalent was <3 years

3.2.2 Typically-developing children (control subjects & peer raters)

Although the children with DS were able to be recruited via the MRC speech motor control study the control group of TD children could not be as the speech motor control study had matched control subjects on the basis of developmental age. As a similar stage of physical development of the vocal tract between groups was deemed a necessity for a study of voice quality, controls needed to be matched for chronological age.

TD children and young people were recruited in a number of ways. An application to the Children and Families Department of The City of Edinburgh Council was granted, allowing the researcher to directly approach schools within the city of Edinburgh. Letters detailing the needs of the study were subsequently sent to six primary schools and ten secondary schools in the city, resulting in participation from a single primary school and two secondary schools. Information packs and consent forms were sent in advance to each school (appendices III, IV & V) and the researcher negotiated suitable times to interview the children within the school premises during the school day.

The head office of the South East Scotland Regional Scout Council in Edinburgh was also approached and a suitable local scout group identified to support the research as peer raters (study 1). The young people from this group were not recorded due to the Scout meeting room being unsuitable for this purpose.

Finally, TD young people were recruited via general e-mail requests sent to all staff and students on the Queen Margaret University e-mail system (in line with university protocols); these individuals were recorded within the Speech Sciences recording studio at the university.

Written consent was received from all participants and their parents/carers prior to taking part in the study. An incentive of a £10 Zavvi gift voucher was given to all the TD children and young people who took part in the study.

The TD children were subject to a simple inclusion criteria:

1. English was required to be the child's first language and the language spoken in the home
2. No identified hearing loss
3. No identified speech and language difficulties
4. No identified intellectual disability
5. Children must have been brought up within the central belt of Scotland in order to be representative of the accent features of the children in the DS study group

3.2.3 Education Staff

The special-needs education staff (SNES) and mainstream education staff (MES) were recruited from schools within the Midlands area of England. Although it is acknowledged that it would have been preferable to recruit teachers from within the Edinburgh area, due to the poor response rate of schools targeted for peer participants the decision was taken to recruit from elsewhere. The researcher had worked in special-schools in the Midlands previously, and consequently had a number of contacts who expressed interest in supporting the study. In total ten mainstream schools and four special-needs schools were approached, from which four mainstream and three special-needs schools agreed to take part in the study.

The term 'education staff' includes teachers, teaching assistants and support workers, as all were deemed to have regular contact with pupils in the classroom environment. All staff were required to have two years experience of working in their respective areas. All received information sheets and consent forms (appendices VI and VII) prior to taking part.

3.3 STUDY 1: QUESTIONNAIRE-BASED ANALYSIS OF LISTENER JUDGEMENTS OF VOICE

3.3.1 Pilot Study

A small-scale pilot study was completed in order to test the methodology of this task, in particular the questionnaire design and the mode of presentation of voice samples, before commencing the full-scale study.

Audio-recordings of two children with DS (11.25 & 14.0 years) and two TD children (aged 11.83 & 12.08 years) were played to three children (ages, 12.08, 11.83 & 10. 0 years) and two adults. The listeners were asked to make judgements about the characteristics that they associated with each recording on a specially designed semantic differential type questionnaire. They were then asked to give feedback on how accessible they found the task. This feedback guided further changes in the finalised layout of the questionnaire, the rate at which the recordings were presented and the duration of the task.

3.3.1.1 The questionnaire design

Following the rationale of Moran, LaBarge and Haynes (1988) the 5-point bipolar adjectival scales used to evaluate parameters such as ‘happy-sad’ and ‘calm-angry’, were originally presented in a random order. Thus ‘happy-sad’ ratings were followed by ratings of ‘angry-calm’ rather than ‘calm-angry’. Some of the participants expressed that having the parameters running in the same direction (e.g. positive-negative) would avoid confusion, especially where the opposite negative adjective read very similarly to the positive one (e.g. intelligent & unintelligent).

Action: All bipolar adjectival parameters were presented in a positive to negative direction.

3.3.1.2 The pace of the task

The recordings of each speaker lasted approximately one minute with a silent pause of ten seconds between each. In order that listeners rated in an instinctive manner, giving their initial 'gut response', once started the playlist of recordings was left to run until the end of the final voice. Listeners found that they did not always have sufficient time to complete the questionnaire for a speaker and then turn the page to prepare for the next recording.

Action: The ten second silent pauses between recordings were increased to fifteen seconds, and where necessary, the researcher paused the recordings to allow individuals to catch-up. At the end of each recording the researcher prompted listeners that the next was about to start.

3.3.1.3 The duration of the task

Although this was a short pilot of only four recordings (lasting approximately five minutes) both adults expressed that they felt it would be difficult to maintain attention and enthusiasm for the full planned duration of the task (over twenty-six minutes). A parent of one of the child participants also felt that the accuracy of her child's answers might suffer in such a long exercise.

Action: The group size of the DS and TD recordings was reduced from ten to eight subjects in each group (plus repetition of two recordings from each to evaluate consistency of repeated judgements). This reduced the length of the task by over five minutes to approximately twenty minutes.

3.3.2 Participants

3.3.2.1 Rater groups

The full-scale study consisted of three groups of listeners:

- **Special-needs education staff raters:** Fifty-two in total, consisting of eighteen support workers (17 females & 1 male) and thirty-four teachers (27 females & 7 males), ranging from early twenties to mid-sixties.

- **Mainstream education staff raters:** Thirteen teaching assistants (11 females & 2 males) and thirty-two teachers (14 females & 18 males), also aged between early twenties and mid-sixties.

- **Peer raters:** In this group there were seventy-five children (43 males & 32 females) aged between 10.25 and 17.92 years.

These are the overall numbers from which data was selected for analysis for each questionnaire parameter based on consistency of judgement scores; raters judged to show poor consistency in ratings for any parameter were excluded from further analysis (see section 3.3.8).

3.3.2.2 DS and TD speakers

Each group of raters was required to make judgements about the personal characteristics and attributes that they associated with a small subset of the total DS and TD audio-recorded speakers. The finalised playlist consisted of the recordings of eight children with DS, seven males and one female, aged between 10.1 and 16.5 years (median age 11.42 years) and eight TD children, seven males and one female aged between 10.0 and 15.0 years (median age 12.17 years).

3.3.3 Audio recording process

The audio-recordings for all the children and young people were elicited using a purpose-designed picture-description exercise. The researcher produced five hand-drawn pictures containing colourful images of familiar vocabulary which represented a series of events which could be made into a story (appendix VIII). The use of pictures which tell a story had the advantage of gaining a relatively naturalistic sample of speech, whilst ensuring that the children produced a high-frequency of the same vocabulary, enabling comparisons to be made more easily between subjects. The task was also considered to be reasonably accessible to the children with DS, all of whom had some degree of intellectual disability. Recordings were completed by the speech and language therapist involved in the MRC speech motor control study as she was already familiar with the children, and thus most likely to elicit speech which was representative of the children's typical abilities.

Although every attempt was made to record the children in a quiet, distraction-free environment, as many of the recordings took place in busy schools, some background noise was to be expected. Extraneous noise and voices other than the target speaker were edited from the audio files before analysis.

Recordings for all the participants were made using the same Marantz PMD670n Compact Flash Digital Audio Recorder with Beyer MPC65 boundary microphone.

3.3.4 Selection of recordings for use in study 1

The recordings of the eight DS children presented to listeners were selected on the basis of their expressive language scores from the 'Clinical Evaluation of Language Fundamentals-Preschool UK' (CELF-P. UK; Wiig, Secord & Semel, 1992) which was completed as part of the MRC speech motor control study; those with the highest scores at the time of the production of the playlist being included (see appendix IX for assessment results and general information about the age, gender and education status of the selected speakers). This measure was introduced in order to reduce the risk of poor expressive language ability (rather than voice quality) influencing ratings made by listeners. The eight TD children were then selected to match the DS group for overall age and gender. From both of the groups two recordings (25% of the total recordings presented) were picked randomly to be played a second time within the playlist in order that reliability of judgements made by individual raters (i.e. consistency of the ratings from each individual listener for the same voice heard a second time) could be checked.

The narrative samples were edited into strings of single utterances, each separated by a one second silent pause (see appendix X for examples of speech rated); the total length of each sample was up to one minute. A silent pause of fifteen seconds was included between each of the twenty voice samples, creating a total playlist of approximately twenty minutes. In order to try to more evenly match the language used by the DS and TD children, long/complex grammar was edited out of the TD samples. Also to avoid any lack of story-telling coherence within the samples of the children with DS becoming a primary focus the individual utterances were presented randomly. Voice samples were not edited within utterances so that the natural prosodic elements of speech were preserved.

3.3.5 Questionnaire Design

Questionnaires, using a 5-point semantic differential scale (appendix XI, child questionnaire & appendix XII, adult questionnaire) were devised to evaluate the judgements which children and adults made about the characteristics and personality traits of DS and TD speakers. In order to enable direct comparison of results both the child and adult questionnaires followed the same format (with one additional question being put to the peer raters).

The parameters investigated were presented in a positive to negative direction as follows:

- Confident-shy
- Calm-angry
- Friendly-unfriendly
- Happy-sad
- Intelligent-unintelligent

These parameters were chosen as they are likely to reflect the type of features or attributes which are considered by children and young people when making decisions about the formation of friendships. Additionally, parameters such as confidence and intelligence in particular, might have an impact on the level of expectations held by education staff about children in their care. More specifically, as people with DS are frequently described as having rough or harsh phonation, which tends to be associated with aggressiveness (Tanner, 2007) and dominance (Laver, 1968), it was of particular interest to evaluate the response to the 'calm-angry' parameter. Finally, as these parameters overlap with those used in other studies of voice quality, such as that of Moran LaBarge and Haynes (1988), also looking at children with DS, they allowed some level of comparison between studies.

For all of the above parameters, listeners were asked to circle the single statement which best described each of the individual recordings played. For example:

Speaker 1 sounds:

'Very happy' *'Quite happy'* *'Neutral'* *'Quite sad'* *'Very sad'*

Listeners also judged the age and gender ('male', 'female' or 'not sure') of each voice, and in addition to the education staff questions, the peer raters were asked if they would feel comfortable spending time in the company of each of the speakers ('yes', 'no' or 'not sure'). For this latter parameter it was emphasized that judgements were to reflect whether or not the speakers' voices might cause the peer listeners to feel any social discomfort or embarrassment which might inhibit their desire to be in the company of the speakers, rather than which speakers would be selected to be close friends.

3.3.6 Completion of questionnaire by rater groups

The listener questionnaire was completed at the various venues from which the listeners were recruited. Rooms were quiet and distraction-free, and the numbers of listeners in groups kept relatively small (up to ten persons at a time) to ensure adequate proximity to the loudspeakers. The volume of the recordings was consistent throughout all sessions.

Prior to listening to the voice samples the researcher explained fully the requirements of the task. Listeners were told that they would hear twenty voices and should complete a short questionnaire for each. The questionnaires were read through and any parameters which were not familiar to the listeners were explained using examples. Listeners were asked to rate according to their first impressions of each voice, and to focus on the overall quality of the voices rather than specific language ability, clarity of articulation, or the degree of sense of the narrative told.

During presentation, the researcher remained in the room so as to be able to provide assistance if required and to indicate when individual recordings were complete and the next about to begin. Ratings by listeners were not directly observed during the task as it was hoped that this would be less inhibiting and encourage the listeners to rate honestly without fear of criticism or comment. Similarly, the finished ratings were not examined in front of the raters at the end of the task.

3.3.7 Data Analysis

In order that the two DS and two TD repeated recordings did not skew results only the data from the first set of ratings by the three listener groups for these four individuals was used in the analysis.

The data from the completed listener questionnaires was first of all coded. For the parameters of 'happy-sad', 'calm-angry', 'confident-shy', 'friendly-unfriendly' and 'intelligent-unintelligent' the following system of numerical coding was used:

- 'Very happy' (code 4)
- 'Quite happy' (code 3)
- 'Neutral' (code 2)
- 'Quite sad' (code 1)
- 'Very sad' (code 0)

For each of the parameters a subjective decision was made to consider those adjectives coded as '4' (very happy, very calm, very confident, very friendly & very intelligent) to be the most positive, whilst those at the other end of the scale coded '0' were deemed the most negative; this method of coding allowed higher scores to be interpreted as more positive judgements made by listeners.

By identifying the spread of ratings made about the DS and TD groups across the codes (0-4) for each parameter in relation to the total number of judgements made by listener groups and the highest total possible, the percentage values of scores allocated to each code could be calculated. For example the 73 PEER raters made a total of 584 ratings for the 8 TD speakers for the parameter happy-sad. Of these 584 ratings 91 were judged as 'very happy' (code 4) which translates to 15.58% of the happy-sad ratings for the TD group, whilst at the other end of the scale only 5 ratings of 'very sad' (code '0') were made, translating to 0.86% of the TD ratings for this parameter.

As it would be clumsy to have to discuss repeatedly the five percentage scores (representing codes '0-4') the above system was extended to create an overall percentage score for the DS and TD groups for each parameter. This was done by determining the highest possible score for all the speakers in a given group by all the raters in a given group and then identifying what percentage of that total score was actually achieved. For example for the happy-sad parameter it was possible for the 73 PEER raters to make an overall highest score of 2336 for the eight speakers in the TD group (73 judgements, all at code '4' for all 8 speakers). However, the actual scores reflected 91 ratings of code '4' (score of 91 x 4 = 364), 209 ratings of code '3' (score of 209 x 3 = 627), 190 of code '2' (score of 190 x 2 = 380), 89 of code '1' (score of 89 x 1 = 89), and 5 of code '0' (score of 5 x 0 = 0) which combined create a total score of 1460, equating to 62.5% of the total 2336 highest possible score. Again this system enables higher overall percentage scores to be interpreted as more positive overall judgements and allows a single overall figure to be discussed for each parameter for each group, thus increasing clarity. This system was repeated to identify the overall percentage scores for individual speakers within the DS and TD groups.

For the judgements of gender, ratings of 'male' were coded '1', 'female' coded '2' and 'not sure', coded '3'. From these codes the percentage of correct and incorrect gender judgements were calculated for each voice and across groups, as well as the percentage of those judged to be ambiguous.

The peer ratings concerning desire to spend time in the company of each speaker ('spend time with') were coded as 'yes, 2', 'no, 0' and 'not sure, 1'; as before, a higher score (and converted percentage) indicating those individuals, or group, who proved to be more socially desirable.

Age judgements did not require to be coded, the actual judged ages forming the raw data for further analysis. The median judged age of individuals and overall groups could then be compared to actual ages.

3.3.8 Consistency of group judgements for repeated samples

Although 52 SNES, 45 MES and 75 PEER raters completed the listener questionnaire, only the judgements of those who demonstrated reasonable consistency between their first and second ratings of the four randomly selected repeated recordings were put forward for analysis, thus creating different numbers of final raters for each parameter. The percentage of consistent ratings for each listener group for each repeated voice, and across the four voices as a whole are reported in the results in order to illustrate the effectiveness of the following 'consistency criteria' in removing data which is unreliable from the final analysis. As a consequence of using only the consistently judged data, results can be considered to be a reliable reflection of the judgements made by the SNES, MES and PEER listener groups.

3.3.8.1 Criteria for consistency of listener judgements

For the parameters of 'happy-sad', 'calm-angry', 'confident-shy', 'friendly-unfriendly', 'intelligent-unintelligent' and 'spend time with' the following exclusion criteria were applied:

Individual raters were excluded where two or more of the four repeated voices were rated >1 scalar degree different on the second rating. (e.g. on the rating scale of 0-4, ratings of 0 on the first rating, followed by 1 on the second, 1 followed by 2, 2 followed 3, or 3 followed by 4 are acceptable, but not for example a rating of 0 followed by 2 or 1 followed by 3); the same principle applied to the smaller scale of 0-2 for the 'spend time with' parameter. The final numbers of raters whose data was deemed suitable for inclusion in the final analysis for each of the above parameters is shown in table 3.1.

Final listener group numbers based on intra-rater reliability judgements			
PARAMETER	**SNES**	**MES**	**PEER**
Calm - angry	51	45	73
Confident - shy	43	39	65
Friendly - unfriendly	52	42	70
Happy - sad	51	44	73
Intelligent - unintelligent	49	41	66
Spend time with	N/A	N/A	72

Table 3.1: Number of SNES, MES and PEER raters having adequate consistency of judgements, whose data was used in the analysis of the 'calm-angry', 'confident-shy', 'friendly-unfriendly', 'happy-sad', 'intelligent-unintelligent' and 'spend time with' parameters

For judgements of 'gender' individual raters were excluded where 2 or more of the 4 repeated voices were rated differently on the second rating (i.e. ratings of 'male' & 'male' or 'female' & 'female' are acceptable, but not 'male' & 'female', 'male' & 'not sure' or 'female' & 'not sure'). Final numbers are shown in table 3.2.

Final listener group numbers based on intra-rater reliability judgements			
PARAMETER	**SNES**	**MES**	**PEER**
Gender	49	40	74

Table 3.2: Number of SNES, MES and PEER raters judged as having adequate consistency of judgements, whose data was used in the analysis of the 'gender' parameter

For the parameter of 'age', a more complex set of criteria were necessary. Individual raters were excluded where 2 or more of the 4 repeated voices failed to meet the following criteria:

- **SET 1** used for judgements **where both repetitions are judged as <19 years:**

Judgements are excluded where the difference in judgements is >2 years (e.g. 17 & 15 or 18 & 16 are acceptable but not 15 & 18, or 13 and 16).

- **SET 2** used for judgements **where both repetitions are judged between 19 and 25 years:**

Judgements are excluded where the difference in judgements is >3 years (e.g. 19 & 22 or 21 & 24 are acceptable, but not 19 and 23, or 21 & 25).

- **SET 3** used for judgements **where both repetitions are judged as >25 years:**

  Judgements are excluded where the difference in judgements is >10 years (i.e. 26 & 36, or 55 and 65 are acceptable, but not 26 & 37, or 55 & 66)

For judgements where judged ages span across the above sets, the average of the sum of the acceptable margins of difference for those sets will be used. Therefore:

- where ratings span SET 1 (<19 years) and SET 2 (19-25 years) the average of the acceptable difference for SET 1 and SET 2 will be applied (2+3 years = **2.5 years**).
- where ratings span SET 2 (19-25 years) and SET 3 (>25 years) the average of the acceptable difference for SET 2 and SET 3 will be applied (3+10 years = **6.5 years**).
- ratings which span SET 1 and SET 3 will automatically be excluded.

Numbers of raters meeting these ‘age’ criteria are shown in table 3.3.

Final listener group numbers based on intra-rater reliability judgements			
PARAMETER	**SNES**	**MES**	**PEER**
Age	30	30	59

Table 3.3: Number of SNES, MES and PEER raters judged as having adequate consistency of judgements, whose data was used in the analysis of the ‘age’ parameter

3.3.8.2 <u>Rationale for consistency of judgements scale for ‘age’ judgements</u>

The increasing acceptable margins for differences in repeated judgements of age between sets 1, 2 and 3 (2, 3 & 10 years respectively) are based on the premise that estimation of the age of some of the older speakers, who may possess adult-like, or approaching adult-like physiology, may be more difficult to judge than for the younger speakers (as adult features naturally reflect patterns which would be associated with a much broader age-band than those of, for example, pre-pubertal voices).

Changes in voice are associated with the development of secondary sexual characteristics and the onset of puberty. Voice break in is no longer considered to be a rapid phenomenon, associated with a sudden drop in acoustic values, rather it is a process which occurs relatively slowly, and across a wide band of ages; the typical onset of adult male voice ranging from 12.5 to 17.5 years, with a mean of 15.0 years (Hägg & Taranger, 1980). It is generally accepted that growth retardation and gonadal deficiency are well-known features of DS (Arnell, Gustafsson, Ivarsson & Annerén, 1996) and these differences might be imagined to have consequences for pubertal development. However, according to Pueschel, Orson, Boylan and Pezullo (1985); Hsiang, Berkovitz, Bland, Migeon and Warren (1987); Sakadamis, Angelopoulou, Matziari, Papameletiou and Souftas (2002) the pattern of development of secondary sexual characteristics in males with DS is consistent with typical development. On this basis, adult-like voice features can be expected to be found within both the TD and DS male voices presented in the listening task.

It is only in recent years that it has come to be accepted that voice-breaking occurs gradually as a consequence of the development of secondary sexual characteristics in girls as well as boys (Charpy, 2002). Although early studies of pubertal development in girls with DS pointed towards a delay in onset (Roche, 1965; Rundle, 1970) more recent studies have failed to replicate these findings; Goldstein (1988), Scola and Pueschel (1992) and Elkins (1992) all finding the average onset of menarche in girls with DS to be not dissimilar to the average age of onset in TD girls. The most recent data on menarcheal onset in the UK identifies a median age of 12.9 years for TD girls (Whincup, Gilg, Odoki, Taylor & Cook, 2001), and only a slightly lower age of 12.5 years has been found for girls with DS (Howard, 1989). As both the DS and TD females in the listening task are younger (DS30, 10.08 years & TD4, 11.83) than the average identified ages of onset of puberty for those groups, it is less likely that they will display adult-like voice features.

3.3.8.3 Method for determining consistency of group judgements: individual repetitions and overall values

Once the data from the excluded listeners had been removed from the data set it was possible to calculate the percentage of consistent ratings that the remaining raters in the SNES, MES and PEER groups as a whole made for the four repeated voices (TD2, TD7, DS13 & DS7). Each rater was scored '0' for a rating that was inconsistent and '1' for a consistent rating, allowing calculation of the percentage that were judged consistently (e.g. for the 'confident-shy' parameter the 65 PEER raters could score a highest possible 65 consistent judgements (100%) for TD2; the actual number (42) represented 64.62% of the total level of consistency possible). Therefore high percentage values indicate voices which listeners have judged with better consistency and low values those judged less consistently.

An overall consistency score was then calculated for each group following the same principle of determining what percentage of the four repeated voices combined were judged as consistent (i.e. the 65 PEER raters could achieve a total of 260 consistent judgements (65 x 4), whilst the actual figure achieved was 221 (TD2, 42 + TD7, 63 + DS13, 59 + DS7, 57) which equated to an overall consistency score of 85%.

3.3.9 Statistical analysis

All the coded (ordinal) data from the listener questionnaire was categorised as qualitative data and therefore treated non-parametrically. The judgements of 'age' (ratio scale) would normally be treated as quantitative data, and subject to parametric hypothesis testing. As a normal distribution is a prerequisite of parametric testing this data was first analysed using a Shapiro-Wilk test. Results showed that the data of all three groups of raters for the DS and TD groups fell outwith the required normal distribution (table 3.4); consequently all ‘age’ judgements were analysed using non-parametric statistical tests.

Results of Shapiro-Wilk test for 'age' judgements		
	DS	**TD**
SNES	W = 0.790, df = 240, p < 0.001	W = 0.983, df = 240, p = 0.006
MES	W = 0.695, df = 240, p < 0.001	W = 0.965, df = 240, p < 0.001
PEER	W = 0.703, df = 470, p < 0.001	W = 0.975, df = 472, p < 0.001

Table 3.4: Results of Shapiro-Wilk test of the normality of distribution of ‘age’ judgements of the DS and TD groups by the SNES, MES and PEER listeners

3.3.9.1 Analysis of differences between DS and TD ratings within listener groups

For each of the listener groups (SNES, MES & PEER) a set of Mann-Whitney U tests were used to determine if there was a significant statistical difference between the judgements made about the DS and TD groups as a whole for each parameter, excluding ‘spend time with’ and ‘gender’ judgements which were analysed using Chi Square tests as the data was categorical rather than ordinal.

Hypothesis: Listeners from all three groups will rate the abilities and characteristics of the children with DS more negatively than those of the age-matched TD peers and the PEER raters will be less likely to want to spend time with the DS speakers than the TD speakers. Judgements of gender will be less accurate for the DS group and their age judged to be younger than their actual age and that of the TD peer group.

Rationale: Findings from earlier research by Moran, LaBarge and Haynes (1988) found listeners to be more negative in their assumptions of a range of personality traits and abilities in children with DS than children without developmental disorder. Judgements of age and gender, based on the voices of children with DS, have been found to be problematic; age being judged as approximately 2 years younger than actual chronological age and the age of TD age-matched control subjects (Montague, 1976).

3.3.9.2 Analysis of differences between DS and TD ratings between listener groups

A second set of analyses sought to investigate whether differences existed between the judgements made about the TD and DS groups independently, between the three listener groups (MES, SNES & PEER).

For each parameter (excluding 'gender' which used a Chi Square test) a Kruskal-Wallis test was first used to identify whether there were statistically significant differences in the ratings of the DS group between the three sets of listeners. For any parameters which yielded a significant result post-hoc Mann-Whitney U tests were then used (comparing the 'MES & SNES', the 'MES & PEER' and the SNES & PEER' groups) to establish between which groups the significant difference lay. This process was repeated for the ratings of the TD speakers. As Bonferroni adjustment is used when making multiple comparisons when searching for significant associations without pre-established hypotheses (Perneger, 1998) a new p value was established by dividing 0.05 by the number of comparisons made (0.05/3 = 0.017)

3.3.9.3 Analysis of the effect of gender and age on PEER ratings

Gender effects within the PEER listening group were evaluated by dividing the group into males and females. Only those judged as consistent in their ratings were used in the final analysis, therefore participant numbers vary between parameters (table 3.5).

Age-related effects on judgments were evaluated by dividing the PEER raters into two groups, split according to the education class they attended. Children from Primary 6 level through to and including Secondary 2 level were classified as the younger listeners, the older group being the remaining raters, who belonged to secondary 3-6 levels. Again, numbers vary depending on consistency of judgements (table 3.5).

Mann-Whitney U tests were used to evaluate differences between the overall ratings of the DS and TD groups independently for all the questionnaire parameters (excluding 'spend time with' & 'gender' which were analysed using Chi Square tests) by the male and female PEER raters and the younger and older PEER raters.

Numbers of PEER raters divided by Gender and Age		
PARAMETER	**GENDER**	**AGE**
Calm-angry	**MALES**: 43 (10.5 to 17.92) median age 14.33 **FEMALES**: 30 (10.26 to 16.17) median age 14.38	**YOUNG**: 22 males & 16 females, median age 12.33 **OLD**: 21 males & 14 females, median age 15.33
Confident-shy	**MALES**: 36 (10.58 to 16.83) median age 14.46 **FEMALES**: 29 (10.25 to 17.0) median age 14.25	**YOUNG**: 18 males & 17 females, median age 12.33 **OLD**: 18 males & 12 females, median age 15.58
Friendly-unfriendly	**MALES**: 38 males (10.5 to 17.92) median age 14.58 **FEMALES**: 32 (10.25 to 17.0) median age 14.33	**YOUNG**: 18 males & 18 females, median age 11.75 **OLD**: 20 males & 14 females, median age 15.38
Happy-sad	**MALES**: 42 (10.5 to 17.92) median age 14.33 **FEMALES**: 31 (10.25 to 17.0) median age 14.33	**YOUNG**: 22 males & 17 females, median age 12.33 **OLD**: 20 males & 14 females, median age 15.38
Intelligent-unintelligent	**MALES**: 36 (10.5 to 17.92) median age 14.33 **FEMALES**: 30 (10.25 to 17.0) median age 14.33	**YOUNG**: 19 males & 17 females, median age 12.33 **OLD**: 17 males & 13 females, median age 15.46
Spend time with	**MALES**: 40 (10.5 to 17.92) median age 14.46 **FEMALES**: 32 (10.25 to 17.0) median age 14.33	**YOUNG**: 20 males & 18 females, median age 12.33 **OLD**: 20 males & 14 females, median age 15.25
Gender	**MALES**: 44 (10.5 to 17.92) median age 14.46 **FEMALES**: 30 (10.25 to 16.17) median age 14.33	**YOUNG**: 22 males & 17 females, median age 12.33 **OLD**: 21 males & 14 females, median age 15.33
Age	**MALES**: 37 (10.5 to 17.92) median age 14.33 **FEMALES**: 22 (10.42 to 17.0) median age 14.38	**YOUNG**: 20 males & 12 females, median age 12.42 **OLD**: 17 males & 10 females, median age 15.50

Table 3.5: Numbers and median ages of consistent PEER raters split by gender and age for all questionnaire parameters

3.4 STUDY 2: ACOUSTIC ANALYSIS OF VOICE

3.4.1 Participants

Acoustic analysis of 6 separate groups of participants was completed:

3.4.1.1 Overall DS and TD groups

This analysis was based on the entire set of DS and TD recordings. It consisted of a group of twenty-two children and young people with DS (13 males and 9 females) aged 10.08 to 20.33, (average age, 14.36) and a group of TD control subjects consisting of fifty-two children (34 males & 18 females) aged between 10.0 and 18.67, (average age, 13.97).

3.4.1.2 Overall DS and TD male speakers

Thirteen males with DS (median age 16.08 years, ranging from 10.58 to 20.33) compared against thirty-four TD males (median age 14.08 years, ranging from 10.0 to 18.8)

3.4.1.3 Overall DS and TD female speakers

Nine females with DS (median age 13.75 years, ranging from 10.08 to 16.92) compared against eighteen TD females (median age 14.67 years, ranging from 10.0 to 17.0)

3.4.1.4 Overall DS female and male speakers

Nine females with DS (median age 13.75 years, ranging from 10.08 to 16.92) compared against thirteen males with DS (median age 16.08 years, ranging from 10.58 to 20.33)

3.4.1.5 Overall TD female and male speakers

Eighteen TD females (median age 14.67 years, ranging from 10.0 to 17.0) compared against the thirty-four TD males (median age 14.08 years, ranging from 10.0 to 18.8)

3.4.1.6 DS and TD speakers presented in study 1

Eight children with DS, seven males and one female, aged between 10.1 and 16.5 years (median age 11.42 years) compared against eight TD children, seven males and one female aged between 10.0 and 15.0 years (median age 12.16 years)

3.4.2 Recordings

The audio-recordings used in the acoustic analysis were obtained using the same method described in study 1 (section 3.3.3). For each speaker, the full (unedited) length of their recording was used (see appendix XIII for examples).

3.4.3 Data Analysis

Acoustic analysis was completed using Praat (Boersma &Weenink, 2009); a computer program which enables analysis of a range of acoustic parameters of voice. It is a widely used package which has been found to be effective in the differentiation of typical and pathological voices, and its results to be reliable and simple to replicate (Oguz, Tarhan, Korkmaz, Yilmaz, Safak, Demirci & Ozluoglu, 2007; Oguz, Tunc, Safak, Inan, Kargın & Demirci, 2006).

Each audio recording was first edited into voiced and voiceless segments of speech in order that the pitch contour of the voiced parts only could be selected for analysis; this was done using Praat software which visualised the voicing properties of the recording alongside the sound wave. Each segment was then adjusted manually to ensure accuracy (any errors in the automated recognition of voiced versus voiceless segments, or in the representation of the pitch contour, such as octave jumps or drops, typically occurring in transitions between voiced and voiceless segments, being corrected and irregularities in the onset and offset of voicing trimmed). This also allowed any unsuitable segments, such as those with background noise, to be excluded from further analysis. During this process each sound file was converted to a ‘textgrid’ file and then to a ‘pitch’ file according to the requirements of Praat analysis.

These edited files were then subject to a Praat script (Schaeffler, see appendix XIV) specially-designed to analyse the acoustic parameters of F0 (mean & standard deviation), perturbation measures (jitter and shimmer), harmonic-to-noise ratio (HNR) and spectral tilt (SPT), based on data extracted from the Praat voice report for each file. The script was adjusted to recognise only those voiced segments which contained fifteen or more pitch periods in order that sufficient information existed within each segment to effectively analyse their acoustic properties. As there are fundamental differences in the duration of voicing between males and females, the selection of segments based on the number of pitch cycles was deemed a more suitable method of selection than one based on duration of voicing.

3.4.3.1 Acoustic analysis parameters

The DS and TD recordings were analysed to gain information about 5 key acoustic parameters:

- **Fundamental Frequency (F0)**

F0 is the lowest component frequency of a complex sound (Fucci and Lass, 1999). According to Kent, Weismer, Kent, Vorperian and Duffy (1999) it is an acoustic parameter which is used to refer to the physical measure of the lowest periodic component of vocal fold vibration and as such it is the acoustic correlate of perceived pitch (increased F0 causing the perception of higher pitch level). Mean F0 and F0 mean standard deviation (stdev) were measured.

F0 mean is a measure of the average F0 across all the voiced segments of a recording, whilst F0 mean (stdev) indicates the average degree of variability around the mean F0 across all analysed segments, a higher stdev value being associated with a greater degree of pitch movement.

- **Frequency perturbation (jitter)**

This is the short-term variation in the frequency (i.e. duration) of cycle-to-cycle vocal-fold opening and closure (Mathieson, 2001) and one of the two main objective measures for micro-instability or irregularity in vibration of the vocal folds (Wolfe & Martin, 1997). It is frequently used to detect voice pathologies (Kreiman & Gerrat, 2005) with increased values being perceived as breathy, rough or hoarse voices (Farrús, Hernando & Ejarque, 2007). 'Jitter rap' and 'jitter ppq5' were measured.

Jitter (relative average perturbation; rap) is the average absolute difference between a period and the average of it and its two neighbours, divided by the average period (Boersma & Weenink, 2009). Jitter (ppq5) is the five-point Period Perturbation Quotient, the average absolute difference between a period and the average of it and its four closest neighbours, divided by the average period (Boersma & Weenink, 2009).

- **Amplitude perturbation (shimmer)**

Shimmer is the "short-term instability of the intensity of the vocal signal" Mathieson (2001, p. 81). Like jitter, shimmer is concerned with irregularities in the cycle-to-cycle phases of the vocal folds, but of volume rather than frequency. It is associated with poor and inconsistent contact of the edges of the vocal folds (Reijonen, Soderlund & Rihkanen, 2002) and is recognised to vary considerably across speaking styles (Slyh, Nelson & Hansen, 1999). 'Shimmer apq3' and 'shimmer apq5' were measured.

Shimmer (amplitude perturbation quotient 3; apq3) is the three-point amplitude perturbation quotient, i.e. the average absolute difference between the amplitude of a period and the average of the amplitudes of its neighbours, divided by the average amplitude (Boersma & Weenink, 2009). Shimmer (amplitude perturbation quotient 5; apq5): is the same formula as for shimmer apq3 but for a pitch period and its four closest neighbours.

- **Harmonic-to-noise ratio (NHR)**

This is the ratio of periodic components to the aperiodic components in the vocal signal. Increased noise is a consequence of insufficiency of vocal fold closure, causing air to leak through the glottis, as well as being influenced by the presence of short-term frequency and amplitude variations (Reijonen, Soderlund & Rihkanen, 2002); thus this parameter "is a general evaluation of noise in the analyzed signal and is not specific to any cyclic parameter and includes contributions from both perturbations of amplitude and frequency" (Oguz, Demirci1, Safak, Arslan, Islam & Kargin, 2007, p. 259). Increased levels of aperiodic components (which equate to lower HNR) are associated with vocal fold pathology (Mathieson, 2001); such voices are often perceived as sounding "rough, gravelly and breathy" (Mathieson, 2001, p. 154).

- **Spectral tilt (SPT)**

Spectral tilt is a ratio of energy (amplitude values) between different frequency ranges. It functions as a measure of how quickly the amplitudes of the harmonics decline (Lofqvist & Mandersson, 1987); thus can be seen as one of the acoustic correlates of vocal loudness (Beauchamp, 2007) and/or degree of muscular tension (Guion, Post & Payne, 2004) as a more flaccid vocal mechanism will absorb acoustic energy faster than a tense system. It is suggested to be an appropriate measure for spontaneous speech analysis (Hillenbrand & Houde, 1996). 'SPT (1-5kHz)' and 'SPT (2-5kHz)' were measured.

SPT (1-5kHz) is a comparison of the mean energy at 1000 and 5000Hz, whilst SPT (2-5kHz) compares the mean energy at higher frequencies, between 2000 and 5000Hz.

According to Lee, Thorpe and Verhoeven (2009) when studying all of the vowels within a speech sample (rather than specific vowels) the different vowel contexts will result in natural variation in, for example, F0 and intensity; however, they note that these differences are applicable equally to all speakers, therefore average values should not be compromised. Likewise, in this voice study where all clearly identifiable voiced segments from the acoustic waveform have been used the effects of individual phoneme segments are expected to average-out.

For each speaker, the mean value of each voiced segment for all five parameters were calculated. These individual means were then combined and averaged to identify five overall means for each of the twenty-two children in the DS group and the fifty-two children in the TD group. Further subsets of data were created by dividing the groups by gender, and also to compare the eight DS and eight TD audio-recordings presented in the Questionnaire study (study 1).

3.4.5 Statistical Analysis

3.4.5.1 Overall DS and TD groups

Independent samples t-tests were used to determine if significant differences existed between the means for the overall DS group (22) and the TD group (52) across the five acoustic analysis parameters studied.

3.4.5.2 Sub-groups of DS and TD speakers divided by gender and speakers from study 1

Due to smaller numbers of DS and TD subjects non-parametric statistical methods (Mann Whitney U-tests) were used to evaluate differences between the groups when divided by gender and between the DS and TD speakers from study 1 across all the acoustic analysis parameters.

3.4.6 Hypotheses

For all analyses the following hypotheses were applied:

3.4.6.1 Fundamental Frequency (F0)

F0 will not differ significantly between the TD and DS groups.

Rationale: Although differences in F0 levels have been found in pre-school-age children with DS (Weinberg & Zlatin, 1970) in adults with DS (Moran & Gilbert, 1978; Lee Thorpe & Verhoeven, 2009) and in Portuguese-speaking children with DS (Moura et al., 2008) studies of English-speaking school-age children with DS have found no significant difference in F0 level in comparison with TD age-matched peers (Michel & Carney, 1964; Hollien & Copeland, 1965; Montague, Brown & Hollien, 1974; Pentz & Gilbert, 1983).

3.4.6.2 Jitter

Jitter values will be significantly higher in the DS group.

Rationale: Significantly greater values of frequency perturbation have been identified in English-speaking (Pentz & Gilbert, 1983) and Portuguese-speaking (Moura et al., 2008) children with DS in comparison with their TD peers.

3.4.6.3 Shimmer

Shimmer values will be significantly higher in DS group

Rationale: Although differences in amplitude perturbation were not found in young adults with DS (Lee, Thorpe & Verhoeven, 2009), significantly higher values have been found in English-speaking (Pentz & Gilbert, 1983) and Portuguese-speaking (Moura et al., 2008) children with DS in relation to their TD peers.

3.4.6.4 Harmonic-to-noise ratio (HNR)

HNR will be significantly lower in the DS group

Rationale: Both English-speaking (Pentz & Gilbert, 1983) and Portuguese-speaking (Moura et al., 2008) children with DS have been found to have higher levels of noise than their TD peers. Increased noise within the acoustic signal equates to a lower harmonic-to-noise ratio.

3.4.6.5 Spectral tilt (SPT)

SPT will be significantly lower in DS group

Rationale: Lower values of spectral tilt have been found in Portuguese-speaking children with DS in comparison to their TD peers (Moura et al., 2008). No values are available presently for English-speaking children with DS.

3.5 Study 3: Perceptual Analysis of Voice

3.5.1 Participants

This study used the same audio-recordings (appendix XIII) as the previous acoustic analysis (study 2) and investigated the same sub-groups of speakers as described in section 3.4.1.

3.5.2 Selection of a perceptual rating scale

According to Shewell (1998) the Vocal Profile Analysis Scheme (VPAS; Laver, Wirz, Mackenzie & Hiller, 1991) is the vocal perceptual rating scale most often used by British speech and language therapists. Aside from this clinical application, it is also frequently used within research of disordered voice quality (Mackenzie Beck, 2005a). It has been utilised in a number of previous studies which have examined the judgements that listeners make about speakers, allowing these attributions to be correlated with perceptual voice analysis findings. Such studies include van Erp (1991) who looked at the character traits of speakers with cleft lip and palate; Irving (1997) who examined physical and personality characteristics of male-to-female transsexual speakers, and also the data from a small scale unpublished study by Thomson (1995) looking at the personality traits of patients with oral cancer, before and after partial glossectomy, was later subject to the VPAS in a separate study by Mackenzie Beck, Wrench, Jackson, Soutar, Robertson and Laver (1998). An example of the VPAS form can be found in appendix XV.

The VPAS is regarded as a sensitive measure of vocal quality as its many parameters give information about the contribution of the whole of the vocal apparatus (Mackenzie Beck, 2005a) rather than a specific focus on vocal fold function (phonation). It has been suggested that the increased complexity of the scheme may be at the expense of reliability which has been found to be poor to moderate (Webb, Carding, Deary, MacKenzie, Steen & Wilson, 2004); however, Mackenzie Beck

(2005a) notes that degree of training is a key factor in confidence and ability to use the scheme and subsequently increases reliability. As this study of voice is concerned with how vocal acoustic and perceptual parameters correlate with judgements of personality and ability an in-depth, multi-dimensional scale such as the VPAS is essential.

According to Mackenzie Beck (2005a) the VPAS considers that voice quality is made up of three key features:

- Configurational settings, which consist of the long-term average positioning of the vocal apparatus, around which the individual segments of speech are produced.
- Range settings, which relate to the amount of movement made away from the neutral position.
- Tension settings, which consider the degree of muscular tension present within the vocal apparatus during speech.

The above are considered in relation to a range of vocal tract features, including labial, mandibular, lingual, pharyngeal, velopharyngeal and laryngeal vocal settings. Temporal and prosodic aspects of voice are also evaluated.

The individual components of voice analysed in the VPAS are considered, not in relation to what is regarded as 'normal' (as this subjective judgement may vary considerably between different language or accent groups) but rather in relation to what is termed a 'neutral' setting. Neutral settings are defined within the VPAS user manual (Mackenzie Beck, 2007) for each parameter analysed. For example, within English, the neutral velopharyngeal setting would reflect audible nasality on those segments that are phonologically classed as nasal (for example /m/ & /n/) with some anticipatory nasality on preceding vowels, and there should be no fricative nasal airflow. As a low level of background nasality affecting vowels in any context is fairly common in English speakers Mackenzie Beck (2007) notes that a neutral velopharyngeal setting is not a common feature of English speakers.

Ratings away from neutral are made on a 6-point scale, where 1-3 is indicative of the presence of a feature that falls within margins expected in non-speech/voice disordered populations (classed as the ‘moderate’ range) and 4-6 being a more severe presentation (classed as the ‘extreme’ range). For each setting the rater is required to consider how identified key segments are affected in individual speakers. For example, in English in analysis of denasality the key segments are again the nasal phonemes which will have an increasingly ‘oral’ quality as the scalar degree increases, with a rating of ‘6’ indicating full oral production (Mackenzie Beck, 2007).

3.5.3 Vocal Profile Analysis Scheme raters

Two qualified speech and language therapists (rater 1, the researcher; and rater 2, the first supervisor of the study) completed the VPAS for all the TD and DS audio-recordings. Training was provided for the researcher by the supervisor, who as a co-author of the VPAS is recognised as an expert in the use of the scheme and wider voice analysis.

Although it is preferable to view speakers when completing the VPAS (in order to identify any visual evidence to back-up auditory perceptions) as this was not possible for the researcher during the initial data collection due to time constraints within the education setting, and impossible for the second rater, all perceptual analysis was completed on the basis of audio-recordings alone. This has the advantage of ensuring that both the VPAS raters were rating under similar conditions. All the audio-recordings were coded numerically to ensure that the raters were blind as to which study group (DS or TD) the speakers belonged (although inevitably the researcher was to some degree familiar with the voices from editing the audio-recordings).

3.5.4 Data analysis

A system which reflected the actual value of the ratings made by the VPAS raters was devised to code the raw data. Where two parameters reflected two ends of a continuum (e.g. ‘lip spreading’ – ‘lip rounding/protrusion’, where a speaker cannot be rated as both) a negative-to-positive scale was adopted. Typically this involved a scale running from -6 to 6, where for example, -6 would represent the maximum degree of spread lip pattern, 6 the maximum degree of rounded/protruded lip pattern and 0 the neutral setting (i.e. not spread and not rounded/protruded). As for the most part the VPAS is organised on a scale of 0 to 6, where 0 represents neutral, a rating of below 3 indicates a moderate degree of the parameter investigated (i.e. within expected typical margins) and a rating of 3 or above, an extreme (or atypical) degree of the parameter, the coding system used was able to retain those boundaries in both the positive and negative scales. Thus, a coded rating of greater than 3 (e.g. in the direction of ‘rounded/protruded lips’) or -3 (‘spread lips’) indicates a severe presentation of that particular parameter, whilst a value close to 0 is considered close to the neutral setting.

Although the majority of VPAS parameters conform to this -6 to 6 scale, there are exceptions. Within the mandibular settings section it is recognised that it is not possible to divide the parameter of ‘close jaw’ into more than two categories, thus the scale for ‘open jaw – close jaw’ runs from -6 to 2. Similarly, where a parameter stands alone (i.e. there is no opposite/negative setting) the scale runs only in a positive direction, indicating the extent to which that parameter is present. For example, in the case of ‘labiodentalisation’ the scale runs from 0 to 6 (0 again reflecting a neutral setting, whilst a value above 3 would indicate a severe presentation of labiodentalisation). The parameter of ‘audible nasal escape’ is treated slightly differently as it is acknowledged in the VPAS that it is not possible to have a moderate degree of this parameter (any nasal escape is considered an atypical feature, and therefore present to a severe degree) thus the scale for ‘audible nasal escape’ consists of 0 (neutral or no nasal escape) and 4 to 6 (indicating the presence of nasal escape).

Categorical data, which represents a feature being ‘present’ or ‘not present’ (as in ratings of ‘falsetto’, ‘creak’, ‘whisper’ and ‘diplophonia’), ‘neutral voice’ or ‘non-neutral voice’ ratings and ‘adequate’ or ‘inadequate’ respiratory support ratings are coded on a scale of 0 to 1.

Parameters assessed and their coding systems are shown within tables 3.6-3.11.

VPAS coding system: VOCAL TRACT FEATURES	
Labial Settings	**CODE**
spreading - rounding/protrusion	-6 to 6
labiodentalisation	0 to 6
minimised range - extensive range	-6 to 6
Mandibular Settings	**CODE**
open - close jaw	-6 to 2
protruded jaw	0 to 6
minimised - extensive range	-6 to 6
Lingual Settings	**CODE**
retracted - advanced tip/blade	-6 to 6
backed - fronted tongue body	-6 to 6
lowered - raised tongue body	-6 to 6
minimised - extensive range	-6 to 6
Pharyngeal Settings	**CODE**
constriction - expansion	-6 to 6
Velopharyngeal Settings	**CODE**
audible nasal escape	(0, 4 to 6*)
denasal - nasal	-6 to 6
Larynx Height Settings	**CODE**
lowered - raised larynx	-6 to 6

Table 3.6: Codes used to label judgements made within the ‘vocal tract features’ section of the VPAS (* Audible nasal escape is considered an extreme setting only, therefore 0 represents neutral or no audible nasal escape, and 4-6 the presence of nasal escape)

VPAS coding system: OVERALL MUSCULAR TENSION	
Vocal Tract Tension Settings	**CODE**
la - tense vocal tract	-6 to 6
Laryngeal Tension Settings	**CODE**
lax - tense larynx	-6 to 6

Table 3.7: Codes used to label judgements made within the 'overall muscular tension' section of the VPAS

VPAS coding system: PHONATION FEATURES	
Voicing Type	**CODE**
voice	0 = neutral, 1 = non-neutral
falsetto	0 = absent, 1 = present
creak	0 = absent, 1 = present
creaky	0 to 6
Laryngeal Friction	**CODE**
whisper	0 = absent, 1 = present
whispery	0 to 6
Laryngeal Irregularity	**CODE**
harsh	0 to 6
tremor	0 to 6

Table 3.8: Codes used to label judgements made within the 'phonation features' section of the VPAS

VPAS coding system: PROSODIC FEATURES	
Pitch	**CODE**
low - high mean	-6 to 6
minimised - extensive range	-6 to 6
low - high variability	-6 to 6
Loudness	**CODE**
low - high mean	-6 to 6
minimised - extensive range	-6 to 6
low - high variability	-6 to 6

Table 3.9: Codes used to label judgements made within the 'prosodic features' section of the VPAS

VPAS coding system: TEMPORAL ORGANIZATION	
Continuity	**CODE**
interrupted	0-6
Rate	**CODE**
slow - fast	-6 to 6

Table 3.10: Codes used to label judgements made within the 'temporal organisation' section of the VPAS

VPAS coding system: OTHER FEATURES	
	CODE
Respiratory Support	0 = adequate, 1 = inadequate
Diplophonia	0 = absent, 1 = present

Table 3.11: Codes used to label judgements made within the 'other features' section of the VPAS

3.5.5 Agreement between raters

For all of the scaled parameters of the VPAS agreement was defined as a difference of no more than 1 scalar degree between ratings. Thus for example, rater 1 and 2 could rate '0' and '-1', or '3' and '4' respectively and still be classed as in agreement. This method echoes that of previous research using the VPAS (Mackenzie Beck, 2005a). For the categorical or dichotomous data ('voice', 'falsetto', 'creak', 'whisper', respiratory support' and 'diplophonia' ratings) exact agreement was deemed necessary. The percentage of agreement between raters for each parameter of the VPAS is reported in the results chapter for the overall DS and TD groups.

3.5.6 Statistical analysis

As the VPAS data is non-parametric, for the overall DS and TD groups, the groups divided by gender and the speakers from study 1, statistical analysis consisted of

Mann Whitney U-tests, and thus median and interquartile ranges were reported rather than means and standard deviation values. The only exception to this is for the categorical data (‘voice’, ‘falsetto’, ‘creak’, ‘whisper’, respiratory support’ and ‘diplophonia’ ratings) which were analysed using Chi Square tests rather than Mann Whitney U-tests.

3.5.7 Hypotheses

The hypotheses for the VPAS for the children with DS are concerned with how established organic differences will impact on voice production. This issue has been considered in depth by Mackenzie Beck (1997) and as such her predictions will form the basis of this study’s VPAS hypotheses (table 3.12). Thus for the statistical analyses the following hypotheses (predicted phonetic consequences) will apply:

VPAS: Hypotheses for results for children with DS in relation to typical settings	
ORGANIC FEATURE	**PREDICTED PHONETIC CONSEQUENCE**
Thick, everted lips	Increased protruded labial setting
Maxillary under-development	Increased protruded jaw setting Tongue more advanced relative to palate and upper teeth
Short, narrow palate and normal or large tongue	More advanced tip/blade articulations Fronted and raised tongue-body setting
Pharynx reduced in anterior-posterior dimension	Increased pharyngeal constriction
Mucosal disorders affecting the vocal folds	More irregular vocal fold vibration and poor adduction, resulting in harshness and/or whisperiness
Generalised muscular hypotonia	More lax tension settings Increased nasality & nasal emission More open jaw More lowered larynx More minimised range of articulation

Table 3.12: Predicted results of the VPAS for children with DS
Adapted from Mackenzie Beck (1997) ‘Characteristic organic features of the vocal apparatus in Down’s syndrome and predicted phonetic consequences’

3.6 CORRELATIONS BETWEEN STUDIES 1, 2 AND 3

The data of the eight DS and eight TD speakers from study 1 was combined (n = 16) in order to determine if there were correlations between the judgements made by listeners in study 1 and the acoustic data (study 2) and perceptual VPAS data (study 3) for these speakers.

To do this the judgements made by the SNES, MES and PEER raters from study 1 were also combined and a median score attributed to each speaker for each of the questionnaire parameters. For the judgements of 'gender' and 'spend time with' any 'unsure'/'not sure' ratings were excluded (leaving only 'male' and 'female' and 'yes' and 'no' judgements respectively), thus higher median values indicated more judgements of the speaker sounding female and a greater desire to spend time in the company of that speaker. Similarly, for study 3 a median of the two expert SLT ratings for each speaker in each section of the VPAS was determined. This data was then subject to analysis using Spearman's correlations alongside the median values from the eight acoustic parameters for each speaker from study 2. As there were so many parameters within the three studies only correlations which were considered to be strong were reported. According to Dancey and Reidy (2007) strong correlations are +/-0.7 or above.

4
RESULTS

4.1 MATCHING PARTICIPANTS FOR AGE (STUDIES 1, 2 & 3)

An independent samples t-test confirmed that the ages of the twenty-two DS and fifty-two TD children whose audio-recordings were analysed acoustically (study 2) and perceptually (study 3) did not differ significantly.

The ages of subsets of these children were analysed non-parametrically, due to smaller numbers. Mann Whitney U-tests revealing no significant difference when split on the basis of gender (13 DS males versus 34 TD males, and 9 DS females versus 18 TD females) or between the ages of the eight DS and eight TD children's recordings selected to be played to listeners in the Questionnaire study (study 1).

4.2 STUDY 1: QUESTIONNAIRE-BASED ANALYSIS OF LISTENER JUDGEMENTS OF VOICE

Findings for each of the parameters addressed in the listener questionnaire are reported. This includes:

- Consistency of group judgements for repeated samples
- Analysis of differences between DS and TD ratings within listener groups
- Analysis of differences between listener groups' ratings of DS and TD groups
- Analysis of the effect of Gender and Age on PEER ratings
- Variability in judgements of individual DS and TD speakers

For the parameters of ‘calm-angry’, ‘confident-shy’, friendly-unfriendly’, ‘happy-sad’, ‘intelligent-unintelligent’ and ‘age’ statistically significant differences from Mann-Whitney U-tests are reported and median and interquartile range (IQR) values given. The level of statistical significance from Chi Square tests are provided for the remaining ‘spend time with’ and ‘gender’ parameters. Where results are non-significant the abbreviation ‘ns’ is used. Where it is judged to be informative values are illustrated in boxplot form.

Ratings by the SNES, MES and PEER listeners for each parameter, with the exception of ‘age’, are also expressed in percentage form, indicating the spread of ratings across the different scales and overall rating scores for whole groups and individual speakers.

For the five semantic differential parameters the overall percentage calculations indicate how positively or negatively the DS and TD groups and individual speakers were judged, with higher percentage values representing more positive judgements. In this instance a subjective judgement as to which end of each scale merits the label of a ‘positive’ characteristic, and which a ‘negative’ characteristic was made. ‘Calm’, ‘confident’, ‘friendly’, ‘happy’ and ‘intelligent’ were considered to be positive attributes, whilst their polar opposites ‘angry’, ‘shy’, ‘unfriendly’, ‘unhappy’ and ‘unintelligent’ were deemed to be negative qualities. Diagrams are used to illustrate the overall percentage value attributed to each individual speaker by each listener group for each parameter.

For the parameter ‘spend time with’ the overall percentage values represent the degree to which the PEER raters consider the DS and TD groups and individual speakers as socially desirable, again a higher percentage indicates a group or speaker that the listeners would choose to socialise with.

In all diagrams showing results for individual speakers, the order of subjects on the x-axis reflects the order of presentation to listeners within the questionnaire task (DS subjects being on the left and TD subjects on the right).

4.2.1 Calm – angry ratings

4.2.1.1 Consistency of group judgements for repeated samples

Table 4.1 shows the number of individuals in each listener group who met the criteria for consistent judgements for this parameter (SNES, 51 from 52, MES, 45 from 45, & PEER, 73 from 75), those with poor consistency having been excluded from further analysis. For each listener group the individual consistency scores for each of these final listeners have been combined to give a group consistency score (percentage) for each of the repeated recordings (DS13, DS7, TD2 & TD7) and an overall score calculated from the combined consistency scores for all four repeated recordings. High percentage scores indicate greater consistency of judgements whilst lower percentages give an indication of voices which were less consistently judged between the first and second set of ratings.

CALM-ANGRY: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (51)	98.00	94.00	100.00	100.00	98.01
MES (45)	95.35	95.56	95.45	97.78	96.05
PEER (73)	89.04	93.15	98.61	94.52	93.81

Table 4.1: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter 'calm-angry'

4.2.1.2 Analysis of differences between DS and TD ratings within listener groups

The spread of ratings (percentage) made by the three listener groups for the parameter of 'calm-angry' are shown in table 4.2.

CALM-ANGRY: Group ratings breakdown %						
	SNES (51)		MES (45)		PEER (73)	
	DS	TD	DS	TD	DS	TD
very calm	6.44	21.92	5.71	30.25	9.09	28.87
quite calm	54.95	57.88	52.57	55.74	36.02	54.12
neutral	31.68	16.50	33.14	11.76	39.97	13.23
quite angry	6.68	3.69	8.57	2.24	13.72	3.44
very angry	0.25	0.00	0.00	0.00	1.20	0.34

Table 4.2: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter ‘calm-angry’

The median and interquartile range (IQR) and the overall ‘calm-angry’ percentage for the DS and TD groups by the SNES, MES and PEER raters are shown in table 4.3 (higher percentages indicating judgements of ‘calm’, lower values representing judgements of ‘angry’, with ‘neutral’ judgements falling at 50%). Also shown are the results of a Mann-Whitney U-test finding that all three of the listener groups rated the DS group significantly more negatively than the TD group. The median and range of ratings of the DS and TD groups by the three listener groups are illustrated as a boxplot in figure 4.1 (neutral, not calm but not angry, being indicated by a dashed line).

CALM-ANGRY: Median rating (IQR) & overall group ratings %			
	DS	TD	statistical significance
SNES (51)	3 (1) 65.16%	3 (0) 74.51%	n = 404 (DS), 406 (TD), U = 60098.5, $p < 0.001$
MES (45)	3 (1) 63.86%	3 (1) 78.50%	n = 350 (DS), 357 (TD), U = *37052.0,* $p < 0.001$
PEER (73)	2 (1) 59.52%	3 (1) 76.93%	n = 583 (DS), 582 (TD), U = 95322.5, $p < 0.001$

Table 4.3: ‘Calm-angry’ median (IQR) and overall percentage ratings attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

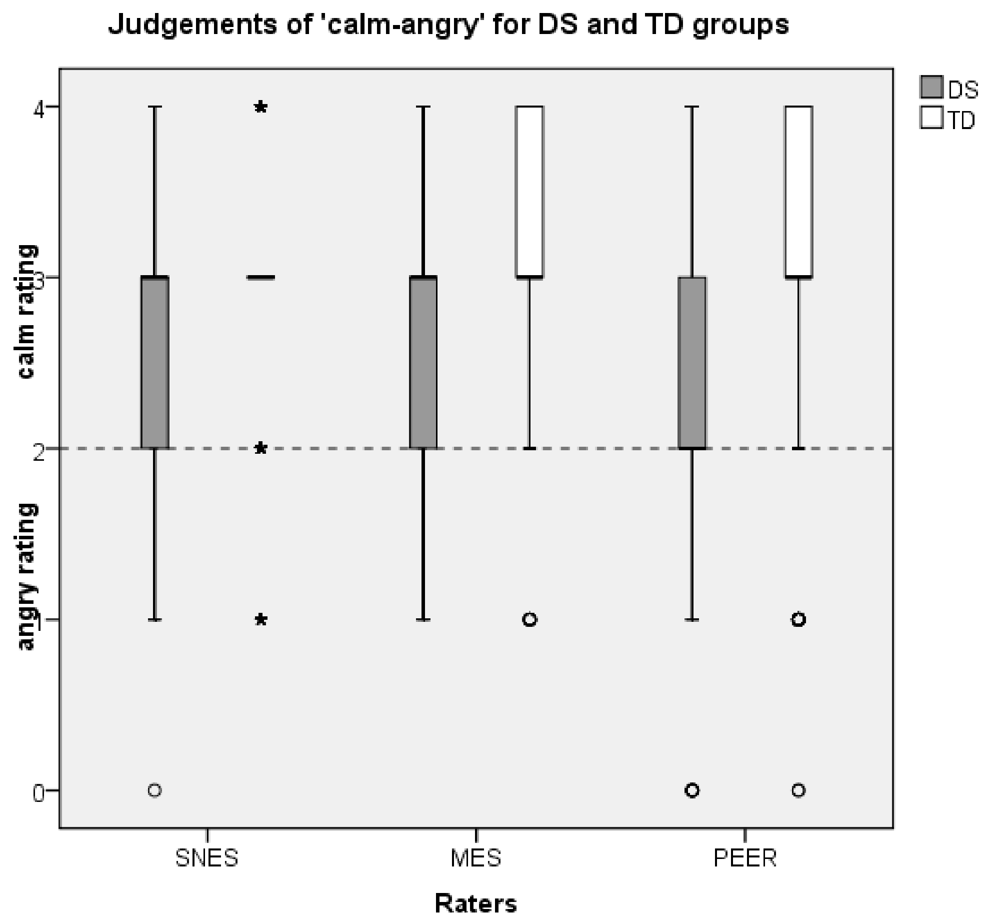


Figure 4.1: Boxplot illustrating the median and range of ‘calm-angry’ judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.1.3 <u>Analysis of differences between listener groups’ ratings of DS and TD groups</u>

A Kruskal Wallis test and the resulting post hoc Mann Whitney U-tests indicated significant differences in the ‘calm-angry’ ratings of the DS and TD children between the three listener groups (table 4.4). The MES listeners were found to judge significantly more positively than SNES raters for the TD group, whilst both groups of education staff rated significantly more positively than the PEER raters for the children with DS. Bonferroni adjustment was employed in this analysis, creating an alternative *p* value of 0.017.

CALM-ANGRY: Differences between raters		
	Statistically significant differences	
Listener Group	DS	TD
SNES versus MES	ns	n = 406 (SNES), 357 (MES), U = 64102.5, p = 0.002
SNES versus PEER	n = 404 (SNES), 583 (PEER), U = 99392.5, p < 0.001	ns
MES versus PEER	n = 350 (MES), 583 (PEER), U = 89704.0, p = 0.001	ns

Table 4.4: Statistically significant differences in ratings of 'calm-angry' for DS and TD groups between SNES, MES and PEER raters

4.2.1.4 Analysis of the effect of gender and age on PEER ratings

No significant differences were found between the PEER ratings for the DS and TD groups when divided on the basis of age and gender.

4.2.1.5 Variability in judgements of individual DS and TD speakers

Table 4.5 shows the overall percentage rating of 'calm-angry' for individual speakers in the DS and TD groups for the SNES, MES and PEER listeners. These values are illustrated in figure 4.2 (neutral being represented by a dashed line at 50%).

CALM-ANGRY: Overall ratings for individual speakers %								
	DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
SNES (51)	69.50	74.00	59.31	58.33	62.50	65.69	67.65	65.00
MES (45)	70.35	71.51	57.32	62.22	60.56	63.64	58.89	66.48
PEER (73)	66.44	76.71	50.68	47.26	61.64	59.38	53.08	63.36
	TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
SNES (51)	77.50	73.53	78.92	79.90	78.43	79.00	64.22	64.71
MES (45)	78.98	81.25	82.22	82.78	84.44	81.11	67.78	69.32
PEER (73)	76.74	75.68	82.53	86.64	81.51	81.94	64.73	66.44

Table 4.5: Overall 'calm-angry' rating (percentage) attributed to individual DS and TD speakers by SNES, MES and PEER raters

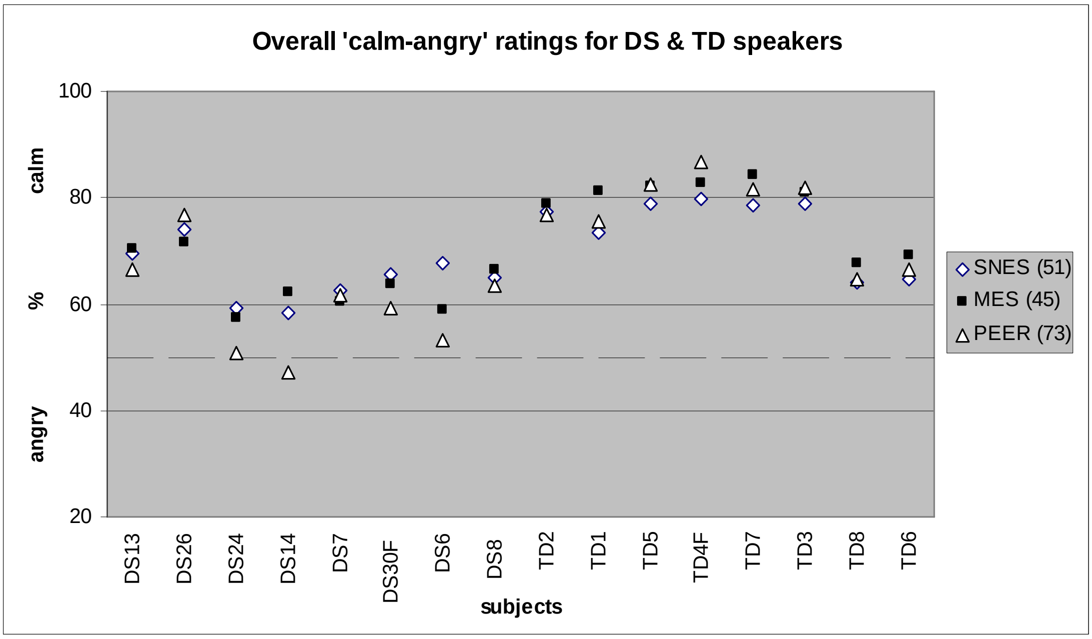


Figure 4.2: Diagram showing the relationship between overall 'calm-angry' ratings (percentage) for individual DS and TD speakers by SNES, MES and PEER raters

4.2.2 Confident – shy ratings

4.2.2.1 Consistency of group judgements for repeated samples

The ratings of forty-three SNES, thirty-nine MES and sixty-five PEER listeners were judged as consistent and therefore included in the analysis of the 'confident-shy' parameter. The degree of consistency for the four individual repeated recordings and the combined results for each listener group are shown in table 4.6.

CONFIDENT-SHY: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (43)	80.95	97.67	93.02	95.35	91.81
MES (39)	79.49	97.44	94.87	92.11	90.97
PEER (65)	64.62	96.92	90.77	87.69	85.00

Table 4.6: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter 'confident-shy'

4.2.2.2 Analysis of differences between DS and TD ratings within listener groups

The percentage breakdown of 'confident-shy' ratings for the DS and TD groups by the SNES, MES and PEER listener groups are shown in table 4.7.

CONFIDENT-SHY: Group ratings breakdown %						
	SNES (43)		**MES (39)**		**PEER (65)**	
	DS	**TD**	**DS**	**TD**	**DS**	**TD**
very confident	4.36	26.24	5.16	35.28	9.44	32.50
quite confident	29.07	47.52	32.26	44.66	17.53	42.69
neutral	30.23	17.20	20.97	11.97	19.85	15.00
quite shy	29.36	9.04	35.81	8.09	34.68	9.42
very shy	6.98	0.00	5.81	0.00	18.50	0.38

Table 4.7: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter 'confident-shy'

The median ratings, IQR and the overall percentage ratings by the SNES, MES and PEER raters are shown in table 4.8, alongside statistically significant differences between the ratings for the DS and TD groups identified by Mann Whitney U-tests. In all three cases the raters judged the TD group more positively than the DS group; this relationship is illustrated as a boxplot in figure 4.3.

CONFIDENT-SHY: median (IQR) & overall ratings %			
	DS	**TD**	**statistical significance**
SNES (43)	2 (2) 48.62%	3 (2) 72.74%	n = 344 (DS), 343 (TD), $U = 29477.5$, $p < 0.001$
MES (39)	2 (2) 48.79%	3 (1) 76.78%	n = 310 (DS), 309 (TD), $U = 21387.0$, $p < 0.001$
PEER (65)	1 (2) 41.18%	3 (1) 74.38%	n = 519 (DS), 520 (TD), $U = 57313.5$, $p < 0.001$

Table 4.8: 'Confident-shy' median (IQR) and overall percentage ratings attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

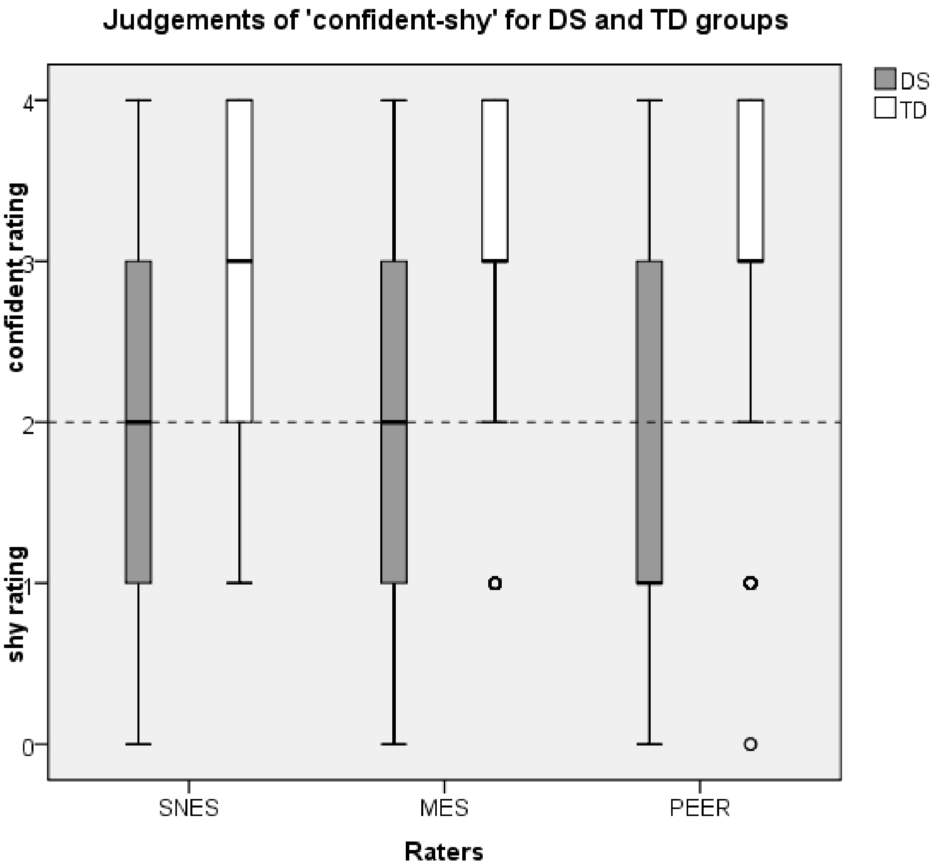


Figure 4.3: Boxplot illustrating the median and range of 'confident-shy' judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.2.3 Analysis of differences between listener groups' ratings of DS and TD groups

A Kruskal Wallis test and post hoc Mann-Whitney U-tests revealed statistically significant differences between the three listener groups for their judgements of confidence for the DS and TD children (table 4.9). Although no differences existed between the SNES and MES ratings of the DS and TD groups, significant differences were found between both groups of education staff and the PEER raters for judgements of the children with DS; the PEER raters rating the DS group significantly more negatively in both cases. Significant results are based on a modified *p* value of 0.017 as Bonferroni adjustment has been used in this analysis.

CONFIDENT-SHY: Differences between raters		
	Statistically significant differences	
Listener Group	DS	TD
SNES versus MES	ns	ns
SNES versus PEER	n = 344 (SNES), 519 (PEER), *U* = 74670.5, p < 0.001	ns
MES versus PEER	n = 310 (MES), 519 (PEER), *U* = 67730.5, p < 0.001	ns

Table 4.9: Statistically significant differences in ratings of ‘confident-shy’ for DS and TD groups between SNES, MES and PEER raters

4.2.2.4 Analysis of the effect of gender and age on PEER ratings

Mann Whitney U-tests revealed a statistically significant difference between the male and female ratings of confidence for the DS group with the females rating more positively (median rating 2, IQR, 1) than males (median rating 1, IQR, 1); (table 4.10 & figure 4.4).

No significant difference was found between the ratings of the younger and older PEER group for either the DS or TD speakers.

CONFIDENT-SHY: Effects of gender and age on PEER judgements				
	male (M) versus female (F)		younger (Y) versus older (O)	
Group	DS	TD	DS	TD
statistical significance	n = 288 (M), 231 (F), *U* = 27384.5, p < 0.001	ns	ns	ns

Table 4.10: Statistically significant differences in ratings of ‘confident-shy’ for DS and TD groups between PEER listeners grouped according to gender and age

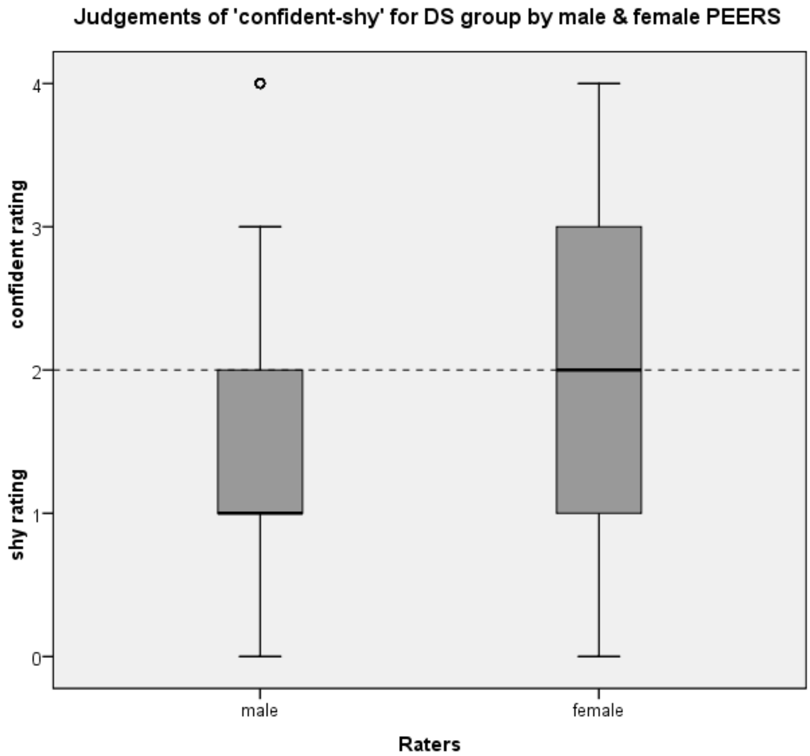


Figure 4.4: Boxplot showing the median and range of 'confident-shy' ratings for the DS group by the PEER raters when split by gender

4.2.2.5 Variability in judgements of individual DS and TD speakers

The ratings (converted to percentage values) by SNES, MES and PEER listeners for the 'confident-shy' parameter, for individual speakers in both the DS and TD groups are shown in table 4.11 and illustrated in figure 4.5

CONFIDENT-SHY: Overall ratings for individual speakers %								
	DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
SNES (43)	29.65	69.19	41.28	50.00	45.93	53.49	40.70	72.67
MES (39)	27.56	73.03	41.03	54.49	41.03	51.28	40.13	73.72
PEER (65)	27.69	75.00	32.31	40.38	38.85	43.46	35.16	73.46
	TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
SNES (43)	61.31	78.49	81.98	89.53	84.88	75.58	56.40	53.49
MES (39)	64.74	80.26	86.18	90.13	91.03	82.69	60.26	59.62
PEER (65)	52.31	80.77	91.15	90.38	83.46	78.08	56.54	63.08

Table 4.11: Overall 'confident-shy' rating (percentage) attributed to individual DS and TD speakers by SNES, MES and PEER raters

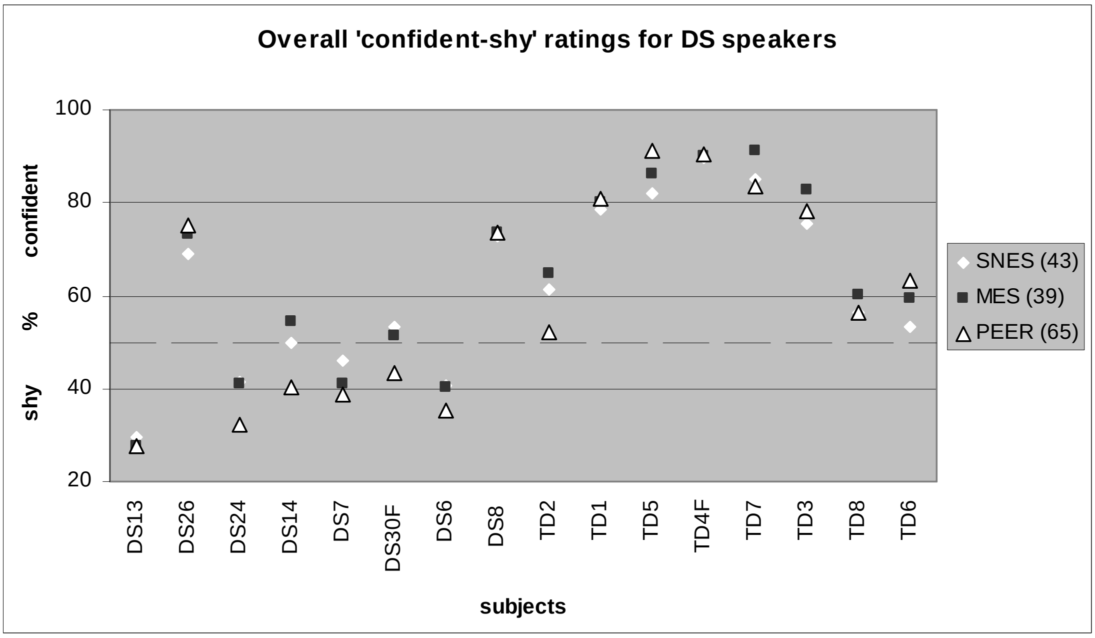


Figure 4.5: Diagram showing the relationship between overall 'confident-shy' ratings (percentage) for individual DS and TD speakers by SNES, MES and PEER raters

4.2.3 Friendly – unfriendly ratings

4.2.3.1 <u>Consistency of group judgements for repeated samples</u>

The consistency scores for each repeated recording and the overall group consistency ratings for the fifty-two SNES, forty-two MES and seventy PEER raters meeting the consistency of judgements criteria for 'friendly-unfriendly' are shown in table 4.12.

FRIENDLY-UNFRIENDLY: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (52)	97.96	96.15	100.00	98.08	98.04
MES (42)	92.86	100.00	100.00	97.56	97.59
PEER (70)	91.30	91.43	91.43	97.14	92.83

Table 4.12: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter 'friendly-unfriendly'

4.2.3.2 Analysis of differences between DS and TD ratings within listener groups

A breakdown of the ratings of friendliness, expressed in percentage form, by the SNES, MES and PEER listeners for the DS and TD groups are shown in table 13.

Median and IQR values and overall friendliness ratings (percentage) are shown in table 4.14, alongside significant statistical differences identified from Mann Whitney U-tests; all three listener groups rating the DS children significantly more negatively than their TD peers. Median scores and range of ratings are illustrated as a boxplot (figure 4.6).

FRIENDLY-UNFRIENDLY: Group ratings breakdown %						
	SNES (52)		MES (42)		PEER (70)	
	DS	TD	DS	TD	DS	TD
very friendly	6.80	15.01	7.88	17.61	10.55	20.43
quite friendly	39.81	40.68	40.91	45.67	32.56	48.21
neutral	46.12	37.77	43.33	31.04	40.97	24.19
quite unfriendly	7.04	6.30	7.27	5.67	12.70	6.63
very unfriendly	0.24	0.24	0.61	0.00	3.22	0.54

Table 4.13: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter 'friendly-unfriendly'

FRIENDLY-UNFRIENDLY: Median, IQR & overall ratings %			
	DS	TD	statistical significance
SNES (52)	2 (1) 61.47%	3 (1) 65.98%	n = 412 (DS), 413 (TD), $U = 74840.5$, $p = 0.001$
MES (42)	2 (1) 62.05%	3 (1) 68.81%	n = 330 (DS), 335 (TD), $U = 45256.5$, $p < 0.001$
PEER (70)	2 (1) 58.63%	3 (1) 70.34%	n = 559 (DS), 558 (TD), $U = 112059.0$, $p < 0.001$

Table 4.14: 'Friendly-unfriendly' median (IQR) and overall percentage ratings attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

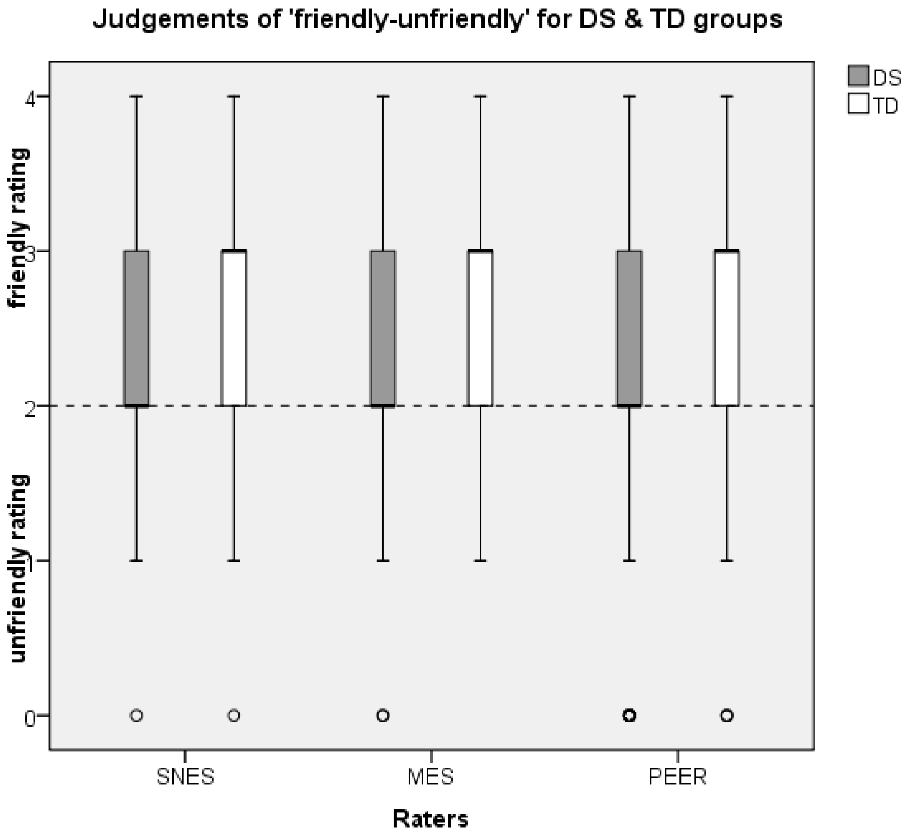


Figure 4.6: Boxplot illustrating the median and range of 'friendly-unfriendly' judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.3.3 <u>Analysis of differences between listener groups' ratings of DS and TD groups</u>

Kruskal Wallis and follow-up Mann Whitney U-tests demonstrated significant statistical differences between the ratings of the SNES group and the PEER raters for the TD children (PEER listeners rating more positively). No difference was found between the ratings of the DS group (table 4.15). Statistically significant results are based on an alternative *p* value of 0.017 due to Bonferroni adjustment.

FRIENDLY-UNFRIENDLY: Differences between raters		
	Statistically significant differences	
Listener Group	**DS**	**TD**
SNES versus MES	ns	ns
SNES versus PEER	ns	n = 413 (SNES), 518 (PEER), $U = 100388.5$, $p < 0.001$
MES versus PEER	ns	ns

Table 4.15: Statistically significant differences in ratings of 'friendly-unfriendly' for DS and TD groups between SNES, MES and PEER raters

4.2.3.4 Analysis of the effect of gender and age on PEER ratings

Ratings of 'friendliness' did not differ between the male and female, or the younger and older PEER raters for either the DS or TD groups.

4.2.3.5 Variability in judgements of individual DS and TD speakers

Overall percentage values for judgements of 'friendly-unfriendly' for individual DS and TD speakers are shown in table 4.16 and illustrated in figure 4.7.

FRIENDLY-UNFRIENDLY: Overall ratings for individual speakers %								
	DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
SNES (52)	54.90	77.88	55.39	63.73	54.33	57.84	53.85	74.04
MES (42)	54.27	79.76	57.50	61.59	55.49	61.31	52.44	74.40
PEER (70)	51.43	87.50	48.19	54.64	57.50	61.07	47.14	67.86
	TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
SNES (52)	63.78	62.50	89.90	75.48	61.54	68.75	52.40	53.85
MES (42)	65.48	66.67	92.07	79.76	66.67	72.62	55.95	51.79
PEER (70)	65.22	63.57	92.86	82.50	71.79	78.26	56.43	53.21

Table 4.16: Overall 'friendly-unfriendly' rating (percentage) attributed to individual DS and TD speakers by SNES, MES and PEER raters

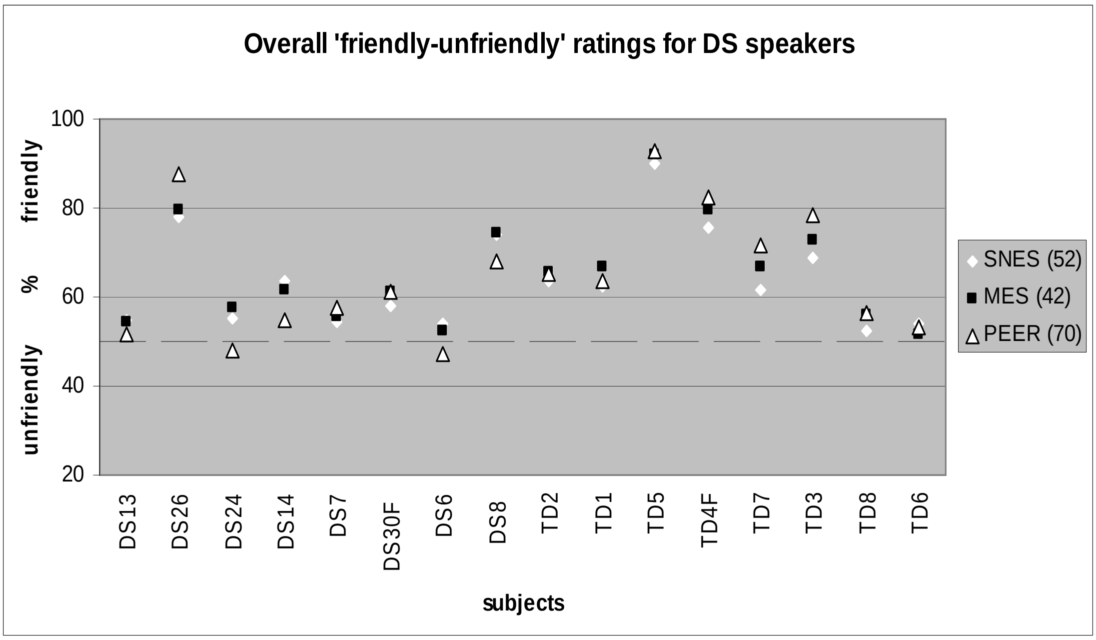


Figure 4.7: Diagram showing the relationship between overall 'friendly-unfriendly' ratings (percentage) for individual DS and TD speakers by SNES, MES and PEER raters

4.2.4 Happy – sad ratings

4.2.4.1 Consistency of group judgements for repeated samples

Table 4.17 shows the consistency of ratings (percentage) for the four repeated recordings, and the overall consistency rating, for the fifty-two SNES, forty-four MES and seventy-three PEER raters meeting the consistency of judgements criteria.

HAPPY-SAD: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (51)	94.12	94.12	96.00	90.20	93.60
MES (44)	90.48	100.00	97.62	90.70	94.74
PEER (73)	84.93	95.89	95.83	95.89	93.13

Table 4.17: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter 'happy-sad'

4.2.4.2 Analysis of differences between DS and TD ratings within listener groups

Table 4.18 shows the spread of judgements, expressed in percentage form, for the DS and TD speakers by all three listener groups.

HAPPY-SAD: Group ratings breakdown %						
	SNES (51)		MES (44)		PEER (73)	
	DS	TD	DS	TD	DS	TD
very happy	3.97	10.07	6.98	12.17	10.34	15.58
quite happy	35.48	31.20	24.71	31.59	20.69	35.79
neutral	33.75	43.00	38.95	40.00	26.72	32.53
quite sad	23.57	15.23	26.74	16.52	31.38	15.24
very sad	3.23	0.49	2.62	0.00	10.86	0.86

Table 4.18: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter 'happy-sad'

Mann Whitney U-tests indicated significant statistical differences between the 'happy-sad' ratings for the DS and TD speakers by all three listener groups; the DS group being judged more negatively than the TD speakers in each case. Table 4.19 shows the degree of statistical significance, median (IQR) judgements, and the overall percentage ratings by each listener group. Median values and range of judgements are illustrated as a boxplot in figure 4.8.

HAPPY-SAD: Median (IQR) & overall ratings %			
	DS	TD	statistical significance
SNES (51)	2 (2) 53.35%	2 (1) 58.78%	n = 403 (DS), 407 (TD), U = 73193.5, $p = 0.005$
MES (44)	2 (2) 51.67%	2 (1) 59.83%	n = 344 (DS), 346 (TD), U = 48513.5, $p < 0.001$
PEER (73)	2 (2) 47.07%	3 (1) 62.50%	n = 580 (DS), 584 (TD), U = 117391.5, $p < 0.001$

Table 4.19: 'Happy-sad' median (IQR) and overall percentage ratings attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

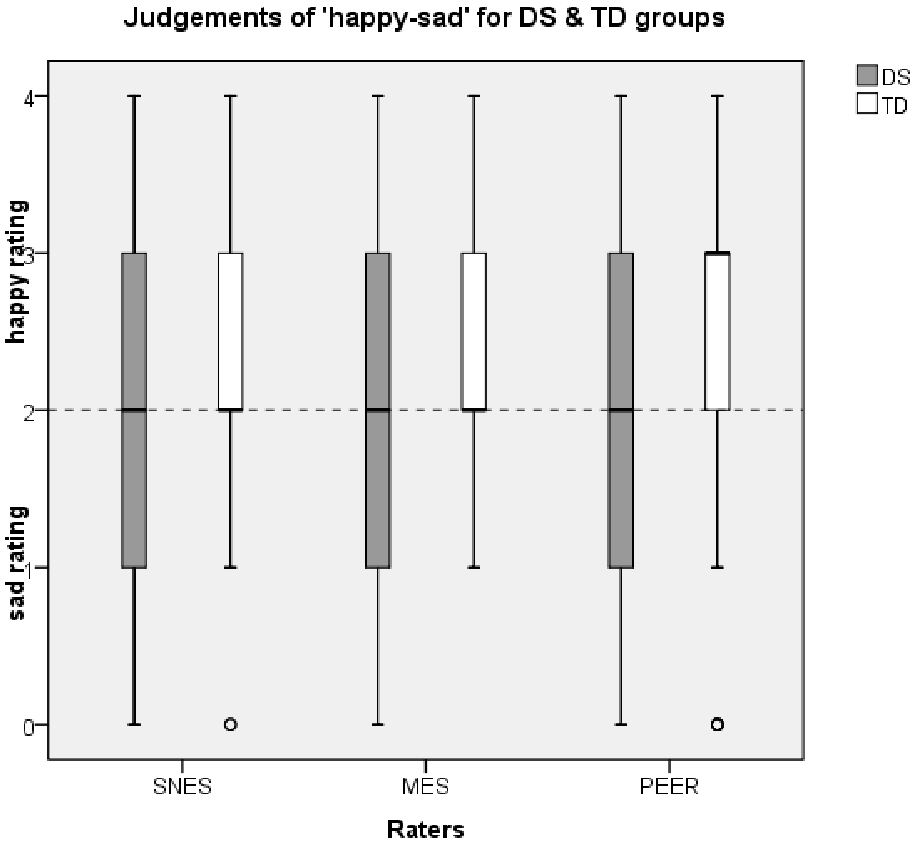


Figure 4.8: Boxplot illustrating the median and range of 'happy-sad' judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.4.3 Analysis of differences between listener groups' ratings of DS and TD groups

The results of a Kruskal-Wallis test and follow-up Mann Whitney U-tests (table 4.20) indicate no significant difference between the ratings of the SNES and MES groups for either the DS or TD children. Both education staff groups differed significantly from the PEER ratings of happiness for the DS group (PEERS rating more negatively) and the SNES group also differed from PEER ratings of the TD group (PEERS rating more positively). Bonferroni adjustment in this analysis created an alternative p value of 0.017.

HAPPY-SAD: Differences between raters		
	Statistically significant differences	
Listener Group	DS	TD
SNES versus MES	ns	ns
SNES versus PEER	n = 403 (SNES), 580 (PEER), *U* 100260.5, p < 0.001	n = 407 (SNES), 584 (PEER), *U* = 107567.0, p = 0.007
MES versus PEER	n = 344 (MES), 580 (PEER), *U* = 89553.5, p = 0.007	ns

Table 4.20: Statistically significant differences in ratings of 'happy-sad' for DS and TD groups between SNES, MES and PEER raters

4.2.4.4 Analysis of the effect of gender and age on PEER ratings

No significant differences were found between the 'happy-sad' ratings of the male and female PEER raters or the younger and older raters for either the DS or TD groups.

4.2.4.5 Variability in judgements of individual DS and TD speakers

A calculation of the overall percentage rating for the 'happy-sad' parameter for individual DS and TD speakers is shown in table 4.21. These percentage values are illustrated in figure 4.9.

HAPPY-SAD: Overall ratings for individual speakers %								
	DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
SNES (51)	37.50	76.50	43.50	48.50	51.47	59.31	42.50	73.53
MES (44)	33.72	77.38	40.48	46.59	46.51	54.07	41.48	79.07
PEER (73)	30.14	86.64	35.56	36.27	48.29	51.03	36.64	72.95
	TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
SNES (51)	50.98	56.37	90.69	72.55	51.96	62.25	43.50	42.65
MES (44)	50.00	53.49	92.44	76.74	54.55	64.77	43.75	43.02
PEER (73)	48.97	56.51	96.23	77.74	61.99	67.47	45.21	47.60

Table 4.21: Overall 'happy-sad' rating (percentage) attributed to individual DS and TD speakers by SNES, MES and PEER raters

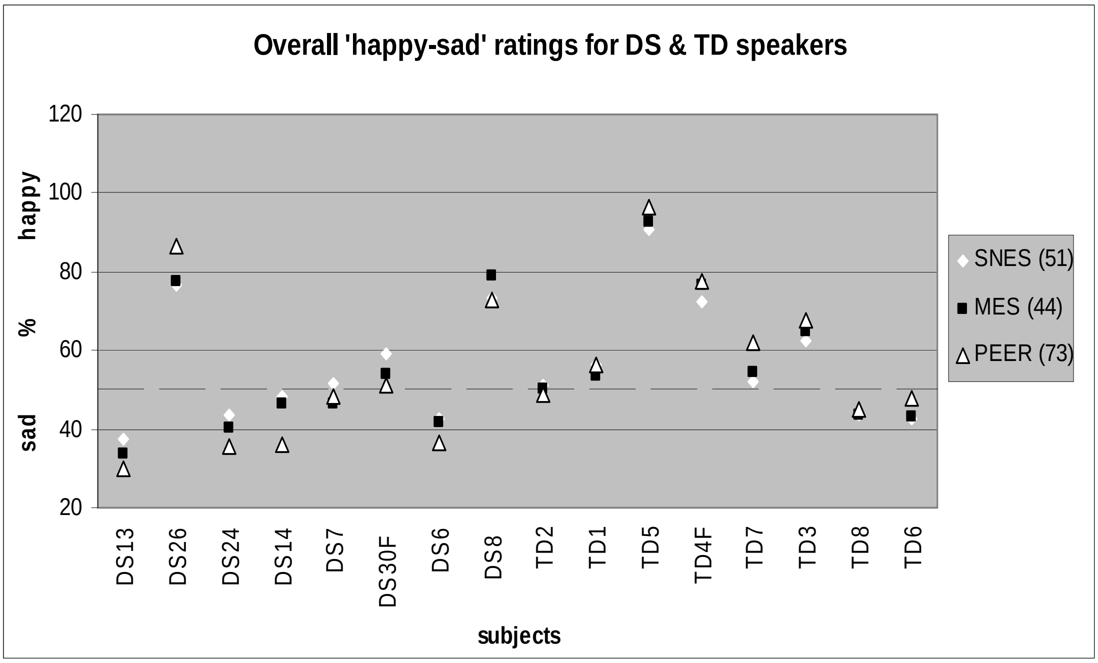


Figure 4.9: Diagram showing the relationship between overall ‘happy-sad’ ratings (percentage) for individual DS & TD speakers by SNES, MES and PEER raters

4.2.5 Intelligent – unintelligent ratings

4.2.5.1 Consistency of group judgements for repeated samples

The number of raters meeting the consistency criteria (SNES, 49, MES, 41, & PEER, 66) together with the percentage scores for consistency of judgements for each group are shown in table 4.22.

INTELLIGENT-UNINTELLIGENT: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (49)	97.92	97.96	91.84	95.83	95.88
MES (41)	75.61	97.56	94.87	97.56	91.36
PEER (66)	84.85	98.48	95.38	95.45	93.54

Table 4.22: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter ‘intelligent-unintelligent’

4.2.5.2 Analysis of differences between DS and TD ratings within listener groups

A breakdown of the 'intelligent-unintelligent' ratings (expressed in percentage form) for the DS and TD groups by the SNES, MES and PEER listener groups are shown in table 4.23.

INTELLIGENT-UNINTELLIGENT: Group ratings breakdown %						
	SNES (49)		MES (41)		PEER (66)	
	DS	TD	DS	TD	DS	TD
very intelligent	1.28	20.72	1.53	16.51	0.38	13.07
quite intelligent	13.55	51.66	12.27	51.07	11.22	53.41
neutral	34.02	23.79	34.66	23.55	30.23	26.52
quite unintelligent	45.01	3.58	36.50	8.26	39.16	6.82
very unintelligent	6.14	0.26	15.03	0.61	19.01	0.19

Table 4.23: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter 'intelligent-unintelligent'

Table 4.24 reports the median (IQR) and overall percentage ratings by the SNES, MES and PEER raters for the intelligent-unintelligent parameter and the degree of statistically significant differences found in Mann Whitney U-tests between ratings of the DS and TD groups. All listener groups rated the DS group more negatively than the TD group. Medians and range of values are represented as a boxplot in figure 4.10.

INTELLIGENT-UNINTELLIGENT: Median (IQR) & overall group ratings %			
	DS	TD	statistical significance
SNES (49)	1 (1) 39.71%	3 (1) 72.25%	n = 391 (DS & TD), U = 22429.0, $p < 0.001$
MES (41)	1 (1) 37.19%	3 (1) 68.65%	n = 326 (DS), 327 (TD), U = 18601.0, $p < 0.001$
PEER (66)	1(1) 33.70%	3 (1) 68.09%	n = 526 (DS), 528 (TD), U = 40726.0, $p < 0.001$

Table 4.24: 'Intelligent-unintelligent' median (IQR) and overall percentage ratings attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

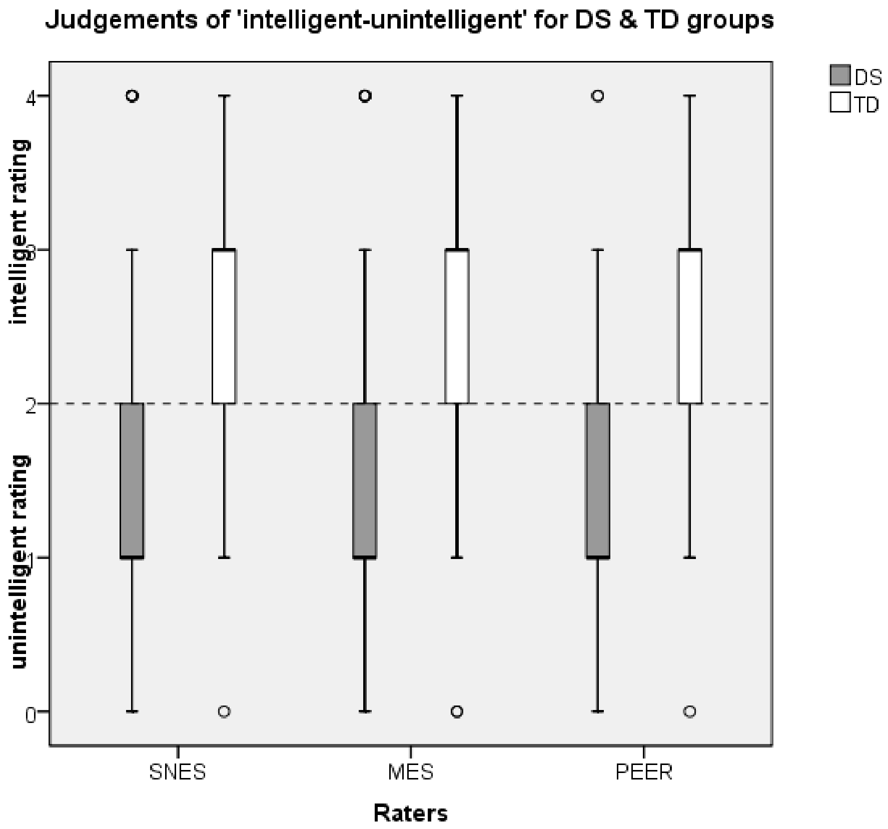


Figure 4.10: Boxplot illustrating the median and range of 'intelligent-unintelligent' judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.5.3 Analysis of differences between listener groups' ratings of DS and TD groups

Analysis by a Kruskal Wallis test and subsequent post hoc Mann-Whitney U-tests identified statistically significant differences in the pattern of ratings for intelligence between the SNES and PEER raters for both the DS and TD groups; in both cases the SNES listeners rated more positively than the PEER listeners (table 4.25). All significant results are based on an alternative p value of 0.017 due to Bonferroni adjustment.

INTELLIGENT-UNINTELLIGENT: Differences between raters		
	Statistically significant differences	
Listener Group	**DS**	**TD**
SNES versus MES	ns	ns
SNES versus PEER	n = 391 (SNES), 526 (PEER), *U* = 88633.0, p < 0.001	n= 391 (SNES), 528 (PEER), *U* = 92021.0, p = 0.002
MES versus PEER	ns	ns

Table 4.25: Statistically significant differences in ratings of 'intelligent-unintelligent' for DS and TD groups between SNES, MES and PEER raters

4.2.5.4 Analysis of the effect of gender and age on PEER ratings

A Mann Whitney U-test found the ratings of the male PEER listeners for the DS group to be significantly more negative than those of the females (male median rating 1.00, IQR, 2.00 compared to 2.00, IQR, 1.00 by the female raters; figure 4.11); no significant difference was found between genders for the TD group (table 4.26).

Statistically significant results were also identified between the ratings of the younger and older PEER listeners for the DS group (table 4.26); the older raters having a wider range of judgements than the older raters (younger, 1.00, IQR, 1.00 versus older, 1.00, IQR, 2.00). Results are illustrated in figure 4.12.

INTELLIGENT-UNINTELLIGENT: Effects of gender and age on PEER judgements				
	male (M) versus female (F)		younger (Y) versus older (O)	
Group	**DS**	**TD**	**DS**	**TD**
statistical significance	n = 288 (M), 238 (F), *U* = 21859.5, p < 0.001	ns	n = 286 (Y), 240 (O), *U* = 27520.0, p < 0.001	ns

Table 4.26: Statistically significant differences in ratings of 'intelligent-unintelligent' for DS and TD groups between PEER listeners grouped according to gender and age

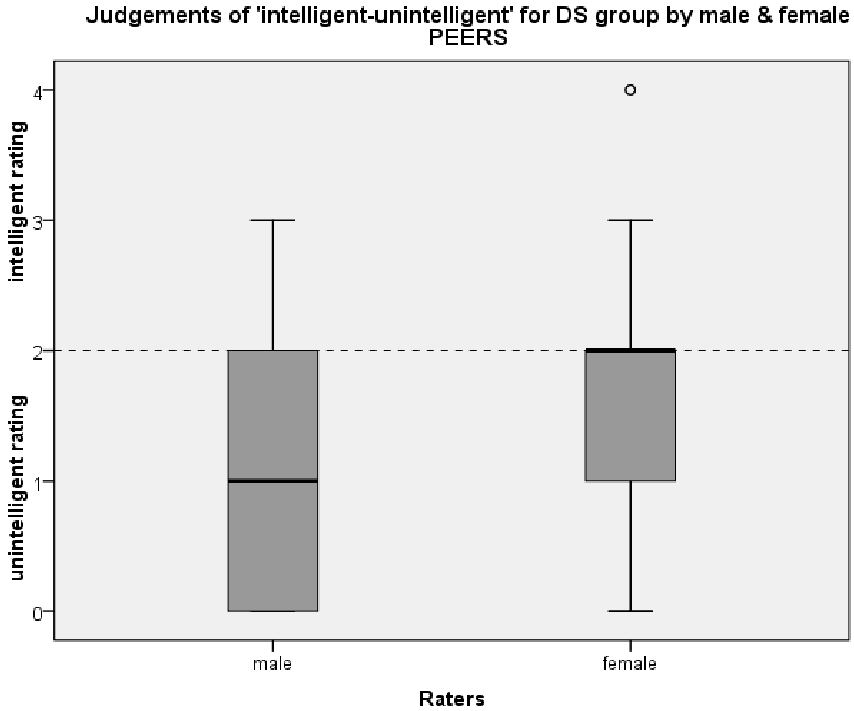


Figure 4.11: Boxplot showing the median and range of 'intelligent-unintelligent' for the DS group by the PEER raters when split by gender

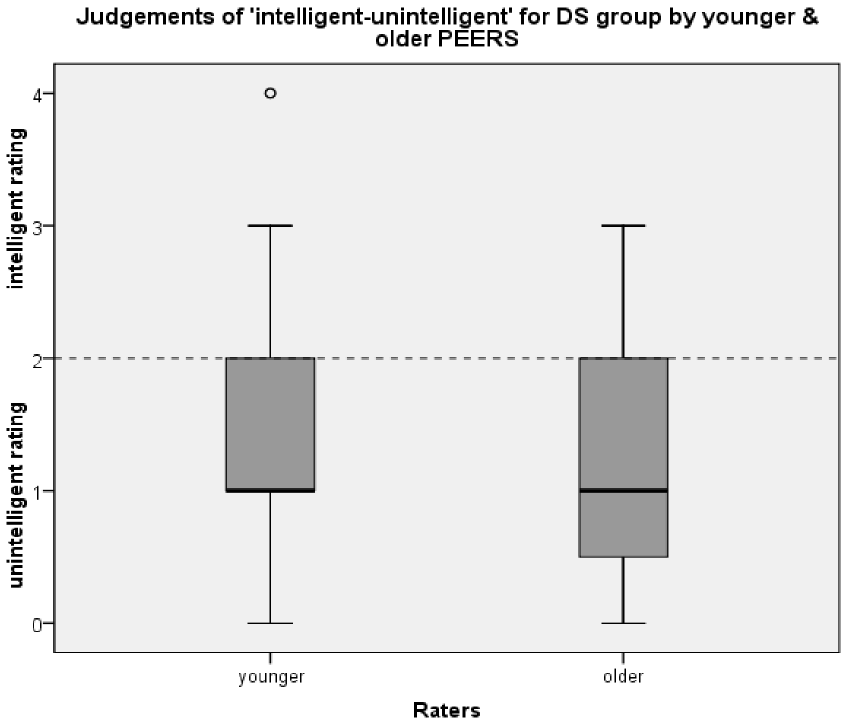


Figure 4.12: Boxplot showing the median and range of 'intelligent-unintelligent' for the DS group by the PEER raters when split by age

4.2.5.5 Variability in judgements of individual DS and TD speakers

Table 4.27 shows the overall intelligent-unintelligent ratings for each individual speaker (DS & TD) expressed in percentage form. These values are illustrated in figure 4.13.

INTELLIGENT-UNINTELLIGENT: Overall ratings for individual speakers %								
	DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
SNES (49)	38.27	53.57	34.69	38.78	44.79	34.69	36.73	48.47
MES (41)	35.00	50.63	37.80	42.07	40.85	37.20	39.02	45.12
PEER (66)	35.77	43.56	34.47	35.23	39.77	36.36	37.50	45.00
	TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
SNES (49)	64.58	78.57	70.92	84.69	85.71	72.45	59.69	61.73
MES (41)	56.71	73.13	72.56	81.10	82.32	73.78	54.88	56.10
PEER (66)	54.55	70.83	68.56	82.20	80.30	72.73	57.58	58.33

Table 4.27: Overall 'intelligent-unintelligent' rating (percentage) attributed to individual DS and TD speakers by SNES, MES and PEER raters

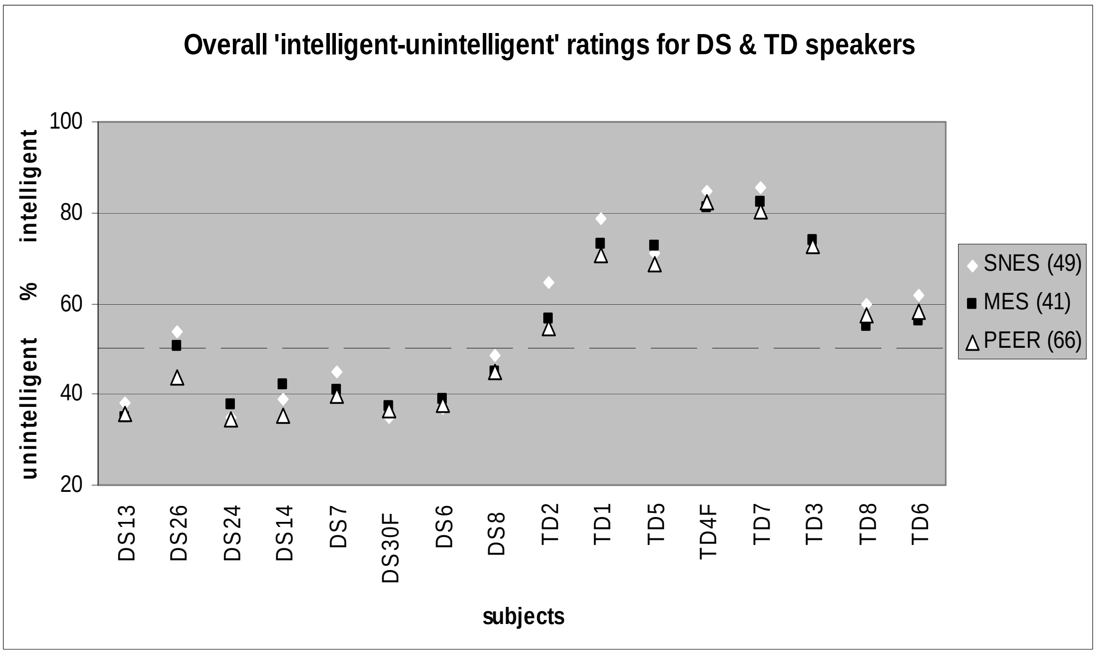


Figure 4.13: Diagram showing the relationship between overall 'intelligent-unintelligent' ratings (percentage) for individual DS and TD speakers by SNES, MES and PEER raters

4.2.6 ‘Spend time with’ ratings

4.2.6.1 Consistency of group judgements for repeated samples

This parameter was rated by the PEER group only. The percentages for consistency of judgements (individual & overall) for the seventy-two PEER raters who met the criteria for consistency are shown in table 4.28.

SPEND TIME WITH: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
PEER (72)	92.96	97.18	95.77	97.18	95.77

Table 4.28: Degree of consistency of the PEER group ratings (percentage) for the four randomly selected repeated samples, and the overall consistency score for the parameter ‘spend time with’

4.2.6.2 Analysis of differences between DS and TD ratings within listener group

The breakdown of ratings by PEER listeners, expressed as percentage values, for the parameter ‘spend time with’ for the DS and TD groups are illustrated in figure 4.14.

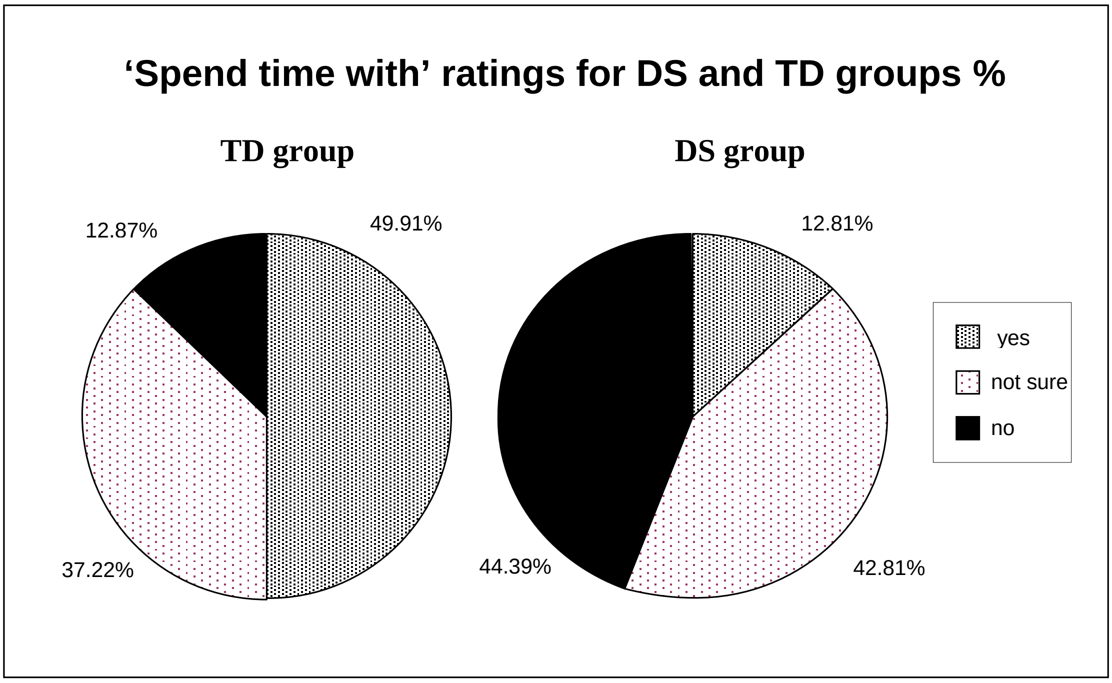


Figure 4.14: Pie chart illustrating the PEER ratings (‘yes’, ‘no’ & ‘not sure’) shown as percentage values, for the ‘spend time with’ parameter, for the DS and TD groups

Overall percentage of ratings are shown in table 4.29 alongside the degree of statistical significance found in a Chi Square test between the ratings of the DS and TD groups (the DS group being rated significantly less positively than their peers).

SPEND TIME WITH: Overall ratings %			
	DS	**TD**	**statistical significance**
PEER (72)	34.21%	68.52%	$\chi^2 = 227$ $df = 2$ $p < 0.001$

Table 4.29: 'Spend time with' overall percentage ratings attributed to DS and TD groups by PEER raters, and test results between DS and TD ratings of 'yes', 'not sure' and 'no' by PEER listeners

4.2.6.3 Analysis of the effect of gender and age on PEER ratings

No significant differences were found between the male and female PEER raters or the younger and older raters for either the DS or TD groups.

4.2.6.4 Variability in judgements of individual DS and TD speakers

Table 4.30 shows the individual breakdown of ratings for the parameter 'spend time with' ('yes', 'no' & 'not sure') and a calculation of the overall degree to which PEER listeners rated that they would like to 'spend time with' each speaker. The 'yes', 'no' and 'not sure' percentage values for each speaker are shown in a 100% stacked column graph in figure 4.15.

SPEND TIME WITH: Overall ratings for individual speakers %				
PEER (72)	yes	not sure	no	overall desirability
DS13	11.11	31.94	56.94	**27.08**
DS26	34.29	41.43	24.29	**55.00**
DS24	6.94	41.67	51.39	**27.78**
DS14	7.04	32.39	60.56	**23.24**
DS7	11.27	49.30	39.44	**35.92**
DS30F	6.94	56.94	36.11	**35.42**
DS6	5.63	46.48	47.89	**28.87**
DS8	19.72	42.25	38.03	**40.85**
TD2	20.83	61.11	18.06	**51.39**
TD1	41.67	40.28	18.06	**61.81**
TD5	65.28	31.94	2.78	**81.25**
TD4F	79.17	18.06	2.78	**88.19**
TD7	70.42	22.54	7.04	**81.69**
TD3	63.89	31.94	4.17	**79.86**
TD8	31.94	50.00	18.06	**56.94**
TD6	26.39	41.67	31.94	**47.22**

Table 4.30: Breakdown of ratings (‘yes’, ‘no’ & ‘not sure’) and overall ‘spend time with’ rating (percentage) attributed to individual DS and TD speakers by PEER raters

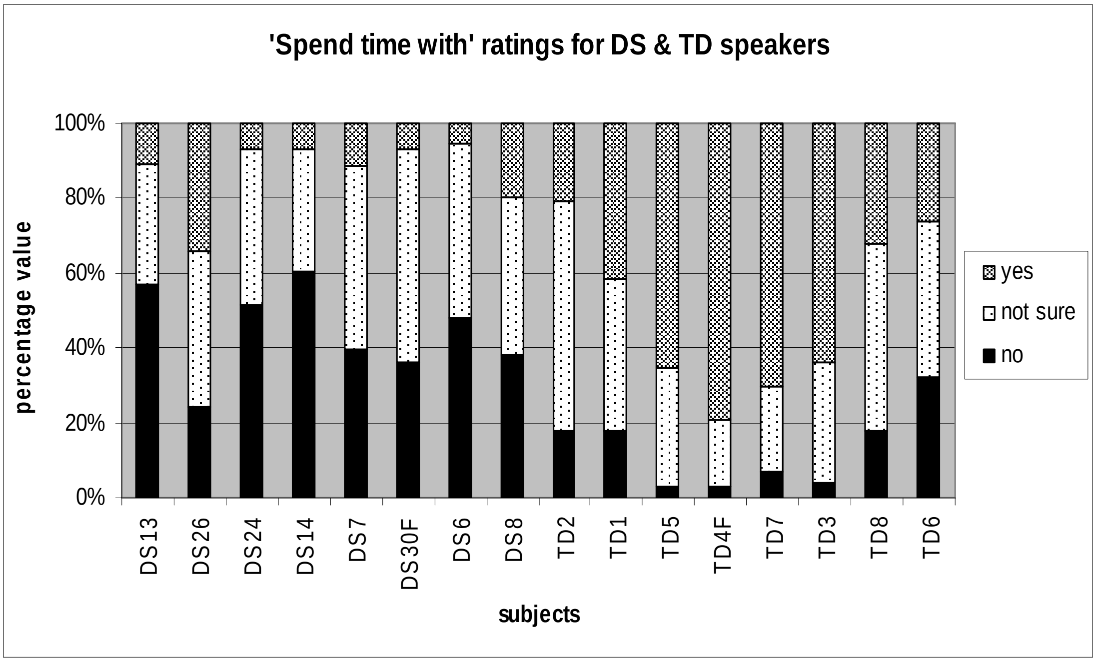


Figure 4.15: Stacked column chart showing the breakdown of ratings (‘yes’, ‘no’ & ‘not sure’) for individual DS and TD speakers by PEER raters

4.2.7 Gender ratings

4.2.7.1 Consistency of group judgements for repeated samples

The consistency ratings of the SNES (49), MES (40) and PEER (74) listeners meeting the consistency of judgements criteria are shown in table 4.31.

GENDER: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (49)	95.92	73.47	95.92	100.00	92.86
MES (40)	100.00	52.50	100.00	100.00	88.75
PEER (74)	100.00	78.08	97.30	100.00	93.90

Table 4.31: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter 'gender'

4.2.7.2 Analysis of differences between DS and TD ratings within listener groups

Table 4.32 shows the breakdown of gender judgements (percentage) for the DS and TD children by all 3 listener groups and the degree of statistically significant differences found in a Chi Square test between both groups. In each case judgements of gender were found to be less accurate for the DS speakers than the TD speakers.

GENDER: Breakdown of ratings (%)			
SNES (49)	**DS**	**TD**	**statistical significance**
correct	67.52	83.63	$\chi^2 = 14.4$
incorrect	24.55	14.32	$df = 2$
unsure	7.93	2.05	$p = 0.001$
MES (40)	**DS**	**TD**	**statistical significance**
correct	66.04	84.33	$\chi^2 = 8.22$
incorrect	27.67	13.17	$df = 2$
unsure	6.29	2.51	$p = 0.016$
PEER (74)	**DS**	**TD**	**statistical significance**
correct	65.99	82.77	$\chi^2 = 8.62$
incorrect	27.07	14.02	$df = 2$
unsure	6.94	3.21	$p = 0.013$

Table 4.32: Breakdown of ratings (percentage) by SNES, MES and PEER listeners for DS and TD groups for the parameter 'gender', and test results between DS and TD ratings within listener groups

4.2.7.3 Analysis of differences between listener groups' ratings of DS and TD groups

No significant differences were identified in the gender ratings of either the DS or TD groups between the SNES, MES and PEER raters.

4.2.7.4 Analysis of the effect of gender and age on PEER ratings

No significant differences were found between the judgements of gender between the male and female or the younger and older PEER raters for either the DS or TD groups.

4.2.7.5 Variability in judgements of individual DS and TD speakers

The percentage of correct judgements of the gender of individual DS and TD speakers by all three rater groups are shown in table 4.33 and illustrated as a bar chart in figure 4.16.

GENDER: Correct judgements for individual speakers %								
DS	**DS13**	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
SNES (49)	97.96	14.29	95.83	100.00	81.63	40.82	100.00	10.20
MES (40)	90.00	10.00	87.18	100.00	80.00	51.28	95.00	15.00
PEER (74)	100.00	8.11	95.95	100.00	78.38	34.25	98.65	12.16
TD	**TD2**	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
SNES (49)	97.96	100.00	2.08	97.96	100.00	69.39	100.00	100.00
MES (40)	100.00	100.00	7.69	100.00	100.00	65.00	100.00	100.00
PEER (74)	100.00	98.65	5.41	98.65	100.00	60.81	98.65	100.00

Table 4.33: Correct judgements of 'gender' (percentage) for individual DS and TD speakers by SNES, MES and PEER raters

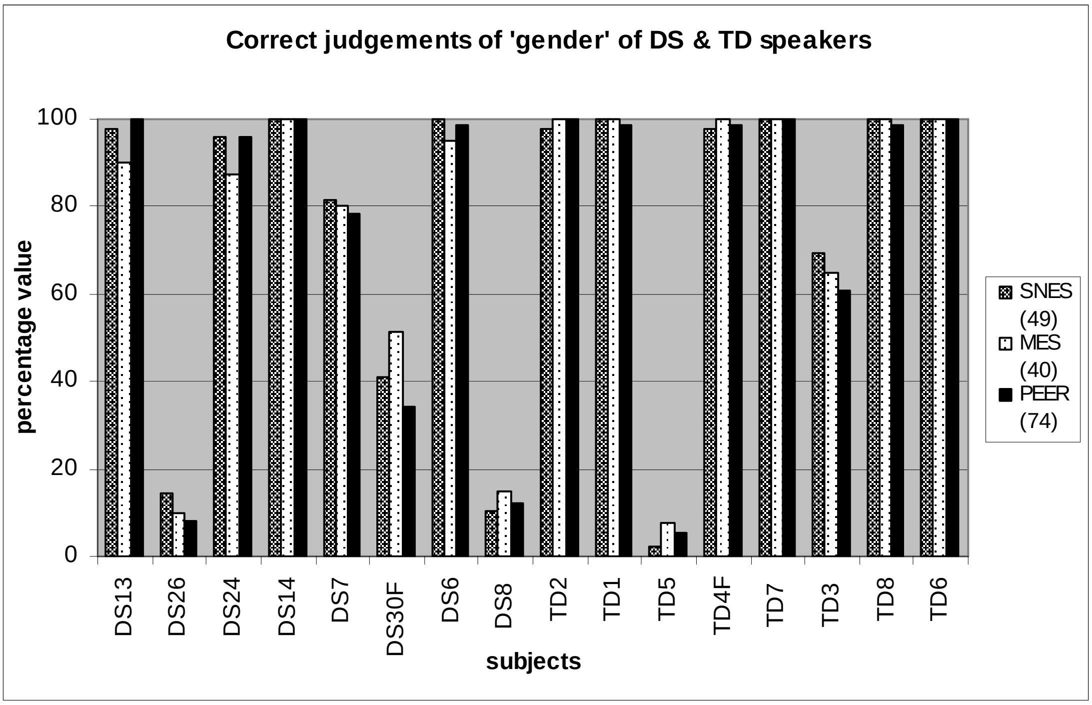


Figure 4.16: Bar chart showing the correct judgements of ‘gender’ (percentage) for individual DS speakers by SNES, MES and PEER raters

4.2.8 Age ratings

4.2.8.1 Consistency of group judgements for repeated samples

The consistency of judgements (percentages) for the raters meeting the consistency criteria (SNES, 30, MES, 30 & PEER, 59) are shown in table 4.34.

AGE: Consistency of repeated ratings %					
	Rep DS13	**Rep DS7**	**Rep TD2**	**Rep TD7**	**Overall consistency (across all 4 reps)**
SNES (30)	83.33	96.67	90.00	80.00	87.50
MES (30)	83.33	96.67	86.67	73.33	85.00
PEER (59)	91.53	100.00	86.44	79.31	89.36

Table 4.34: Degree of consistency of the SNES, MES and PEER group ratings (percentage) for the four randomly selected repeated recordings, and the overall consistency score for the parameter ‘age’

4.2.8.2 Analysis of differences between DS and TD ratings within listener groups

The median (IQR) age judgements for the DS and TD groups by the SNES, MES and PEER listeners, and the actual median age of the groups are reported in table 4.35 along with the degree of statistically significant differences found in Mann Whitney U-tests between the two groups. All three listener groups judged both the DS and TD groups to be older than their actual median age, with a significantly greater range in the judgements of age for the DS group. The median value, range and relationship between judgements by the three listener groups are illustrated as a boxplot in figure 4.17.

AGE: Overall median and interquartile range (IQR)			
	DS group	**TD group**	**statistical significance**
Actual median	11.42	12.17	N/A
SNES Ratings (30)	14.00 (6.00)	14.00 (4.00)	n = 240 (DS & TD), *U* = 24290.0, p = 0.03
MES Ratings (30)	14.00 (6.00)	14.00 (3.00)	n = 240 (DS & TD), *U* = 25873.0, p = 0.053
PEER Ratings (59)	13.00 (7.00)	13.00 (3.00)	n = 470 (DS), 472 (TD), *U* = 99071.5, p = 0.004

Table 4.35: Median (IQR) 'age' judgements attributed to DS and TD groups by SNES, MES and PEER raters, and test results between DS and TD ratings within listener groups

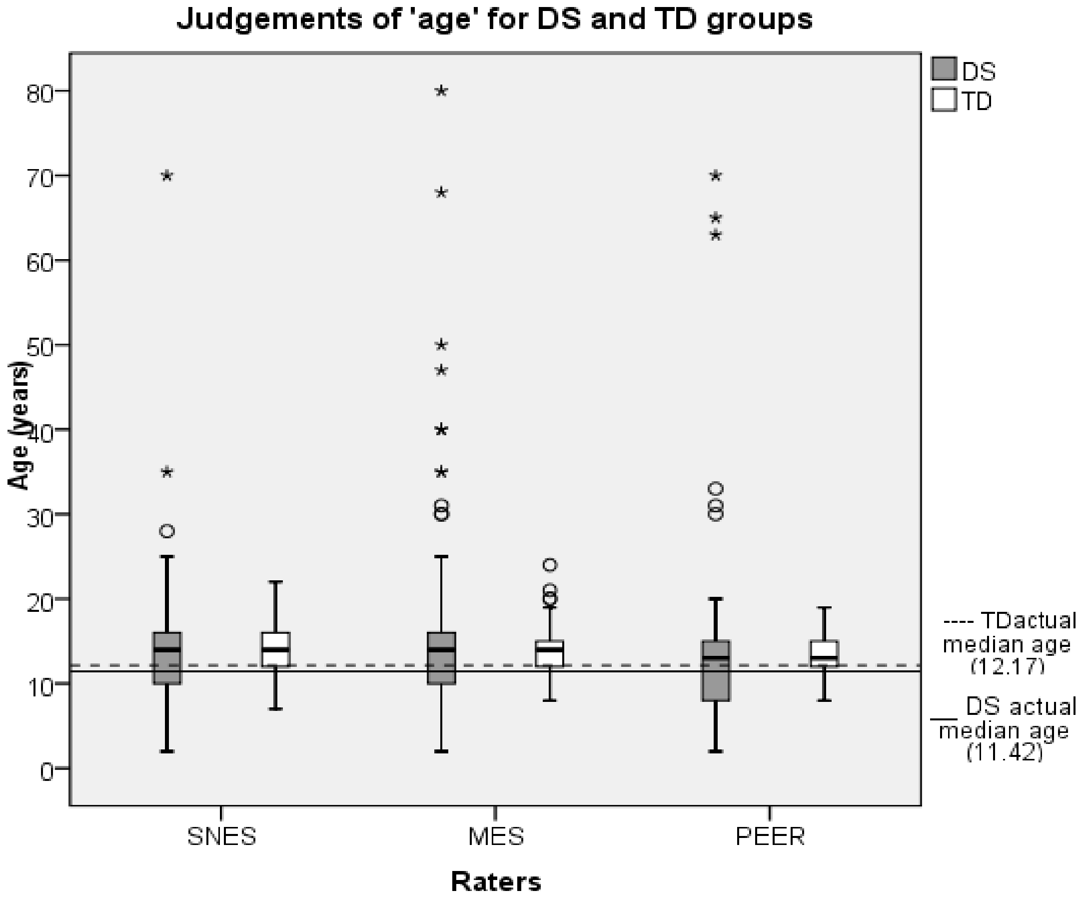


Figure 4.17: Boxplot illustrating the median and range of 'age' judgements attributed to DS and TD groups by SNES, MES and PEER raters

4.2.8.3 Analysis of differences between listener groups' ratings of DS and TD groups

A Kruskal Wallis and post hoc Mann Whitney U-tests identified significant differences in the judgements of age between the SNES and PEER raters and the MES and PEER raters for both the DS and TD groups (table 4.36). In both cases, the PEER raters judged the age of the speakers as younger than the median judgements by the education staff. Bonferroni adjustment created an alternative *p* value of 0.017.

AGE: Differences between raters		
	Statistically significant differences	
Listener Group	**DS**	**TD**
SNES versus MES	ns	ns
SNES versus PEER	n = 240 (SNES), 470 (PEER), *U* = 49787.0, p = 0.01	n = 240 (SNES), 472 (PEER), *U* = 42197.5, p < 0.001
MES versus PEER	n = 240 (MES), 470 (PEER), *U* = 49605.5, p = 0.008	n = 240 (MES), 472 (PEER), *U* = 46353.0, p < 0.001

Table 4.36: Statistically significant differences in judgements of 'age' for DS and TD groups between SNES, MES and PEER raters

4.2.8.4 Analysis of the effect of gender and age on PEER ratings

Mann-Whitney U-tests identified statistically significant differences within the PEER listener group when adjusted for gender and age (table 4.37).

The female PEER raters were found to have a significantly higher (older) range of age judgements than those of the male PEER raters for the TD group, although no differences were found in the median & IQR values (male median judgement, 13.00, IQR, 3.00 years and female median judgement, 13.00, IQR, 3.00 years); values are illustrated as a boxplot in figure 4.18. No significant difference was found between judgements of the DS group.

Significant statistical differences were also found between the younger and older raters for judgements of the TD group, with the younger raters being more accurate in their judgements of age (younger median 13.00, IQR 3.00 compared to the older median of 14.00, IQR 3.00; illustrated as a boxplot in figure 4.19). Again, no difference was found between the judgements of the DS group.

AGE: Effects of gender and age on PEER judgements				
	male (M) versus female (F)		younger (Y) versus older (O)	
PEER (59)	**DS**	**TD**	**DS**	**TD**
statistical significance	ns	n = 296 (M), 176 (F), *U* = 22880.5, p = 0.03	ns	n = 256 (Y), 216 (O), *U* = 22057.0, p < 0.001

Table 4.37: Statistically significant differences in judgements of 'age' for DS and TD groups between PEER listeners grouped according to gender and age

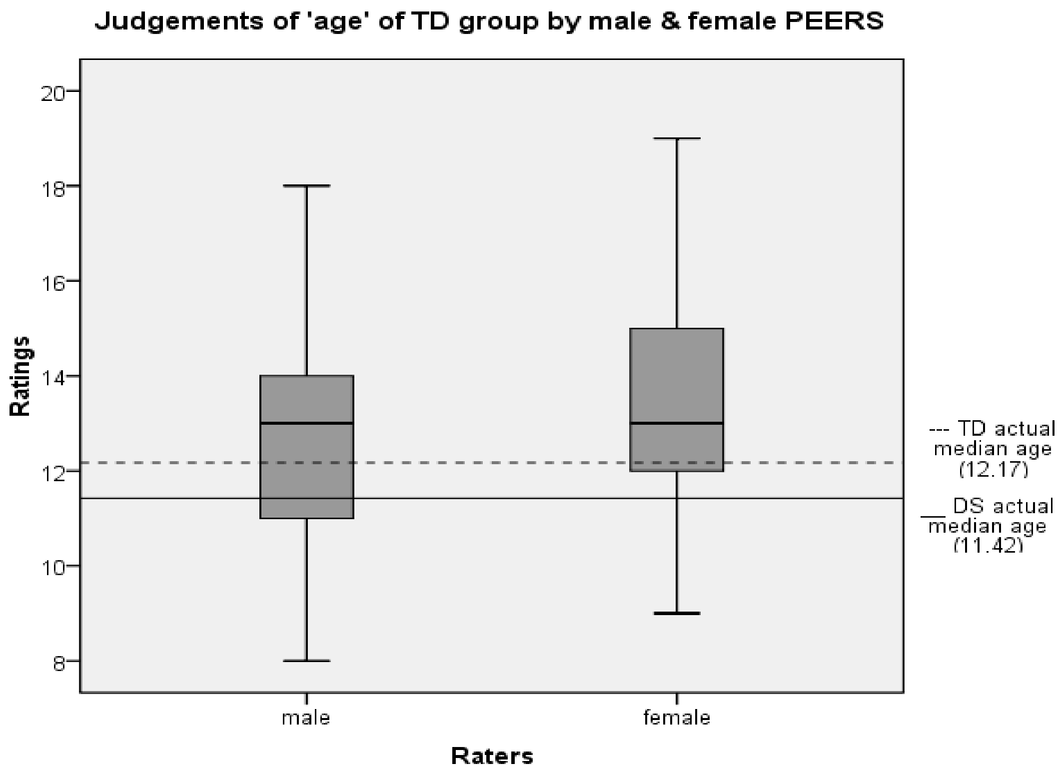


Figure 4.18: Boxplot showing the median and range of 'age' judgements for the TD group by the PEER raters when split by gender

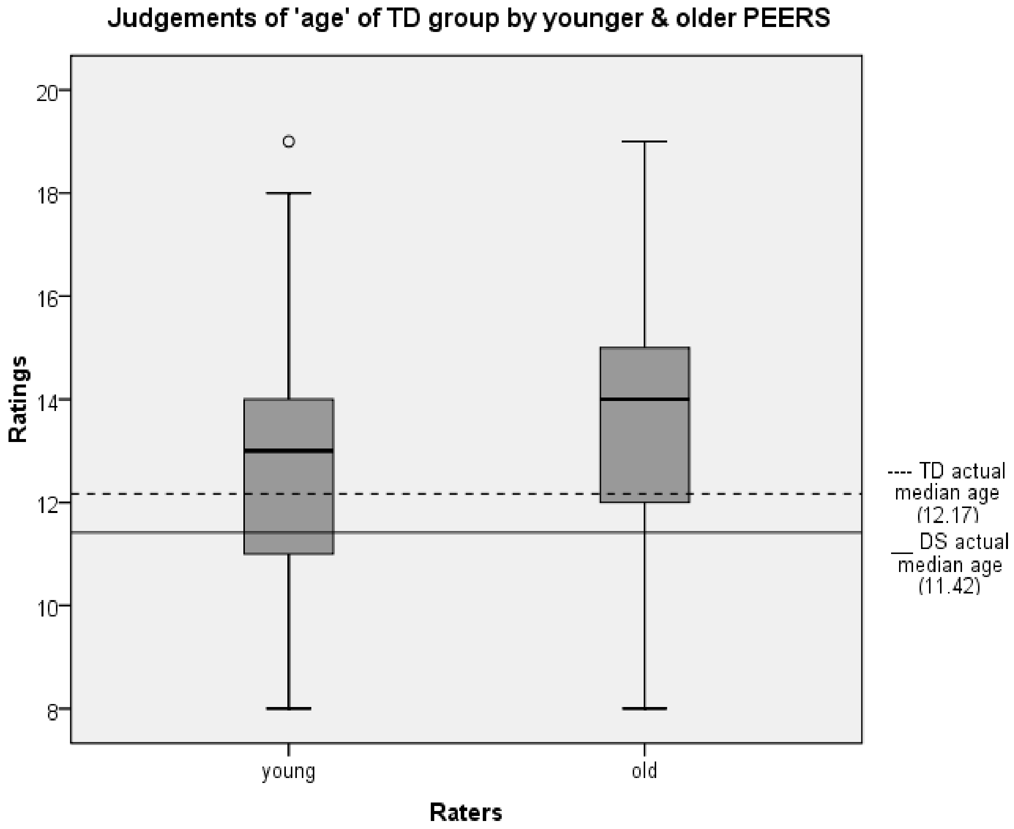


Figure 4.19: Boxplot showing the median and range of ‘age’ judgements for the TD group by the PEER raters when split by age

4.2.8.5 Variability in judgements of individual DS and TD speakers

The actual age of individual speakers within the DS and TD groups, alongside the median judged age and IQR and the full range of judgements (minimum to maximum) by each of the three listener groups are presented in table 4.38. Judgements are illustrated in figure 4.20 in relation to actual ages.

AGE: Individual median (IQR) & min-max ratings for DS & TD speakers				
	actual age	**SNES (30)**	**MES (30)**	**PEER (59)**
DS13	14.00	**15.5** (4) 9-35	**16** (3) 13-47	**15** (1) 10-19
DS26	10.58	**4.75** (2) 2-12	**5** (2.2) 2-8	**4** (2) 2-13
DS24	11.58	**15** (4) 7-25	**15** (3) 9-40	**14** (3) 8-31
DS14	16.08	**16** (4) 8-70	**16.5** (5) 10-80	**15** (3) 5-70
DS7	11.00	**14** (2) 10-19	**13** (2) 9-20	**13** (4) 7-33
DS30	10.08	**10.25** (5) 4-15	**10** (4) 4-20	**9** (4) 7-33
DS6	16.50	**16.5** (4) 11-25	**15.5** (5) 12-40	**15** (2) 11-20
DS8	11.25	**10** (4) 6-17	**10** (4) 5-15	**8** (3) 4-14
TD2	12.00	**12** (2) 7-16	**12** (3) 8-15	**11** (3) 8-16
TD1	12.25	**13** (2) 10-16	**12.5** (2) 9-16	**12** (2) 8-14
TD5	10.00	**13.5** (2) 9-16	**12** (2) 10-16	**12** (3) 8-15
TD4	11.83	**14** (3) 9-19	**14** (3) 12-21	**13** (2) 10-19
TD7	14.00	**16** (2) 12-20	**16** (2) 13-18	**15** (2) 11-18
TD3	12.08	**13** (3) 9-18	**12** (2) 10-17	**12** (2) 8-15
TD8	14.50	**16** (2) 10-22	**16** (2) 13-24	**15** (1) 12-19
TD6	15.00	**16** (2) 12-20	**15** (2) 12-20	**14** (1) 12-18

Table 4.38: Actual age of DS and TD speakers with median (IQR) and maximum - minimum judgements of 'age' attributed to speakers by SNES, MES and PEER raters

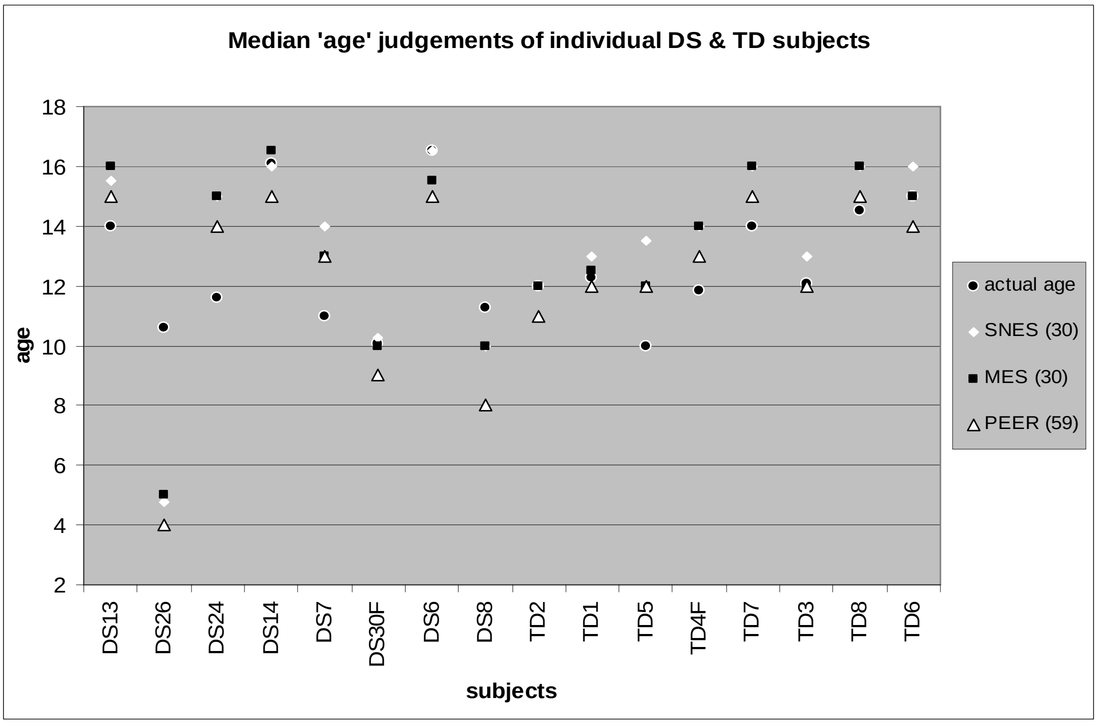


Figure 4.20: Diagram showing the overall median 'age' judgements for individual DS and TD speakers by SNES, MES and PEER raters in comparison to actual age

4.3 STUDY 2: ACOUSTIC ANALYSIS OF VOICE

This section will report the findings of fundamental frequency (F0) mean and F0 mean (stdev), jitter (rap & ppq5), shimmer (apq3 & apq5), harmonic-to-noise ratio (HNR), and spectral tilt (SPT, 1-5kHz & 2-5kHz) analysis of recordings. This includes:

- Results between overall DS and TD groups
- Results between DS and TD male speakers
- Results between DS and TD female speakers
- Results between DS female and male speakers
- Results between TD female and male speakers
- Results between DS and TD speakers presented in study 1
- Variability between individual DS and TD speakers presented in study 1

Where statistical results are given, mean and standard deviation values are reported for the overall groups and median and IQR values for the smaller subsets. Non-significant results are abbreviated as 'ns'.

4.3.1 Results between overall DS and TD groups

Independent samples t-tests identified that within the overall DS (22) and TD (52) groups the only parameters to differ significantly between groups were those of SPT (1-5kHz & 2-5kHz); in both instances the SPT values were higher for the DS speakers. Table 4.39 shows the mean (stdev) values of each parameter for both groups and the degree of the statistically significant differences. SPT values are illustrated as boxplots in figures 4.21 and 4.22.

Acoustic analysis: Mean (stdev) values of overall DS & TD groups			
Parameter	**DS (22)**	**TD (52)**	**statistical significance**
F0 mean	192.10 (60.17)	184.53 (52.13)	ns
F0 mean (stdev)	13.10 (6.55)	10.90 (3.97)	ns
jitter (rap)	1.04 (0.36)	0.93 (0.26)	ns
jitter (ppq5)	1.04 (0.28)	0.98 (0.25)	ns
shimmer (apq3)	4.36 (1.02)	4.43 (1.10)	ns
shimmer (apq5)	5.69 (1.12)	5.90 (1.37)	ns
HNR	13.56 (1.86)	12.79 (2.42)	ns
SPT 1-5kHz	16.53 (3.08)	13.89 (2.56)	$t = 3.786$, $df = 72$, $p < 0.001$
SPT 2-5kHz	22.61 (2.63)	19.56 (2.88)	$t = 4.228$, $df = 72$, $p < 0.001$

Table 4.39: Mean (stdev) values and statistical differences between overall DS and TD groups across all acoustic analysis parameters

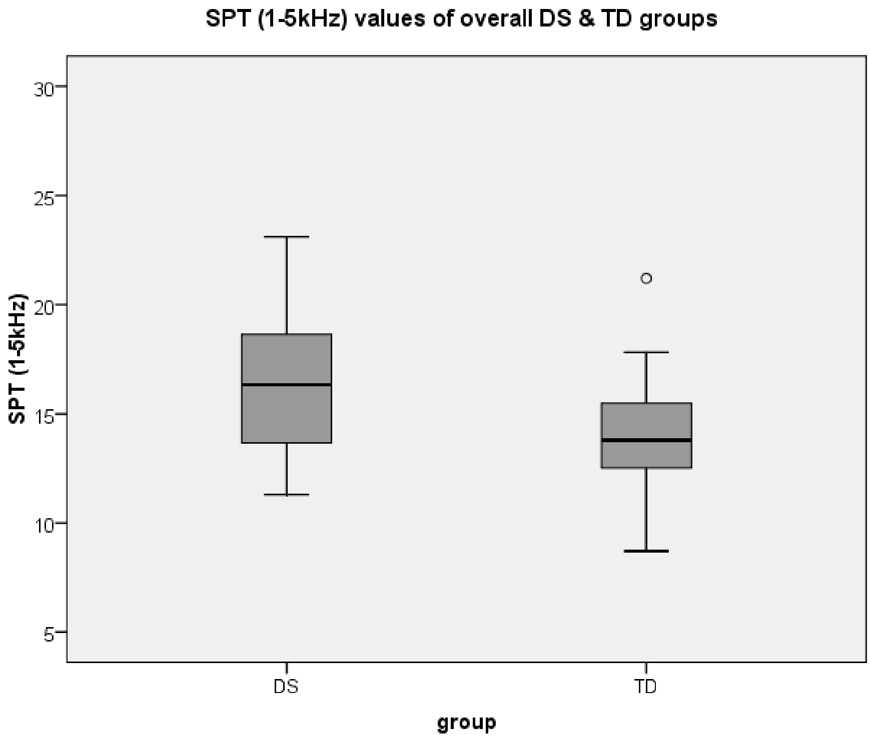


Figure 4.21: Boxplot showing SPT (1-5kHz) values of the overall DS and TD groups

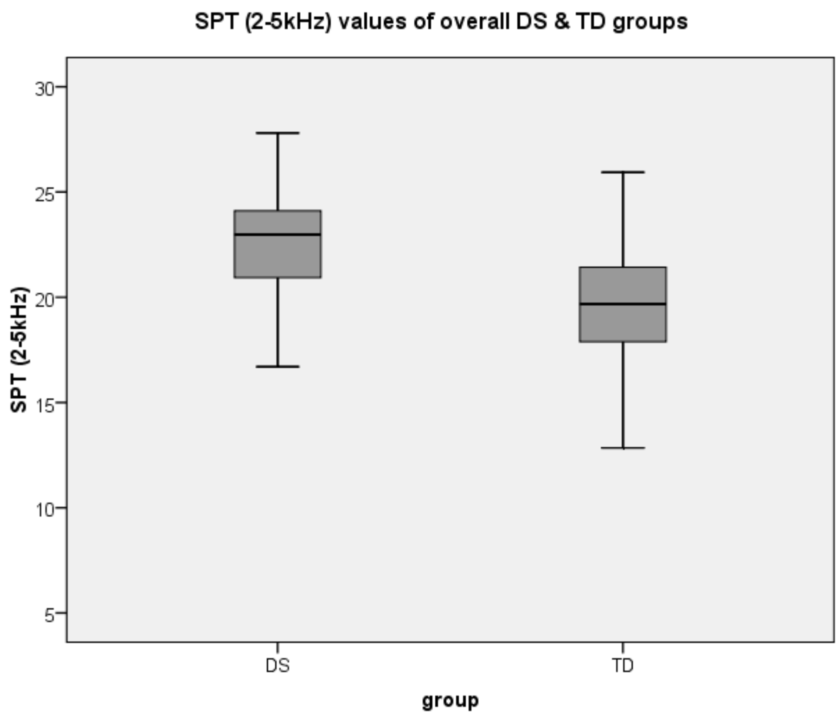


Figure 4.22: Boxplot showing SPT (2-5kHz) values of the overall DS and TD groups

4.3.2 Results between DS and TD male speakers

Mann Whitney U-tests found the same pattern of results as in the overall groups between DS males (13) and TD males (34), with only SPT values differing significantly; again values were higher in the DS males than the TD males (table 4.40). SPT findings are illustrated as boxplots in figures 4.23 and 4.24.

Acoustic analysis: Median (IQR) values of male DS & TD speakers			
Parameter	**DS (13)**	**TD (34)**	**statistical significance**
F0 mean	136.63 (75.06)	174.73 (104.66)	ns
F0 mean (stdev)	9.48 (7.14)	9.14 (6.52)	ns
jitter (rap)	0.95 (0.34)	0.89 (0.36)	ns
jitter (ppq5)	1.00 (0.25)	0.97 (0.30)	ns
shimmer (apq3)	4.04 (1.18)	4.54 (1.30)	ns
shimmer (apq5)	5.51 (1.06)	6.25 (1.84)	ns
HNR	12.91 (1.66)	12.21 (3.47)	ns
SPT (1-5kHz)	16.91 (4.66)	13.47 (2.54)	n = 13 (DS), 34 (TD), $U = 79.5$, $p = 0.001$
SPT (2-5kHz)	23.33 (3.13)	19.68 (4.22)	n = 13 (DS), 34 (TD), $U = 96.0$, $p = 0.003$

Table 4.40: Median (IQR) values and statistical differences between male DS and TD speakers across all acoustic analysis parameters

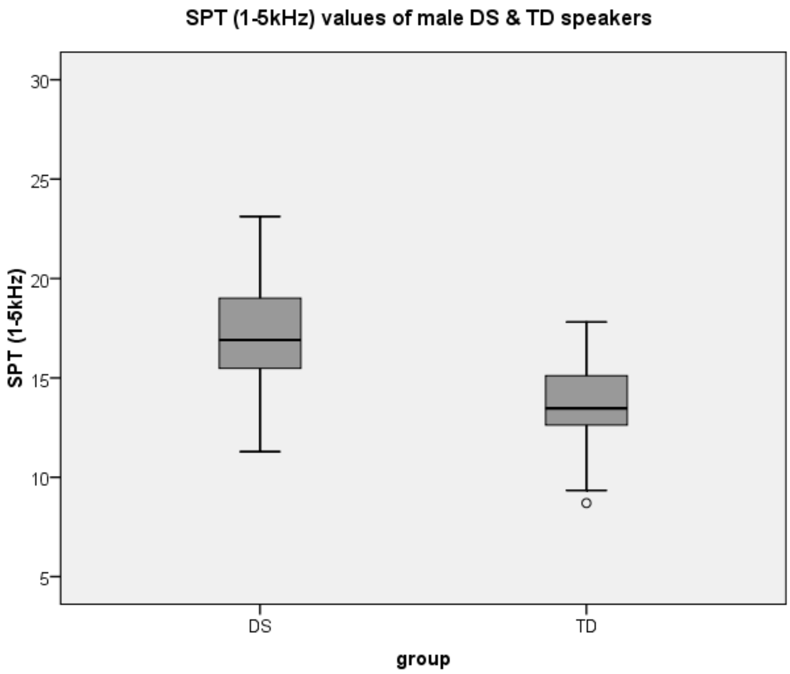


Figure 4.23: Boxplot showing SPT (1-5kHz) values of the male DS and TD speakers

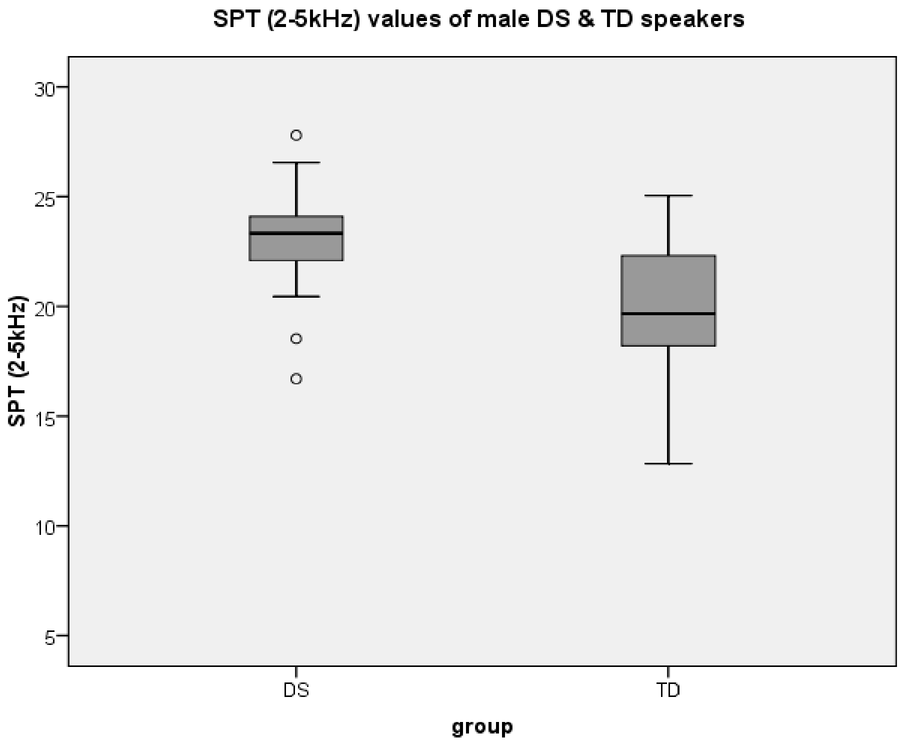


Figure 4.24: Boxplot showing SPT (2-5kHz) values of the male DS and TD speakers

4.3.3 Results between DS and TD female speakers

Table 4.41 shows the median (IQR) values and results of Mann Whitney U-tests between the female DS speakers (9) and the female TD speakers (18). Only SPT (2-5 kHz) was found to be significantly different (illustrated as a boxplot in figure 4.25).

Acoustic analysis: Median (IQR) values of female DS & TD speakers			
Parameter	**DS (9)**	**TD (18)**	**statistical significance**
F0 mean	236.71 (72.32)	226.40 (33.66)	ns
F0 mean (stdev)	18.28 (10.49)	12.60 (2.93)	ns
jitter (rap)	1.03 (0.40)	0.87 (0.36)	ns
jitter (ppq5)	1.05 (0.42)	0.89 (0.33)	ns
shimmer (apq3)	4.14 (1.76)	4.15 (1.37)	ns
shimmer (apq5)	5.52 (1.53)	5.15 (1.38)	ns
HNR	14.94 (2.59)	14.08 (2.70)	ns
SPT (1-5kHz)	15.28 (4.85)	14.92 (4.67)	ns
SPT (2-5kHz)	22.67 (3.32)	19.59 (4.59)	n = 9 (DS), 18 (TD), $U = 32.0$, $p = 0.011$

Table 4.41: Median (IQR) values and statistical differences between female DS and TD speakers across all acoustic analysis parameters

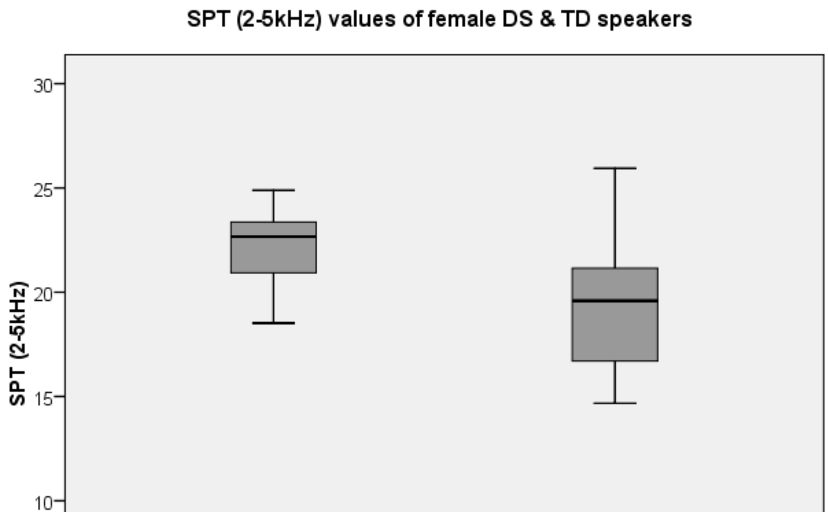


Figure 4.25: Boxplot showing SPT (2-5kHz) values of the female DS and TD speakers

4.3.4 Results between DS female and male speakers

The median (IQR) values and results of statistical analysis using Mann Whitney U-tests between DS females and males are shown in table 4.42. F0 mean, F0 mean (stdev) and HNR were all found to be higher in DS females than males (illustrated as boxplots in figures 4.26, 4.27 & 4.28).

Acoustic analysis: Median (IQR) values of DS female & male speakers			
Parameter	**DS females (9)**	**DS males (13)**	**statistical significance**
F0 mean	236.71 (72.32)	136.63 (75.06)	n = 9 (female), 13 (male), $U = 18.0$, $p = 0.006$
F0 mean (stdev)	18.28 (10.49)	9.48 (7.14)	n = 9 (female), 13 (male), $U = 25.0$, $p = 0.025$
jitter (rap)	1.03 (0.40)	0.95 (0.34)	ns
jitter (ppq5)	1.05 (0.42)	1.00 (0.25)	ns
shimmer (apq3)	4.14 (1.76)	4.04 (1.18)	ns
shimmer (apq5)	5.52 (1.53)	5.51 (1.06)	ns
HNR	14.94 (2.59)	12.91 (1.66)	n = 9 (female), 13 (male), $U = 25.0$, $p = 0.025$
SPT (1-5kHz)	15.28 (4.85)	16.91 (4.66)	ns
SPT (2-5kHz)	22.67 (3.32)	23.33 (3.13)	ns

Table 4.42: Median (IQR) values and statistical differences between female and male DS speakers across all acoustic analysis parameters

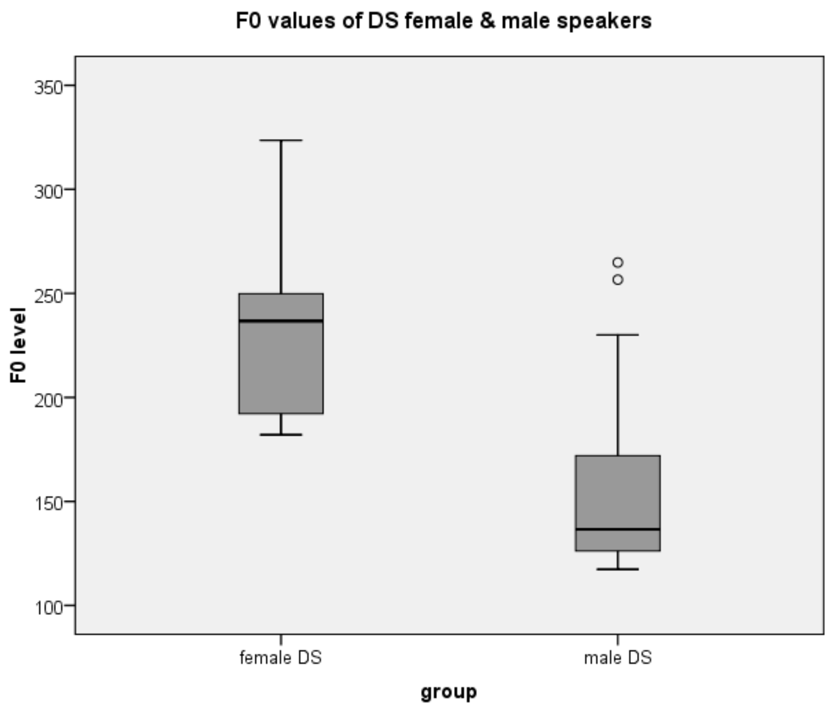


Figure 4.26: Boxplot showing F0 mean levels of DS female and male speakers

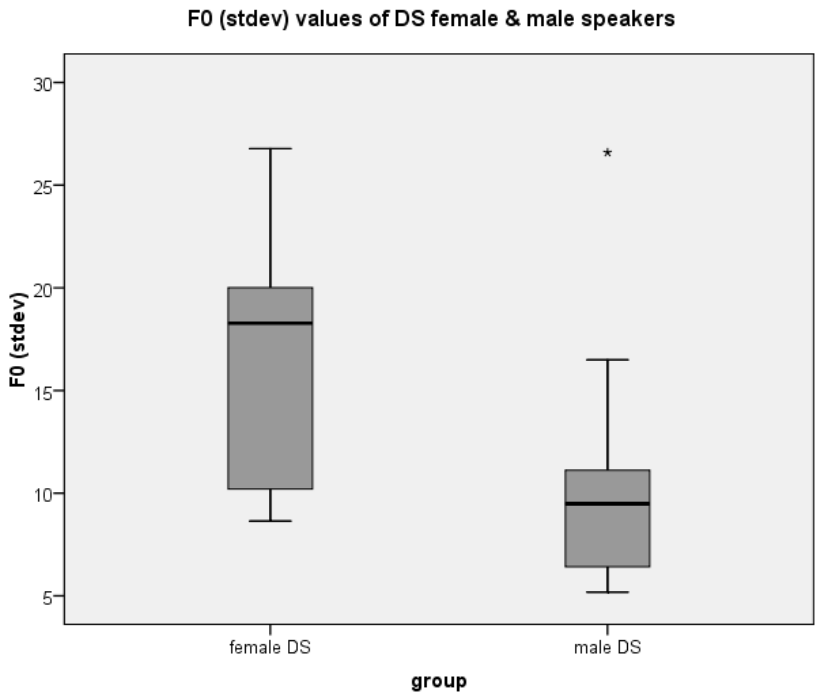


Figure 4.27: Boxplot showing F0 mean (stdev) levels of DS female and male speakers

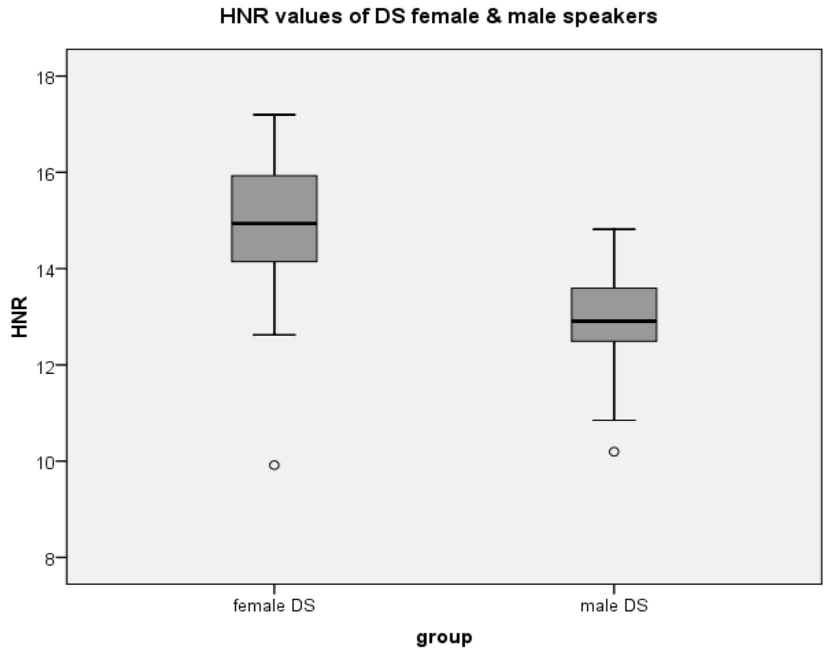


Figure 4.28: Boxplot showing HNR values of DS female and male speakers

4.3.5 Results between TD female and male speakers

Table 4.43 shows the median (IQR) values and results of Mann Whitney U-tests between TD females and males. TD females demonstrate significantly higher F0 mean, F0 mean (stdev) and HNR values but lower shimmer (apq5) values than male TD speakers. Significant results are shown as boxplots (figures 4.29-4.32).

Acoustic analysis: Median (IQR) values of TD female & male speakers			
Parameter	**TD females (18)**	**TD males (34)**	**statistical significance**
F0 mean	226.40 (33.66)	174.73 (104.66)	n = 18 (female), 34 (male), U = 124.0, p < 0.001
F0 mean (stdev)	12.60 (2.93)	9.14 (6.52)	n = 18 (female), 34 (male), U = 139.0, p = 0.001
jitter (rap)	0.87 (0.36)	0.89 (0.36)	ns
jitter (ppq5)	0.89 (0.33)	0.97 (0.30)	ns
shimmer (apq3)	4.15 (1.37)	4.54 (1.30)	ns
shimmer (apq5)	5.15 (1.38)	6.25 (1.84)	n = 18 (female), 34 (male), U = 163.0, p = 0.006
HNR	14.08 (2.70)	12.21 (3.47)	n = 18 (female), 34 (male), U = 149.0, p = 0.003
SPT (1-5kHz)	14.92 (4.67)	13.47 (2.54)	ns
SPT (2-5kHz)	19.59 (4.59)	19.68 (4.22)	ns

Table 4.43: Median (IQR) values and statistical differences between female and male TD speakers across all acoustic analysis parameters

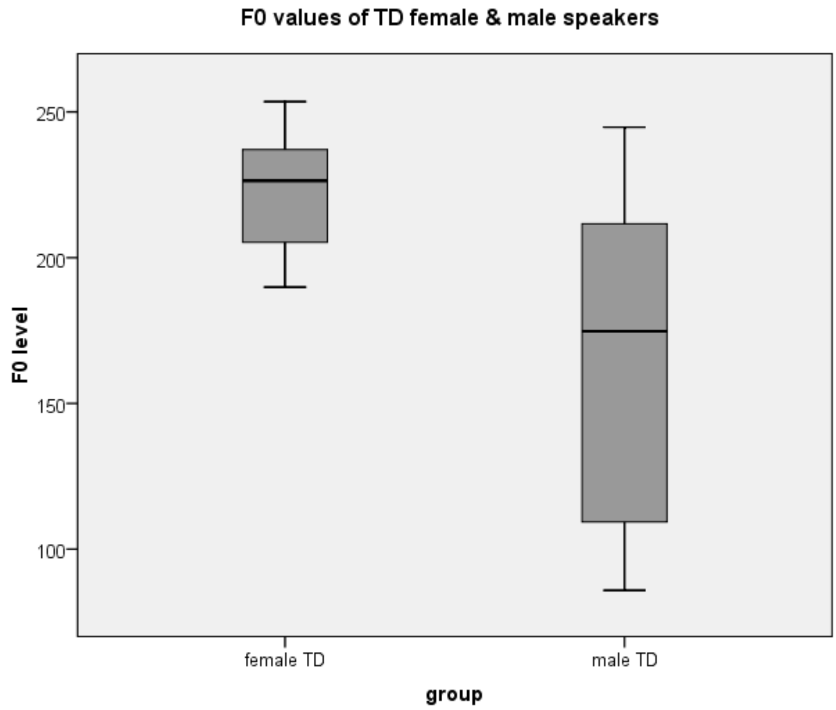


Figure 4.29: Boxplot showing F0 mean levels of TD female and male speakers

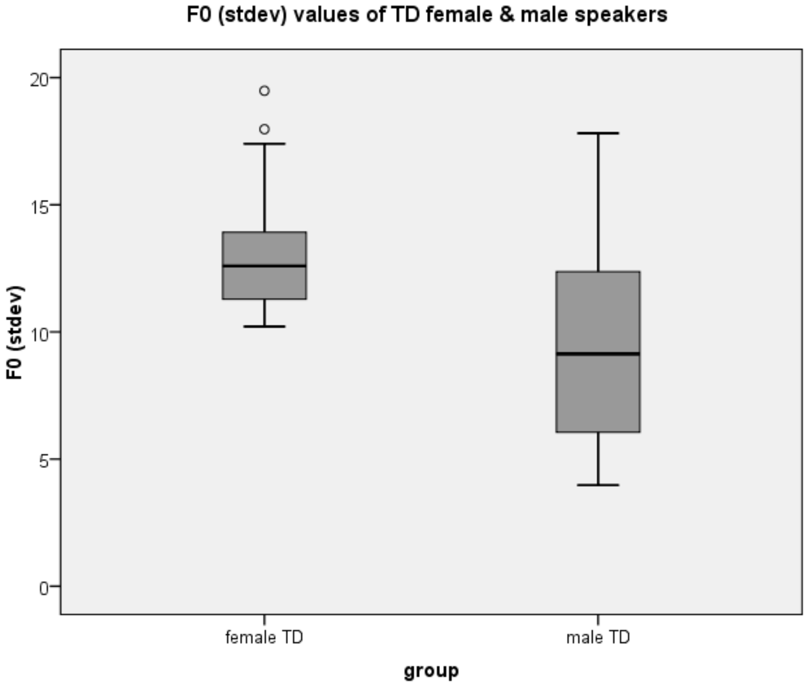


Figure 4.30: Boxplot showing F0 mean (stdev) levels of TD female and male speakers

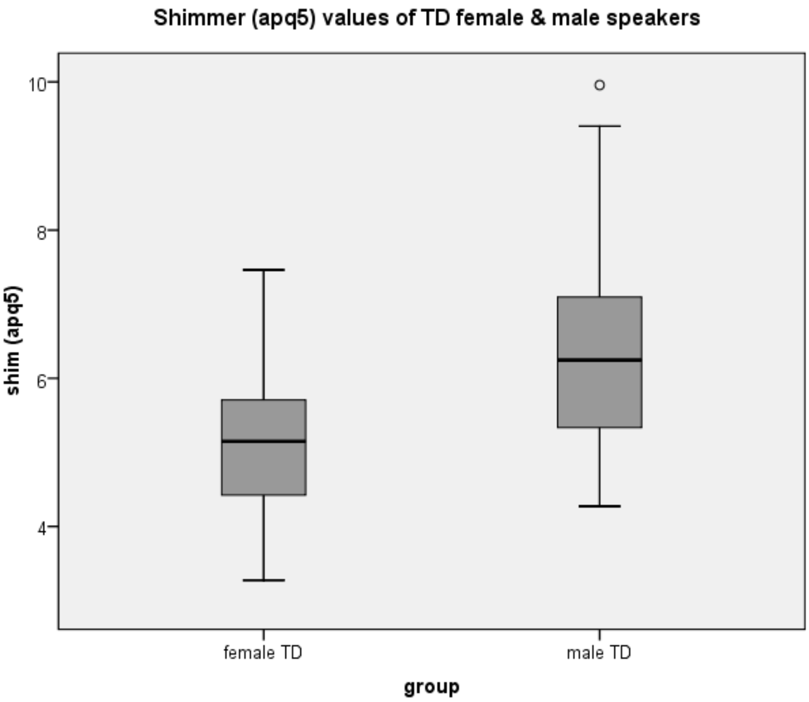


Figure 4.31: Boxplot showing shimmer (apq5) levels of TD female and male speakers

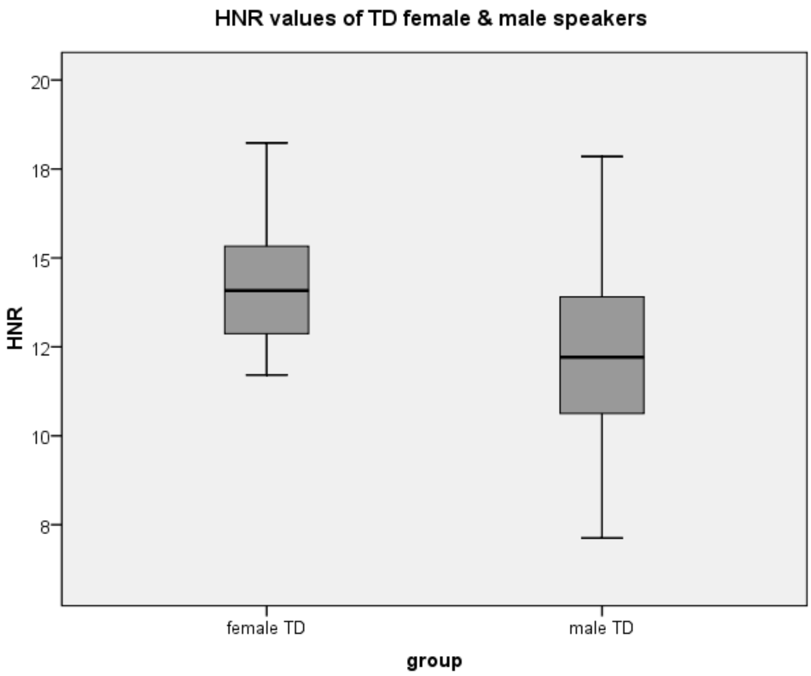


Figure 4.32: Boxplot showing HNR levels of TD female and male speakers

4.3.6 Results between DS and TD speakers presented in study 1

Within the eight DS and eight TD speakers presented to listeners in study 1, only jitter (rap) was found to significantly different (table 4.44). The jitter (rap) values are shown as a boxplot in figure 4.33.

Acoustic analysis: Median (IQR) values of DS & TD speakers from study 1			
Parameter	**DS (8)**	**TD (8)**	**statistical significance**
F0 mean	166.77 (107.39)	208.60 (100.00)	ns
F0 mean (stdev)	9.79 (11.92)	9.61 (5.96)	ns
jitter (rap)	1.01 (0.35)	0.83 (0.26)	n = 8 (DS & TD), $u = 13.5$, $p = 0.05$
jitter (ppq5)	1.03 (0.24)	0.91 (0.28)	ns
shimmer (apq3)	4.41 (1.46)	4.07 (1.98)	ns
shimmer (apq5)	5.71 (0.96)	6.02 (2.68)	ns
HNR	13.09 (2.25)	13.37 (2.42)	ns
SPT (1-5kHz)	17.11 (16.05)	12.98 (4.06)	ns
SPT (2-5kHz)	23.12 (5.10)	20.04 (5.28)	ns

Table 4.44: Median (IQR) values and statistical differences between DS and TD speakers from study 1 across all acoustic analysis parameters

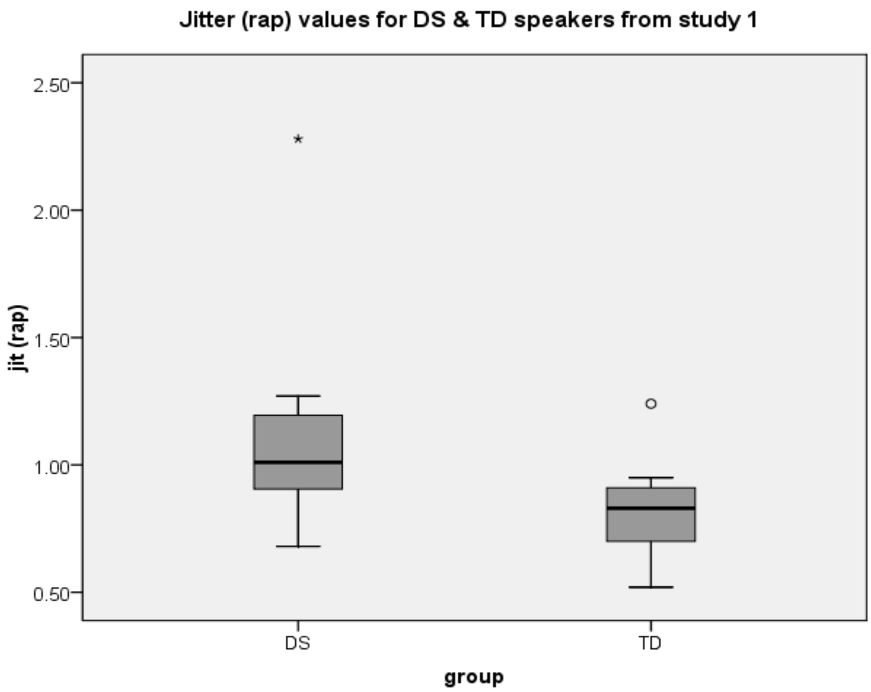


Figure 4.33: Boxplot of jitter (rap) values of the DS and TD speakers from study 1

4.3.7 Variability between individual DS and TD speakers presented in study 1

Median (IQR) values for each of the acoustic analysis parameters for the individual DS (8) and TD (8) speakers from study 1 are shown in table 4.45, and illustrated as boxplots in figures 4.34-4.42.

Acoustic analysis: Median (IQR) values of individual DS & TD speakers from study 1									
	F0 mean	**F0 (stdev)**	**jitter (rap)**	**jitter (ppq5)**	**shim' (apq3)**	**shim' (apq5)**	**HNR**	**SPT 1-5 kHz**	**SPT 2-5 kHz**
DS13	129.06 (5.89)	4.74 (3.22)	1.68 (2.12)	1.18 (1.52)	5.41 (3.61)	5.67 (2.37)	12.31 (6.30)	19.27 (6.33)	24.32 (4.90)
DS26	268.43 (39.62)	17.16 (28.64)	0.77 (0.58)	0.76 (0.60)	4.50 (2.62)	5.09 (2.97)	13.00 (5.50)	12.93 (7.57)	20.37 (7.40)
DS24	162.28 (13.22)	8.07 (5.48)	0.71 (0.96)	0.82 (0.87)	3.86 (1.61)	5.90 (2.70)	13.75 (4.20)	18.69 (4.45)	23.81 (6.12)
DS14	126.88 (23.98)	8.91 (6.99)	0.69 (0.67)	0.76 (0.62)	4.07 (1.61)	5.15 (1.61)	12.88 (5.90)	18.69 (6.77)	22.80 (5.60)
DS7	173.76 (16.21)	7.02 (4.80)	0.84 (0.84)	0.83 (0.83)	3.55 (1.91)	5.20 (2.90)	13.41 (4.90)	16.98 (7.45)	23.61 (6.01)
DS30F	199.59 (75.82)	12.35 (27.26)	1.20 (1.17)	1.02 (1.10)	4.68 (2.78)	5.48 (3.75)	9.75 (6.10)	13.19 (5.94)	18.04 (3.43)
DS6	116.16 (7.14)	5.56 (3.41)	0.66 (0.41)	0.68 (0.34)	2.66 (0.78)	3.90 (1.68)	14.02 (4.00)	17.58 (4.71)	24.28 (4.97)
DS8	247.63 (38.39)	13.43 (10.78)	0.68 (0.83)	0.69 (0.73)	4.20 (2.49)	4.87 (2.56)	13.05 (5.90)	10.82 (7.41)	16.57 (8.45)
TD2	221.53 (26.43)	10.45 (7.42)	0.84 (0.51)	0.94 (0.51)	4.51 (2.53)	6.20 (3.28)	13.57 (4.30)	15.21 (6.16)	24.15 (6.01)
TD1	207.93 (18.12)	5.56 (3.68)	0.69 (0.45)	0.78 (0.53)	4.06 (2.54)	6.27 (3.18)	12.45 (4.40)	12.60 (5.51)	19.63 (7.71)
TD5	237.69 (30.82)	13.66 (15.05)	0.59 (0.56)	0.64 (0.62)	3.71 (2.55)	5.05 (2.87)	14.89 (5.20)	9.47 (5.22)	12.22 (7.29)
TD4F	231.95 (22.66)	9.49 (5.65)	0.38 (0.37)	0.41 (0.36)	2.26 (1.11)	3.40 (1.57)	17.01 (3.90)	10.56 (7.50)	15.82 (5.89)
TD7	144.96 (12.99)	5.51 (3.71)	0.48 (0.52)	0.56 (0.56)	2.59 (1.47)	4.15 (2.43)	15.48 (4.30)	15.01 (6.60)	22.67 (4.16)
TD3	207.37 (16.45)	8.57 (5.35)	0.52 (0.70)	0.61 (0.76)	3.61 (2.02)	5.10 (3.36)	15.10 (5.80)	14.24 (4.17)	20.28 (5.04)
TD8	109.21 (4.64)	4.52 (3.65)	0.77 (0.69)	0.74 (0.60)	5.94 (3.77)	6.88 (3.27)	9.65 (4.30)	13.19 (3.87)	22.98 (5.18)
TD6	128.01 (8.35)	6.14 (7.00)	0.62 (0.60)	0.79 (0.68)	3.20 (2.07)	4.69 (2.14)	12.70 (4.20)	12.62 (4.34)	20.16 (7.32)

Table 4.45: Median (IQR) values for all acoustic analysis parameters for the individual DS and TD speakers from study 1

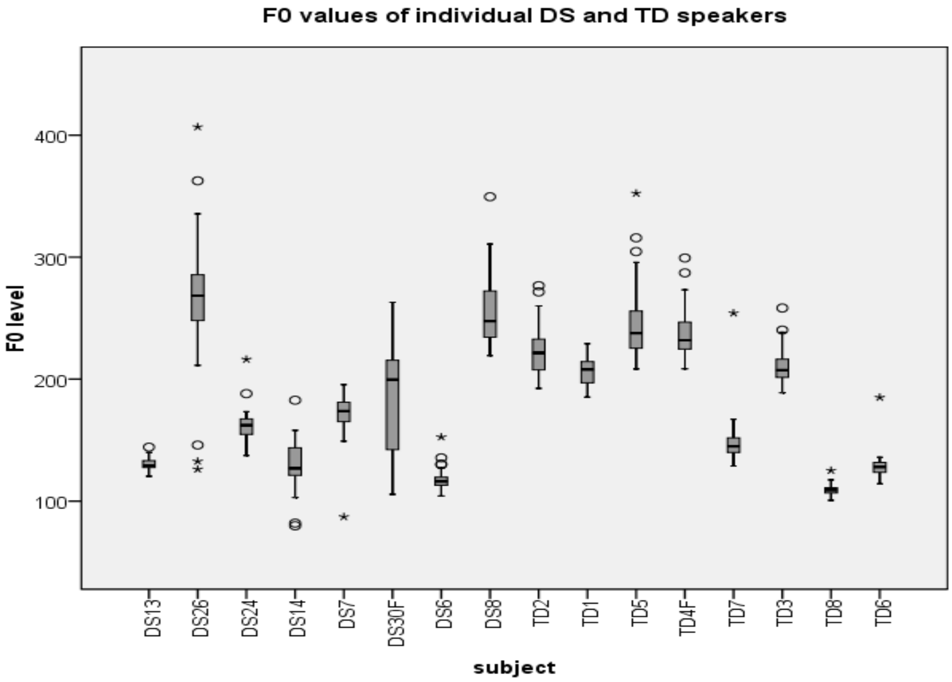


Figure 4.34: Boxplot showing F0 (mean) values of individual DS and TD speakers presented in study 1

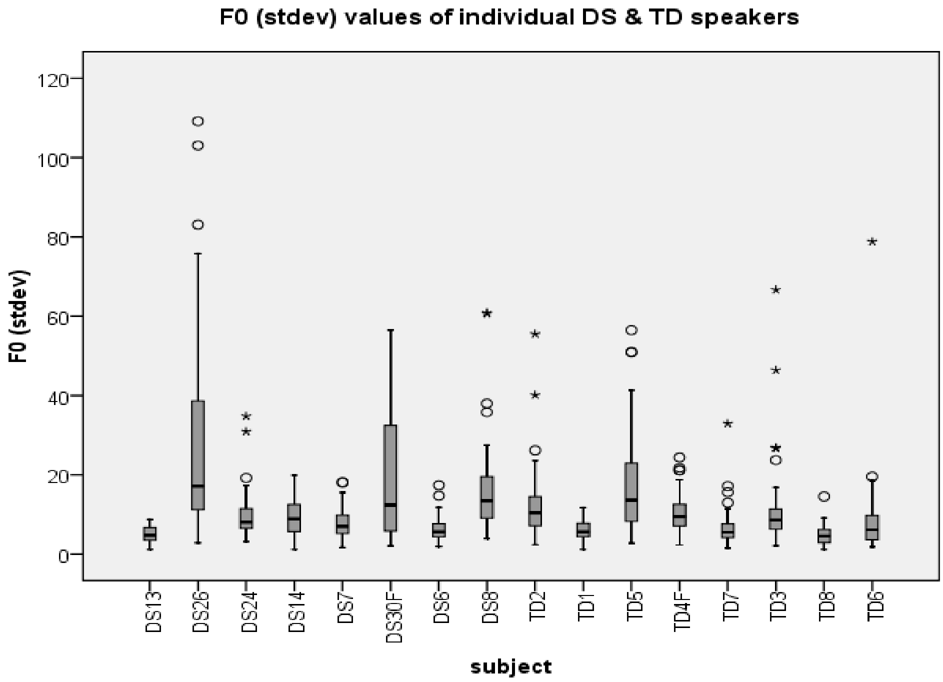


Figure 4.35: Boxplot showing F0 (stdev) values of individual DS and TD speakers presented in study 1

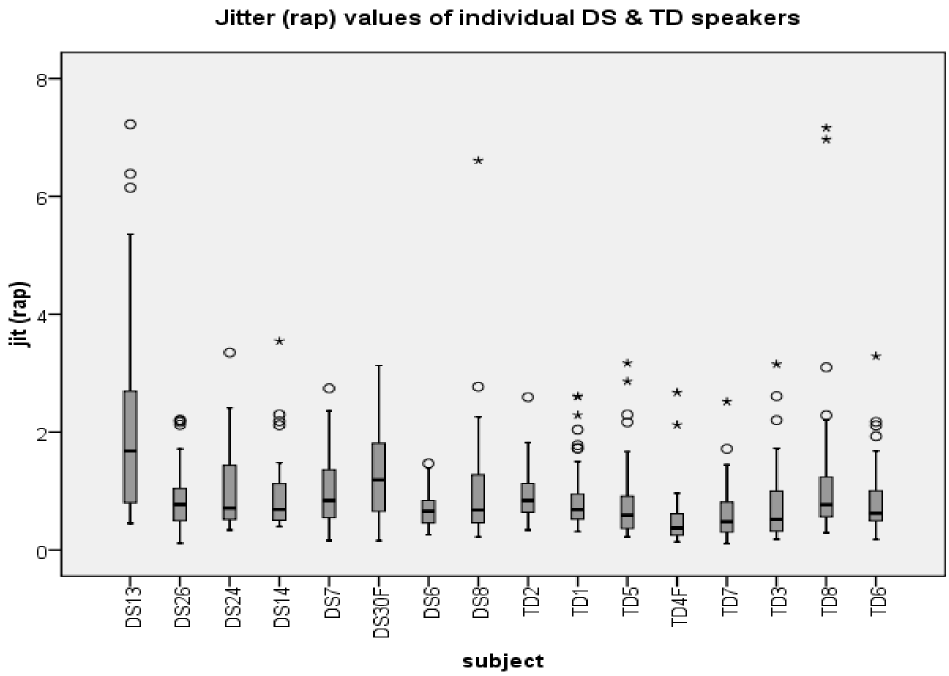


Figure 4.36: Boxplot showing jitter (rap) values of individual DS and TD speakers presented in study 1

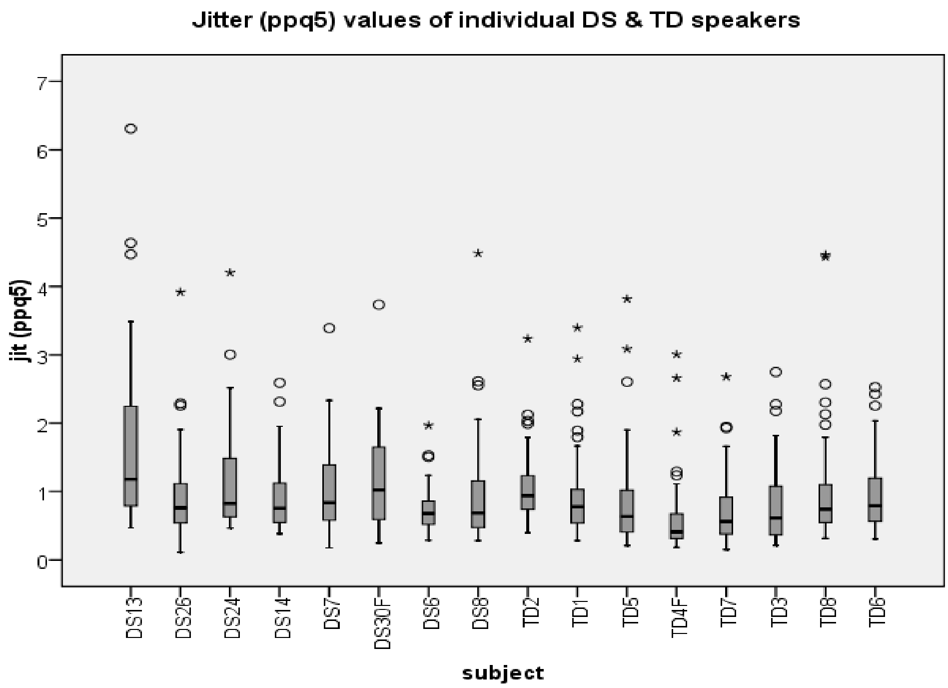


Figure 4.37: Boxplot showing jitter (ppq5) values of individual DS and TD speakers presented in study 1

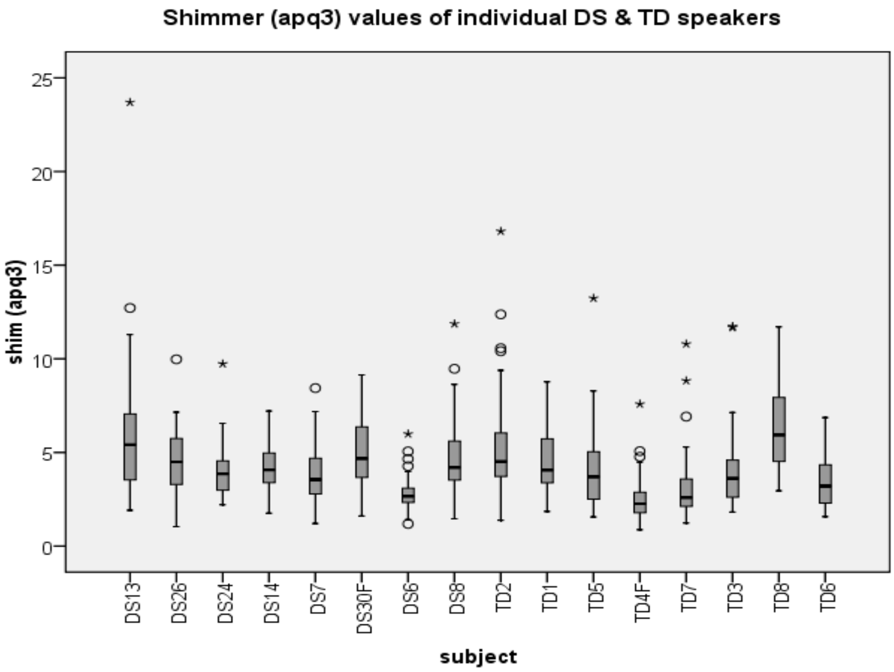


Figure 4.38: Boxplot showing shimmer (apq3) values of individual DS and TD speakers presented in study 1

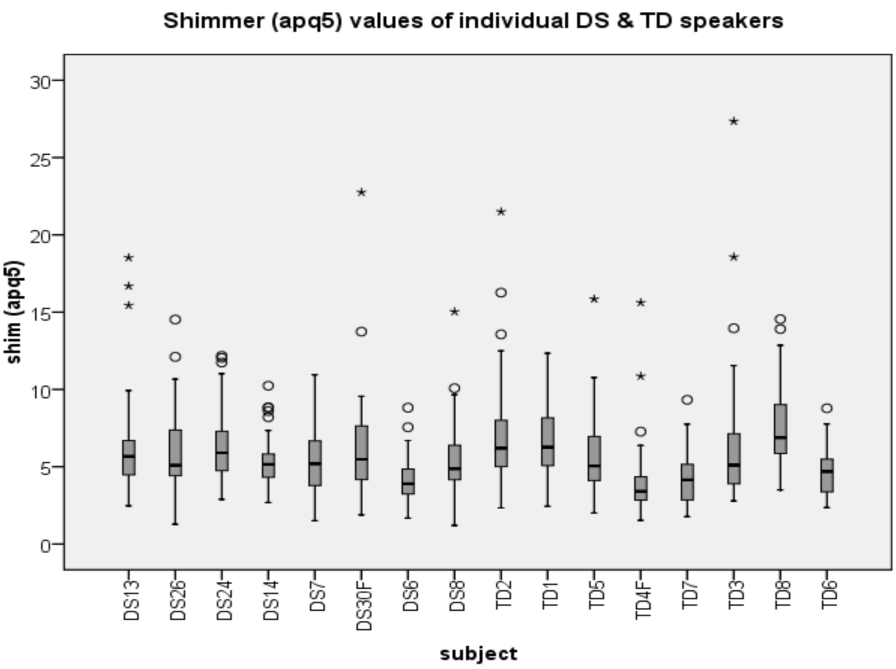


Figure 4.39: Boxplot showing shimmer (apq5) values of individual DS and TD speakers presented in study 1

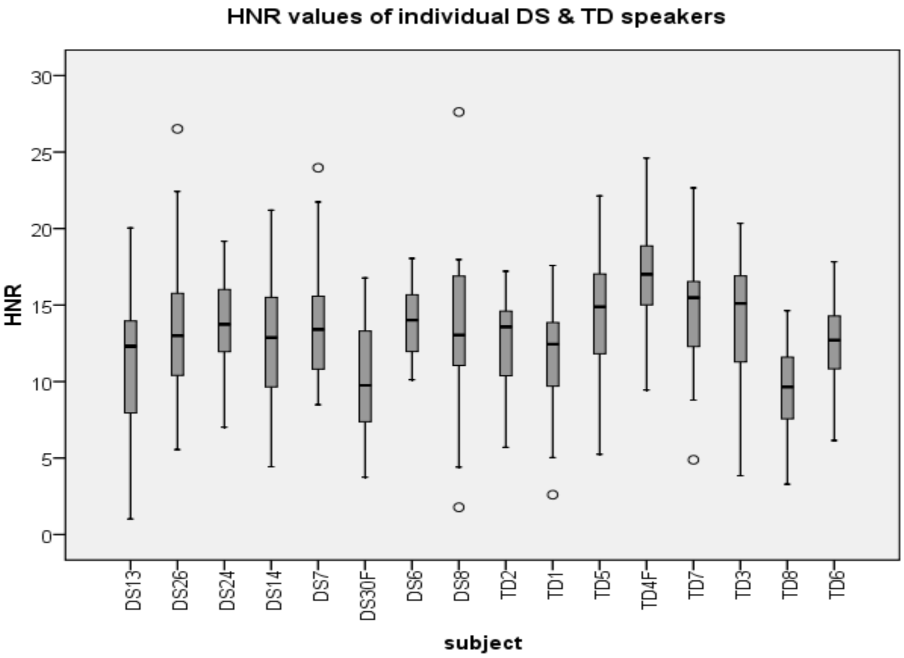


Figure 4.40: Boxplot showing HNR values of individual DS and TD speakers presented in study 1

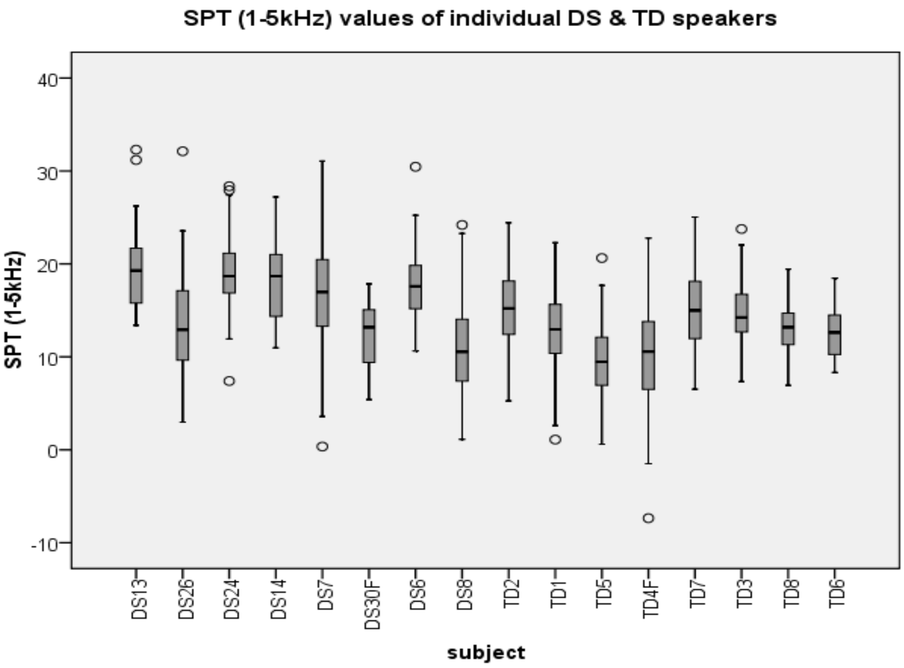


Figure 4.41: Boxplot showing SPT (1-5kHz) values of individual DS and TD speakers presented in study 1

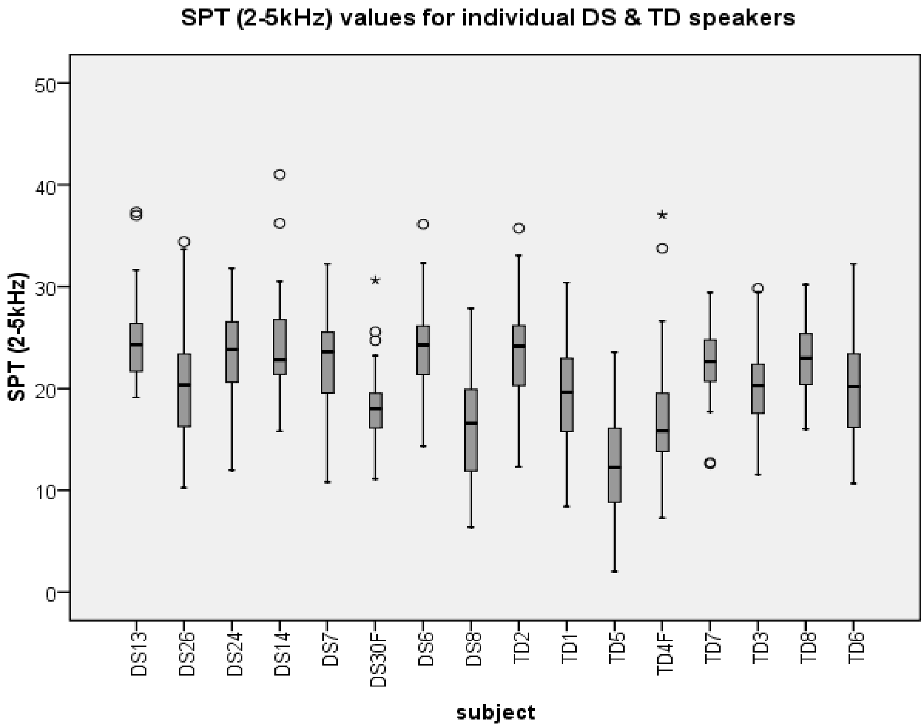


Figure 4.42: Boxplot showing SPT (2-5kHz) values of individual DS and TD speakers presented in study 1

4.4 Study 3: Perceptual Analysis of Voice

In this section the results of the VPAS ratings by both the trained speech and language therapist raters are presented. The same groups as analysed in the acoustic study are reported:

- Results between overall DS and TD groups
- Results between DS and TD male speakers
- Results between DS and TD female speakers
- Results between DS female and male speakers
- Results between TD female and male speakers
- Results between DS and TD speakers presented in study 1
- Variability between individual DS and TD speakers presented in study 1

Rater agreement and the spread of ratings for both groups are reported for each section of the VPAS for the overall DS and TD groups, and the results of statistical tests reported for all groups.

For all the statistically significant results the median and IQR values are given and non-significant results abbreviated as 'ns'. All significant results are illustrated as boxplots where neutral is represented by a short dashed line at zero, and long dashed lines at '3' and '-3' represent the boundaries of moderate presentation of features (i.e. a rating between '3' and '-3' is within the typical or moderate range whilst one above '3' or below '-3' indicates a rating within the severe or atypical range).

4.4.1 Results between overall DS and TD groups

4.4.1.1 Labial settings

The degree of consistency in labial judgements (percentage) between raters for the overall DS and TD groups are shown in table 4.46.

LABIAL Settings: Rater agreement %		
	DS (22)	**TD (52)**
rounding/protrusion	86.36	94.23
spreading	86.36	90.38
labiodentalisation	90.91	96.15
extensive range	95.45	100.00
minimised range	95.45	100.00

Table 4.46: Agreement in judgements (percentage) between raters for labial settings of overall DS and TD groups

Table 4.47 shows the spread of ratings in percentage form for both raters combined for spread to rounding/protrusion, labiodentalisation and minimised to excessive range for the DS and TD groups. Negative ratings indicate spread lip pattern and minimised range, a rating of zero shows a neutral setting, and positive ratings indicate rounding/protrusion, labiodentalisation and excessive range of lip movements.

LABIAL SETTINGS: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	spread							**rounding/protrusion**					
DS	0	0	0	0	6.8	5.7	59.1	6.8	19.3	2.3	0	0	0
TD	0	0	0	1.0	3.4	6.7	75.5	9.6	3.4	0.5	0	0	0
								labiodentalisation					
DS							84.6	13.5	1.0	1.0	0	0	0
TD							75.0	9.1	11.4	4.5	0	0	0
	minimised range							**extensive range**					
DS	0	0	2.3	12.5	12.5	9.1	62.5	0	1.1	0	0	0	0
TD	0	0	0	0	1.0	13.0	85.6	0.5	0	0	0	0	0

Table 4.47: Spread of ratings (percentage) by both raters combined for the labial settings of the overall DS and TD groups

Results of Mann Whitney statistical tests are shown in table 4.48, alongside the median and IQR of ratings for both the DS and TD groups. The DS group were found to have a significantly greater degree of rounding/protrusion and a more minimised range than the TD group (illustrated as boxplots in figures 4.43 and 4.44). No significant difference was found between the two for ‘labiodentalisation’ ratings.

LABIAL SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (22)	**TD (52)**	**statistical significance**
(-) spreading - rounding/protrusion (+)	1 (3)	0 (1)	n = 44 (DS), 104 (TD) *U* = 1640.5 p = 0.005
labiodentalisation	0 (1)	0 (0)	ns
(-) minimised – extensive (+) range	-2 (3)	0 (1)	n = 44 (DS), 104 (TD) *U* = 965.0 p < 0.001

Table 4.48: Results of Mann Whitney U-tests and median (IQR) values for labial settings of overall DS and TD groups

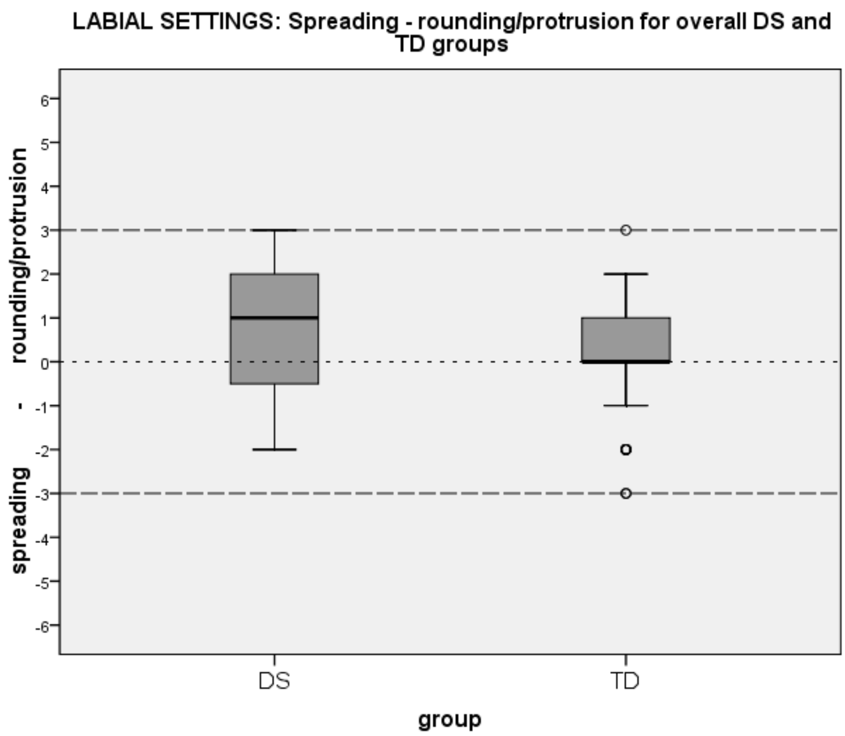


Figure 4.43: Boxplot of lip spreading – rounding/protrusion values of overall DS and TD groups

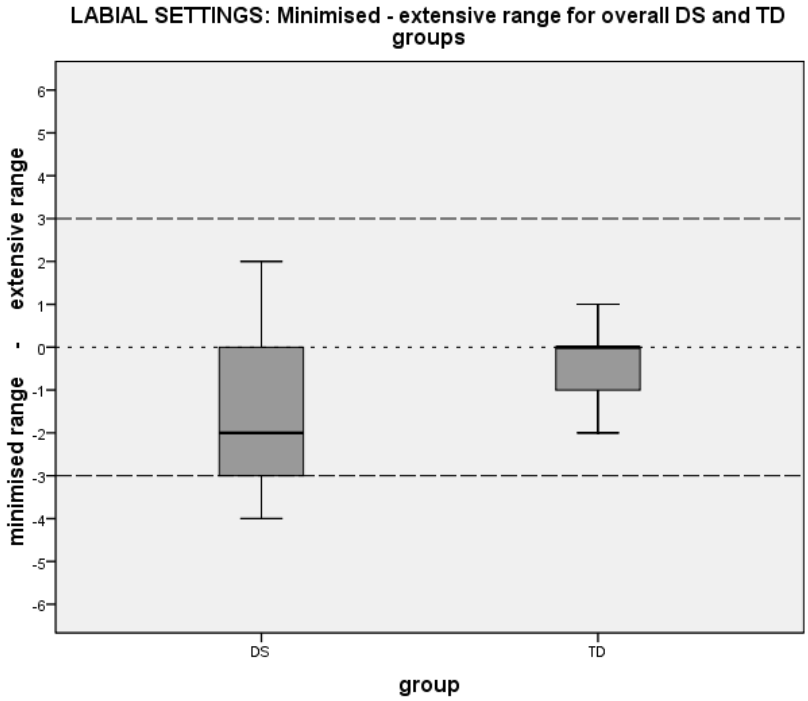


Figure 4.44: Boxplot of minimised – extensive lip range values of overall DS and TD groups

4.4.1.2 Mandibular settings

Table 4.49 shows the percentage of agreement between the VPAS ratings of both raters for the mandibular settings of the DS and TD groups and table 4.50 indicates the spread of ratings (in percentage form).

MANDIBULAR SETTINGS: Rater agreement (%)		
	DS (21)	**TD (52)**
close jaw	100.00	100.00
open jaw	90.48	100.00
protruded jaw	90.48	100.00
extended range	100.00	100.00
minimised range	90.48	100.00

Table 4.49: Agreement in judgements (percentage) between raters for mandibular settings of overall DS and TD groups

MANDIBULAR SETTINGS: Spread of ratings (%) for overall DS (21) and TD (52) groups													
code	-6	-5	-4	-3	-2	-1	0	1	2	3	4	5	6
	open jaw							close jaw					
DS	0	0	0	0	21.4	38.1	35.7	4.8	0				
TD	0	0	0	0	0	29.8	70.2	0	0				
								protruded jaw					
DS							11.9	16.7	26.2	33.3	11.9	0	0
TD							99.0	1.0	0	0	0	0	0
	minimised range							extensive range					
DS	0	0	0	0	7.14	31.0	59.5	2.4	0	0	0	0	0
TD	0	0	0	0	0	8.7	91.3	0	0	0	0	0	0

Table 4.50: Spread of ratings (percentage) by both raters combined for the mandibular settings of the overall DS and TD groups

The results of Mann Whitney U-tests are shown in table 4.51 with median (IQR) values. The overall DS group were found to have significantly greater presentation of open jaw and protruded jaw and a more minimised jaw range than the overall TD group. Values are illustrated as boxplots in figures 4.45-4.47.

MANDIBULAR SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (21)	TD (52)	statistical significance
(-) open - close (+) jaw	-1 (1)	0 (1)	n = 42 (DS), 104 (TD) U = 724.5 $p < 0.001$
protruded jaw	2 (2)	0 (0)	n = 42 (DS), 104 (TD) U = 272.0 $p < 0.001$
(-) minimised - extensive (+) range	0 (1)	0 (0)	n = 42 (DS), 104 (TD) U = 1575.0 $p < 0.001$

Table 4.51: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of overall DS and TD groups

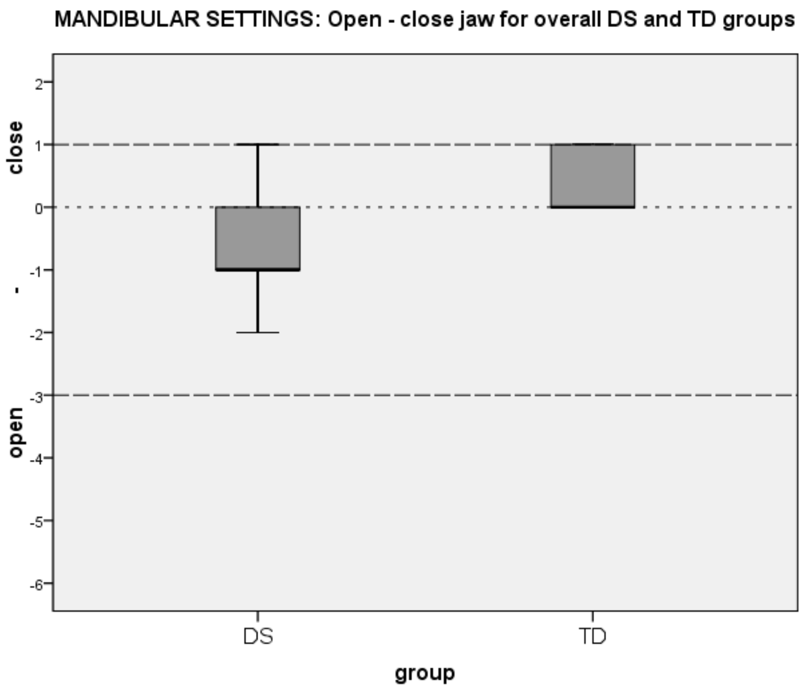


Figure 4.45: Boxplot of open – close jaw values of overall DS and TD groups

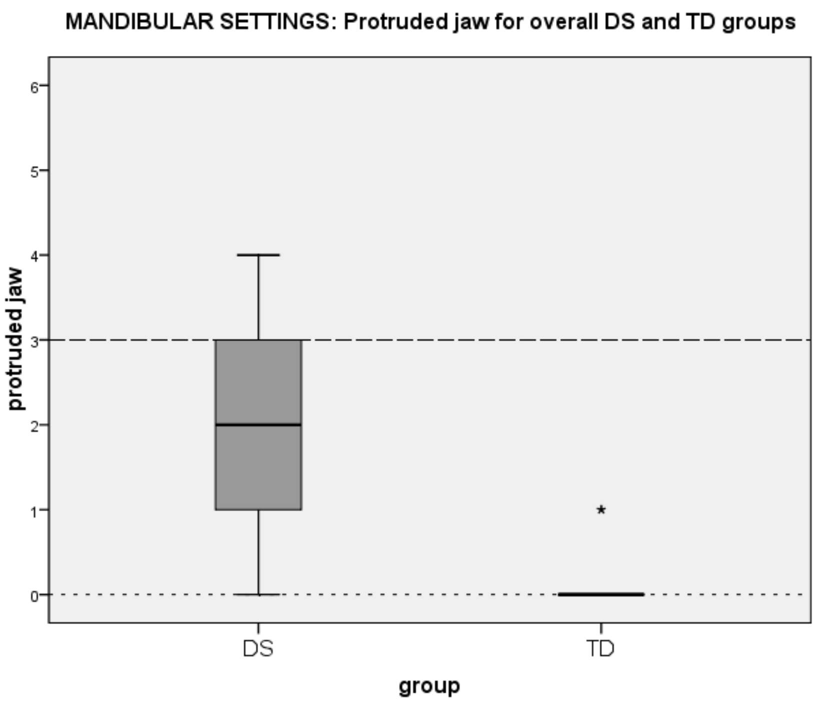


Figure 4.46: Boxplot of protruded jaw values of overall DS and TD groups

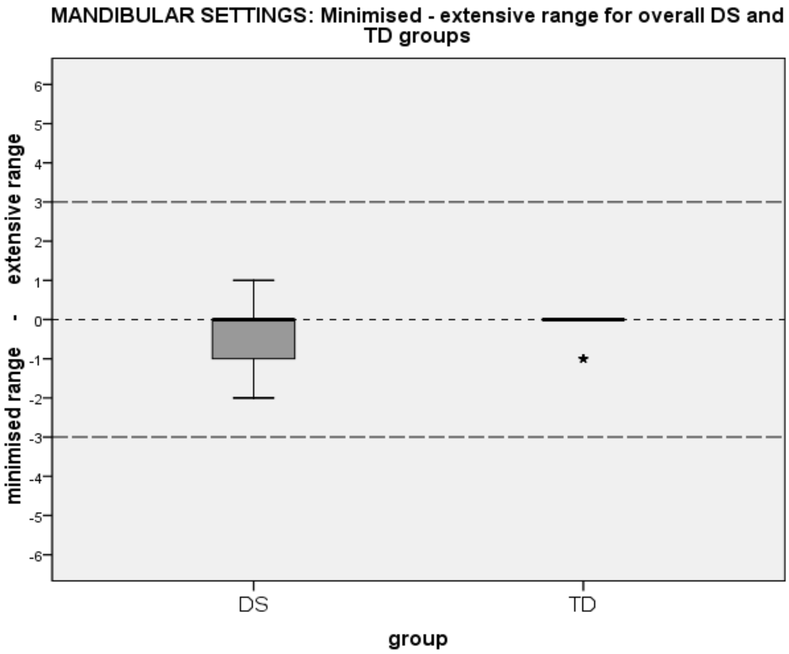


Figure 4.47: Boxplot of minimised – extensive jaw range values of overall DS and TD groups

4.4.1.3 Lingual settings

Rater agreement for lingual VPAS settings between the two raters are shown in percentage form in table 4.52.

LINGUAL SETTINGS: Rater agreement (%)		
	DS (21)	**TD (52)**
tip/blade advanced	100.00	92.31
tip/blade retracted	100.00	96.15
body fronted	95.24	92.31
body backed	100.00	96.15
body raised	85.71	92.31
body lowered	100.00	100.00
extensive range	100.00	100.00
minimised range	100.00	100.00

Table 4.52: Agreement in judgements (percentage) between raters for lingual settings of overall DS and TD groups

The spread of lingual ratings for the overall DS and TD groups are shown in table 4.53. Statistically significant differences between the two groups in Mann Whitney U-tests are reported in table 4.54 and show a significantly more advanced tongue tip, fronted tongue body, raised tongue body and minimised range in the DS group. The median (IQR) values are illustrated in figures 4.48-4.51.

LINGUAL SETTINGS: Spread of ratings (%) for overall DS (21) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	retracted tip							**advanced tip**					
DS	0	0	0	0	0	2.4	2.4	14.3	16.7	35.7	26.2	2.4	0
TD	0	0	0	0	2.9	8.7	36.5	34.6	14.4	2.9	0	0	0
	backed body							**fronted body**					
DS	0	0	0	2.4	11.9	9.5	7.1	2.4	35.7	31.0	0	0	0
TD	0	0	0	0	7.7	28.8	36.5	16.3	9.6	1.0	0	0	0
	lowered body							**raised body**					
DS	0	0	0	0	0	0	19.0	26.2	42.9	9.5	2.4	0	0
TD	0	0	0	0	0	1.9	41.3	45.2	11.5	0	0	0	0
	minimised range							**extensive range**					
DS	0	2.3	25.0	29.5	29.5	4.5	9.1	0	0	0	0	0	0
TD	0	0	0	0	1.0	9.6	89.4	0	0	0	0	0	0

Table 4.53: Spread of ratings (percentage) by both raters combined for the lingual settings of the overall DS and TD groups

LINGUAL SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (21)	TD (52)	statistical significance
(-) retracted – advanced (+) tip	3 (2)	1 (1)	n = 42 (DS), 104 (TD) $U = 481.5$ $p < 0.001$
(-) backed – fronted (+) body	2 (3)	0 (2)	n = 42 (DS), 104 (TD) $U = 1185.0$ $p < 0.001$
(-) lowered - raised (+) body	2 (1)	1 (1)	n = 42 (DS), 104 (TD) $U = 1142.5$ $p < 0.001$
(-) minimised – extensive (+) range	-3 (2)	0 (0)	n = 42 (DS), 104 (TD) $U = 248.5$ $p < 0.001$

Table 4.54: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of overall DS and TD group

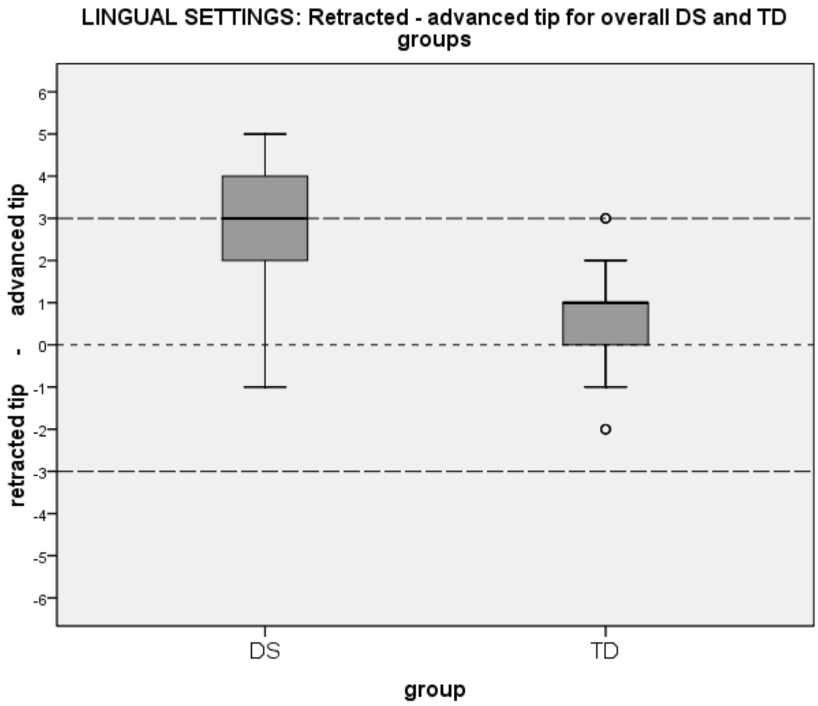


Figure 4.48: Boxplot of retracted – advanced tongue tip values of overall DS and TD groups

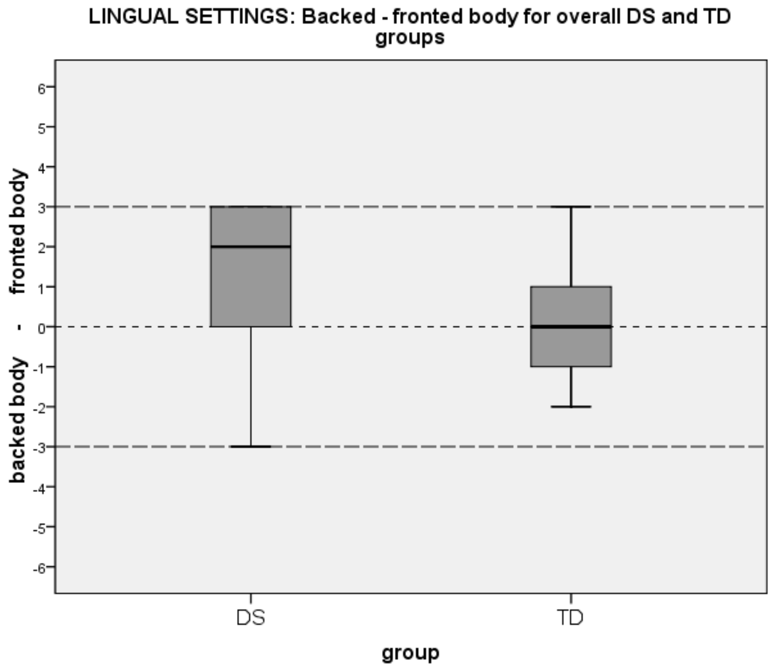


Figure 4.49: Boxplot of backed – fronted tongue body values of overall DS and TD group

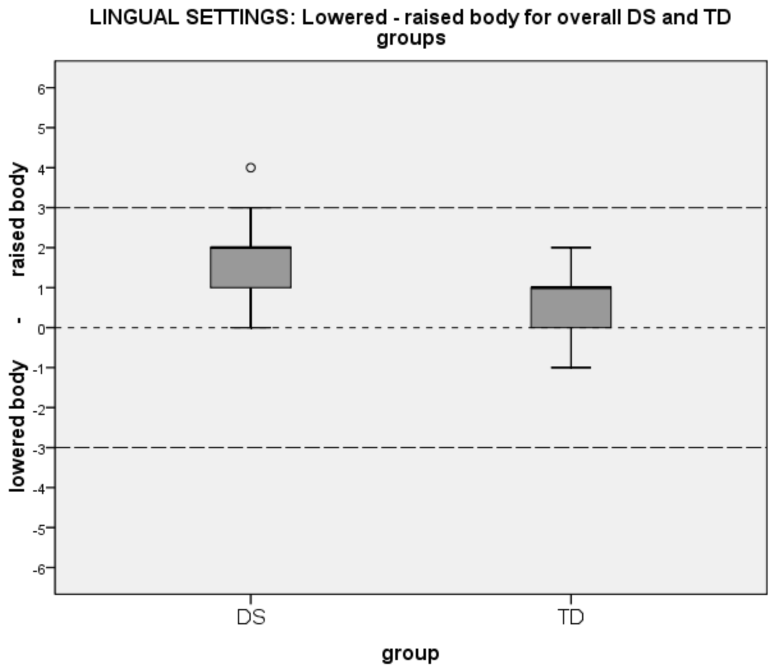


Figure 4.50: Boxplot of lowered – raised tongue body values of overall DS and TD groups

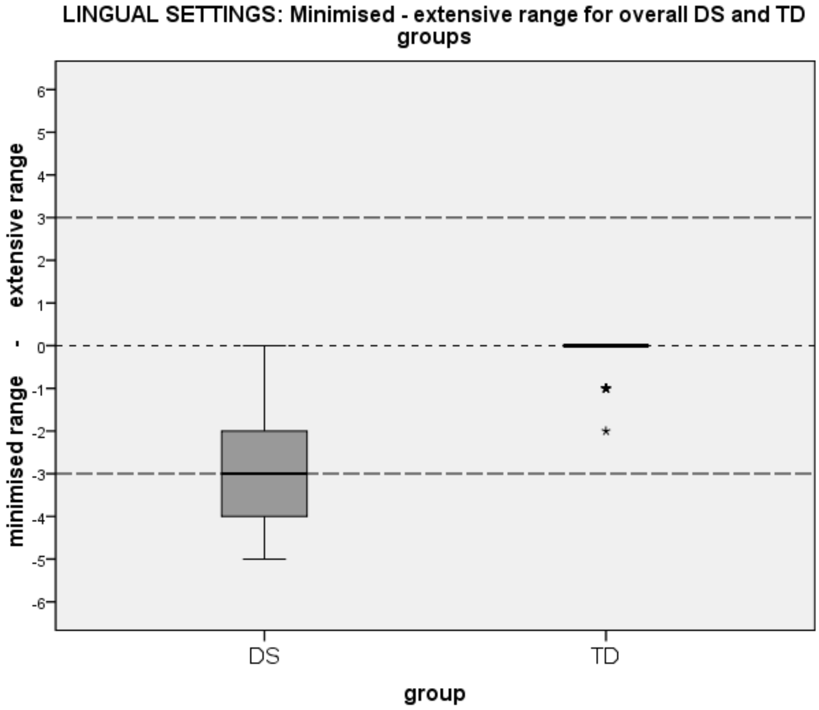


Figure 4.51: Boxplot of minimised – extensive tongue range values of overall DS and TD groups

4.4.1.4 Pharyngeal settings

Rater agreement (percentage) for the pharyngeal settings of the VPAS are shown in table 4.55 and the spread of ratings for pharyngeal constriction – expansion are reported in table 4.56.

PHARYNGEAL SETTINGS: Rater agreement (%)		
	DS (22)	**TD (52)**
pharyngeal constriction	86.36	100.00
pharyngeal expansion	100.00	100.00

Table 4.55: Agreement in judgements (percentage) between raters for pharyngeal settings of overall DS and TD groups

PHARYNGEAL SETTINGS: Spread of ratings (%) for overall DS (22) and TD (22) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	constriction							**expansion**					
DS	0	0	2.3	6.8	20.5	47.7	22.7	0	0	0	0	0	0
TD	0	0	2.3	6.8	20.5	47.7	22.7	0	0	0	0	0	0

Table 4.56: Spread of ratings (percentage) by both raters combined for the pharyngeal settings of the overall DS and TD groups

Table 4.57 reports the results of statistical testing between groups using a Mann Whitney U-test, where the DS group have been found to have significantly greater pharyngeal constriction than the TD group. Median (IQR) values are shown in figure 4.52.

PHARYNGEAL SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (22)	**TD (52)**	**statistical significance**
(-) constriction – expansion (+)	-1 (1)	0 (0)	n = 44 (DS), 104 (TD) $U = 701.0$ $p < 0.001$

Table 4.57: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of overall DS and TD groups

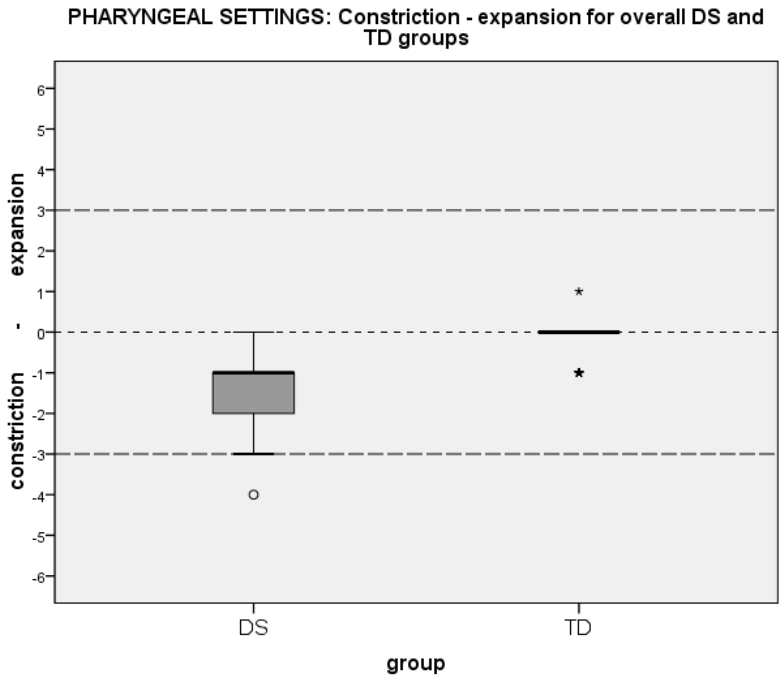


Figure 4.52: Boxplot of pharyngeal constriction – expansion values of overall DS and TD groups

4.4.1.5 Velopharyngeal settings

The level of rater agreement (percentage) is shown for the velopharyngeal settings in table 4.58. Table 4.59 reports the spread of ratings for audible nasal escape and denasal – nasal features.

VELOPHARYNGEAL SETTINGS: Rater agreement (%)		
	DS (22)	**TD (52)**
audible nasal escape	100.00	100.00
nasal	95.45	96.15
denasal	100.00	100.00

Table 4.58: Agreement in judgements (percentage) between raters for velopharyngeal settings of overall DS and TD groups

VELOPHARYNGEAL SETTINGS: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
											audible nasal escape		
DS							72.7				25.0	2.3	0
TD							100				0	0	0
	denasal							**nasal**					
DS	0	0	0	0	2.3	4.5	2.3	2.3	9.1	36.4	29.5	13.6	0
TD	0	0	0	0	0	0.0	3.8	30.8	43.3	22.1	0	0	0

Table 4.59: Spread of ratings (percentage) by both raters combined for the velopharyngeal settings of the overall DS and TD groups

Mann Whitney U-test results are reported in table 4.60 and show a statistically greater degree of audible nasal escape and increased nasality in the DS group in comparison to their TD peers. The median (IQR) values are shown as boxplots in figures 4.53 and 4.54.

VELOPHARYNGEAL SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (22)	**TD (52)**	**statistical significance**
audible nasal escape	0 (1)	0 (0)	n = 44 (DS), 104 (TD) U = 1664.0 $p < 0.001$
(-) denasal – nasal (+)	3 (1)	2 (1)	n = 44 (DS), 104 (TD) U = 864.0 $p < 0.001$

Table 4.60: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of overall DS and TD groups

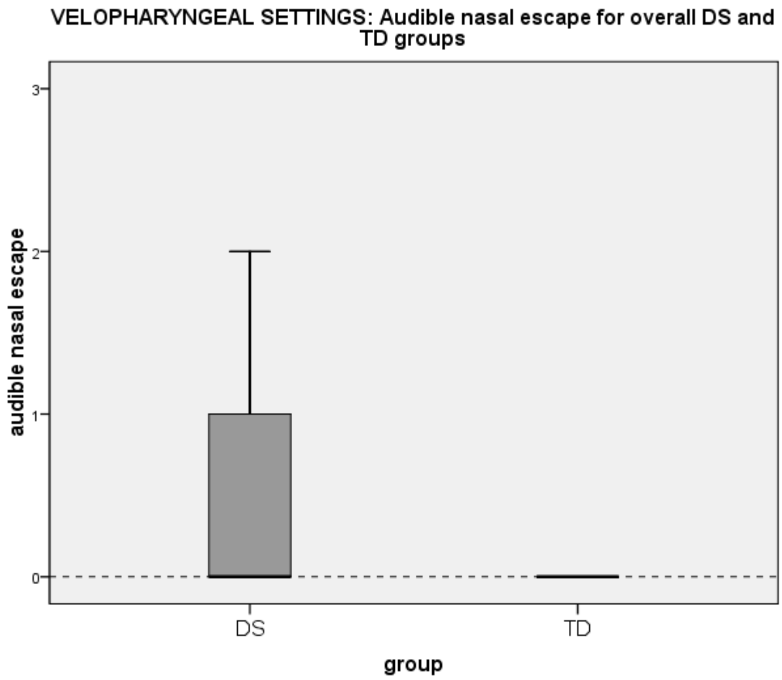


Figure 4.53: Boxplot of audible nasal escape values of overall DS and TD groups

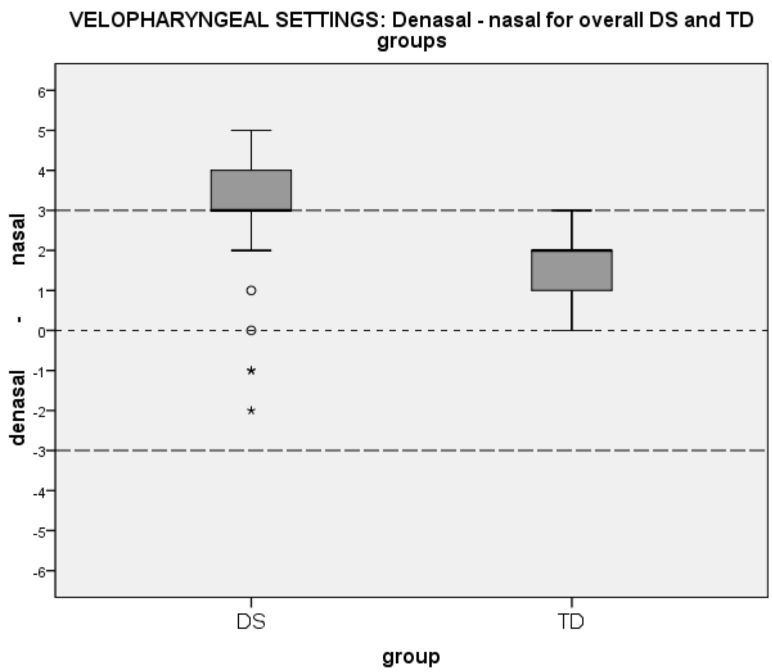


Figure 4.54: Boxplot of denasal – nasal values of overall DS and TD groups

4.4.1.6 Larynx height settings

The level of rater agreement (percentage) is shown in table 4.61 and the spread of ratings for lowered – raised larynx shown in table 4.62. No significant difference was found between the DS and TD groups in this parameter (table 4.63).

LARYNX HEIGHT SETTINGS: Rater agreement (%)		
	DS (22)	**TD (52)**
Raised larynx	95.45	98.08
Lowered larynx	100.00	98.08

Table 4.61: Agreement in judgements (percentage) between raters for larynx height settings of overall DS and TD groups

LARYNX HEIGHT SETTINGS: Spread of ratings (%) for overall DS (22) and TD (22) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	lowered larynx							**raised larynx**					
DS	0	0	0	4.5	22.7	9.1	34.1	9.1	9.1	11.4	0	0	0
TD	0	0	0	2.9	2.9	19.2	53.8	18.3	2.9	0	0	0	0

Table 4.62: Spread of ratings (percentage) by both raters combined for the larynx height settings of the overall DS and TD groups

LARYNX HEIGHT SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (22)	**TD (52)**	**statistical significance**
(-) lowered – raised (+) larynx	0 (3)	0 (1)	ns

Table 4.63: Results of Mann Whitney U-tests and median (IQR) values for larynx height settings of overall DS and TD groups

4.4.1.7 Tension settings

Rater agreement (percentage) is shown in table 4.64 and the spread of ratings for lax – tense vocal tract and lax – tense larynx are shown in table 4.65.

MUSCULAR TENSION SETTINGS: Rater agreement (%)		
	DS (22)	**TD (52)**
tense vocal tract	100.00	100.00
lax vocal tract	95.45	100.00
tense larynx	90.91	94.23
lax larynx	95.45	100.00

Table 4.64: Agreement in judgements (percentage) between raters for muscular tension settings of overall DS and TD groups

MUSCULAR TENSION SETTINGS: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	lax vocal tract							**tense vocal tract**					
DS	0	0	0	0	13.6	20.5	50.0	15.9	0	0	0	0	0
TD	0	0	0	0	1.0	1.9	92.3	4.8	0	0	0	0	0
	lax larynx							**tense larynx**					
DS	0	0	0	0	2.3	2.3	29.5	15.9	27.3	15.9	2.3	4.5	0
TD	0	0	0	0	0	0	34.6	45.2	13.5	6.7	0	0	0

Table 4.65: Spread of ratings (percentage) by both raters combined for the muscular tension settings of the overall DS and TD groups

Table 4.66 shows the results of statistical testing by Mann Whitney U-tests showing that the DS group have been rated as having a significantly more lax vocal tract and a significantly more tense larynx than the TD group (values are illustrated as boxplots in figures 4.55 and 4.56).

MUSCULAR TENSION SETTINGS: Mann Whitney results for overall DS and TD groups			
	DS (22)	**TD (52)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (1)	0 (0)	n = 44 (DS), 104 (TD) U = 1853.5 p = 0.009
(-) lax – tense (+) larynx	1.5 (2)	1 (1)	n = 44 (DS), 104 (TD) U = 183.0 p = 0.044

Table 4.66: Results of Mann Whitney U-tests and median (IQR) values for muscular tension settings of overall DS and TD groups

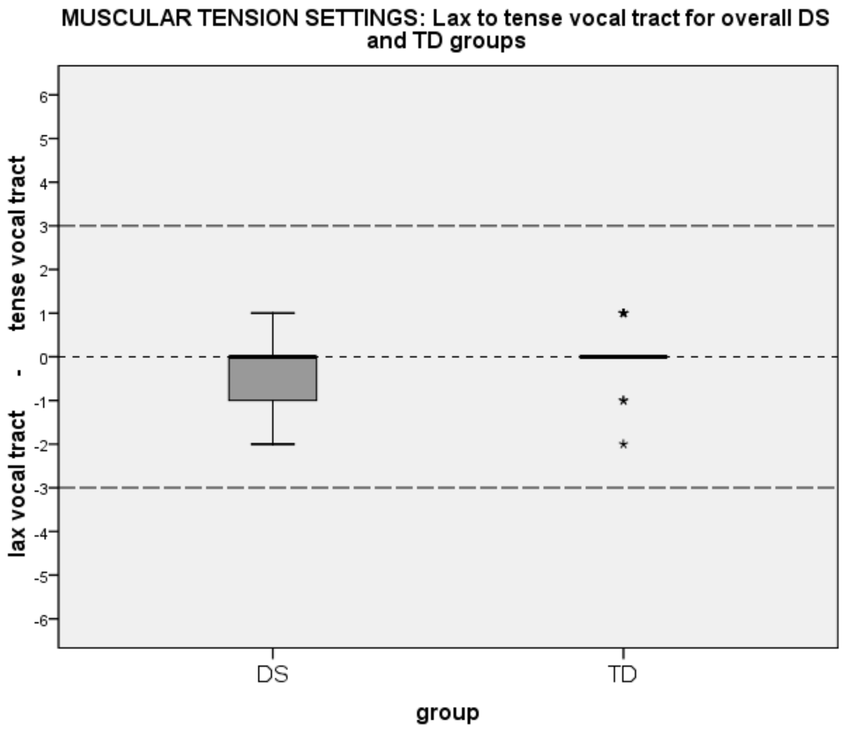


Figure 4.55: Boxplot of lax – tense vocal tract values of overall DS and TD groups

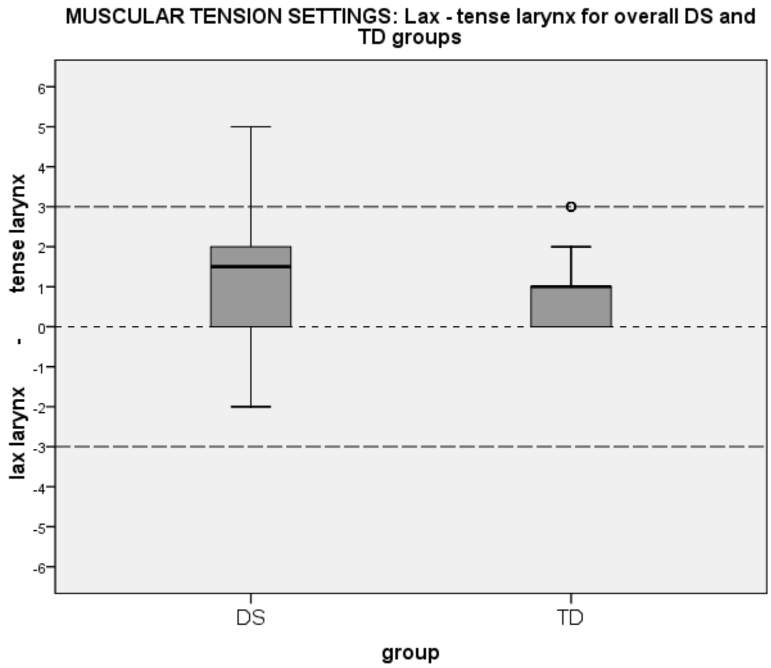


Figure 4.56: Boxplot of lax – tense larynx values of overall DS and TD groups

4.4.1.8 Phonation features

Rater agreement (percentage) for the phonation features of the VPAS is shown in table 4.67 and the spread of ratings in table 4.68.

PHONATION FEATURES: Rater agreement (%)		
	DS (22)	**TD (52)**
non neutral - neutral voice	100.00	100.00
falsetto	100.00	100.00
creak	100.00	100.00
creaky	100.00	100.00
whisper	100.00	100.00
whispery	100.00	98.08
harsh	90.91	96.15
tremor	100.00	99.04

Table 4.67: Agreement in judgements (percentage) between raters for phonation features of overall DS and TD group

PHONATION FEATURES: Spread of ratings (%) for overall DS (22) and TD (52) groups							
code	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	neutral voice	**non-neutral voice**					
DS	0	100					
TD	0	100					
	falsetto absent	**falsetto present**					
DS	100	0					
TD	100	0					
	creak absent	**creak present**					
DS	100	0					
TD	100	0					
	creaky						
DS	88.6	2.3	4.5	4.5	0	0	0
TD	72.1	8.7	10.6	7.7	1.0	0	0
	whisper absent	**whisper present**					
DS	100	0					
TD	100	0					
	whispery						
DS	2.3	6.8	29.5	45.5	15.9	0	0
TD	0	13.5	53.8	30.8	1.9	0	0
	harsh						
DS	25.0	45.5	20.5	0.0	9.1	0	0
TD	47.1	36.5	8.7	7.7	0	0	0
	tremor						
DS	100	0	0	0	0	0	0
TD	99.0	0	1.0	0	0	0	0

Table 4.68: Spread of ratings (percentage) by both raters combined for the phonation features of the overall DS and TD groups

Table 4.69 shows the results of statistical tests (Mann Whitney and Chi square) for phonation features. The DS group were found to have significantly more whispery and harsh phonation and to have significantly less creaky phonation than the TD group. All other features were non-significant. Significant results are shown as boxplots in figures 4.57-4.59.

PHONATION FEATURES: Mann Whitney and Chi Square* results for overall DS and TD groups				
		DS (22)	TD (52)	statistical significance
voicing type	neutral - non neutral voice	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (0)	0 (1)	n = 44 (DS), 104 (TD) *U* = 1919.0 p = 0.035
laryngeal frication	whisper	n/a	n/a	ns*
	whispery	3 (1)	2 (1)	n = 44 (DS), 104 (TD) *U* = 1568.0 p = 0.001
laryngeal irregularity	harsh	1 (2)	1 (1)	n = 44 (DS), 104 (TD) *U* = 1707.0 p = 0.009
	tremor	0 (0)	0 (0)	ns

Table 4.69: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of overall DS and TD groups

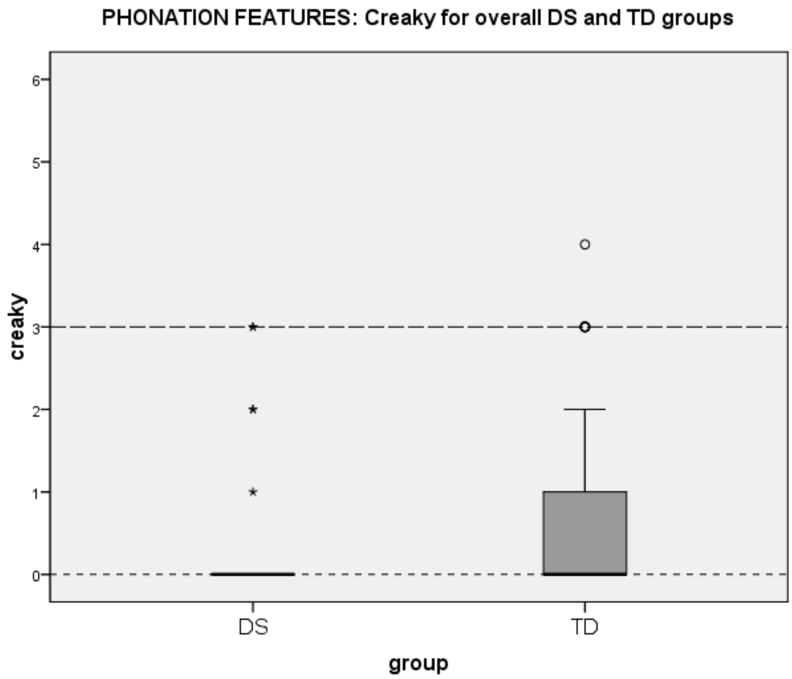


Figure 4.57: Boxplot of creaky phonation values of overall DS and TD groups

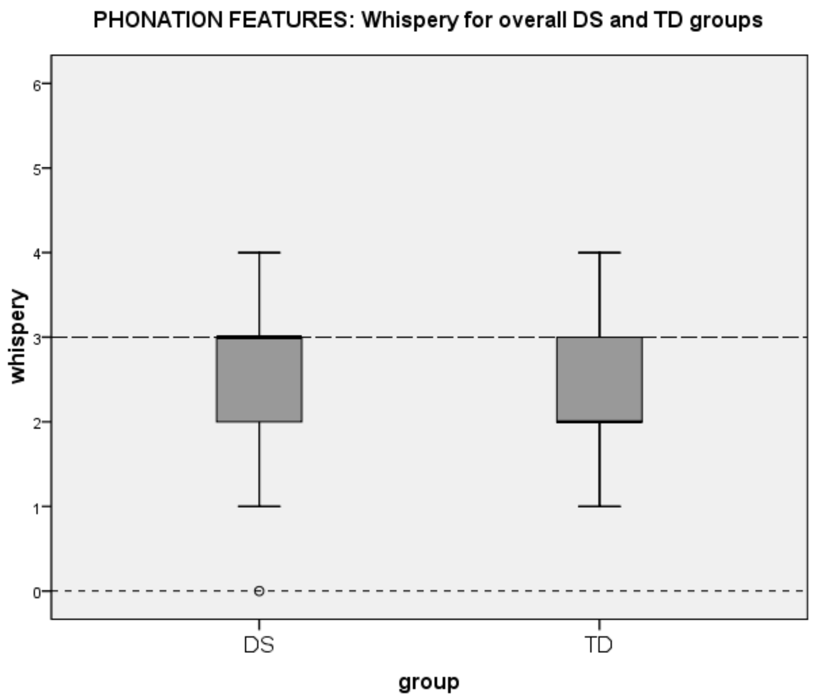


Figure 4.58: Boxplot of whispery phonation values of overall DS and TD groups

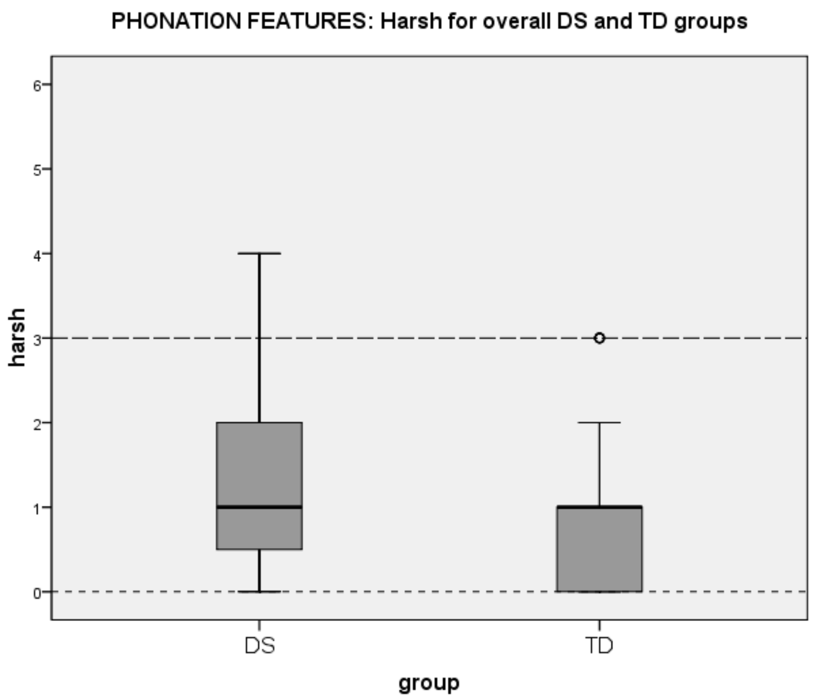


Figure 4.59: Boxplot of harsh phonation values of overall DS and TD groups

4.4.1.9 Pitch

Table 4.70 shows the rater agreement for the pitch section of the VPAS. The spread of ratings are given in table 4.71.

PITCH: Rater agreement (%)		
	DS (22)	**TD (52)**
high mean	95.24	96.15
low mean	90.48	100.00
extensive range	100.00	100.00
minimised range	100.00	96.15
high variability	100.00	100.00
low variability	100.00	94.23

Table 4.70: Agreement in judgements (percentage) between raters for pitch features of overall DS and TD groups

PITCH: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
	low mean							**high mean**					
DS	0	0	2.4	9.5	31.0	21.4	9.5	0	16.7	4.8	4.8	0	0
TD	0	0	0	2.9	4.8	13.5	57.7	15.4	4.8	1.0	0	0	0
	minimised range							**extensive range**					
DS	0	0	4.5	18.2	9.1	18.2	25.0	9.1	9.1	6.8	0	0	0
TD	0	1.0	1.9	3.8	2.9	11.5	76.0	2.9	0	0	0	0	0
	low variability							**high variability**					
DS	0	0	4.5	6.8	11.4	18.2	38.6	4.5	11.4	4.5	0	0	0
TD	0	0	1.0	2.9	6.7	11.5	75.0	2.9	0	0	0	0	0

Table 4.71: Spread of ratings (percentage) by both raters combined for pitch features of the overall DS and TD groups

The results of Mann Whitney U-tests for pitch are shown in table 4.72, with the DS group being found to have significantly lower mean pitch levels than their TD peers (illustrated in boxplot form in figure 4.60). No significant difference was found between pitch range or variability.

PITCH: Mann Whitney results for overall DS and TD groups			
	DS (22)	TD (52)	statistical significance
(-) low – high (+) mean	-1 (4)	0 (0)	n = 44 (DS), 104 (TD) *U* = 1539.0 p = 0.003
(-) minimised - extensive (+) range	-1 (2)	0 (0)	ns
(-) low - high (+) variability	0 (1)	0 (0)	ns

Table 4.72: Results of Mann Whitney U-tests and median (IQR) values for pitch features of overall DS and TD groups

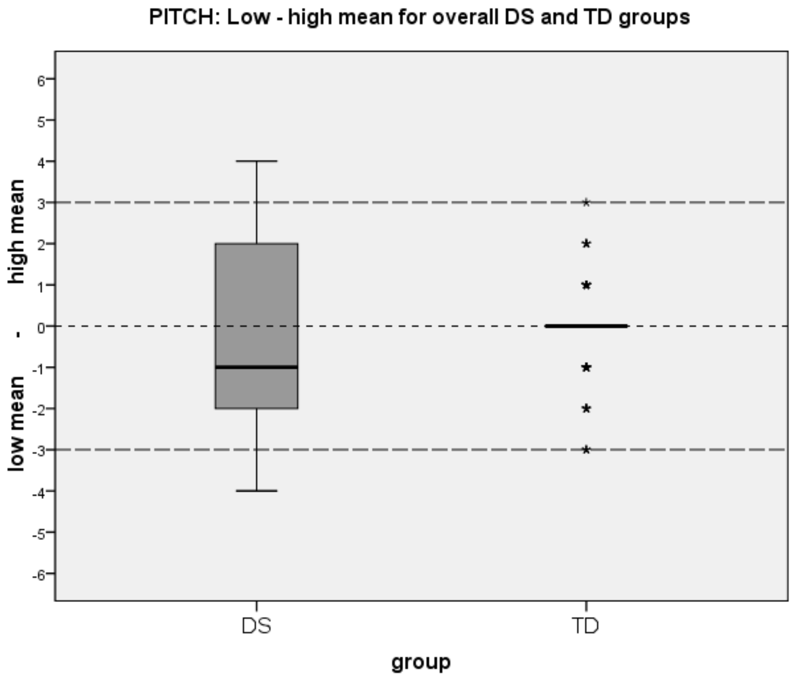


Figure 4.60: Boxplot of low – high mean pitch values of overall DS and TD groups

4.4.1.10 Loudness

The levels of rater agreement (percentage) for loudness ratings are shown in table 4.73. Table 4.73 shows the spread of loudness ratings.

LOUDNESS: Rater agreement (%)		
	DS (22)	TD (52)
high mean	100.00	100.00
low mean	100.00	95.15
extensive range	100.00	100.00
minimised range	95.45	94.23
high variability	100.00	100.00
low variability	95.45	94.23

Table 4.73: Agreement in judgements (percentage) between raters for loudness features of overall DS and TD groups

LOUDNESS: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	-6	-5	-4	-3	-2	-1	0	1	2	3	4	5	6
	low mean							high mean					
DS	0	0	0	0	25.0	11.4	47.7	9.1	6.8	0	0	0	0
TD	0	0	0	0	2.9	3.8	93.3	0	0	0	0	0	0
	minimised range							extensive range					
DS	0	0	0	2.3	13.6	13.6	56.8	4.5	9.1	0	0	0	0
TD	0	0	0	0	2.9	2.9	94.2	0	0	0	0	0	0
	low variability							high variability					
DS	0	0	0	2.3	18.2	4.5	56.8	6.8	9.1	2.3	0	0	0
TD	0	0	0	1.0	2.9	3.8	92.3	0	0	0	0	0	0

Table 4.74: Spread of ratings (percentage) by both raters combined for the loudness features of the overall DS and TD groups

The results of Mann Whitney U-tests in table 4.75 show a significant difference between the DS and TD groups for mean loudness level (illustrated as a boxplot in figure 4.61).

LOUDNESS: Mann Whitney results for overall DS and TD groups			
	DS (22)	TD (52)	statistical significance
(-) low - high (+) mean	0 (2)	0 (0)	n = 44 (DS), 104 (TD) *U* = 1935.0 p = 0.035
(-) minimised – extensive (+) range	0 (1)	0 (0)	ns
(-) low - high (+) variability	0 (1)	0 (0)	ns

Table 4.75: Results of Mann Whitney U-tests and median (IQR) values for loudness features of overall DS and TD groups

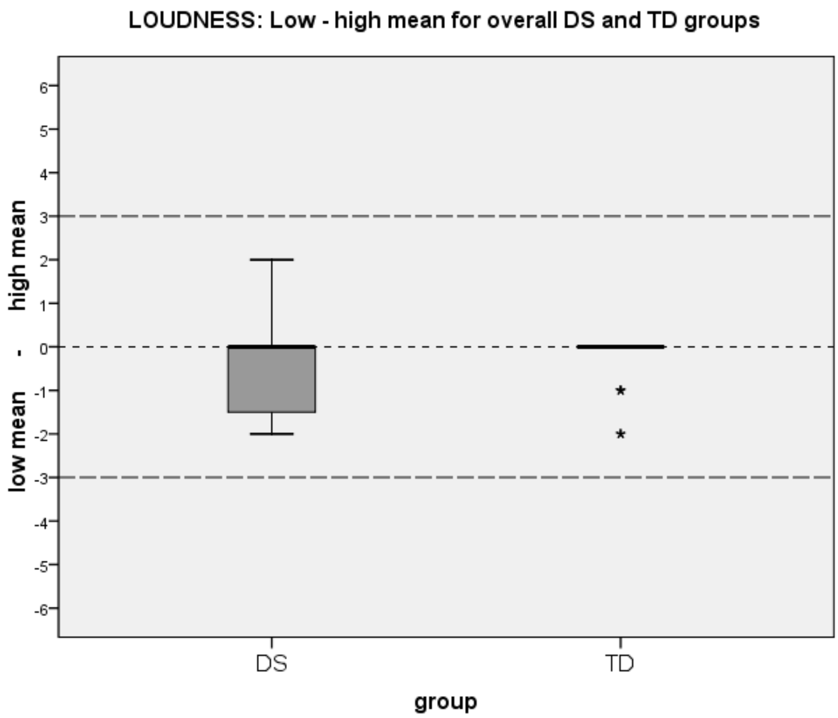


Figure 4.61: Boxplot of low – high mean loudness values of overall DS and TD groups

4.4.1.11 Temporal organization

The rater agreement for the temporal organization categories of continuity and rate are shown in table 4.76, and the spread of ratings in table 4.77.

TEMPORAL ORGANIZATION: Rater agreement (%)			
		DS (22)	TD (52)
continuity	**interrupted**	95.45	100.00
rate	**fast**	100.00	100.00
	slow	100.00	100.00

Table 4.76: Agreement in judgements (percentage) between raters for temporal organization features of overall DS and TD groups

TEMPORAL ORGANIZATION: Spread of ratings (%) for overall DS (22) and TD (52) groups													
code	**-6**	**-5**	**-4**	**-3**	**-2**	**-1**	**0**	**1**	**2**	**3**	**4**	**5**	**6**
							interrupted						
DS							27.3	25.0	22.7	11.4	6.8	6.8	0
TD							100	0	0	0	0	0	0
	slow rate							**fast rate**					
DS	0	0	0	9.1	13.6	25.0	47.7	0	0	2.3	2.3	0	0
TD	0	0	0	0	0	1.0	99.0	0	0	0	0	0	0

Table 4.77: Spread of ratings (percentage) by both raters combined for temporal organization features of the overall DS and TD groups

Table 4.78 shows significant differences in statistical tests (Mann Whitney) between the two groups for both continuity and rate, with the DS group having significantly more interrupted continuity and slower rate (shown as boxplots in figures 4.62 and 4.63).

TEMPORAL ORGANIZATION: Mann Whitney results for overall DS and TD groups				
		DS (22)	TD (52)	statistical significance
continuity	**interrupted**	1 (3)	0 (0)	n = 44 (DS), 104 (TD) $U = 624.0$ $p < 0.001$
rate	**(-) slow – fast (+)**	0 (1)	0 (0)	n = 44 (DS), 104 (TD) $U = 1316.0$ $p < 0.001$

Table 4.78: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of overall DS and TD groups

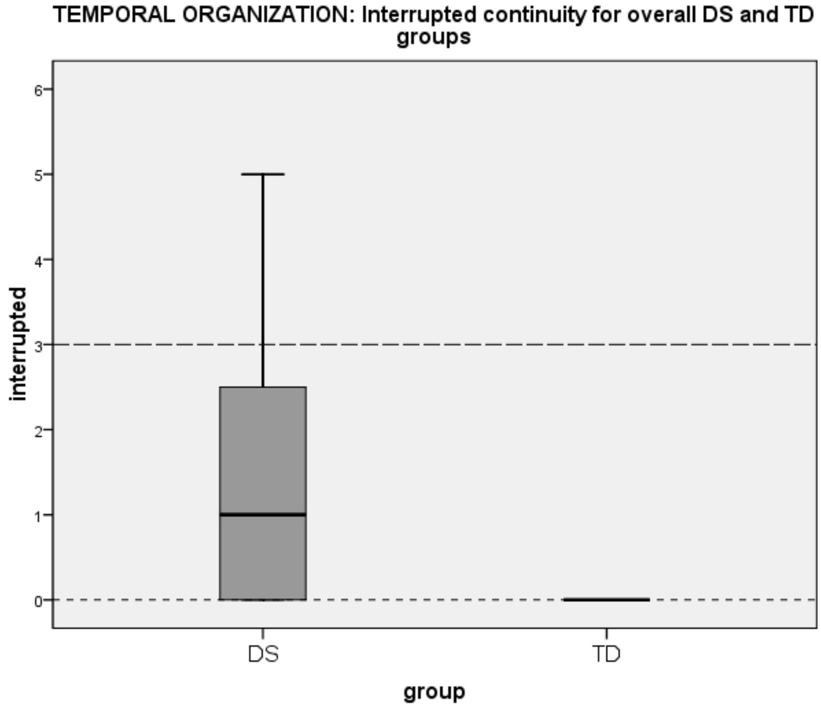


Figure 4.62: Boxplot of interrupted continuity values of overall DS and TD groups

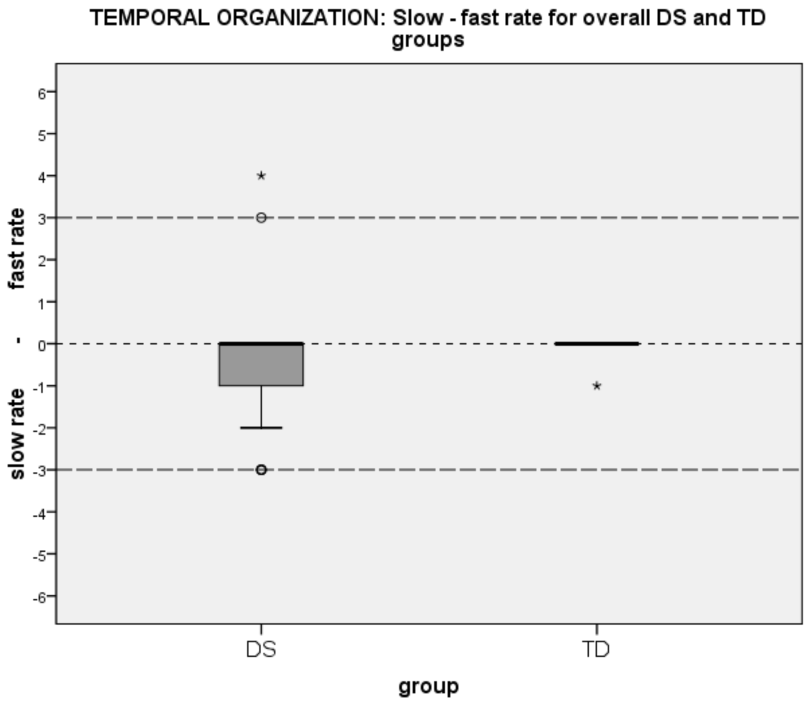


Figure 4.63: Boxplot of slow – fast rate values of overall DS and TD groups

4.4.1.12 Other features

Table 4.79 shows the rater agreement for judgements of respiratory support and diplophonia. The spread of ratings is shown in table 4.80.

OTHER FEATURES: Rater agreement (%)		
	DS (22)	**TD (52)**
respiratory support	91.91	100.00
diplophonia	100.00	100.00

Table 4.79: Agreement in judgements (percentage) between raters for respiratory support and diplophonia features of overall DS and TD groups

OTHER FEATURES: Spread of ratings (%) for overall DS (22) and TD (52) groups		
code	**0**	**1**
	respiratory support adequate	**respiratory support inadequate**
DS	22.7	77.3
TD	0	100
	diplophonia absent	**diplophonia present**
DS	100	0
TD	100	0

Table 4.80: Spread of ratings (percentage) by both raters combined for respiratory support and diplophonia features of the overall DS and TD groups

Results of Chi Square tests (table 4.81) show a significant difference between the DS and TD groups for respiratory support with the DS group having less adequate respiration than their TD peers.

OTHER FEATURES: Chi Square results for overall DS (22) and TD (52) groups	
	statistical significance
respiratory support	$\chi^2 = 25.35$ df = 1 $p < 0.001$
diplophonia	ns

Table 4.81: Results of Chi Square tests for respiratory support and diplophonia features of overall DS and TD groups

4.4.2 Results between DS and TD male speakers

4.4.2.1 Labial settings

Mann Whitney U-tests for labial settings identified significantly minimised range of lip movement in the DS males in comparison to the TD males (values reported in table 4.82 and as a boxplot in figure 4.64).

LABIAL SETTINGS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) spreading - rounding/protrusion (+)	0.5 (3)	0 (1)	ns
labiodentalisation	0 (0)	0 (0)	ns
(-) minimised – extensive (+) range	-2 (3)	0 (1)	n = 26 (DS), 68 (TD) $U = 415.0$ $p < 0.001$

Table 4.82: Results of Mann Whitney U-tests and median (IQR) values for labial settings of DS males and TD males

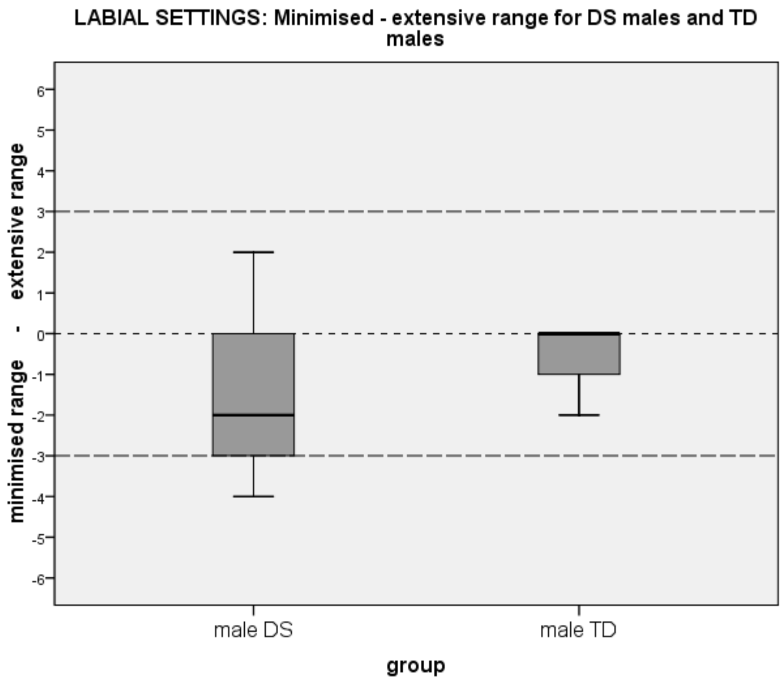


Figure 4.64: Boxplot of minimised – extensive lip range values of DS males and TD males

4.4.2.2 Mandibular settings

Table 4.83 shows the results of Mann Whitney U-tests indicating significant differences between DS and TD males across all of the mandibular sections of the VPAS with the DS males having significantly more open jaw, more protruded jaw and more minimised range of movement (values shown as boxplots in figures 4.65, 4.66 and 4.67).

MANDIBULAR SETTINGS: Mann Whitney results for DS males and TD males			
	DS (12)	**TD (34)**	**statistical significance**
(-) open - close (+) jaw	-1 (2)	0 (1)	n = 24 (DS), 68 (TD) *U* = 182.0 p < 0.001
protruded jaw	3 (1)	0 (0)	n = 24 (DS), 68 (TD) *U* = 38.0 p < 0.001
(-) minimised – extensive (+) range	0 (1)	0 (0)	n = 24 (DS), 68 (TD) *U* = 570.5 p = 0.003

Table 4.83: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of DS males and TD males

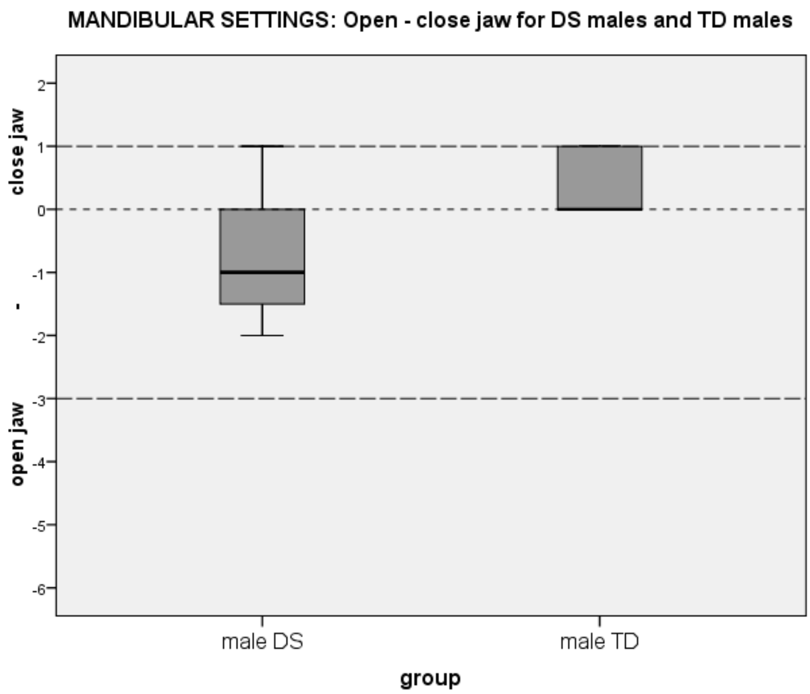


Figure 4.65: Boxplot of open – close jaw values of DS males and TD males

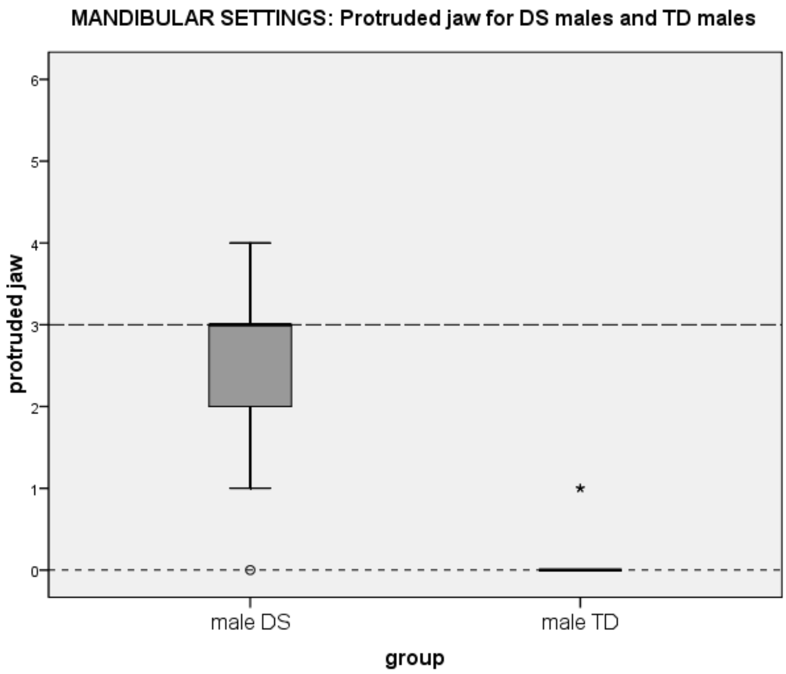


Figure 4.66: Boxplot of protruded jaw values of DS males and TD males

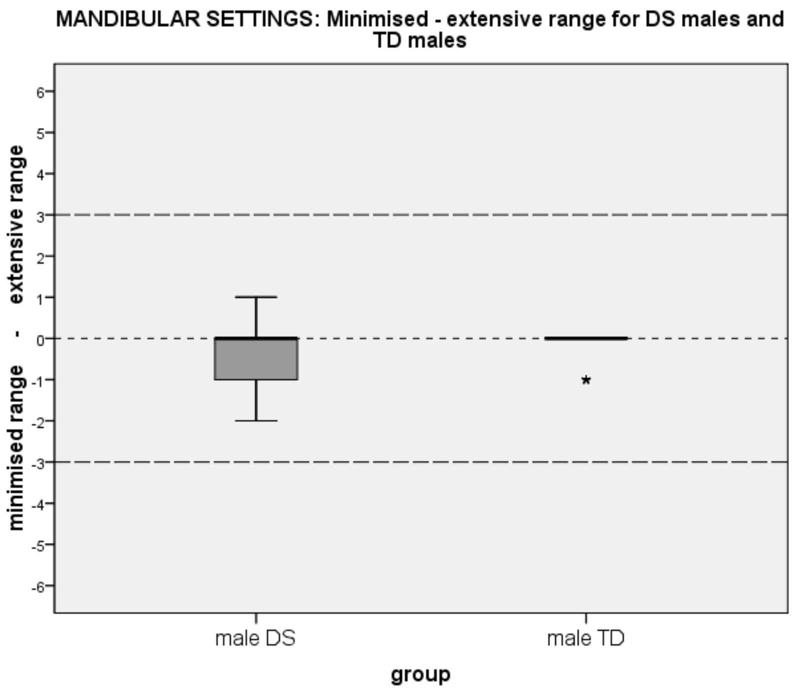


Figure 4.67: Boxplot of minimised – extensive jaw range values of DS males and TD males

4.4.2.3 Lingual settings

In Mann Whitney U-tests the DS males were found to have a significantly advanced tongue tip, raised tongue body and minimised tongue range in comparison to their male TD peers (table 4.84). Values are illustrated as boxplots in figures 4.68-4.70

LINGUAL SETTINGS: Mann Whitney results for DS males and TD males			
	DS (12)	**TD (34)**	**statistical significance**
(-) retracted – advanced (+) tip	3 (2)	0 (1)	n = 24 (DS), 68 (TD) $U = 159.5$ $p < 0.001$
(-) backed – fronted (+) body	0 (4)	-1 (1)	ns
(-) lowered – raised (+) body	1.5 (1)	0 (1)	n = 24 (DS), 68 (TD) $U = 328.0$ $p < 0.001$
(-) minimised – extensive (+) range	-3 (2)	0 (0)	n = 24 (DS), 68 (TD) $U = 165.0$ $p < 0.001$

Table 4.84: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of DS males and TD males

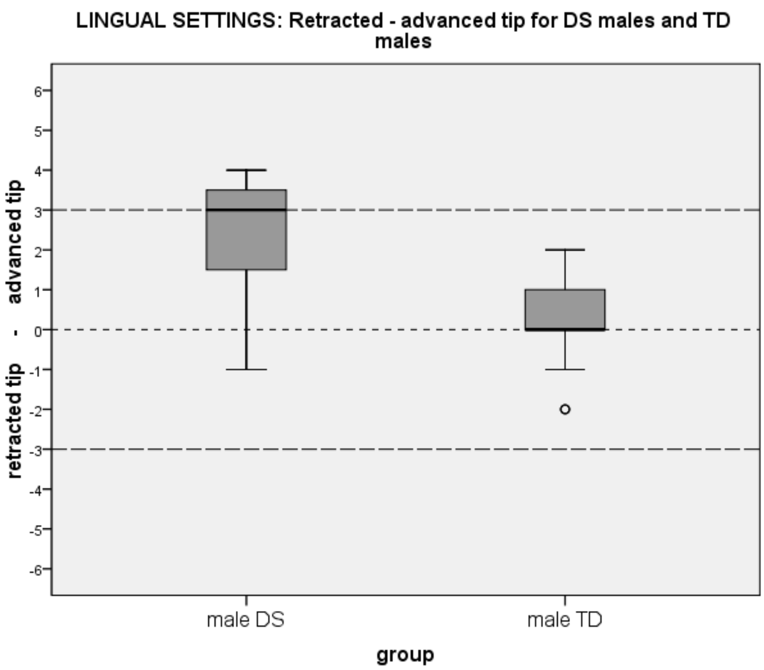


Figure 4.68: Boxplot of retracted – advanced tongue tip values of DS males and TD males

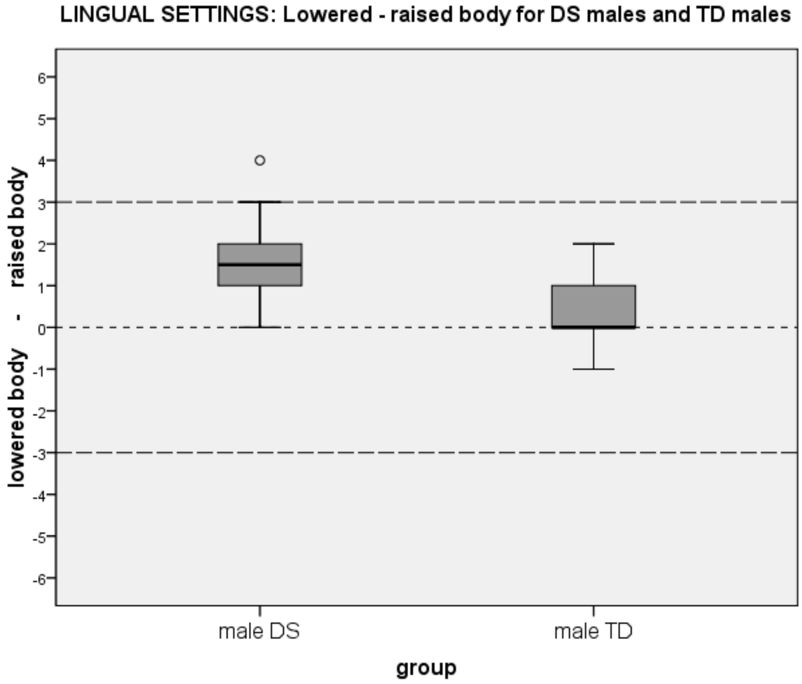


Figure 4.69: Boxplot of lowered – raised tongue body values of DS males and TD males

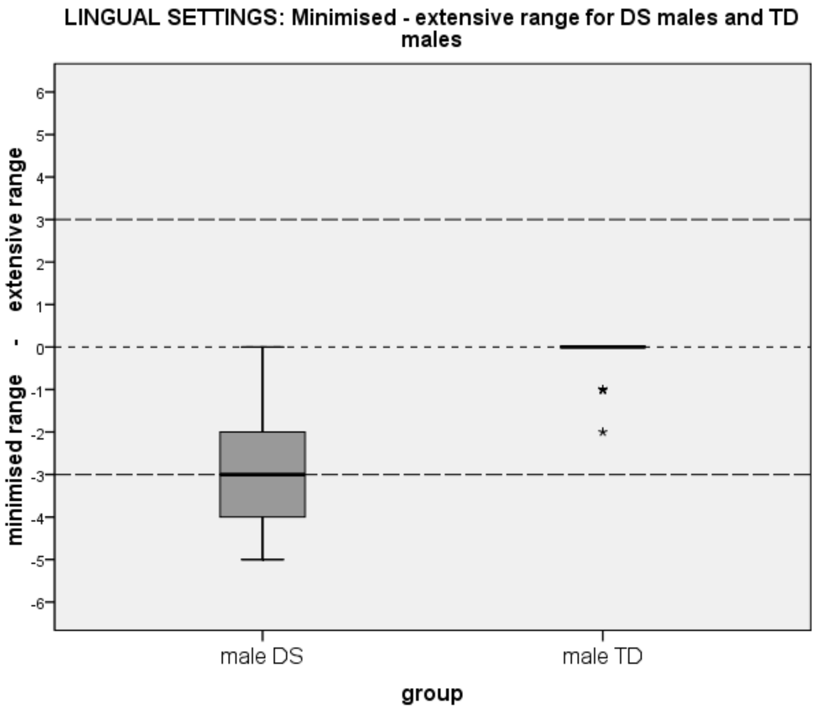


Figure 4.70: Boxplot of minimised – extensive tongue range values of DS males and TD males

4.4.2.4 Pharyngeal settings

A significant difference in pharyngeal constriction was found in a Mann Whitney U-test with DS males having more constriction than TD males (table 4.85 and figure 4.71).

PHARYNGEAL SETTINGS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) constriction – expansion (+)	-1 (1)	0 (0)	n = 26 (DS), 68 (TD) *U* = 300.0 $p < 0.001$

Table 4.85: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of DS males and TD males

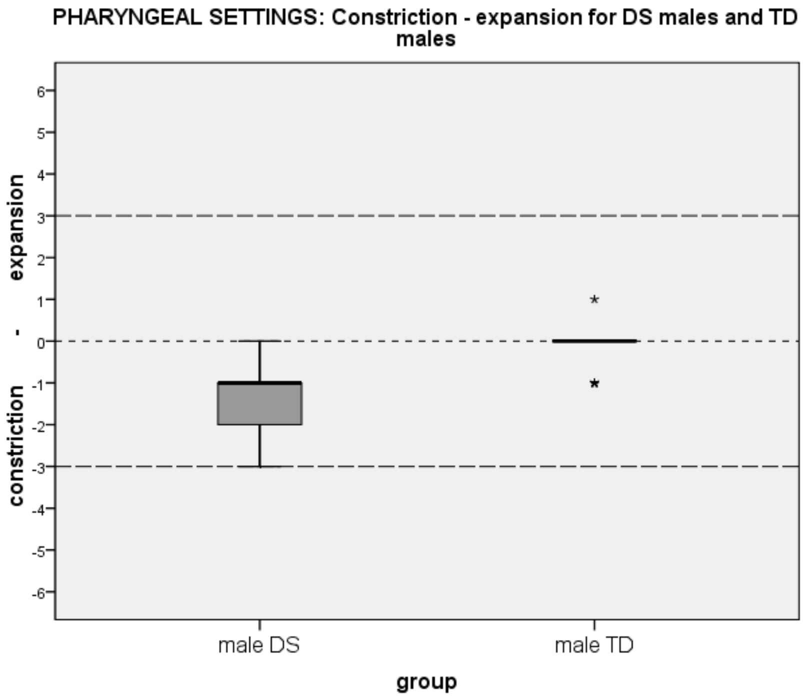


Figure 4.71: Boxplot of pharyngeal constriction – expansion values of DS males and TD males

4.4.2.5 Velopharyngeal settings

Table 4.86 shows the results of Mann Whitney U-tests which identified significant differences in audible nasal escape and nasality ratings between groups. DS males were found to have more nasal escape and higher levels of nasality than the TD males (illustrated in figures 4.72 and 4.73).

VELOPHARYNGEAL SETTINGS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
audible nasal escape	0 (1)	0 (0)	n = 26 (DS), 68 (TD) *U* = 544.0 p < 0.001
(-) denasal – nasal (+)	4 (1)	2 (1)	n = 26 (DS), 68 (TD) *U* = 324.0 p < 0.001

Table 4.86: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of DS males and TD males

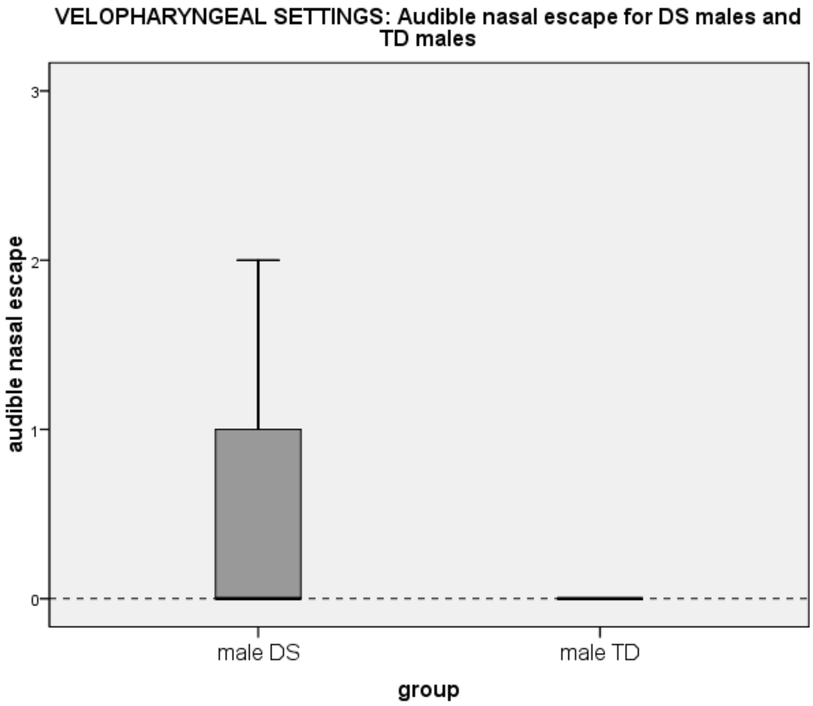


Figure 4.72: Boxplot of audible nasal escape values of DS males and TD males

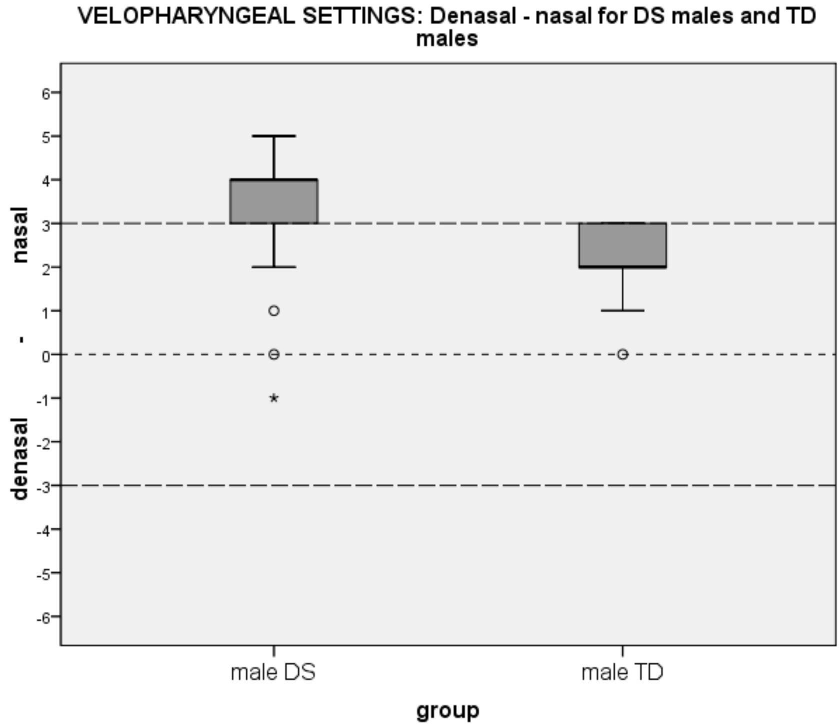


Figure 4.73: Boxplot of denasal – nasal values of DS males and TD males

4.4.2.6 Larynx height settings

No significant differences were found between DS and TD males in larynx height (table 4.87).

LARYNX HEIGHT SETTINGS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) lowered – raised (+) larynx	0 (3)	0 (1)	ns

Table 4.87: Results of Mann Whitney U-tests and median (IQR) values for larynx height settings of DS males and TD males

4.4.2.7 Tension settings

Table 4.88 shows results of Mann Whitney U-tests finding a significant difference in vocal tract tension between DS males and TD males, where DS males have been rated as having reduced tension (illustrated in figure 4.74).

MUSCULAR TENSION SETTINGS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (1)	0 (0)	n = 26 (DS), 68 (TD) *U* = 583.0 p = 0.001
(-) lax – tense (+) larynx	0.5 (2)	1 (1)	ns

Table 4.88: Results of Mann Whitney U-tests and median (IQR) values for tension settings of DS males and TD males

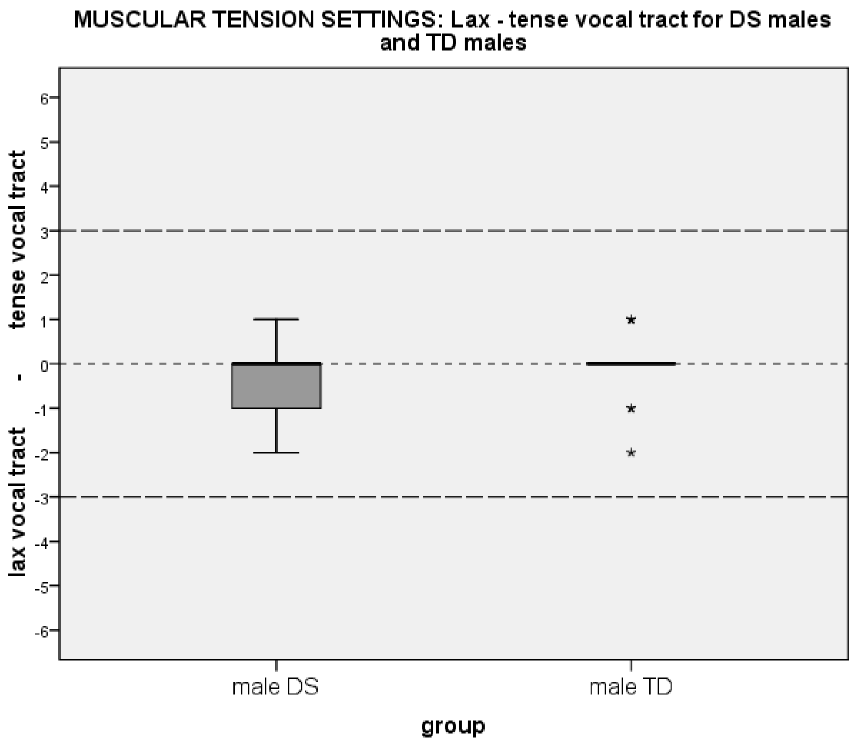


Figure 4.74: Boxplot of lax – tense vocal tract values of DS males and TD males

4.4.2.8 Phonation features

Mann Whitney U-tests and Chi Square tests revealed that only whispery voice differed significantly between the groups with DS males having higher levels of this feature (table 4.89 and figure 4.75).

PHONATION FEATURES: Mann Whitney and Chi Square* results for DS males and TD males				
		DS (13)	**TD (34)**	**statistical significance**
voicing type	**neutral - non neutral voice**	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (0)	0 (2)	ns
laryngeal frication	**whisper**	n/a	n/a	ns*
	whispery	3 (1)	2 (0.5)	n = 26 (DS), 68 (TD) $U = 498.0$ $p < 0.001$
laryngeal irregularity	**harsh**	1 (1)	0 (1)	ns
	tremor	0 (0)	0 (0)	ns

Table 4.89: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of DS males and TD males

Figure 4.75: Boxplot of whispery phonation values of DS males and TD males

4.4.2.9 Pitch

Table 4.90 shows that in Mann Whitney U-tests males with DS had significantly lower mean pitch than TD males. Values are illustrated as a boxplot in figure 4.76.

PITCH: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) low – high (+) mean	-2 (3)	0 (1)	n = 26 (DS), 68 (TD) $U = 521.5$ $p = 0.006$
(-) minimised - extensive (+) range	-1 (4)	0 (1)	ns
(-) low - high (+) variability	0 (3)	0 (1)	ns

Table 4.90: Results of Mann Whitney U-tests and median (IQR) values for pitch features of DS males and TD males

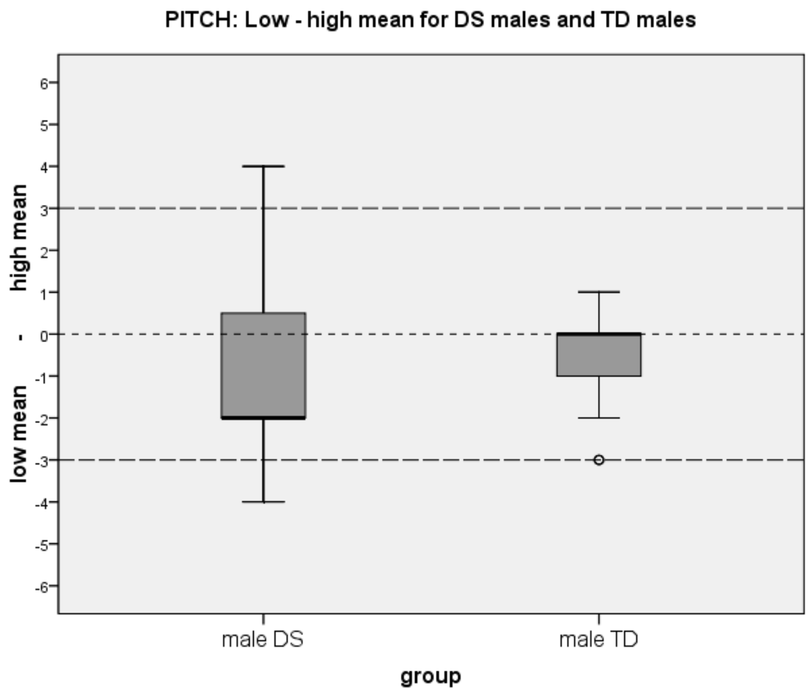


Figure 4.76: Boxplot of low – high mean pitch values of DS males and TD males

4.4.2.10 Loudness

In Mann Whitney U-tests all three loudness measures were found to be significantly lower in DS males compared to TD males (table 4.91 and figures 4.77-4.79).

LOUDNESS: Mann Whitney results for DS males and TD males			
	DS (13)	**TD (34)**	**statistical significance**
(-) low - high (+) mean	0 (2)	0 (0)	n = 26 (DS), 68 (TD) $U = 596.5$ $p < 0.001$
(-) minimised – extensive (+) range	0 (2)	0 (0)	n = 26 (DS), 68 (TD) $U = 687.0$ $p = 0.013$
(-) low - high (+) variability	0 (2)	0 (0)	n = 26 (DS), 68 (TD) $U = 675.0$ $p = 0.01$

Table 4.91: Results of Mann Whitney U-tests and median (IQR) values for loudness features of DS males and TD males

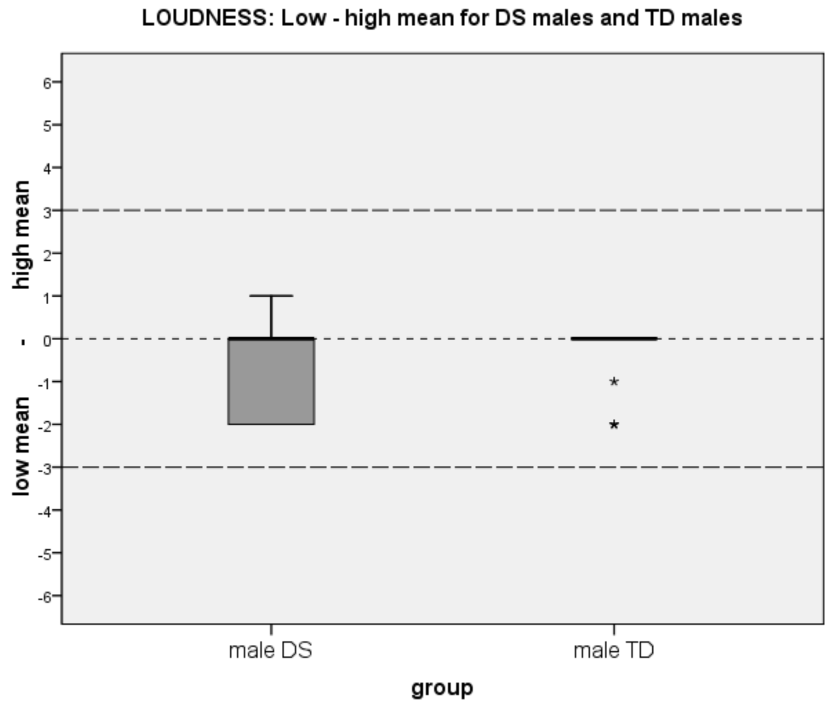


Figure 4.77: Boxplot of low – high mean loudness values of DS males and TD males

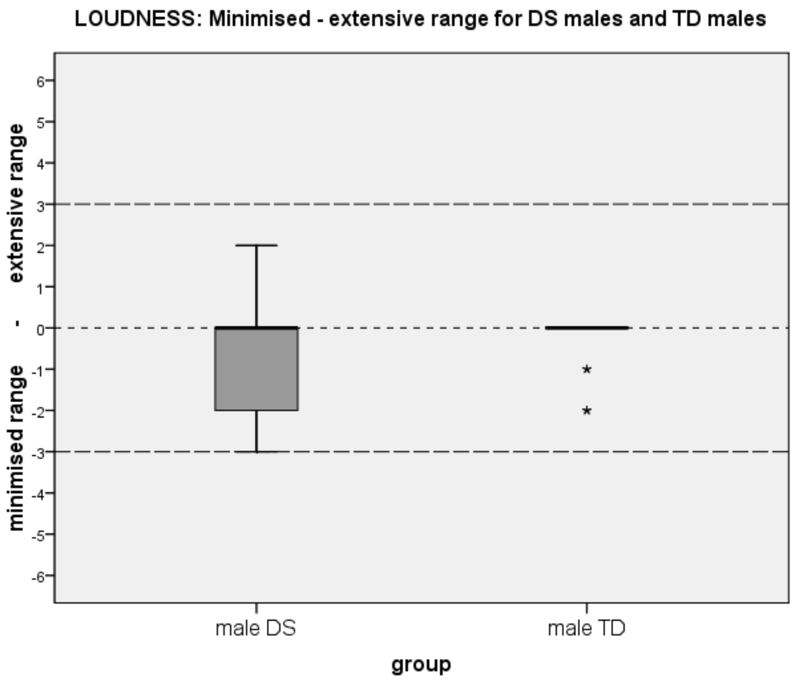


Figure 4.78: Boxplot of minimised - extensive loudness range values of DS males and TD males

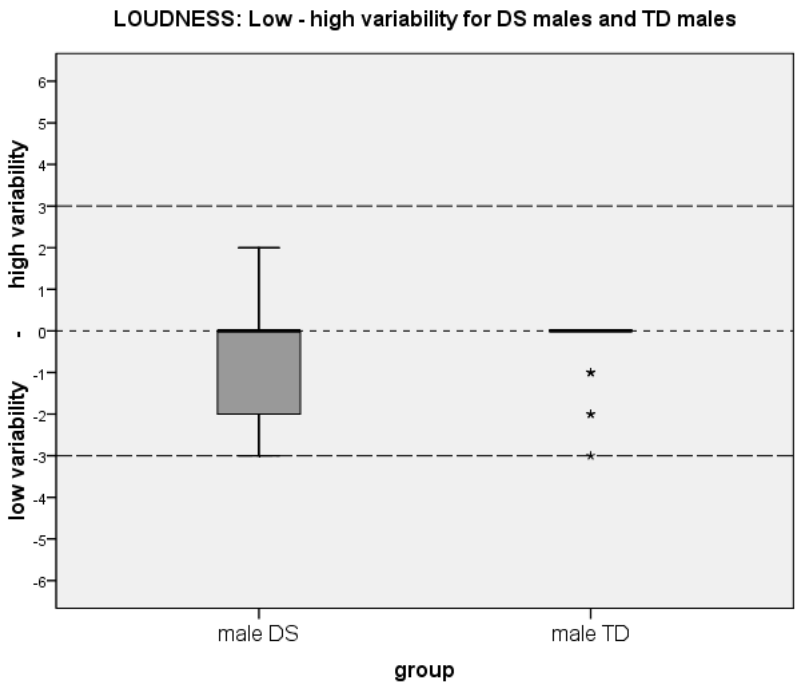


Figure 4.79: Boxplot of low – high loudness variability values of DS males and TD males

4.4.2.11 Temporal organization

Table 4.92 shows the results of Mann Whitney U-tests finding that continuity is more interrupted and rate slower in DS males compared to TD males. Values are illustrated as boxplots in figures 4.80 and 4.81.

TEMPORAL ORGANIZATION: Mann Whitney results for DS males and TD males				
		DS (13)	**TD (34)**	**statistical significance**
continuity	**interrupted**	1 (2)	0 (0)	n = 26 (DS), 68 (TD) *U* = 374.0 $p < 0.001$
rate	**(-) slow – fast (+)**	0 (1)	0 (0)	n = 26 (DS), 68 (TD) *U* = 587.5 $p < 0.001$

Table 4.92: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of DS males and TD males

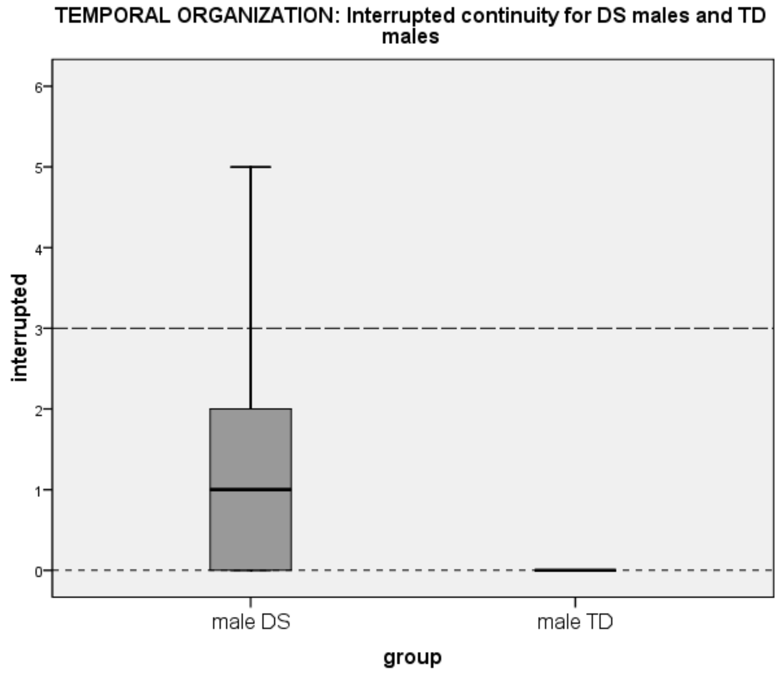


Figure 4.80: Boxplot of interrupted continuity values of DS males and TD males

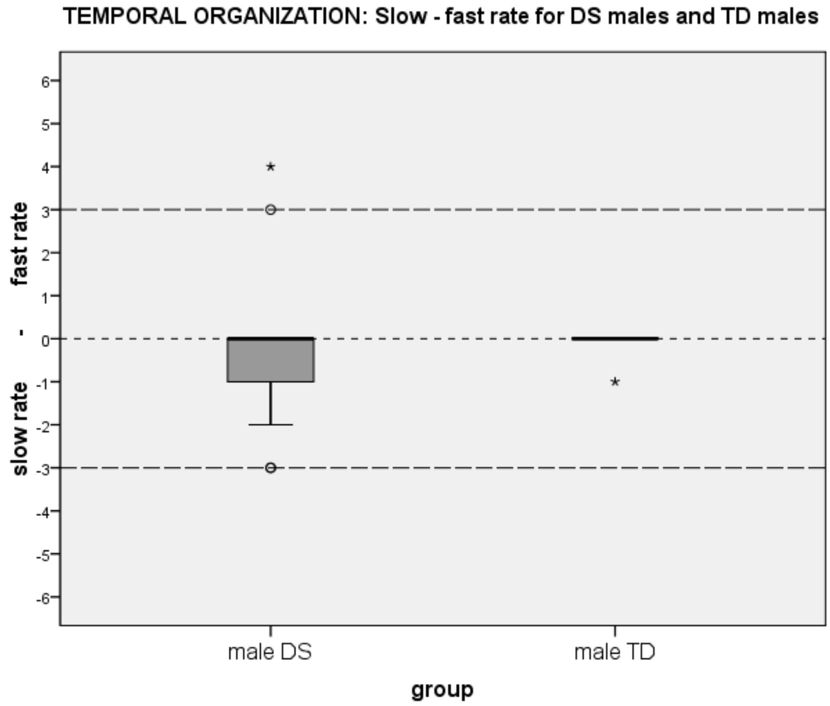


Figure 4.81: Boxplot of slow – fast rate values of DS males and TD males

4.4.2.12 Other features

A Chi square test identified reduced respiratory support in males with DS compared to their TD male peers (table 4.93).

OTHER FEATURES: Chi Square results for DS males (13) and TD males (34)	
	statistical significance
respiratory support	$\chi^2 = 19.78$ df = 1 $p < 0.001$
diplophonia	ns

Table 4.93: Results of Chi Square tests for respiratory support and diplophonia features of DS males and TD males

4.4.3 Results between DS and TD female speakers

4.4.3.1 Labial settings

Mann Whitney U-tests reveal DS females were judged as having significantly more rounding/protrusion and more minimised lip range than TD females (table 4.94 and figures 4.82 and 4.83).

LABIAL SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) spreading - rounding/protrusion (+)	1.5 (2)	-0.5 (1)	n = 18 (DS), 36 (TD) $U = 149.5$ p = 0.001
labiodentalisation	0 (1)	0 (0)	ns
(-) minimised – extensive (+) range	-1.5 (2)	0 (0)	n = 18 (DS), 36 (TD) $U = 96.5$ p < 0.001

Table 4.94: Results of Mann Whitney U-tests and median (IQR) values for labial settings of DS females and TD females

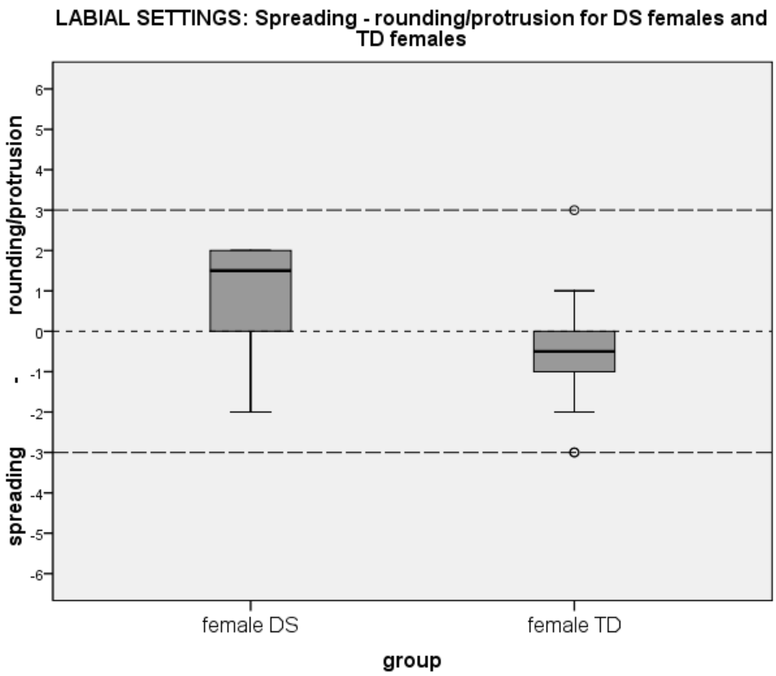


Figure 4.82: Boxplot of lip spreading – rounding/protrusion values of DS females and TD females

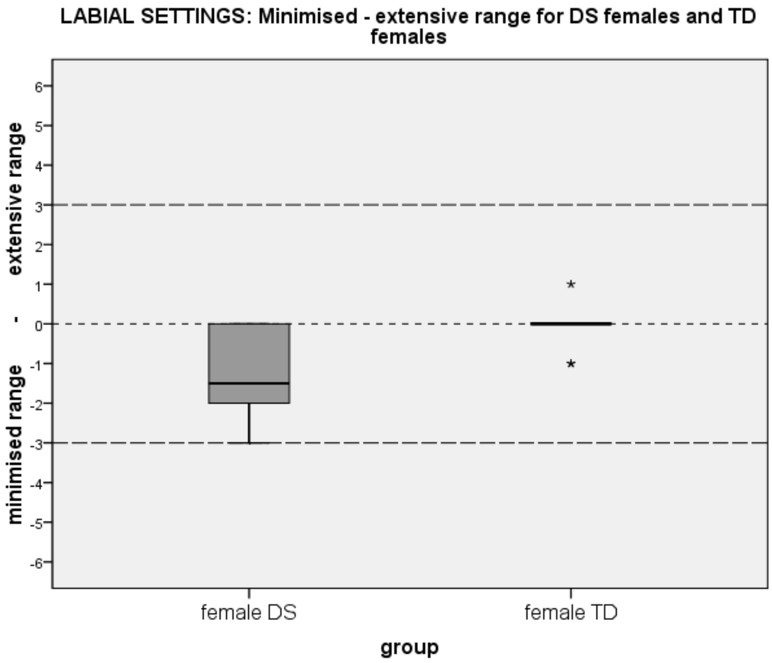


Figure 4.83: Boxplot of minimised – extensive lip range values of DS females and TD females

4.4.3.2 Mandibular settings

Table 4.95 shows significant results between DS females and TD females in Mann Whitney U-tests for all mandibular settings. DS females having more open jaw, more protruded jaw and more minimised jaw range than their TD female peers. Values are illustrated in boxplots in figures 4.84-4.86.

MANDIBULAR SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) open - close (+) jaw	-0.5 (1)	0 (0)	n = 18 (DS), 36 (TD) $U = 180.0$ $p < 0.001$
protruded jaw	2 (2)	0 (0)	n = 18 (DS), 36 (TD) $U = 72.0$ $p < 0.001$
(-) minimised – extensive (+) range	0 (1)	0 (0)	n = 18 (DS), 36 (TD) $U = 234.0$ $p = 0.001$

Table 4.95: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of DS females and TD females

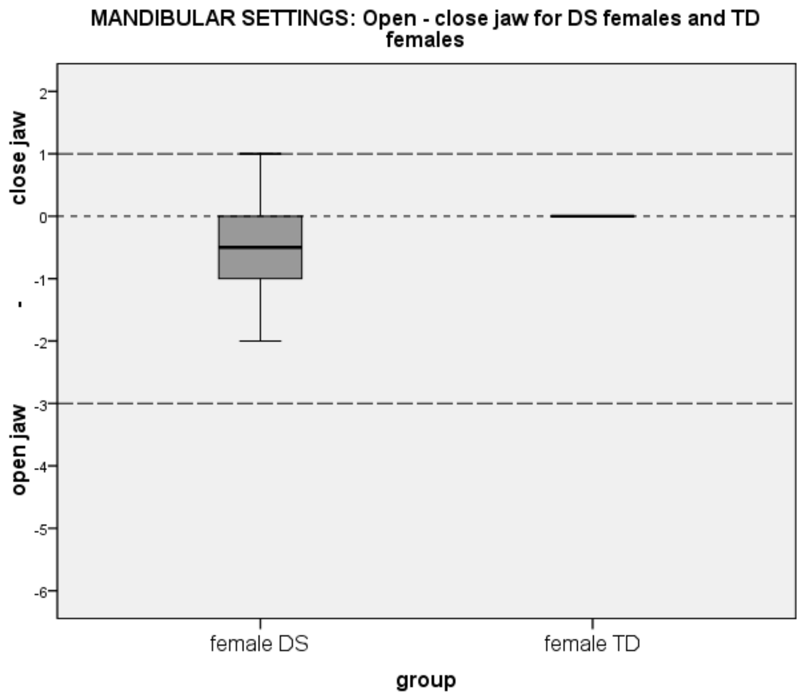


Figure 4.84: Boxplot of open – close jaw values of DS females and TD females

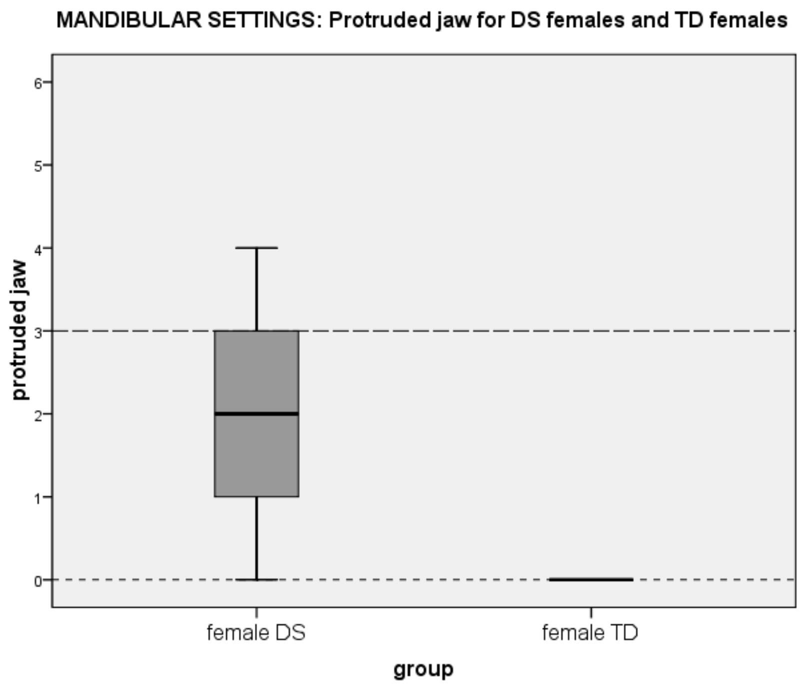


Figure 4.85: Boxplot of protruded jaw values of DS females and TD females

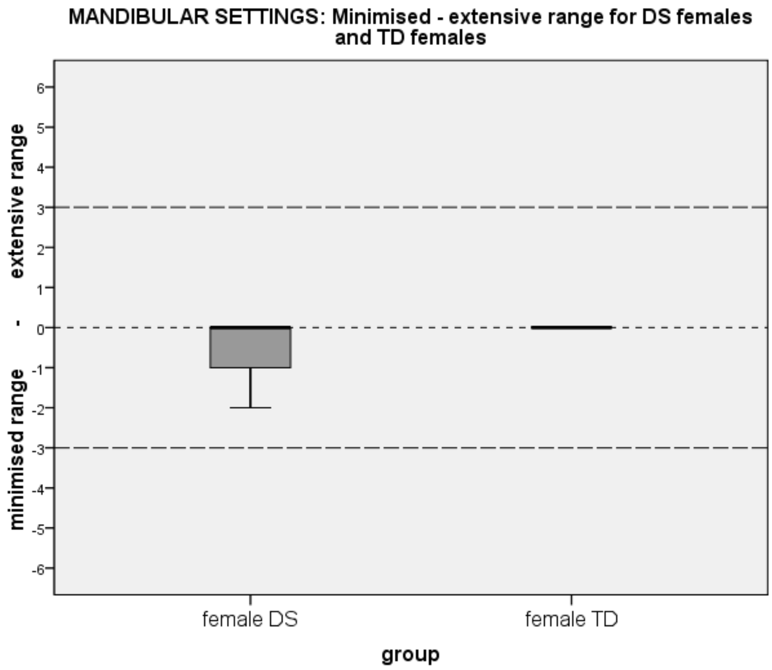


Figure 4.86: Boxplot of minimised – extensive jaw range values of DS females and TD females

4.4.3.3 Lingual settings

Significant differences in Mann Whitney U-tests were found between the groups in several lingual settings, DS females having a more advanced tongue tip position, a more fronted tongue body and a more minimised range than the TD females (table 4.96 and figures 4.87-4.89).

LINGUAL SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) retracted – advanced (+) tip	3 (2)	1 (1)	n = 18 (DS), 36 (TD) $U = 82.0$ $p < 0.001$
(-) backed - fronted (+) body	3 (1)	1 (2)	n = 18 (DS), 36 (TD) $U = 53.0$ $p < 0.001$
(-) lowered - raised (+) body	2 (1)	1 (1)	ns
(-) minimised – extensive (+) range	-2 (1)	0 (0)	n = 18 (DS), 36 (TD) $U = 0$ $p < 0.001$

Table 4.96: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of DS females and TD females

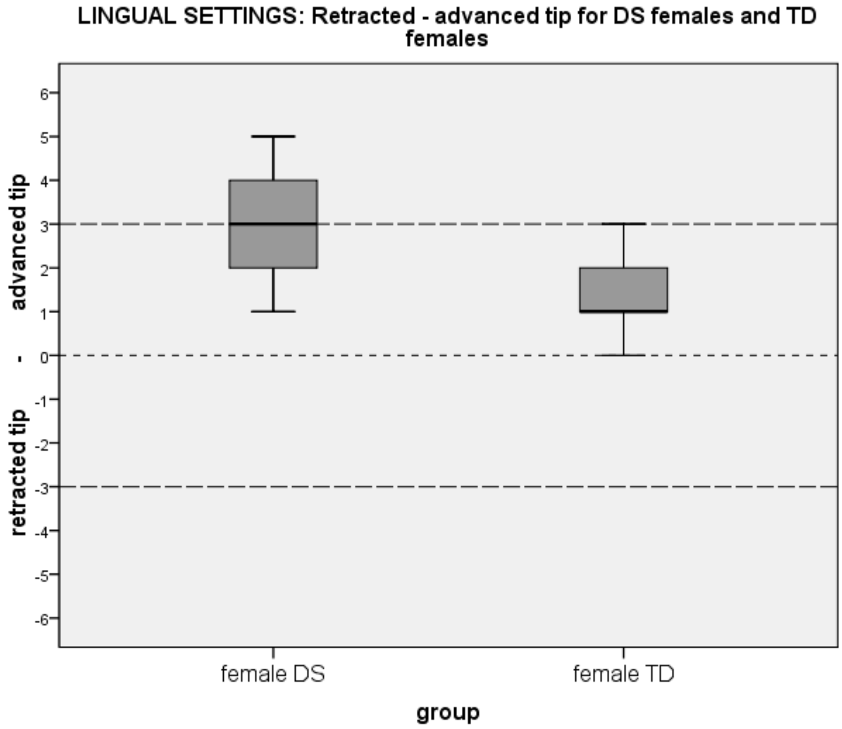


Figure 4.87: Boxplot of retracted – advanced tongue tip values of DS females and TD females

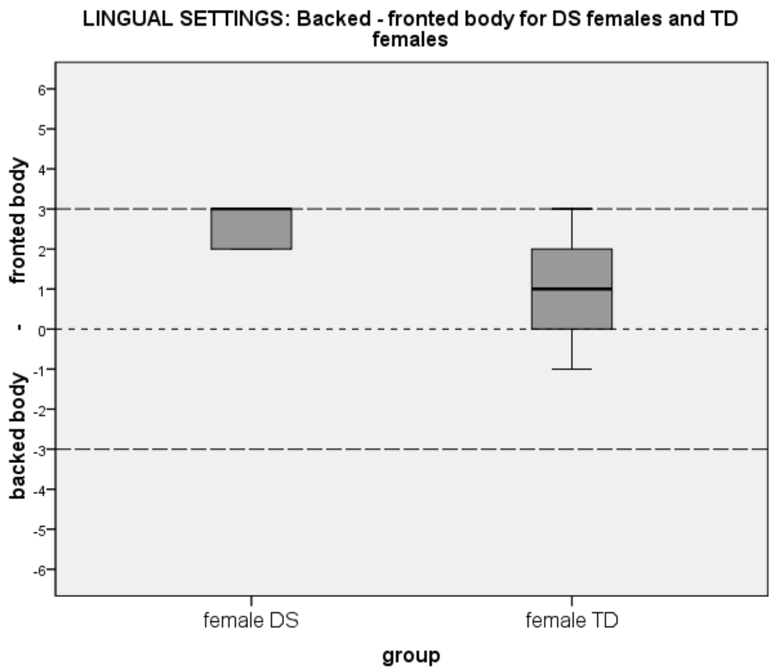


Figure 4.88: Boxplot of backed – fronted tongue body values of DS females and TD females

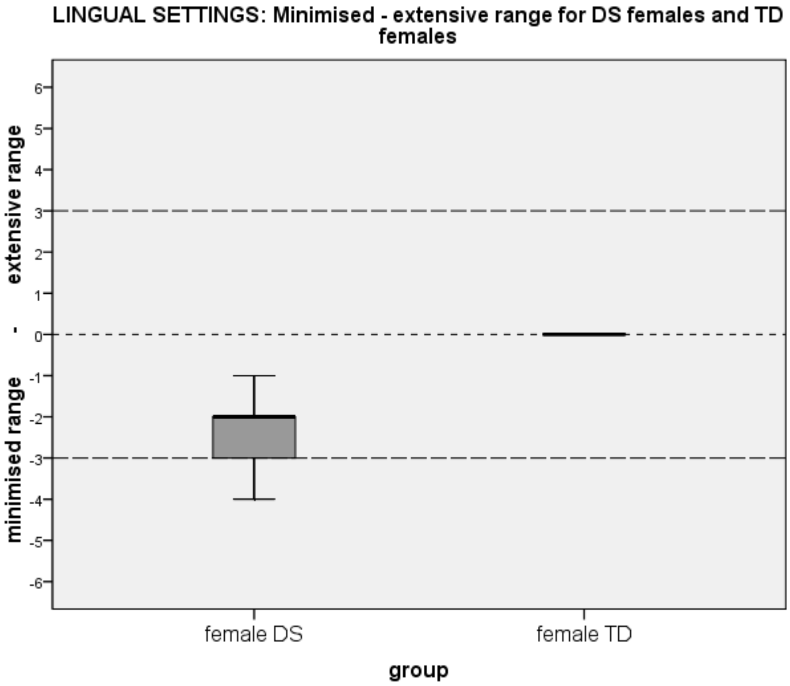


Figure 4.89: Boxplot of minimised – extensive tongue range values of DS females and TD females

4.4.3.4 Pharyngeal settings

A significant difference between the DS and TD females was found for pharyngeal constriction – expansion, with the DS females having a greater degree of constriction (table 4.97 and figure 4.90).

PHARYNGEAL SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) constriction – expansion (+)	-1 (1)	0 (0)	n = 18 (DS), 36 (TD) $U = 78.5$ $p < 0.001$

Table 4.97: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of DS females and TD females

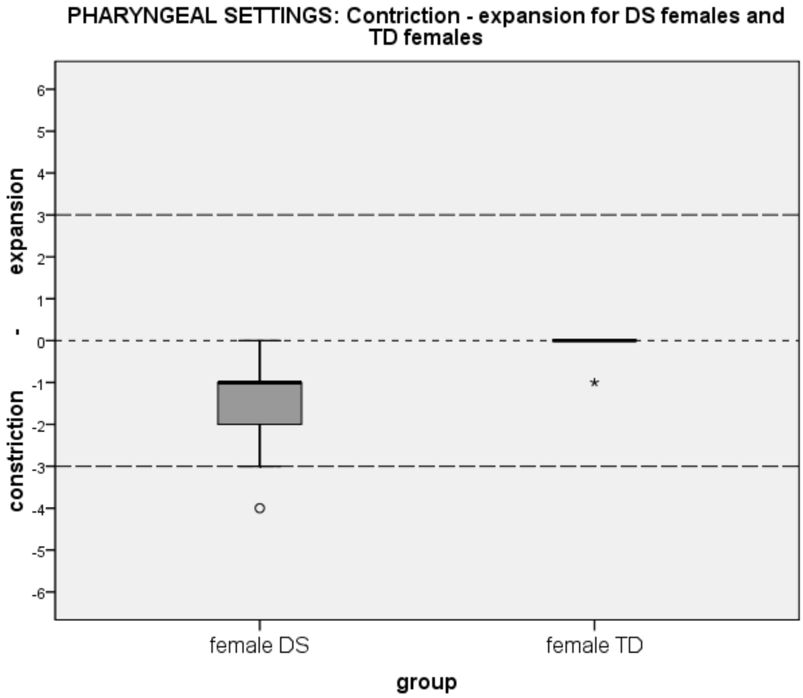


Figure 4.90: Boxplot of pharyngeal constriction – expansion values of DS females and TD females

4.4.3.5 Velopharyngeal settings

Table 4.98 shows the results of Mann Whitney U-tests from the velopharyngeal settings of the VPAS. Females with DS were found to have increased nasal escape and increased nasality in comparison to TD females (illustrated as boxplots in figures 4.91 and 4.92).

VELOPHARYNGEAL SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
audible nasal escape	0 (0)	0 (0)	n = 18 (DS), 36 (TD) U = 288.0 p = 0.043
(-) denasal – nasal (+)	3 (0)	1 (1)	n = 18 (DS), 36 (TD) U = 94.5 p < 0.001

Table 4.98: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of DS females and TD females

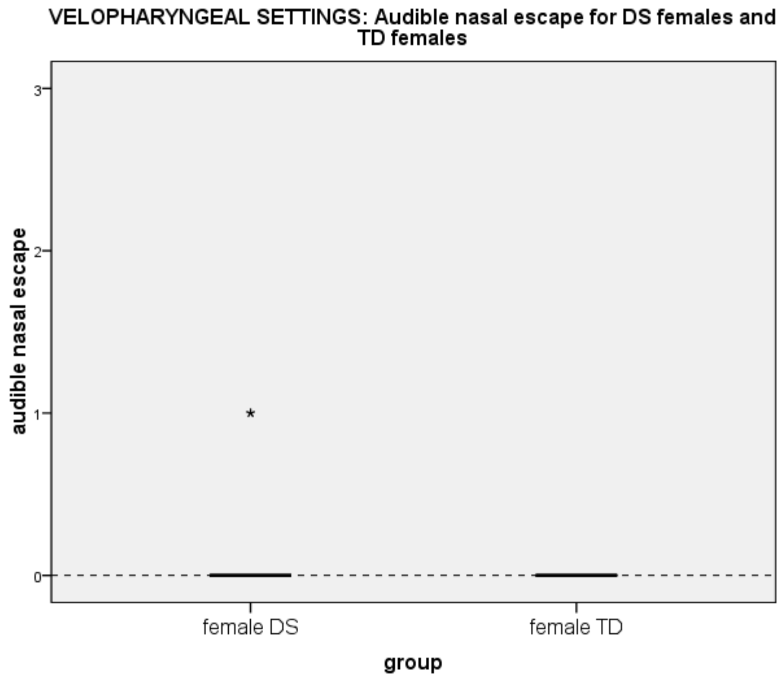


Figure 4.91: Boxplot of audible nasal escape values of DS females and TD females

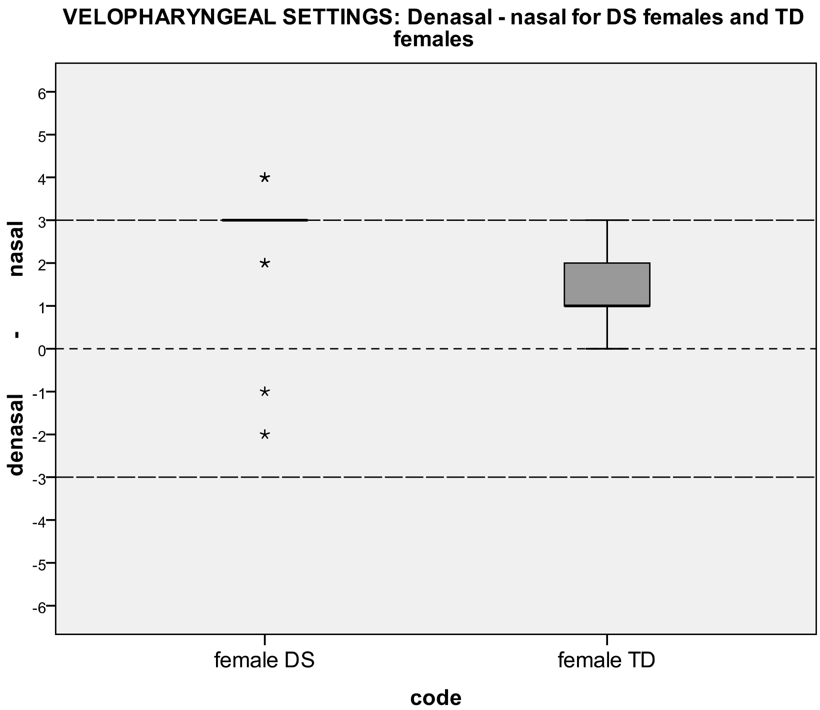


Figure 4.92: Boxplot of denasal – nasal values of DS females and TD females

4.4.3.6 Larynx height settings

No significant differences were found in larynx height between the DS and TD females (table 4.99).

LARYNX HEIGHT SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) lowered – raised (+) larynx	0 (1)	0 (1)	ns

Table 4.99: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of DS females and TD females

4.4.3.7 Tension settings

A significant difference was found in a Mann Whitney U-test between DS and TD females for laryngeal tension, with DS females having increased tension (table 4.100 and figure 4.93).

MUSCULAR TENSION SETTINGS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (0)	0 (0)	ns
(-) lax – tense (+) larynx	2 (2)	1 (1)	n = 18 (DS), 36 (TD) $U = 160.0$ $p = 0.002$

Table 4.100: Results of Mann Whitney U-tests and median (IQR) values for tension settings of DS females and TD females

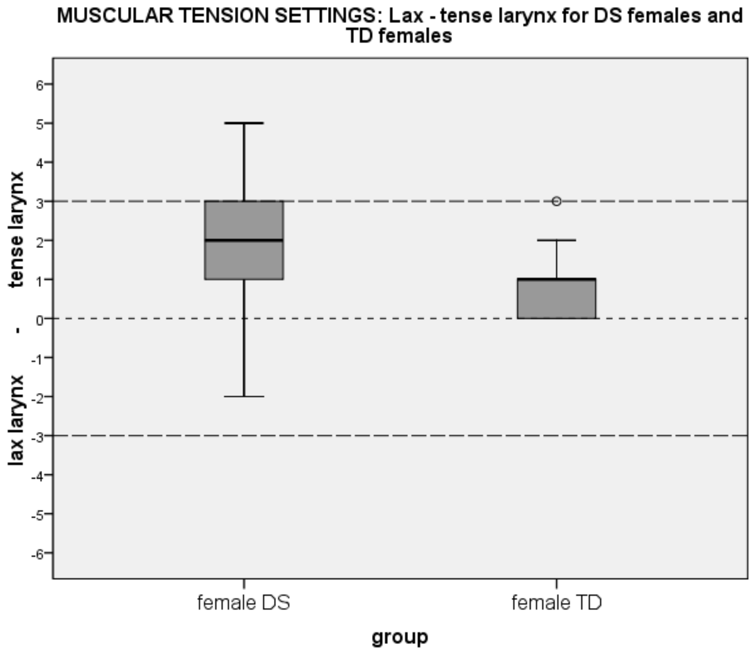


Figure 4.93: Boxplot of lax – tense larynx values of DS females and TD females

4.4.3.8 Phonation features

Table 4.101 shows the results of Mann Whitney U-tests and Chi Square tests where DS females were found to have significantly more harshness than their TD female peers. Values are illustrated as a boxplot in figure 4.94.

PHONATION FEATURES: Mann Whitney and Chi Square* results for DS females and TD females				
		DS (9)	**TD (18)**	**statistical significance**
voicing type	**neutral - non neutral voice**	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (0)	0 (0)	ns
laryngeal frication	**whisper**	n/a	n/a	ns*
	whispery	3 (1)	2 (1)	ns
laryngeal irregularity	**harsh**	1 (2)	1 (1)	n = 18 (DS), 36 (TD) *U* = 216.5 p = 0.03
	tremor	0 (0)	0 (0)	ns

Table 4.101: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of DS females and TD females

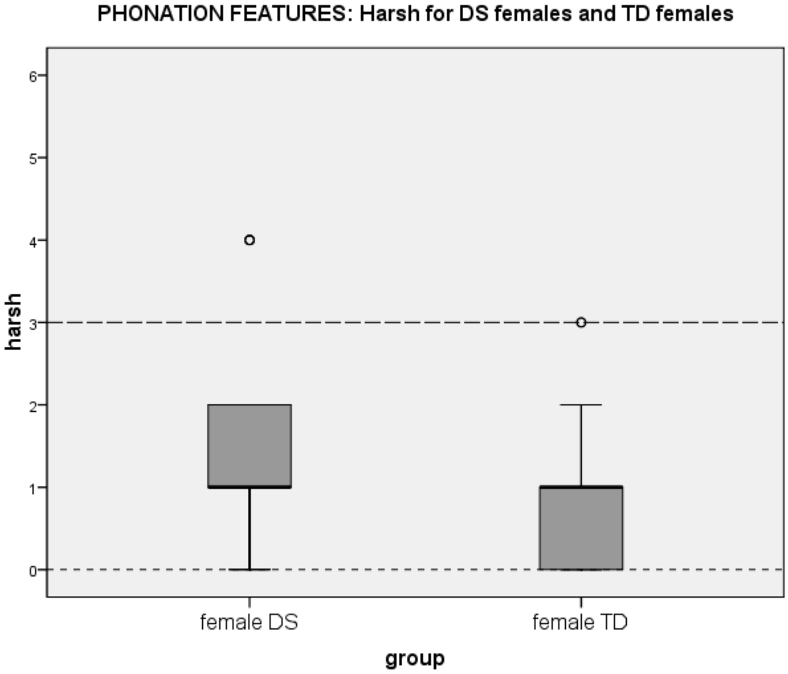


Figure 4.94: Boxplot of harsh phonation values of DS females and TD females

4.4.3.9 Pitch

A significant difference in mean pitch was found in a Mann Whitney U-test between DS and TD females with the DS females having significantly lower pitch (table 4.102 and figure 4.95).

PITCH: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) low – high (+) mean	-0.5 (3)	0 (1)	n = 18 (DS), 36 (TD) *U* = 216.0 p = 0.035
(-) minimised - extensive (+) range	0 (2)	0 (0)	ns
(-) low - high (+) variability	0 (1)	0 (0)	ns

Table 4.102: Results of Mann Whitney U-tests and median (IQR) values for pitch features of DS females and TD females

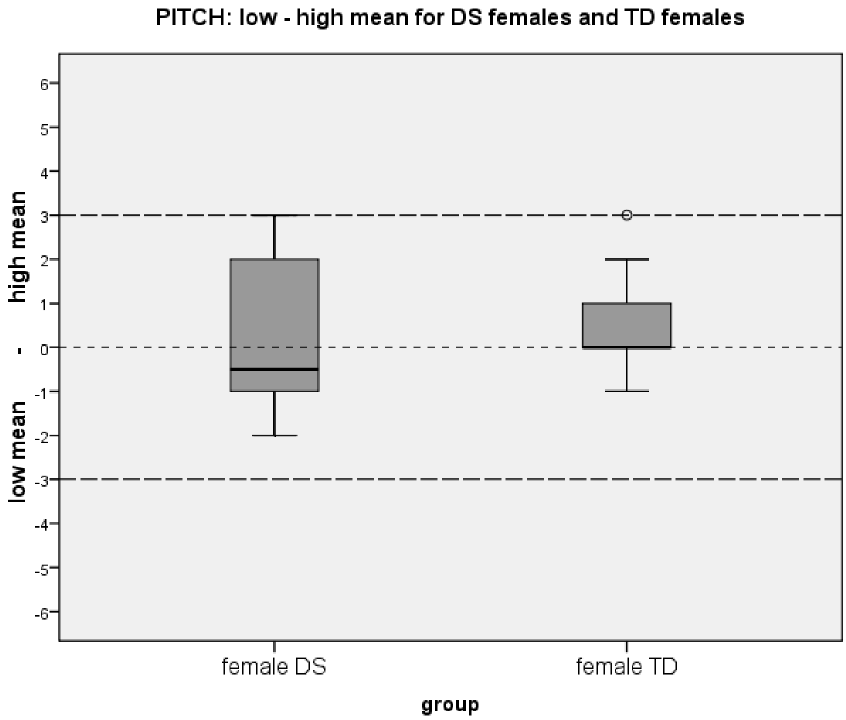


Figure 4.95: Boxplot of low – high mean pitch values of DS females and TD females

4.4.3.10 Loudness

Table 4.103 shows a significant Mann Whitney U-test result between DS and TD females for loudness variability (DS females having higher variability). Values are illustrated as a boxplot in figure 4.96.

LOUDNESS: Mann Whitney results for DS females and TD females			
	DS (9)	**TD (18)**	**statistical significance**
(-) low - high (+) mean	0 (2)	0 (0)	ns
(-) minimised – extensive (+) range	0 (0)	0 (0)	ns
(-) low - high (+) variability	0 (1)	0 (0)	n = 18 (DS), 36 (TD) *U* = 221.5 p = 0.004

Table 4.103: Results of Mann Whitney U-tests and median (IQR) values for loudness features of DS females and TD females

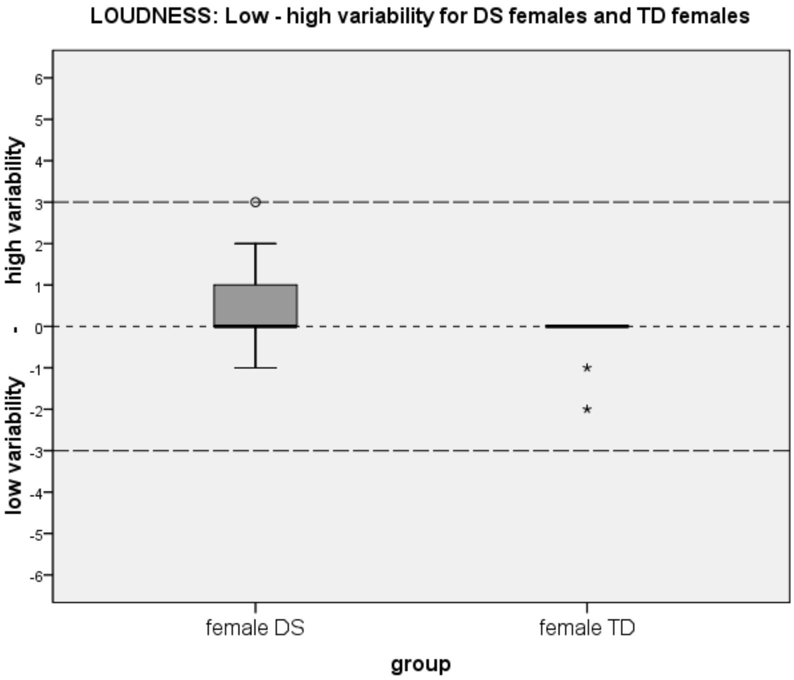


Figure 4.96: Boxplot of low – high loudness variability values of DS females and TD females

4.4.3.11 Temporal organization

Mann Whitney U-tests revealed that the DS females were rated as having significantly more interrupted continuity and a significantly slower rate than their TD female peers (table 4.104 and figures 4.97 and 4.98).

TEMPORAL ORGANIZATION: Mann Whitney results for DS females and TD females				
		DS (9)	**TD (18)**	**statistical significance**
continuity	**interrupted**	2 (2)	0 (0)	n = 18 (DS), 36 (TD) U = 18.0 $p < 0.001$
rate	**(-) slow – fast (+)**	-1 (2)	0 (0)	n = 18 (DS), 36 (TD) U = 144.0 $p < 0.001$

Table 4.104: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of DS females and TD females

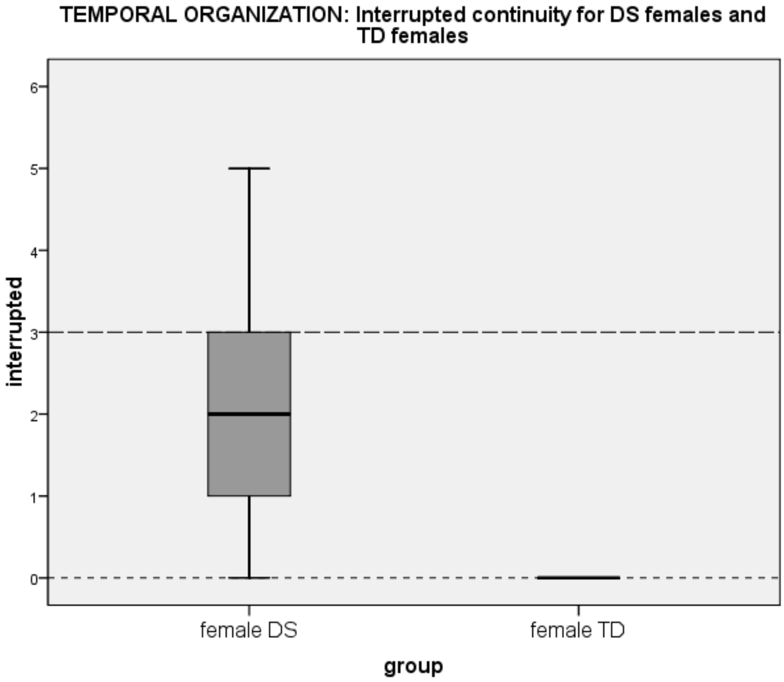


Figure 4.97: Boxplot of interrupted continuity values of DS females and TD females

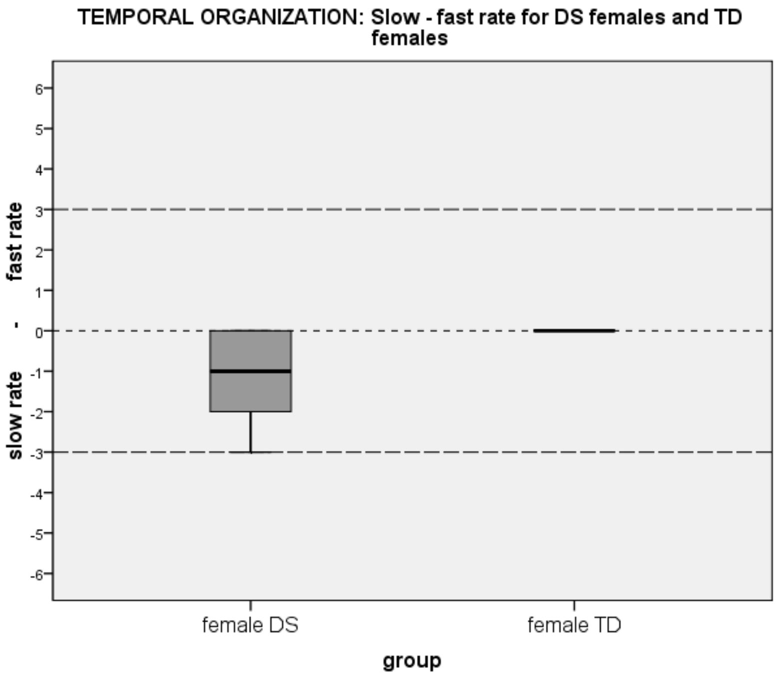


Figure 4.98: Boxplot of slow – fast rate values of DS females and TD females

4.4.3.12 Other features

A Chi Square test found the females with DS to have reduced respiratory support in comparison with their TD female peers (table 4.105).

OTHER FEATURES: Chi Square results for DS females (9) and TD females (18)	
	statistical significance
respiratory support	$\chi^2 = 6.35$ df = 1 p = 0.012
diplophonia	ns

Table 4.105: Results of Chi Square tests for respiratory support and diplophonia features of DS females and TD females

4.4.4 Results between DS female and male speakers

4.4.4.1 Labial settings

No significant differences were found between the females with DS and males with DS in any of the labial settings of the VPAS (table 4.106).

LABIAL SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) spreading – rounding/protrusion (+)	1.5 (2)	0.5 (3)	ns
labiodentalisation	0 (1)	0 (0)	ns
(-) minimised - extensive (+) range	-1.5 (2)	-2 (3)	ns

Table 4.106: Results of Mann Whitney U-tests and median (IQR) values for labial settings of DS females and DS males

4.4.4.2 Mandibular settings

No significant differences were found in Mann Whitney U-tests between the two groups for the mandibular settings (table 4.107).

MANDIBULAR SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (12)**	**statistical significance**
(-) open - close jaw (+)	-0.5 (1)	-1 (2)	ns
protruded jaw	2 (2)	3 (1)	ns
(-) minimised – extensive (+) range	0 (1)	0 (1)	ns

Table 4.107: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of DS females and DS males

4.4.4.3 Lingual settings

Table 4.108 shows significant results between the DS females and DS males for two of the four lingual settings. Females with DS were found to have significantly more fronted tongue position and a less minimised tongue range than the males with DS. Values are shown as boxplots in figures 4.99 and 4.100.

LINGUAL SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (12)**	**statistical significance**
(-) retracted – advanced (+) tip	3 (2)	3 (2)	ns
(-) backed - fronted (+) body	3 (1)	0 (4)	n = 18 (F), 24 (M) $U = 67.0$ $p < 0.001$
(-) lowered - raised (+) body	2 (1)	1.5 (1)	ns
(-) minimised – extensive (+) range	-2 (1)	-3 (2)	n = 18 (F), 24 (M) $U = 145.5$ $p = 0.029$

Table 4.108: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of DS females and DS males

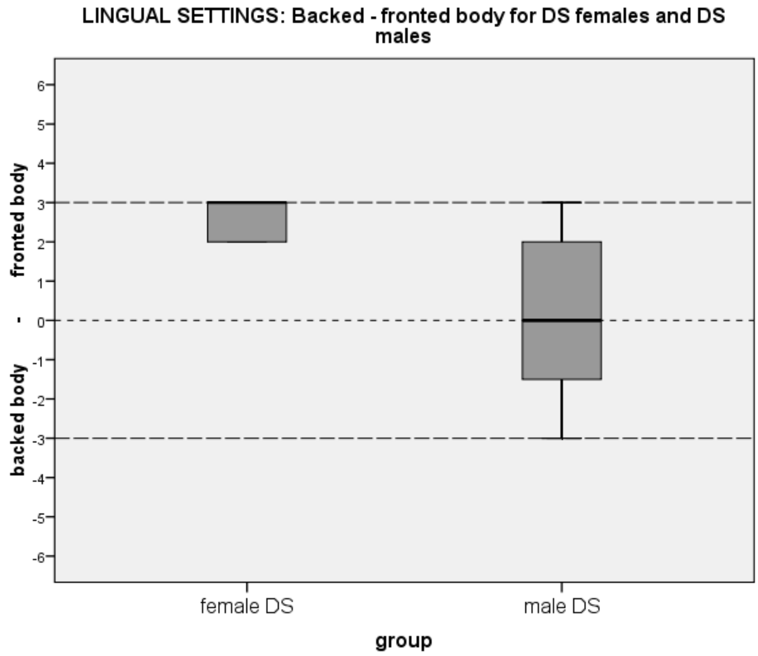


Figure 4.99: Boxplot of backed – fronted tongue body values of DS females and DS males

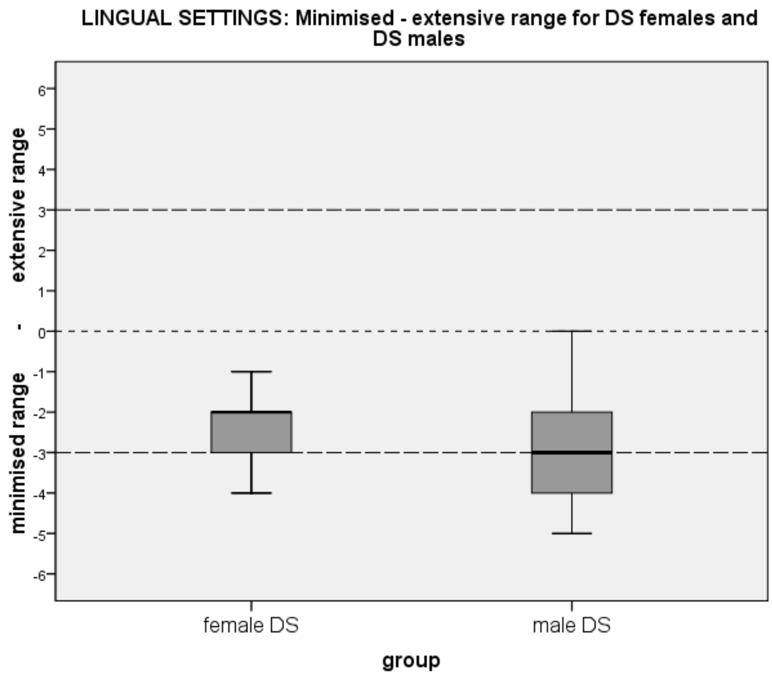


Figure 4.100: Boxplot of minimised – extensive tongue range values of DS females and DS males

4.4.4.4 Pharyngeal settings

No significant difference was found in the pharyngeal setting of females and males with DS (table 4.109).

PHARYNGEAL SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) constriction – expansion (+)	-1 (1)	-1 (1)	ns

Table 4.109: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of DS females and DS males

4.4.4.5 Velopharyngeal settings

Mann Whitney U-tests revealed statistically significant differences between DS females and males in both the velopharyngeal settings, with DS males having significantly higher levels of audible nasal escape and higher nasality (table 4.110, figure 4.101 and 4.102).

VELOPHARYNGEAL SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
audible nasal escape	0 (0)	0 (1)	n = 18 (F), 26 (M) U = 169.0 p = 0.045
(-) denasal – nasal (+)	3 (0)	4 (1)	n = 18 (F), 26 (M) U = 136.0 p = 0.015

Table 4.110: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of DS females and DS males

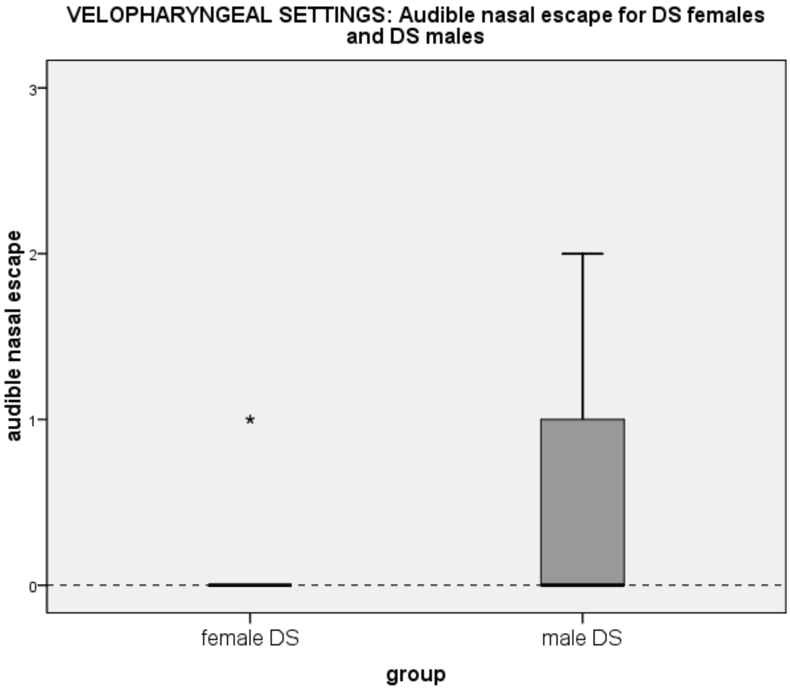


Figure 4.101: Boxplot of audible nasal escape values of DS females and DS males

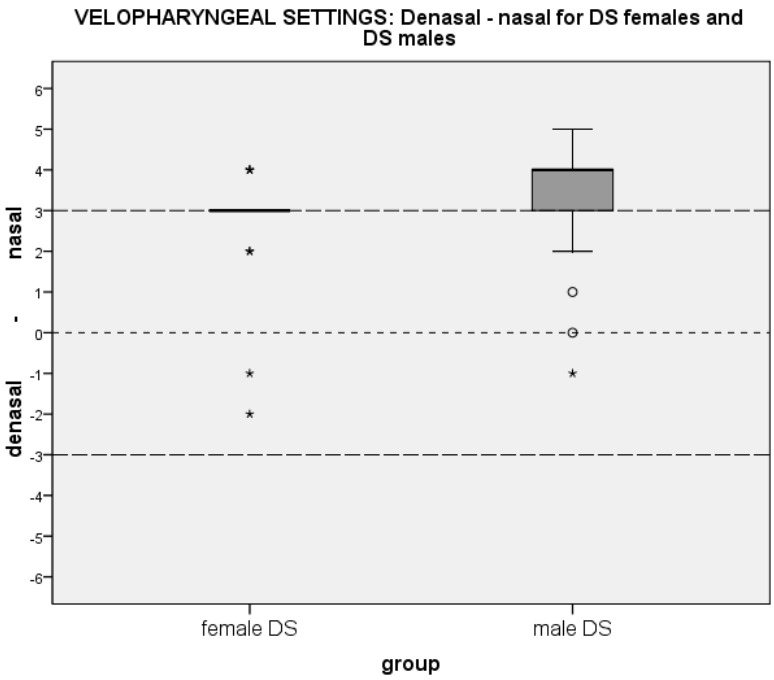


Figure 4.102: Boxplot of denasal – nasal values of DS females and DS males

4.4.4.6 Larynx height settings

No significant difference was found in larynx height between the DS females and DS males (table 4.111).

LARYNX HEIGHT SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) lowered – raised (+) larynx	0 (1)	0 (3)	ns

Table 4.111: Results of Mann Whitney U-tests and median (IQR) values for larynx height settings of DS females and DS males

4.4.4.7 Tension settings

A significant difference in a Mann Whitney U-test was found between the two groups in vocal tract tension, with males with DS being rated as having significantly more lax larynx (table 4.112 and figure 4.103).

MUSCULAR TENSION SETTINGS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (0)	0 (1)	n = 18 (F), 26 (M) U = 158.0 p = 0.05
(-) lax – tense (+) larynx	2 (2)	0.5 (2)	ns

Table 4.112: Results of Mann Whitney U-tests and median (IQR) values for tension settings of DS females and DS males

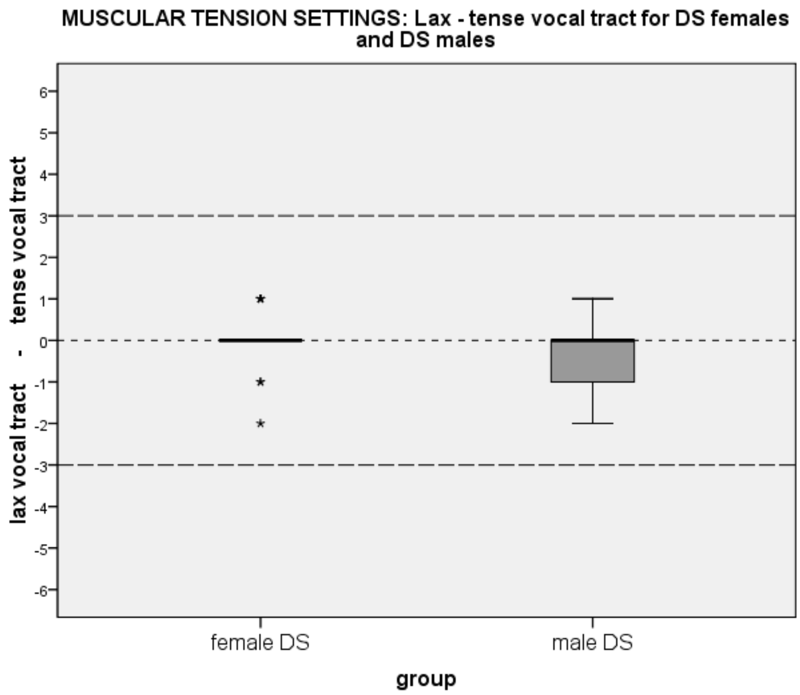


Figure 4.103: Boxplot of lax – tense vocal tract values of DS females and DS males

4.4.4.8 Phonation features

No significant differences were found between the phonation features of females and males with DS (table 4.113).

PHONATION FEATURES: Mann Whitney and Chi Square* results for DS females (F) and DS males (M)				
		F (9)	**M (13)**	**statistical significance**
voicing type	**neutral - non neutral voice**	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (0)	0 (0)	ns
laryngeal frication	**whisper**	n/a	n/a	ns*
	whispery	3 (1)	3 (1)	ns
laryngeal irregularity	**harsh**	1 (2)	1 (1)	ns
	tremor	0 (0)	0 (0)	ns

Table 4.113: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of DS females and DS males

4.4.4.9 Pitch

No significant differences were found between the two groups for the pitch section of the VPAS (table 4.114).

PITCH: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) low – high (+) mean	-0.5 (3)	-2 (3)	ns
(-) minimised - extensive (+) range	0 (2)	-1 (4)	ns
(-) low - high (+) variability	0 (1)	0 (3)	ns

Table 4.114: Results of Mann Whitney U-tests and median (IQR) values for pitch features of DS females and DS males

4.4.4.10 Loudness

Table 4.115 shows a significant difference in a Mann Whitney U-test in loudness variability between DS females and males, with males having increased variability. Values are shown as a boxplot in figure 4.104.

LOUDNESS: Mann Whitney results for DS females (F) and DS males (M)			
	F (9)	**M (13)**	**statistical significance**
(-) low - high (+) mean	0 (2)	0 (2)	ns
(-) minimised – extensive (+) range	0 (0)	0 (2)	ns
(-) low - high (+) variability	0 (1)	0 (2)	n = 18 (F), 26 (M) U = 123.5 p = 0.003

Table 4.115: Results of Mann Whitney U-tests and median (IQR) values for loudness features of DS females and DS males

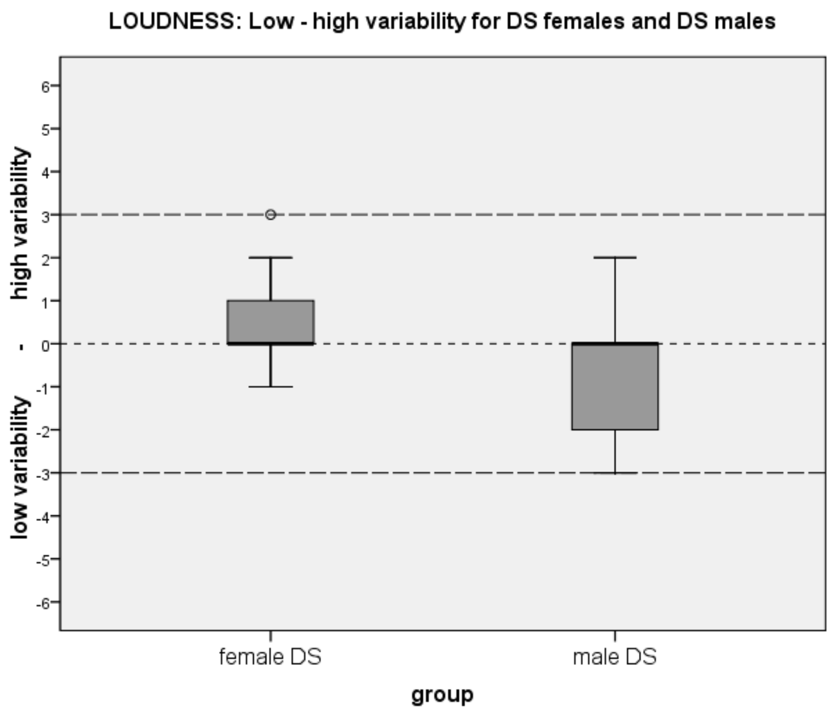


Figure 4.104: Boxplot of low – high loudness variability values of DS females and DS males

4.4.4.11 Temporal organization

No significant difference was found between DS females and males for the temporal organization features of continuity and rate (table 4.116).

TEMPORAL ORGANISATION: Mann Whitney results for DS females (F) and DS males (M)				
		F (9)	**M (13)**	**statistical significance**
continuity	**interrupted**	2 (2)	1 (2)	ns
rate	**(-) slow – fast (+)**	-1 (2)	0 (1)	ns

Table 4.116: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of DS females and DS males

4.4.4.12 Other features

No significant differences were found in the respiratory support or diplophonia ratings of the DS females and DS males (table 4.117).

OTHER FEATURES: Chi Square results for DS females (9) and DS males (13)	
	statistical significance
respiratory support	ns
diplophonia	ns

Table 4.117: Results of Chi Square tests for respiratory support and diplophonia features of DS females and DS males

4.4.5 Results between TD female and male speakers

4.4.5.1 Labial settings

Mann Whitney U-tests revealed significant differences between TD females and males in two of the three labial settings, TD females having significantly more spread lips and TD males having more minimised range of lip movements (table 4.118 and figures 4.105 and 4.106).

LABIAL SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) spreading - rounding/protrusion (+)	-0.5 (1)	0 (1)	n = 36 (F), 68 (M) $U = 549.5$ $p < 0.001$
labiodentalisation	0 (0)	0 (0)	ns
(-) minimised – extensive (+) range	0 (0)	0 (1)	n = 36 (F), 68 (M) $U = 783.5$ $p < 0.001$

Table 4.118: Results of Mann Whitney U-tests and median (IQR) values for labial settings of TD females and TD males

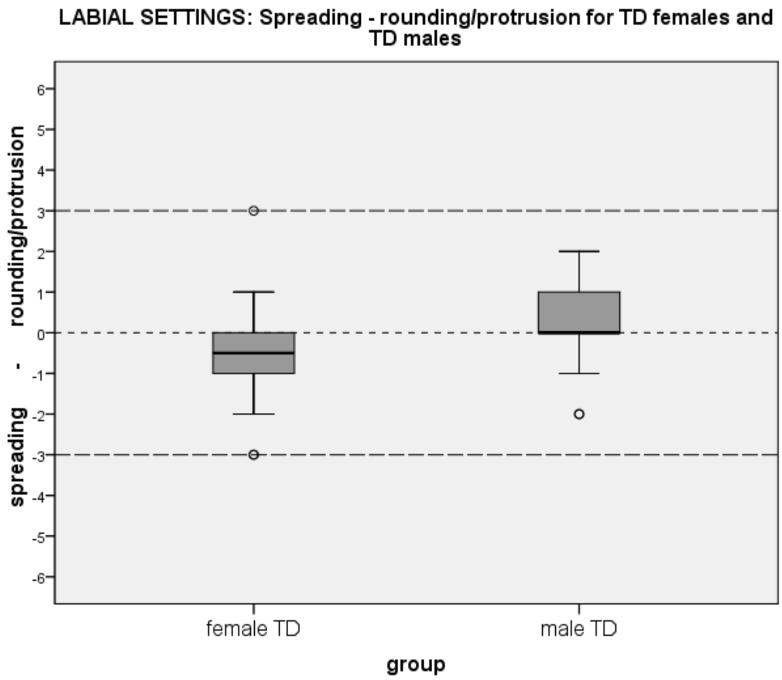


Figure 4.105: Boxplot of lip spreading – rounding/protrusion values of TD females and TD males

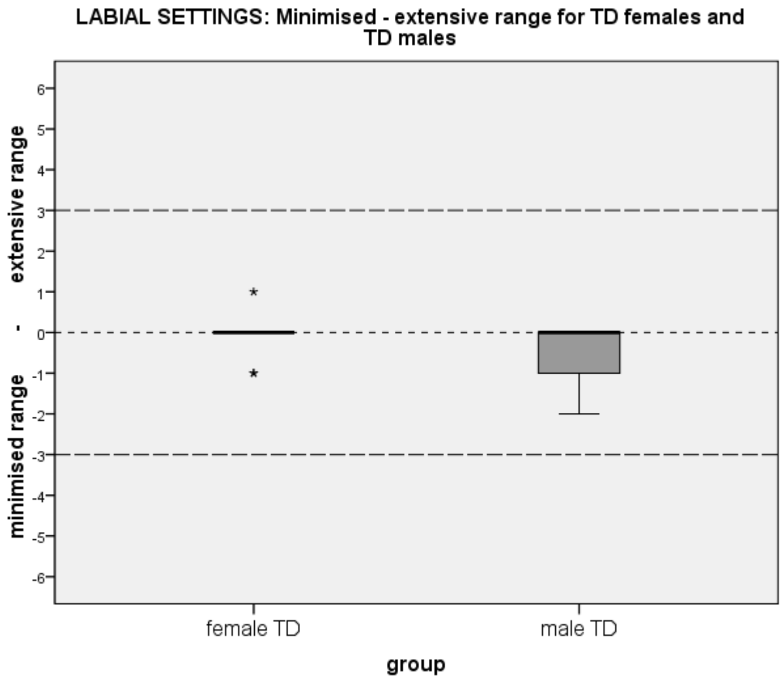


Figure 4.106: Boxplot of minimised – extensive lip range values of TD females and TD males

4.4.5.2 Mandibular settings

Table 4.119 shows significant differences in the jaw settings of TD females and males with males having a more close jaw setting and a more minimised range of jaw movements. Values are illustrated as boxplots in figures 4.107 and 4.108.

MANDIBULAR SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) open - close (+) jaw	0 (0)	0 (1)	n = 36 (F), 68 (M) *U* = 666.0 p < 0.001
protruded jaw	0 (0)	0 (0)	ns
(-) minimised - extensive (+) range	0 (0)	0 (0)	n = 36 (F), 68 (M) *U* = 1062.0 p = 0.023

Table 4.119: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of TD females and TD males

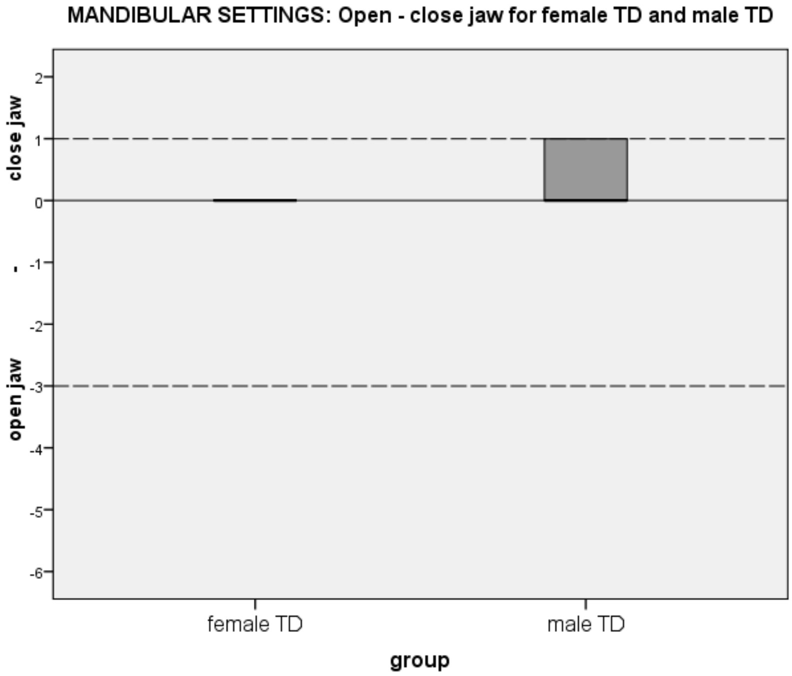


Figure 4.107: Boxplot of open – close jaw values of TD females and TD males

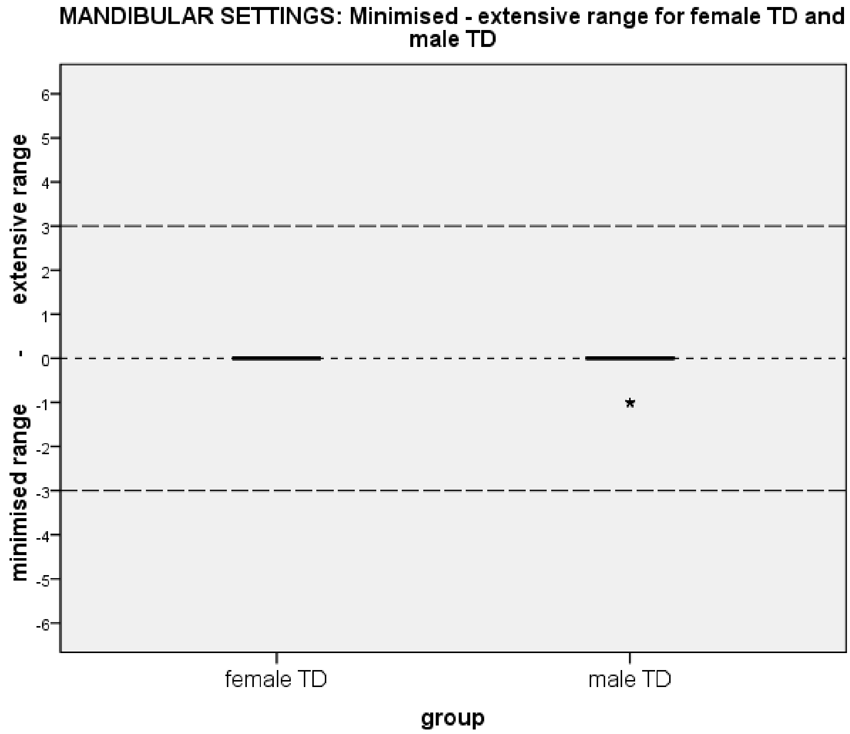


Figure 4.108: Boxplot of minimised – extensive jaw range values of TD females and TD males

4.4.5.3 Lingual settings

Significant differences in Mann Whitney U-tests were found between the TD females and males in all of the lingual settings of the VPAS (table 4.120). Females were found to have significantly more advanced tongue tip and fronted and raised tongue body, whilst males had a significantly more minimised range. Values are shown as boxplots in figures 4.109-4.112.

LINGUAL SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) retracted – advanced (+) tip	1 (1)	0 (1)	n = 36 (F), 68 (M) *U* = 412.0 $p < 0.001$
(-) backed - fronted (+) body	1 (2)	-1 (1)	n = 36 (F), 68 (M) *U* = 264.0 $p < 0.001$
(-) lowered - raised (+) body	1 (1)	0 (1)	n = 36 (F), 68 (M) *U* = 650.0 $p < 0.001$
(-) minimised – extensive (+) range	0 (0)	0 (0)	n = 36 (F), 68 (M) *U* = 1026.0 $p = 0.011$

Table 4.120: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of TD females and TD males

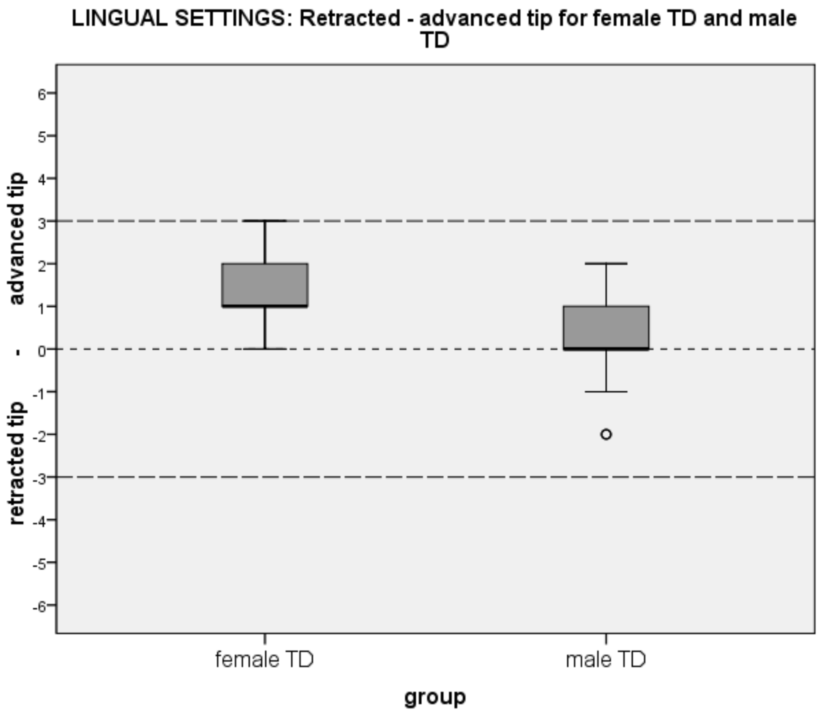


Figure 4.109: Boxplot of retracted – advanced tongue tip values of TD females and TD males

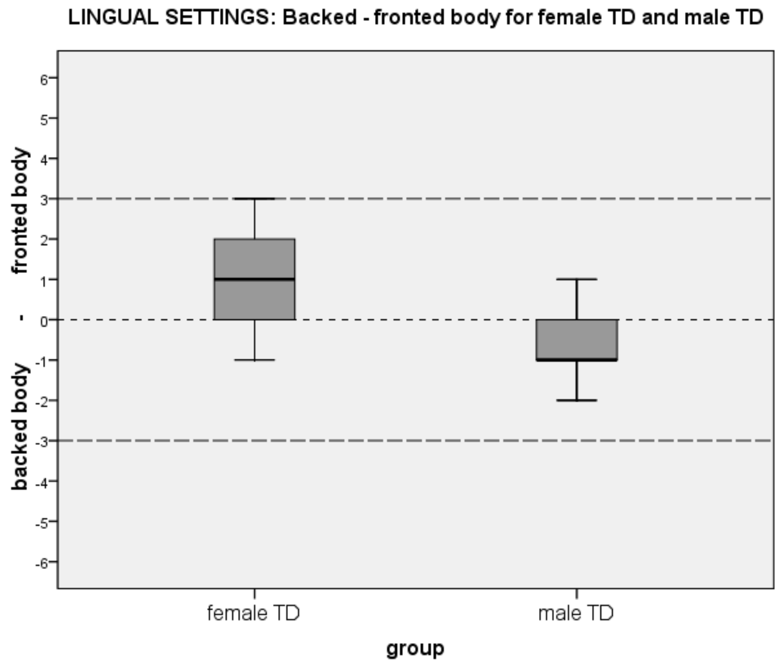


Figure 4.110: Boxplot of backed – fronted tongue body values of TD females and TD males

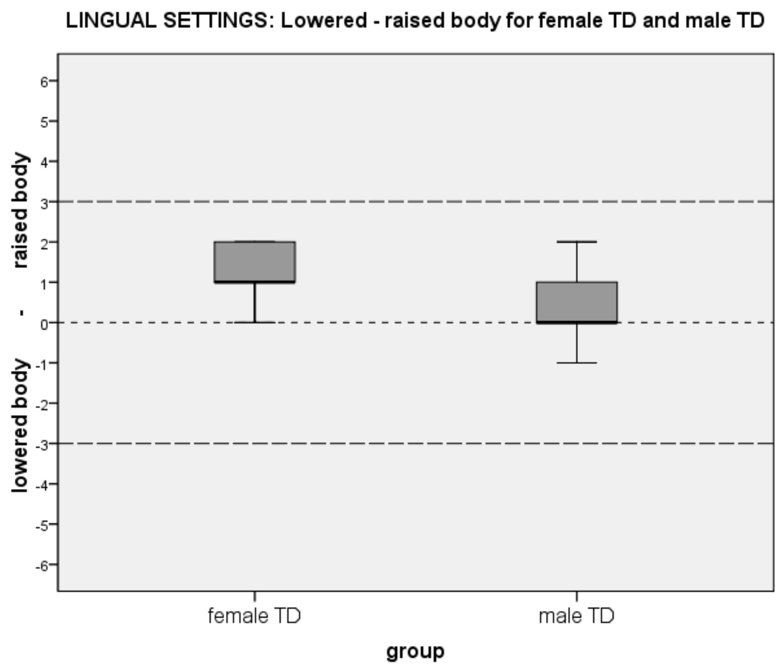


Figure 4.111: Boxplot of lowered – raised tongue body values of TD females and TD males

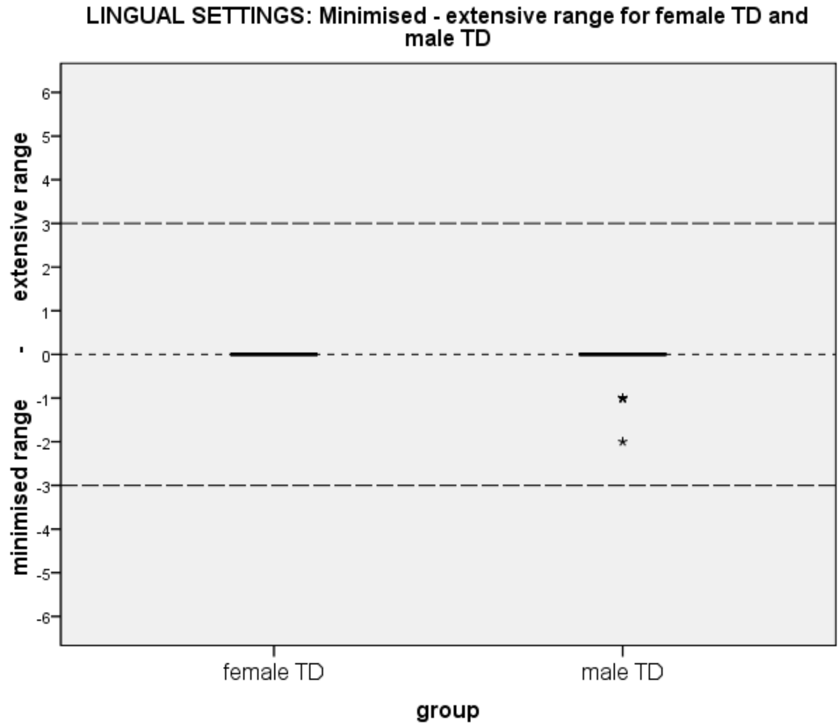


Figure 4.112: Boxplot of minimised – extensive tongue range values of TD females and TD males

4.4.5.4 <u>Pharyngeal settings</u>

No significant difference was found between the two groups for the pharyngeal settings of the VPAS (table 4.121).

PHARYNGEAL SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) constriction – expansion (+)	0 (0)	0 (0)	ns

Table 4.121: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of TD females and TD males

4.4.5.5 Velopharyngeal settings

A Mann Whitney U-test identified significantly greater levels of nasality in TD males in comparison to TD females (table 4.122 and figure 4.113).

VELOPHARYNGEAL SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
audible nasal escape	0 (0)	0 (0)	ns
(-) denasal – nasal (+)	1 (1)	2 (1)	n = 36 (F), 64 (M) *U* = 668.0 $p < 0.001$

Table 4.122: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of TD females and TD males

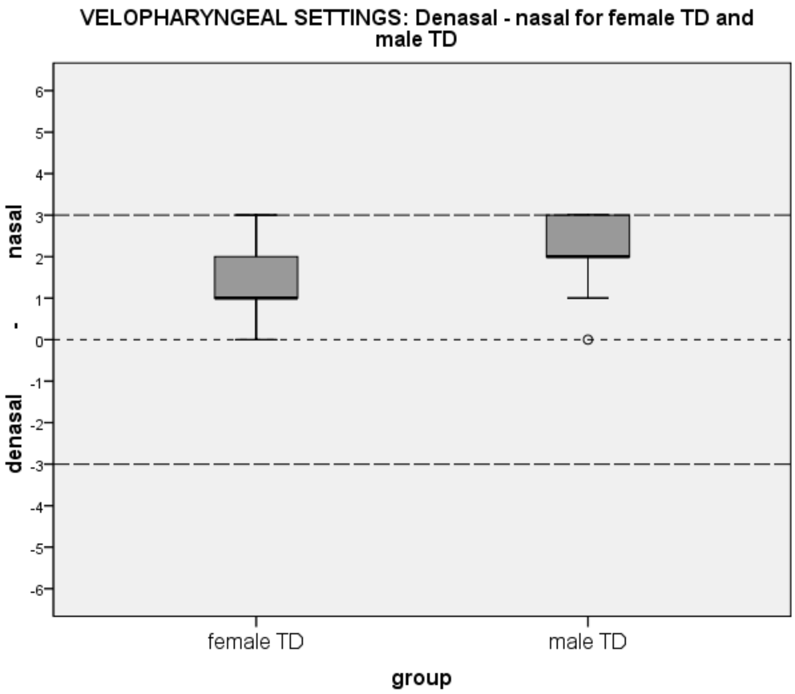


Figure 4.113: Boxplot of denasal – nasal values of TD females and TD males

4.4.5.6 Larynx height settings

A significant difference was found in a Mann Whitney U-test for laryngeal height between TD females and males with males having a significantly lower larynx position (table 4.123 and figure 4.114).

LARYNX HEIGHT SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) lowered – raised (+) larynx	0 (1)	0 (1)	n = 36 (F), 64 (M) *U* = 668.0 p < 0.001

Table 4.123: Results of Mann Whitney U-tests and median (IQR) values for larynx height settings of TD females and TD males

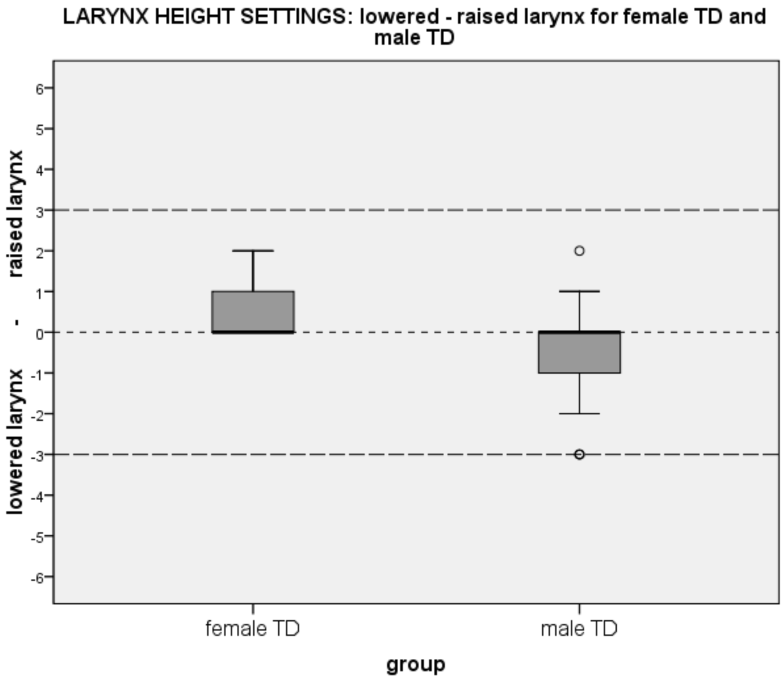


Figure 4.114: Boxplot of lowered – raised larynx values of TD females and TD males

4.4.5.7 Tension settings

No significant differences were found between the muscular tension settings of the TD females and males (table 4.124).

MUSCULAR TENSION SETTINGS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (0)	0 (0)	ns
(-) lax – tense (+) larynx	1 (1)	1 (1)	ns

Table 4.124: Results of Mann Whitney U-tests and median (IQR) values for muscular tension settings of TD females and TD males

4.4.5.8 Phonation features

According to Mann Whitney U-tests significant differences in creaky and whispery phonation were found between TD females and males, with females having significantly less creaky phonation and significantly more whispery phonation (table 4.125) than males. Values are illustrated in figures 4.115 and 4.116.

PHONATION FEATURES: Mann Whitney and Chi Square* results for TD females (F) and TD males (M)				
		F (18)	**M (34)**	**statistical significance**
voicing type	**neutral - non neutral voice**	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (0)	0 (2)	n = 36 (F), 64 (M) $U = 796.0$ $p < 0.001$
laryngeal frication	**whisper**	n/a	n/a	ns*
	whispery	2 (1)	2 (0.5)	n = 36 (F), 64 (M) $U = 836.0$ $p = 0.003$
laryngeal irregularity	**harsh**	1 (1)	0 (1)	ns
	tremor	0 (0)	0 (0)	ns

Table 4.125: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of TD females and TD males

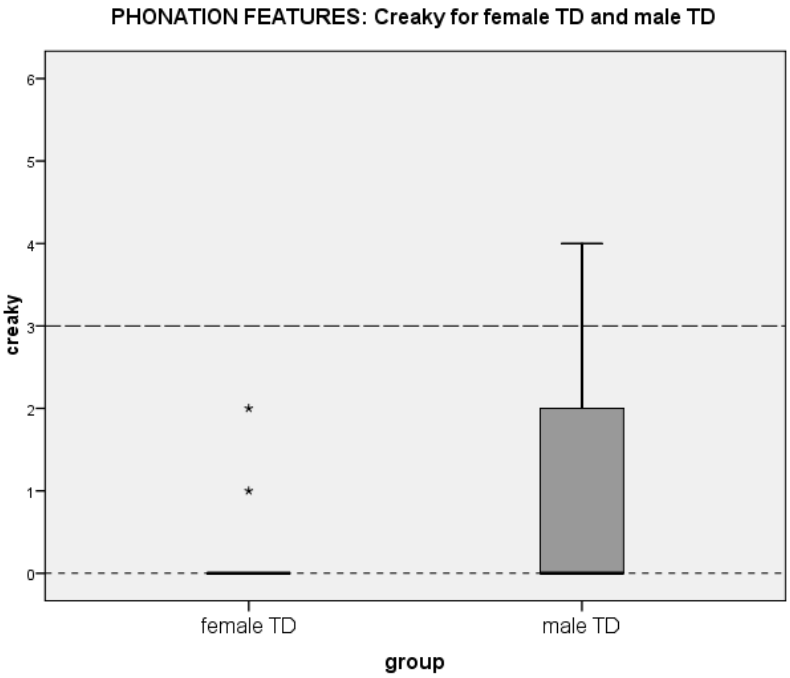


Figure 4.115: Boxplot of creaky phonation values of TD females and TD males

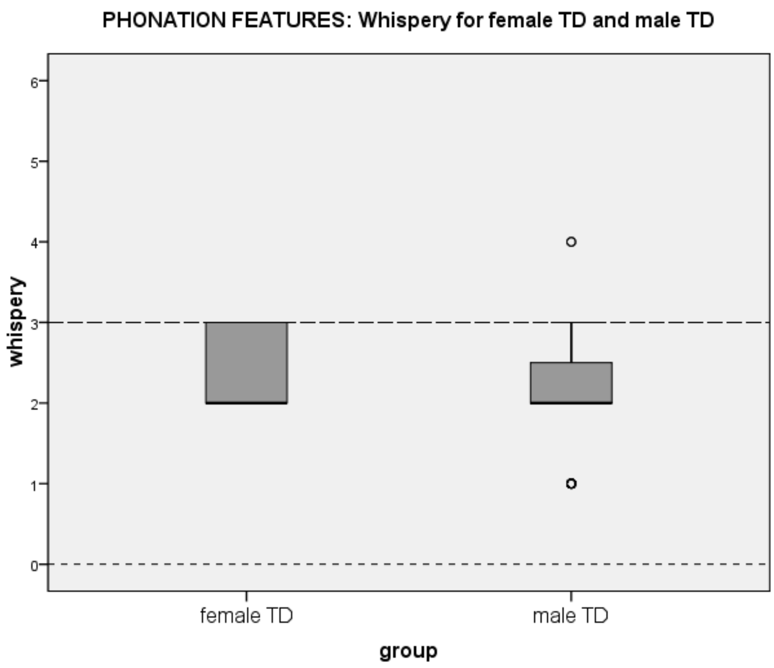


Figure 4.116: Boxplot of whispery phonation values of TD females and TD males

4.4.5.9 Pitch

Significant differences between TD females and males in all the pitch measures of the VPAS were found in Mann Whitney U-tests (table 4.126). Values are illustrated in figures 4.117-4.119.

PITCH: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) low – high (+) mean	0 (1)	0 (1)	n = 36 (F), 68 (M) *U* = 701.0 p < 0.001
(-) minimised - extensive (+) range	0 (0)	0 (1)	n = 36 (F), 68 (M) *U* = 1011.5 p = 0.05
(-) low - high (+) variability	0 (0)	0 (1)	n = 36 (F), 68 (M) *U* = 999.0 p = 0.043

Table 4.126: Results of Mann Whitney U-tests and median (IQR) values for pitch features of TD females and TD males

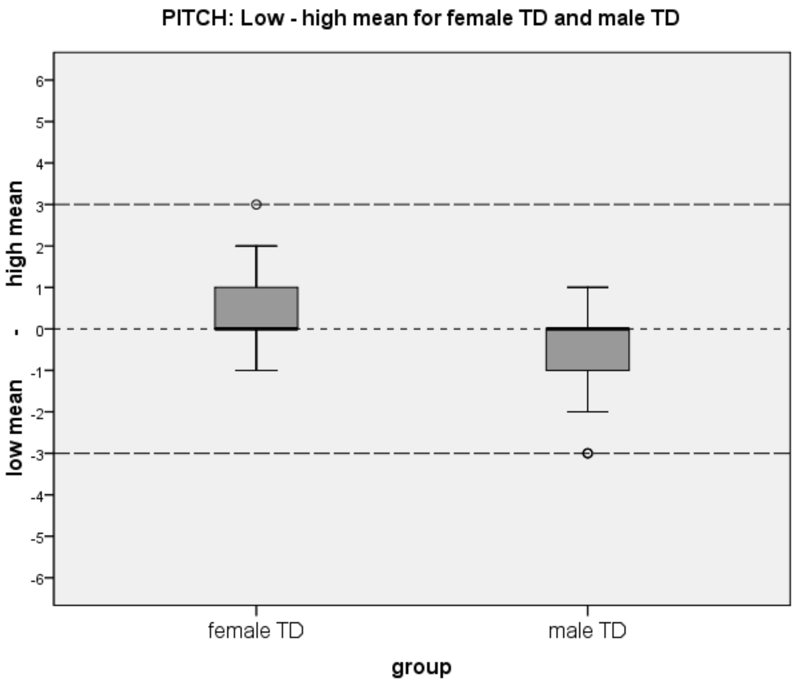


Figure 4.117: Boxplot of low – high mean pitch values of TD females and TD males

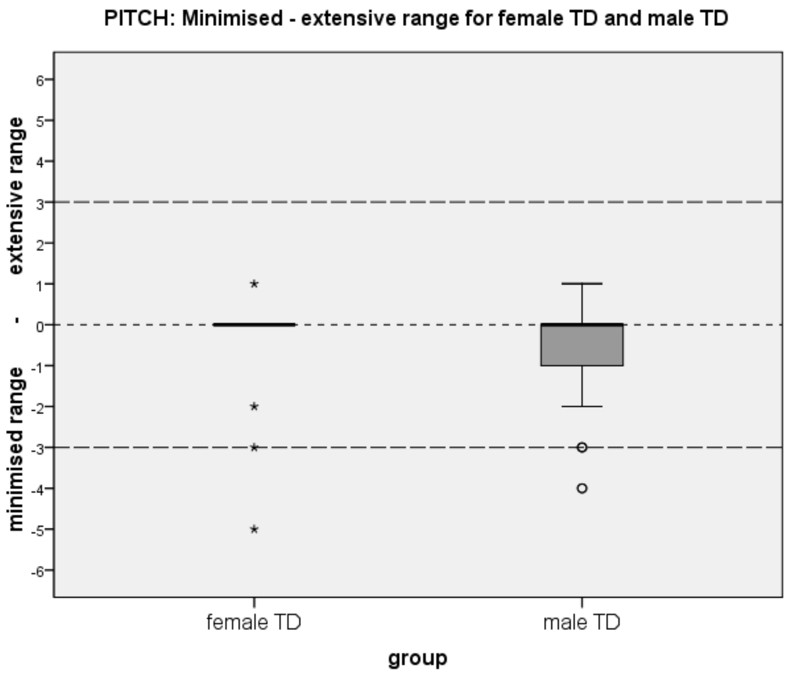


Figure 4.118: Boxplot of minimised – extensive pitch range values of TD females and TD males

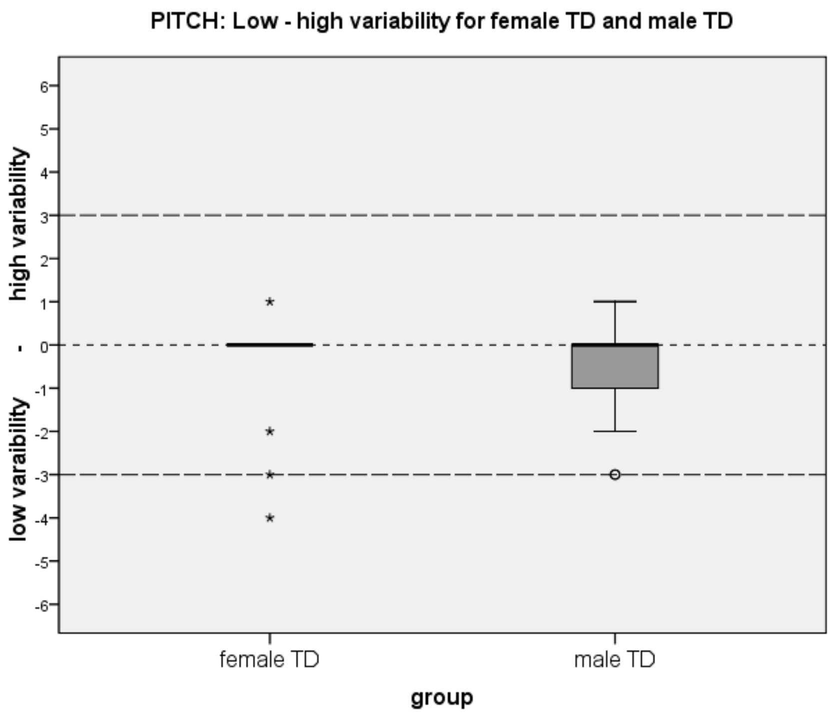


Figure 4.119: Boxplot of low – high pitch variability values of TD females and TD males

4.4.5.10 Loudness

No significant differences were found in measures of loudness between the TD females and males (table 4.127).

LOUDNESS: Mann Whitney results for TD females (F) and TD males (M)			
	F (18)	**M (34)**	**statistical significance**
(-) low - high (+) mean	0 (0)	0 (0)	ns
(-) minimised – extensive (+) range	0 (0)	0 (0)	ns
(-) low - high (+) variability	0 (0)	0 (0)	ns

Table 4.127: Results of Mann Whitney U-tests and median (IQR) values for loudness features of TD females and TD males

4.4.5.11 Temporal organization

No significant differences were found between the two groups for continuity and rate (table 4.128).

TEMPORAL ORGANISATION: Mann Whitney results for TD females (F) and TD males (M)				
		F (18)	**M (34)**	**statistical significance**
continuity	**interrupted**	0 (0)	0 (0)	ns
rate	**(-) slow – fast (+)**	0 (0)	0 (0)	ns

Table 4.128: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of TD females and TD males

4.4.5.12 Other features

No significant differences were found between the TD females and males for respiratory support or ratings of diplophonia (table 4.129).

OTHER FEATURES: Chi Square results for TD females (18) and TD males (34)	
	statistical significance
respiratory support	ns
diplophonia	ns

Table 4.129: Results of Chi Square tests for respiratory support and diplophonia features of TD females and TD males

4.4.6 Results between DS and TD speakers presented in study 1

4.4.6.1 Labial settings

Table 4.130 shows the results of Mann Whitney U-tests showing significant differences between the DS and TD speakers from study 1 for labial settings. The DS speakers have significantly more rounded/protruded lip pattern and significantly more minimised lip range. Values are illustrated as boxplots in figures 4.120 and 4.121.

LABIAL SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) spreading - rounding/protrusion (+)	2 (3)	0 (3)	n = 16 (DS), 16 (TD) $U = 77.5$ $p = 0.05$
labiodentalisation	0 (2)	0 (1)	ns
(-) minimised – extensive (+) range	-2 (3)	-0.5 (1)	n = 16 (DS), 16 (TD) $U = 71$ $p = 0.026$

Table 4.130: Results of Mann Whitney U-tests and median (IQR) values for labial settings of DS and TD speakers from study 1

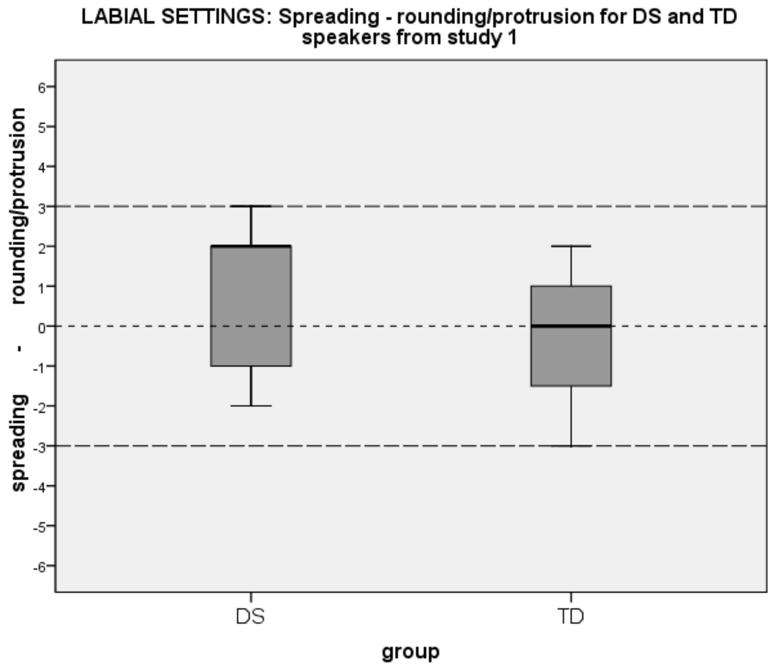


Figure 4.120: Boxplot of lip spreading – rounding/protrusion values of DS and TD speakers from study 1

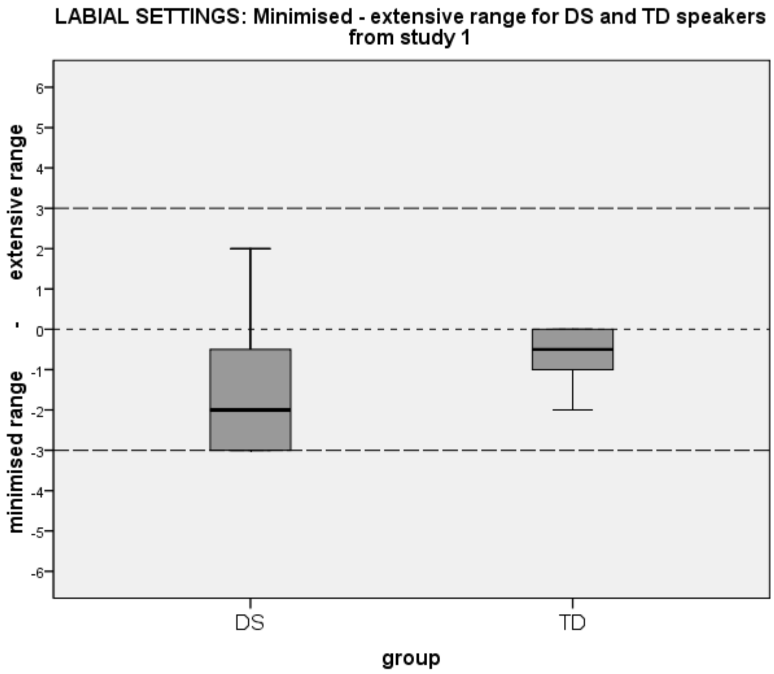


Figure 4.121: Boxplot of minimised – extensive lip range values of DS and TD speakers from study 1

4.4.6.2 Mandibular settings

Results of Mann Whitney U-tests for mandibular ratings are shown in table 4.131 and illustrated in figure 4.122 and 4.123. The DS speakers were shown to have significantly more open jaw and more protruded jaw than the TD speakers.

MANDIBULAR SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) open - close (+) jaw	-1 (1)	0 (1)	n = 16 (DS), 16 (TD) $U = 9.0$ $p < 0.001$
protruded jaw	3 (2)	0 (0)	n = 16 (DS), 16 (TD) $U = 20.0$ $p < 0.001$
(-) minimised - extensive (+) range	0 (1)	0 (0)	ns

Table 4.131: Results of Mann Whitney U-tests and median (IQR) values for mandibular settings of DS and TD speakers from study 1

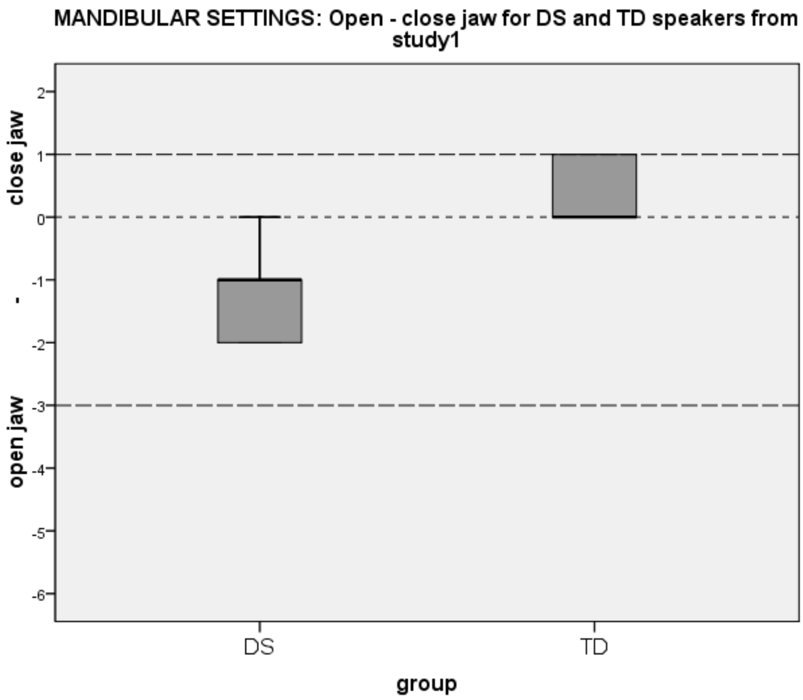


Figure 4.122: Boxplot of open – close jaw values of DS and TD speakers from study 1

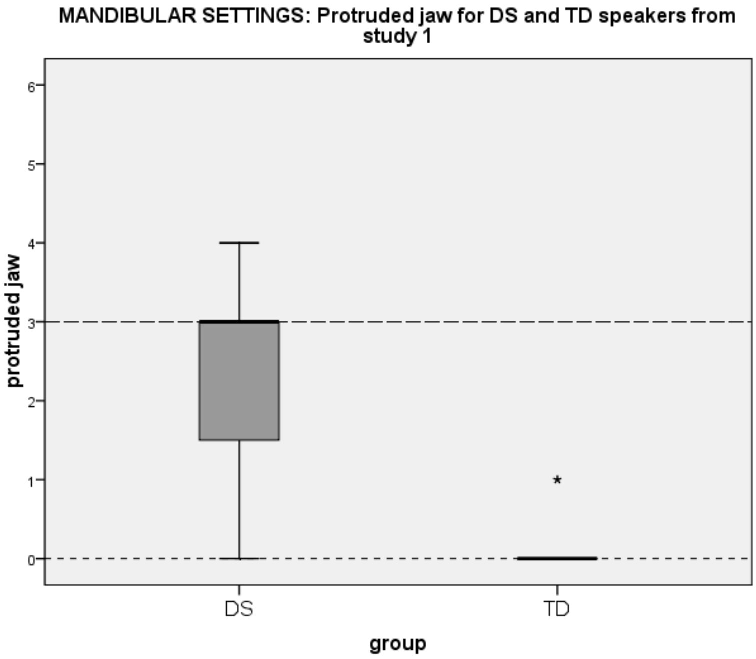


Figure 4.123: Boxplot of protruded jaw values of DS and TD speakers from study 1

4.4.6.3 Lingual settings

Statistically significant results from Mann Whitney U-tests are shown in table 4.132 and illustrated in figures 4.124 and 4.125. The DS speakers have significantly more fronted tongue tip and more minimised tongue range than the TD speakers.

LINGUAL SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) retracted – advanced (+) tip	3 (2)	1 (2)	n = 16 (DS), 16 (TD) $U = 19.0$ $p < 0.001$
(-) backed - fronted (+) body	2 (5)	0 (2)	ns
(-) lowered - raised (+) body	1 (1)	1 (1)	ns
(-) minimised – extensive (+) range	-2.5 (4)	0 (1)	n = 16 (DS), 16 (TD) $U = 44.0$ $p = 0.001$

Table 4.132: Results of Mann Whitney U-tests and median (IQR) values for lingual settings of DS and TD speakers from study 1

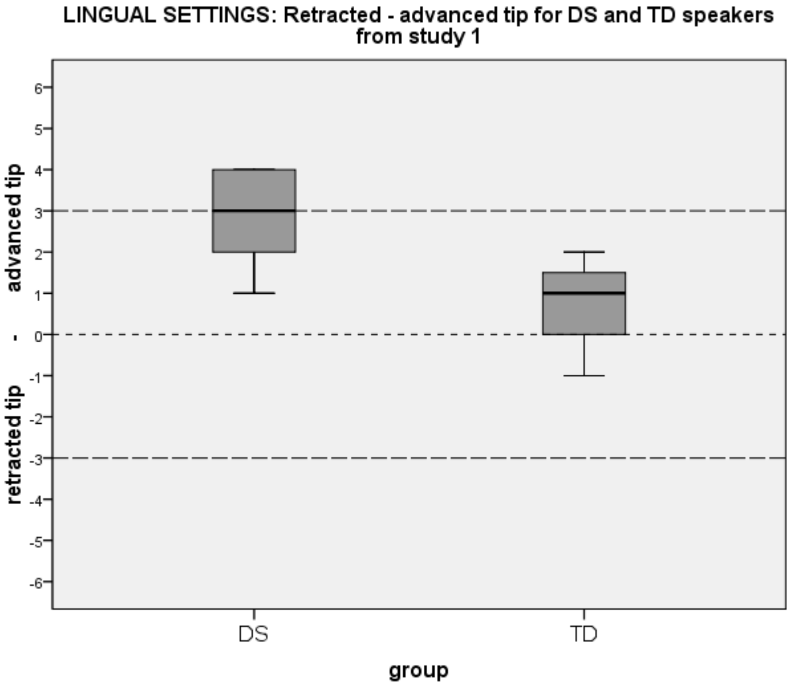


Figure 4.124: Boxplot of retracted – advanced tongue tip values of DS and TD speakers from study 1

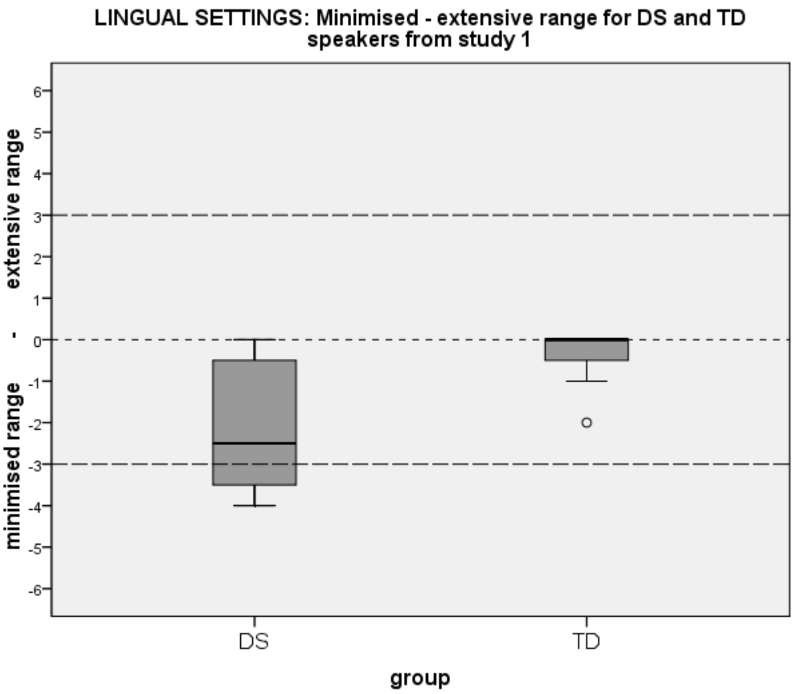


Figure 4.125: Boxplot of minimised – extensive tongue range values of DS and TD speakers from study 1

4.4.6.4 Pharyngeal settings

A significant difference in a Mann Whitney U-test for pharyngeal constriction – expansion was found between the DS and TD speakers, with the DS speakers having significantly more constriction (table 4.133 and figure 4.126).

PHARYNGEAL SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) constriction – expansion (+)	-1 (1)	0 (0)	n = 16 (DS), 16 (TD) U = 39.0 $p < 0.001$

Table 4.133: Results of Mann Whitney U-tests and median (IQR) values for pharyngeal settings of DS and TD speakers from study 1

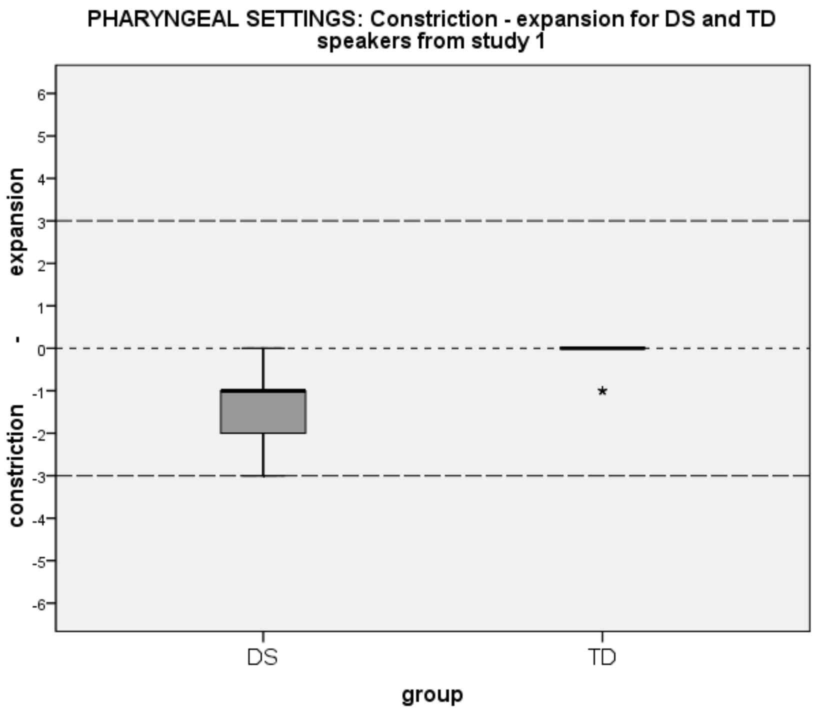


Figure 4.126: Boxplot of pharyngeal constriction – expansion values of DS and TD speakers from study 1

4.4.6.5 Velopharyngeal settings

Table 4.134 shows the results of Mann Whitney U-tests, where the DS speakers can be seen to have significantly higher levels of nasality than the TD speakers. Values are shown as boxplots in figure 4.127.

VELOPHARYNGEAL SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
audible nasal escape	0 (1)	0 (0)	ns
(-) denasal – nasal (+)	3.5 (3)	2 (2)	n = 16 (DS), 16 (TD) $U = 55.0$ $p < 0.005$

Table 4.134: Results of Mann Whitney U-tests and median (IQR) values for velopharyngeal settings of DS and TD speakers from study 1

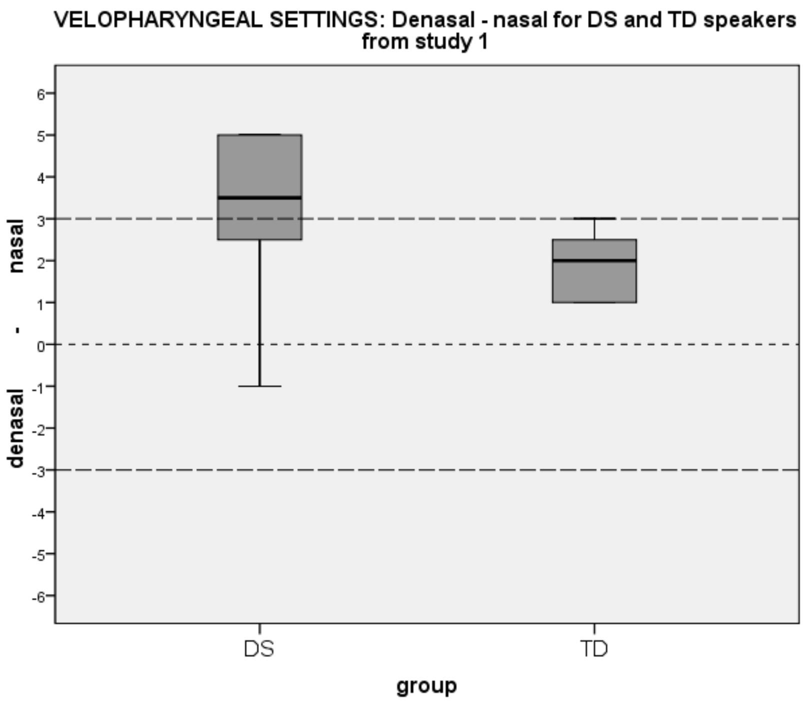


Figure 4.127: Boxplot of denasal – nasal values of DS and TD speakers from study 1

4.4.6.6 Larynx height settings

No significant difference was found between the larynx height ratings of the DS and TD speakers from study 1 (table 4.135).

LARYNX HEIGHT SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) lowered – raised (+) larynx	-1.5 (4)	0 (2)	ns

Table 4.135: Results of Mann Whitney U-tests and median (IQR) values for larynx height settings of DS and TD speakers from study 1

4.4.6.7 Tension settings

No significant differences were found in muscular tension between the DS and TD speakers (table 4.136).

MUSCULAR TENSION SETTINGS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) lax – tense (+) vocal tract	0 (1)	0 (0)	ns
(-) lax – tense (+) larynx	0 (2)	1 (1)	ns

Table 4.136: Results of Mann Whitney U-tests and median (IQR) values for tension settings of DS and TD speakers from study 1

4.4.6.8 Phonation features

Table 4.137 shows that the only significant result from Mann Whitney U-tests and Chi Square tests between the DS and TD speakers from study 1 is for whispery phonation, where the DS speakers have significantly higher levels of whisperiness. Values are shown as a boxplot in figure 4.128.

PHONATION FEATURES: Mann Whitney and Chi Square* results for DS and TD speakers from study 1				
		DS (8)	**TD (8)**	**statistical significance**
voicing type	**neutral - non neutral voice**	n/a	n/a	ns*
	falsetto	n/a	n/a	ns*
	creak	n/a	n/a	ns*
	creaky	0 (2)	0 (1)	ns
laryngeal frication	**whisper**	n/a	n/a	ns*
	whispery	3 (0)	2 (1)	n = 16 (DS), 16 (TD) $U = 62.5$ $p = 0.007$
laryngeal irregularity	**harsh**	1 (1)	1 (2)	ns
	tremor	0 (0)	0 (0)	ns

Table 4.137: Results of Mann Whitney U-tests (with median & IQR values) and Chi square tests for phonation features of DS and TD speakers from study 1

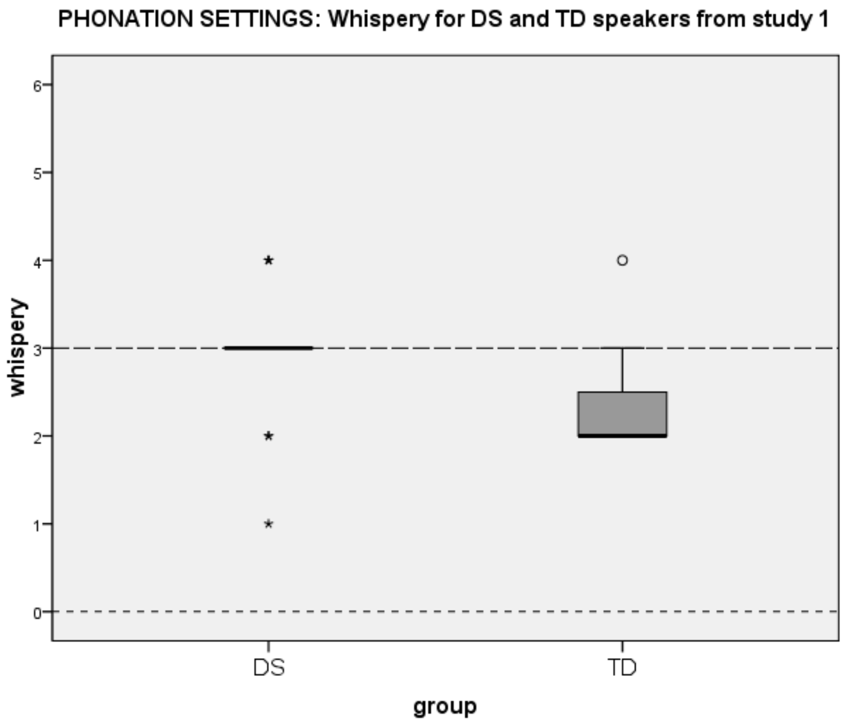


Figure 4.128: Boxplot of whispery phonation values of DS and TD speakers from study 1

4.4.6.9 Pitch

No significant differences were found between the groups for any of the pitch measures (table 4.138).

PITCH: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) low – high (+) mean	-2 (4)	0 (2)	ns
(-) minimised - extensive (+) range	-0.5 (2)	-0.5 (1)	ns
(-) low - high (+) variability	0 (4)	-0.5 (2)	ns

Table 4.138: Results of Mann Whitney U-tests and median (IQR) values for pitch features of DS and TD speakers from study 1

4.4.6.10 Loudness

No significant differences were found in measures of loudness between the DS and TD speakers (table 4.139).

LOUDNESS: Mann Whitney results for DS and TD speakers from study 1			
	DS (8)	**TD (8)**	**statistical significance**
(-) low - high (+) mean	0 (1)	0 (0)	ns
(-) minimised – extensive (+) range	0 (0)	0 (0)	ns
(-) low - high (+) variability	0 (1)	0 (1)	ns

Table 4.139: Results of Mann Whitney U-tests and median (IQR) values for loudness features of DS and TD speakers from study 1

4.4.6.11 Temporal organization

Table 4.140 shows the results of Mann Whitney statistical tests finding that the DS speakers have significantly greater levels of interrupted continuity and significantly slower rate than the TD speakers. Values are illustrated as boxplots in figures 4.129 and 4.130.

TEMPORAL ORGANIZATION: Mann Whitney results for DS and TD speakers from study 1				
		DS (8)	**TD (8)**	**statistical significance**
continuity	**interrupted**	1 (2)	0 (0)	n = 16 (DS), 16 (TD) *U* = 56.0 p = 0.001
rate	**(-) slow – fast (+)**	0 (1)	0 (0)	n = 16 (DS), 16 (TD) *U* = 78.5 p = 0.014

Table 4.140: Results of Mann Whitney U-tests and median (IQR) values for temporal organization features of DS and TD speakers from study 1

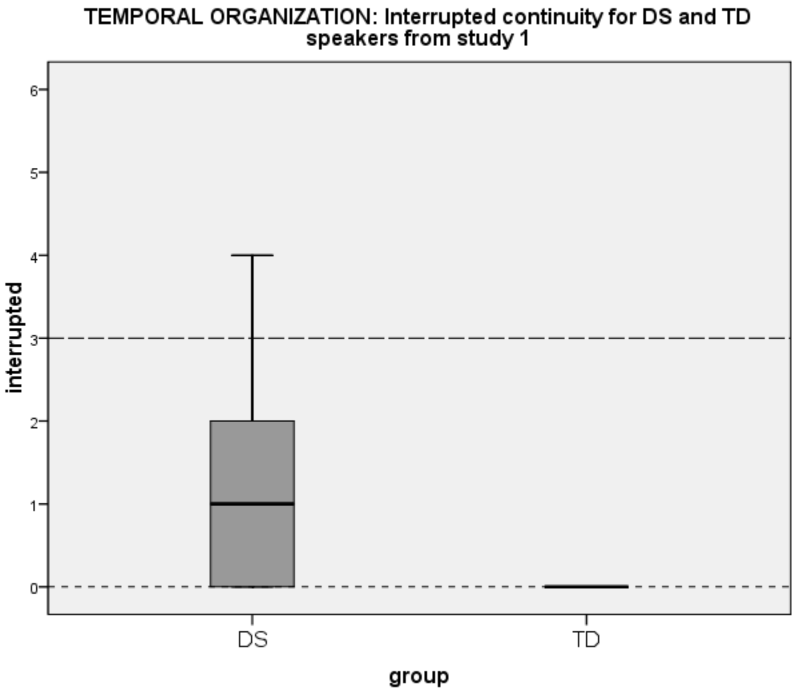


Figure 4.129: Boxplot of interrupted continuity values of DS and TD speakers from study 1

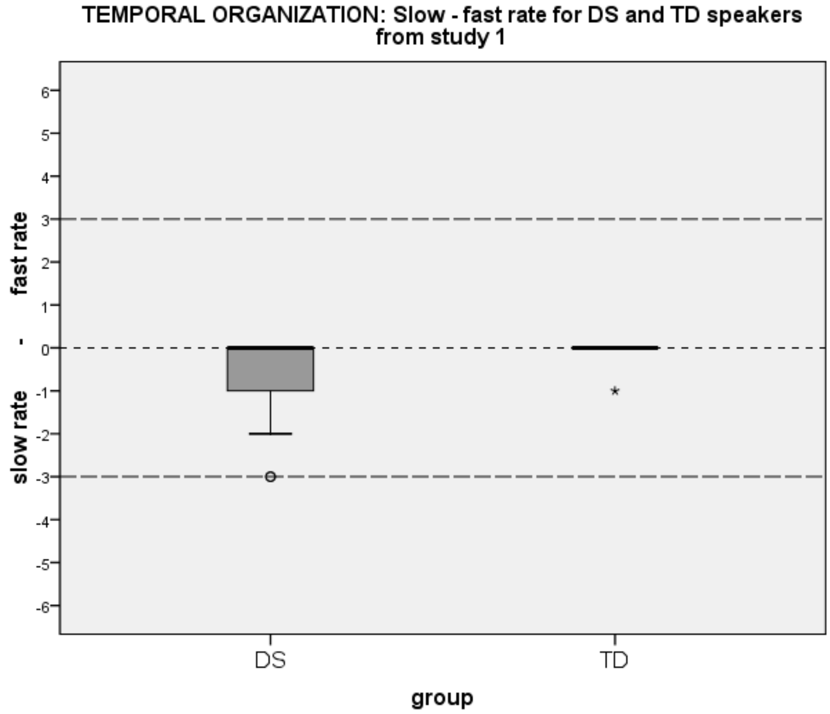


Figure 4.130: Boxplot of slow – fast rate values of DS and TD speakers from study 1

4.4.6.12 Other features

A Chi Square test identified a significant difference between the respiratory support of the DS and TD speakers, the DS speakers having reduced capacity (table 4.141).

OTHER FEATURES: Chi Square results for DS (8) and TD (8) speakers from study 1	
	statistical significance
respiratory support	$\chi^2 = 4.57$ df = 1 p = 0.033
diplophonia	ns

Table 4.141: Results of Chi Square tests for respiratory support and diplophonia features of DS and TD speakers from study 1

4.4.7 Variability between individual DS and TD speakers presented in study 1

4.4.7.1 Labial settings

The ratings by both raters for the individual DS and TD speakers from study 1 for all the labial settings are shown in table 4.142.

LABIAL SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
(-) spreading - rounding/ protrusion (+)	rater 1	1	-2	2	2	1	2	3	-1
	rater 2	2	-1	2	3	-2	2	2	-2
labio-dentalisation	rater 1	0	0	2	0	0	0	0	1
	rater 2	0	1	3	0	2	0	0	2
(-) minimised – extensive (+) range	rater 1	-2	1	-3	-2	-1	-2	-1	0
	rater 2	-3	2	-2	-3	0	-3	-3	0
		TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
(-) spreading - rounding/ protrusion (+)	rater 1	1	0	-2	-2	0	0	1	0
	rater 2	2	1	-2	-3	-1	0	2	0
labio-dentalisation	rater 1	0	0	1	0	0	0	0	0
	rater 2	1	1	3	1	0	0	0	0
(-) minimised – extensive (+) range	rater 1	0	0	0	0	0	0	-1	-1
	rater 2	-1	-1	0	0	-1	-1	-2	-2

Table 4.142: Labial ratings of individual DS and TD speakers from study 1

4.4.7.2 Mandibular settings

Table 4.143 shows the individual ratings for the DS and TD speakers for the mandibular section of the VPAS.

MANDIBULAR SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) open – close (+) jaw	**rater 1**	-2	-1	-2	-2	-2	-2	-1	0
	rater 2	-1	-1	-1	-1	-1	-2	-2	0
protruded jaw	**rater 1**	1	3	2	3	0	0	3	2
	rater 2	3	3	3	4	1	2	4	3
(-) minimised - extensive (+) range	**rater 1**	-1	-1	-1	-1	0	-1	-2	0
	rater 2	0	0	0	1	0	0	0	0
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) open – close (+) jaw	**rater 1**	0	1	0	0	1	0	1	1
	rater 2	1	0	0	0	1	0	0	1
protruded jaw	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	1	0	0	0	0	0	1	0
(-) minimised – extensive (+) range	**rater 1**	0	0	0	0	0	0	0	-1
	rater 2	0	0	0	0	0	0	0	-1

Table 4.143: Mandibular ratings of individual DS and TD speakers from study 1

4.4.7.3 Lingual settings

The individual lingual ratings for the DS and TD speakers from study 1 are shown in table 4.144.

LINGUAL SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
(-) retracted – advanced (+) tip	rater 1	4	3	2	3	1	3	3	2
	rater 2	4	4	3	4	1	3	4	2
(-) backed – fronted (+) body	rater 1	2	2	-2	3	2	3	-1	-1
	rater 2	2	3	-2	3	2	3	-2	-2
(-) lowered – raised (+) body	rater 1	1	2	2	1	2	1	3	1
	rater 2	0	1	1	0	2	0	2	2
(-) minimised – extensive (+) range	rater 1	-4	-1	-3	-4	0	-2	-3	0
	rater 2	-4	-2	-2	-4	0	-3	-3	0
		TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
(-) retracted – advanced (+) tip	rater 1	1	1	2	1	0	1	0	0
	rater 2	-1	-1	2	2	2	1	0	0
(-) backed – fronted (+) body	rater 1	-1	0	1	1	0	0	-1	0
	rater 2	-2	-1	1	2	0	1	-2	-1
(-) lowered – raised (+) body	rater 1	0	0	1	1	1	0	1	1
	rater 2	1	1	1	2	2	1	0	0
(-) minimised – extensive (+) range	rater 1	0	0	0	0	0	0	-1	0
	rater 2	-1	0	0	0	0	0	-2	-1

Table 4.144: Lingual ratings of individual DS and TD speakers from study 1

4.4.7.4 Pharyngeal settings

The individual ratings for pharyngeal constriction – expansion for the DS and TD speakers from study 1 are shown in table 4.145.

PHARYNGEAL SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) constriction – expansion (+)	**rater 1**	-1	-2	-3	-2	-1	-1	-2	-2
	rater 2	0	0	-1	-1	0	-1	-1	-2
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) constriction – expansion (+)	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	-1	0	-1	-1

Table 4.145: Pharyngeal ratings of individual DS and TD speakers from study 1

4.4.7.5 Velopharyngeal settings

Table 4.146 reports the individual velopharyngeal ratings of the DS and TD speakers from study 1.

VELOPHARYNGEAL SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
audible nasal escape	**rater 1**	0	0	5	0	4	0	4	0
	rater 2	0	0	4	0	4	0	4	0
(-) denasal – nasal (+)	**rater 1**	2	3	5	5	3	2	5	0
	rater 2	3	5	4	5	4	3	5	-1
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
audible nasal escape	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
(-) denasal – nasal (+)	**rater 1**	1	1	2	1	2	2	1	2
	rater 2	2	1	3	2	3	3	2	3

Table 4.146: Velopharyngeal ratings of individual DS and TD speakers from study 1

4.4.7.6 Larynx height settings

The individual ratings for lowered – raised larynx for the DS and TD speakers from study 1 are shown in table 4.147.

LARYNX HEIGHT SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) lowered - raised (+) larynx	**rater 1**	-2	3	0	-2	-2	1	-2	2
	rater 2	-3	2	-1	-2	-2	1	-2	2
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) lowered - raised (+) larynx	**rater 1**	-1	-1	1	1	0	1	-3	-1
	rater 2	-1	-1	1	0	2	0	-3	0

Table 4.147: Larynx height ratings of individual DS and TD speakers from study 1

4.4.7.7 Tension settings

Individual muscular tension ratings for the DS and TD speakers from study 1 are reported in table 4.148.

MUSCULAR TENSION SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) lax - tense (+) vocal tract	**rater 1**	-1	0	0	-2	0	0	0	1
	rater 2	-2	-1	0	-2	-1	0	-1	0
(-) lax - tense (+) larynx	**rater 1**	0	2	2	0	2	0	1	3
	rater 2	0	0	-1	0	0	1	0	3
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) lax - tense (+) vocal tract	**rater 1**	0	0	0	0	0	0	-1	-1
	rater 2	0	0	0	0	0	0	-2	0
(-) lax - tense (+) larynx	**rater 1**	0	0	0	1	2	1	1	0
	rater 2	1	0	0	0	3	1	1	1

Table 4.148: Tension ratings of individual DS and TD speakers from study 1

4.4.7.8 Phonation features

Table 4.149 shows the individual phonation ratings for the DS speakers from study 1, and table 4.150 shows those for the TD speakers.

PHONATION FEATURES: Individual ratings of DS speakers from study 1									
		DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
neutral – non neutral	rater 1	1	1	1	1	1	1	1	1
	rater 2	1	1	1	1	1	1	1	1
falsetto	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
creak	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
creaky	rater 1	2	0	0	0	0	0	0	2
	rater 2	3	0	0	0	0	0	1	3
whisper	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
whispery	rater 1	3	4	4	3	3	3	3	2
	rater 2	3	3	3	3	3	2	4	1
harsh	rater 1	0	1	1	2	1	2	1	1
	rater 2	1	0	1	2	0	0	0	0
tremor	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0

Table 4.149: Phonation ratings of individual DS speakers from study 1

PHONATION FEATURES: Individual ratings of TD speakers from study 1									
		TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
neutral – non neutral	**rater 1**	1	1	1	1	1	1	1	1
	rater 2	1	1	1	1	1	1	1	1
falsetto	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
creak	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
creaky	**rater 1**	0	0	0	0	1	0	1	0
	rater 2	0	0	0	0	2	0	2	0
whisper	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
whispery	**rater 1**	2	2	2	2	2	2	3	2
	rater 2	2	3	3	2	2	2	4	2
harsh	**rater 1**	1	0	0	0	2	1	3	3
	rater 2	1	0	1	0	1	1	3	1
tremor	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0

Table 4.150: Phonation ratings of individual TD speakers from study 1

4.4.7.9 Pitch

The individual pitch ratings for the DS and TD speakers from study 1 are shown in table 4.151.

PITCH SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) low – high (+) mean	**rater 1**	-2	4	-2	-4	-2	-2	-2	2
	rater 2	-3	4	-1	-2	-1	0	-2	2
(-) minimised – extensive (+) range	**rater 1**	-4	1	-2	0	-1	0	-1	0
	rater 2	-4	2	-1	1	-1	0	-2	1
(-) low – high (+) variability	**rater 1**	-4	3	-2	1	0	-1	0	2
	rater 2	-4	2	-2	2	0	-1	0	2
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) low – high (+) mean	**rater 1**	0	-1	1	1	0	0	-3	-1
	rater 2	0	-1	1	1	1	0	-3	0
(-) minimised – extensive (+) range	**rater 1**	-1	-1	1	0	0	-1	0	-4
	rater 2	-1	-1	1	1	0	-1	0	-4
(-) low – high (+) variability	**rater 1**	-1	-2	1	0	0	-1	0	-3
	rater 2	-1	-3	1	1	0	-1	0	-2

Table 4.151: Pitch ratings of individual DS and TD speakers from study 1

4.4.7.10 Loudness

Table 4.152 reports the individual ratings attributed to the DS and TD speakers from study 1 for the parameter of loudness.

LOUDNESS SETTINGS: Individual ratings of DS and TD speakers from study 1									
		DS13	**DS26**	**DS24**	**DS14**	**DS7**	**DS30F**	**DS6**	**DS8**
(-) low - high (+) mean	**rater 1**	-2	0	-2	0	0	1	0	0
	rater 2	-2	-1	-1	1	0	1	0	0
(-) minimised – extensive (+) range	**rater 1**	-2	0	-1	0	0	0	0	2
	rater 2	-2	0	0	0	0	0	0	2
(-) low - high (+) variability	**rater 1**	0	0	-2	0	0	1	0	2
	rater 2	0	0	0	0	0	1	0	2
		TD2	**TD1**	**TD5**	**TD4F**	**TD7**	**TD3**	**TD8**	**TD6**
(-) low - high (+) mean	**rater 1**	0	0	0	0	0	0	-1	0
	rater 2	0	0	0	0	0	0	-2	0
(-) minimised - extensive (+) range	**rater 1**	0	0	0	0	0	0	0	0
	rater 2	0	-1	0	0	0	0	0	-2
(-) low - high (+) variability	**rater 1**	0	-1	0	0	0	0	0	-1
	rater 2	0	-3	0	0	0	0	0	-2

Table 4.152: Loudness ratings of individual DS and TD speakers from study 1

4.4.7.11 Temporal organization

The individual continuity and rate ratings for the individual speakers from study 1 are shown in table 4.153.

TEMPORAL ORGANIZATION: Individual ratings of DS and TD speakers from study 1									
		DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
continuity interrupted	rater 1	1	0	3	0	1	2	0	0
	rater 2	2	0	4	0	2	3	1	0
(-) slow - fast (+) rate	rater 1	-1	0	-1	-3	0	-1	0	0
	rater 2	-1	0	0	-3	0	-2	0	0
		TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
continuity interrupted	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
(-) slow - fast (+) rate	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	-1	0

Table 4.153: Temporal organization ratings of individual DS and TD speakers from study 1

4.4.7.12 Other features

Individual ratings for respiratory support and diplophonia for the DS and TD speakers from study 1 are reported in table 4.154.

OTHER FEATURES: Individual ratings of DS and TD speakers from study 1									
		DS13	DS26	DS24	DS14	DS7	DS30F	DS6	DS8
respiratory support	rater 1	0	0	1	0	1	0	0	0
	rater 2	0	0	1	0	1	0	0	0
diplophonia	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
		TD2	TD1	TD5	TD4F	TD7	TD3	TD8	TD6
respiratory support	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0
diplophonia	rater 1	0	0	0	0	0	0	0	0
	rater 2	0	0	0	0	0	0	0	0

Table 4.154: Respiratory support and diplophonia ratings of individual DS and TD speakers from study 1

4.5 Correlations between studies 1, 2 and 3

This section will report the correlations between the combined SNES, MES and PEER ratings of the 8 questionnaire parameters from study 1 and the findings of the acoustic analysis (study 2) and the perceptual analysis (study 3). The data for the 8 DS and 8 TD speakers in all three studies has been combined (n = 16). Only correlations which are considered to be strong are reported (+/-0.7 and above).

4.5.1 Calm – angry correlations

Table 4.155 shows the strong correlations between the questionnaire parameter of 'calm-angry' and the various parameters of studies 2 and 3. Negative correlations were found for lip spreading-rounding/protrusion and denasality-nasality VPAS ratings.

Correlation between studies: Calm – angry ratings		
Study 3	**LABIAL spread-rounding/protrusion**	n = 16, r = -0.7, p = 0.005
Study 3	**VELOPHARYNGEAL denasality-nasality**	n = 16, r = -0.7, p = 0.003

Table 4.155: Correlations between the ratings of 'calm-angry' by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.2 Confident – shy correlations

Positive correlations between confident-shy ratings and friendly-unfriendly, happy-sad, intelligent-unintelligent and spend time with ratings in study 1 and minimised-extensive lip range, close-open jaw, minimised-extensive tongue range, lowered-raised larynx and low-high mean pitch in study 3 were found. Whilst negative correlations were observed in both the SPT measures of study 2 and in perceptual ratings from study 3 of lip spreading-rounding/protrusion, protruded jaw, whispery voice and interrupted continuity (table 4.156).

Correlation between studies: Confident - shy ratings		
Study 1	**Friendly-unfriendly**	n = 16, r = 0.8, p < 0.001
Study 1	**Happy-sad**	n = 16, r = 0.8, p < 0.001
Study 1	**Intelligent-unintelligent**	n = 16, r = 0.9, p < 0.001
Study 1	**Spend time with**	n = 16, r = 0.7, p = 0.006
Study 2	**SPT (1-5kHz)**	n = 16, r = -0.7, p = 0.002
Study 2	**SPT (2-5kHz)**	n = 16, r = -0.8, p < 0.001
Study 3	**LABIAL spread-rounding/protrusion**	n = 16, r = -0.8, p < 0.001
Study 3	**LABIAL minimised-extensive range**	n = 16, r = 0.8, p < 0.001
Study 3	**MANDIBULAR close-open jaw**	n = 16, r = 0.7, p = 0.006
Study 3	**MANDIBULAR protruded jaw**	n = 16, r = -0.7, p = 0.003
Study 3	**LINGUAL minimised-extensive range**	n = 16, r = 0.8, p < 0.001
Study 3	**LARYNX HEIGHT lowered-raised**	n = 16, r = 0.7, p = 0.006
Study 3	**PHONATION whispery voice**	n = 16, r = -0.7, p = 0.005
Study 3	**PITCH low-high mean**	n = 16, r = 0.8, p < 0.001
Study 3	**CONTINUITY interrupted**	n = 16, r = -0.7, p = 0.001

Table 4.156: Correlations between the ratings of ‘confident-shy’ by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.3 Friendly – unfriendly correlations

Positive correlations were found for confident-shy, happy-sad, intelligent-unintelligent and spend time with parameters (study 1), F0 mean (study 2) and minimised-extensive lip and tongue range and low-high mean pitch (study 3). A negative correlation was found for lip spreading-rounding/protrusion (table 4.157).

Correlation between studies: Friendly - unfriendly ratings		
Study 1	**Confident-shy**	n = 16, r = 0.8, p < 0.001
Study 1	**Happy-sad**	n = 16, r = 0.8, p = 0.001
Study 1	**Intelligent-unintelligent**	n = 16, r = 0.8, p < 0.001
Study 1	**Spend time with**	n = 16, r = 0.7, p = 0.001
Study 2	**F0 mean**	n = 16, r = 0.8, p < 0.001
Study 3	**LABIAL spread-rounding/protrusion**	n = 16, r = -0.7, p = 0.002
Study 3	**LABIAL minimised-extensive range**	n = 16, r = 0.8, p < 0.001
Study 3	**LINGUAL minimised-extensive range**	n = 16, r = 0.7, p = 0.002
Study 3	**PITCH low-high mean**	n = 16, r = 0.8, p < 0.001

Table 4.157: Correlations between the ratings of ‘friendly-unfriendly’ by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.4 Happy – sad correlations

Ratings of happy-sad were found to correlate positively with the confident-shy, friendly-unfriendly, intelligent-unintelligent and gender parameters of study 1 and the lip spreading-rounding/protrusion, minimised-extensive lip and tongue range, larynx height, mean pitch and pitch range settings of study 3. Negative correlations existed for the F0 mean and both SPT acoustic parameters of study 2 and the interrupted continuity ratings of study 3 (table 4.158).

Correlation between studies: Happy - sad ratings		
Study 1	**Confident-shy**	n = 16, r = 0.8, p < 0.001
Study 1	**Friendly-unfriendly**	n = 16, r = 0.8, p = 0.001
Study 1	**Intelligent-unintelligent**	n = 16, r = 0.7, p = 0.003
Study 1	**Gender**	n = 16, r = 0.7, p = 0.004
Study 2	**F0 mean**	n = 16, r = 0.8, p = 0.001
Study 2	**SPT (1-5kHz)**	n = 16, r = -0.8, p = 0.001
Study 2	**SPT (2-5kHz)**	n = 16, r = -0.8, p < 0.001
Study 3	**LABIAL spread-rounding/protrusion**	n = 16, r = -0.8, p < 0.001
Study 3	**LABIAL minimised-extensive range**	n = 16, r = 0.8, p < 0.001
Study 3	**LINGUAL minimised-extensive range**	n = 16, r = 0.7, p = 0.005
Study 3	**LARYNX HEIGHT lowered-raised**	n = 16, r = 0.7, p = 0.005
Study 3	**PITCH low-high mean**	n = 16, r = 0.8, p = 0.001
Study 3	**PITCH minimised-extensive range**	n = 16, r = 0.8, p = 0.001
Study 3	**CONTINUITY interrupted**	n = 16, r = -0.7, p = 0.006

Table 4.158: Correlations between the ratings of ‘happy-sad’ by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.5 Intelligent-unintelligent correlations

Table 4.159 shows the correlations for judgements of intelligent-unintelligent. Positive correlations were found in study 1 with confident-shy, friendly-unfriendly, happy-sad and spend time with ratings and in study 3 for minimised-extensive lip and tongue range, close-open jaw, pharyngeal constriction-expansion and slow-fast rate. Negative correlations were identified for the lip spreading-rounding/protrusion, protruded jaw and interrupted continuity settings of study 3.

Correlation between studies: Intelligent - unintelligent ratings		
Study 1	**Confident-shy**	n = 16, r = 0.9, p < 0.001
Study 1	**Friendly-unfriendly**	n = 16, r = 0.8, p < 0.001
Study 1	**Happy-sad**	n = 16, r = 0.7, p = 0.003
Study 1	**Spend time with**	n = 16, r = 0.8, p < 0.001
Study 3	**LABIAL spread-rounding/protrusion**	n = 16, r = -0.8, p = 0.001
Study 3	**LABIAL minimised-extensive range**	n = 16, r = 0.8, p = 0.001
Study 3	**MANDIBULAR close-open jaw**	n = 16, r = 0.7, p = 0.002
Study 3	**MANDIBULAR protruded jaw**	n = 16, r = -0.8, p < 0.001
Study 3	**LINGUAL minimised-extensive range**	n = 16, r = 0.9, p < 0.001
Study 3	**PHARYNGEAL constriction-expansion**	n = 16, r = 0.7, p = 0.001
Study 3	**CONTINUITY interrupted**	n = 16, r = -0.7, p = 0.004
Study 3	**RATE slow-fast**	n = 16, r = 0.7, p = 0.003

Table 4.159: Correlations between the ratings of ‘intelligent-unintelligent’ by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.6 ‘Spend time with’ correlations

Positive correlations were found with the confident-shy, friendly-unfriendly and intelligent-unintelligent ratings from study 1 and for pharyngeal constriction-expansion in study 3 whilst a single negative correlation was found for the setting of interrupted continuity (table 4.160).

Correlation between studies: Spend time with ratings		
Study 1	**Confident-shy**	n = 16, r = 0.7, p = 0.006
Study 1	**Friendly-unfriendly**	n = 16, r = 0.7, p = 0.001
Study 1	**Intelligent-unintelligent**	n = 16, r = 0.8, p < 0.001
Study 3	**PHARYNGEAL constriction-expansion**	n = 16, r = 0.7, p = 0.002
Study 3	**CONTINUITY interrupted**	n = 16, r = -0.7, p = 0.005

Table 4.160: Correlations between the ratings of ‘spend time with’ by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.7 Gender correlations

Table 4.161 shows positive correlations for the happy-sad ratings of study 1, the F0 mean and standard deviation measures of study 2 and the larynx height, pitch mean and pitch range settings of study 3. Both the SPT measures of study 2 were found to correlate negatively.

Correlation between studies: Gender ratings		
Study 1	**Happy-sad**	n = 16, r = 0.7, p = 0.004
Study 2	**F0 mean**	n = 16, r = 0.7, p = 0.002
Study 2	**F0 mean (stdev)**	n = 16, r = 0.7, p = 0.001
Study 2	**SPT (1-5kHz)**	n = 16, r = -0.7, p = 0.002
Study 2	**SPT (2-5kHz)**	n = 16, r = -0.7, p = 0.002
Study 3	**LARYNX HEIGHT lowered-raised**	n = 16, r = 0.7, p = 0.001
Study 3	**PITCH low-high mean**	n = 16, r = 0.7, p = 0.004
Study 3	**PITCH minimised-extensive range**	n = 16, r = 0.7, p = 0.001

Table 4.161: Correlations between the ratings of 'gender' by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

4.5.8 Age correlations

Only negative correlations were found for age judgements. In study 2 these were for F0 mean and standard deviation measures, and in study 3 for the settings of larynx height and mean pitch level (table 4.162).

Correlation between studies: Age ratings		
Study 2	**F0 mean**	n = 16, r = -0.9, p < 0.001
Study 2	**F0 mean (stdev)**	n = 16, r = -0.7, p = 0.001
Study 3	**LARYNX HEIGHT lowered-raised**	n = 16, r = -0.7, p = 0.002
Study 3	**PITCH low-high mean**	n = 16, r = -0.7, p = 0.002

Table 4.162: Correlations between the ratings of 'age' by the SNES, MES and PEER raters combined and the parameters evaluated in studies 2 (acoustic analysis) and 3 (perceptual analysis) for the combined DS and TD speakers from study 1

5
DISCUSSION

5.1 INTRODUCTION

The results of studies 1, 2 and 3 will be discussed initially followed by examination of any identified correlations between the studies. Finally, variability in judgements of individual speakers from study 1 will be addressed in short case studies, where the acoustic vocal profile (study 2) and the expert perceptual ratings of voice (study 3) can be examined in relation to speakers who have been rated particularly negatively or positively, allowing some qualitative interpretation of the relationship between the studies.

5.2 STUDY 1: QUESTIONNAIRE-BASED ANALYSIS OF LISTENER JUDGEMENTS OF VOICE

The following sections reported within the results chapter will be discussed in relation to the 8 questionnaire parameters:

- Consistency of group judgements for repeated samples
- Analysis of differences between DS and TD ratings within listener groups
- Analysis of differences between DS and TD ratings between listener groups
- Analysis of the effect of Gender and Age on PEER ratings

For clarity and ease of reading all percentage values have been rounded up.

5.2.1 Consistency of group judgements for repeated samples

The overall percentage of consistent judgements by all three listener groups (SNES, MES & PEER) across all four repeated recordings for all 8 parameters (table 5.1) range from 85% to 98%. These high values are a consequence of the criteria devised to exclude data from raters found to show poor intra-rater reliability. As such the data included in the final analysis can be accepted as a reliable measure of the judgements made by listeners, accurately reflecting their perception of DS and TD voice quality. Consequently, the intra-rater reliability protocol devised for this study is considered to have been an appropriate measure.

Overall intra-rater reliability (across all 4 repetitions)			
	SNES	**MES**	**PEER**
calm-angry	98	96	94
confident-shy	92	91	85
friendly-unfriendly	98	98	93
happy-sad	94	95	93
intelligent-unintelligent	96	91	94
spend time with	N/A	N/A	96
gender	93	89	94
age	88	85	89

Table 5.1: Summary of overall intra-rater reliability percentages for SNES, MES and PEER raters for all questionnaire parameters

Within the four repeated voices there was some variation in the overall group percentage scores for consistency of ratings, with some voices being judged considerably less consistently than others. For example, DS13 received the most variable judgements, ranging from only 65% consistency ('confident-shy' ratings by PEERS) to 100% consistency (gender; MES & PEER ratings) in contrast to TD2, who had a much narrower and thus more consistent range across the parameters, from 86% (age; PEER ratings) to 100% (gender; MES ratings). Lower percentages are a consequence of reduced consistency between the first and second ratings of voices in any parameter by individual listeners, which are then reflected in the overall group consistency scores. It is likely that where speakers receive lower scores that there are perceptual aspects of those voices that listeners find problematic to quantify or label when making social judgements of this type, hence the disparities between repeated judgements.

The numbers of raters who were judged to demonstrate sufficient intra-rater reliability to be included in the analysis of each parameter varies considerably also. Within the parameter ‘calm-angry’ only one of the SNES and two of the PEER raters were excluded compared to the ‘confident-shy’ parameter, where nine SNES, six MES and ten PEER raters did not meet the intra-rater reliability criteria. Confidence is a more abstract concept than anger therefore it may be the case that listeners find it more difficult to judge than a parameter which reflects emotional state.

The most raters were excluded for judgements of ‘age’ (22 SNES, 15 MES and 16 PEER raters). However, this is unsurprising since judgements within this parameter were open-ended (i.e. listeners could select any age) whilst in the semantic differential parameters they were limited to a 5-point scale, and in the ‘gender’ and ‘spend time with’ parameters there were a choice of only three possible responses. By not restricting the ratings of age in this way, the questionnaire was able to identify judgements which were considerably outside of the expected margins, such as the misperception of school-age speakers as adult or even elderly speakers, as in the case of DS14 where age judgements ranged from 8-70 years (SNES), 10-80 years (MES) and 5-70 years (PEERS).

5.2.2 Analysis of differences between DS and TD ratings within listener groups

5.2.2.1 <u>SNES raters</u>

Across the five semantic differential parameters of ‘calm-angry’, confident-shy’, ‘friendly-unfriendly’, ‘happy-sad’ and ‘intelligent-unintelligent’ (summarised in figure 5.1) the SNES group consistently rated the DS group significantly more negatively than the TD group.

The most positive judgements of the DS group were within the 'calm-angry' and 'friendly-unfriendly' parameters but even here the children with DS were judged as sounding less calm and less friendly than their peers. Despite the stereotypical perception of individuals with DS as "open, easy-going, happy-go-lucky, sociable people" (Pryce, 1994, p. 109) overall ratings of 'happy-sad' were 53%, only marginally on the 'happy' side of the 'neutral' rating of 50%; however it is to be noted that the perception of happiness for the TD group was also low (59% - the lowest score for this group). It may be the case that the SNES listeners, who were recruited from schools within the Midlands, may perceive some quality within both the DS and TD speakers from the Edinburgh area that they associate with sounding less happy. In common with the finding of Moran, Labarge and Haynes (1988) who report that the most negative ratings in their study related to 'confidence' and 'capability', ratings of confidence were low for the children with DS in this study falling just short of the neutral rating (49%) compared to very strong ratings of confidence in the TD group (73%). This finding is again at odds with the often remarked social confidence associated with this group. When asked about how others viewed their levels of confidence and sociability Pryce (2004) found that 60% of individuals with DS felt that others believed them to be introverted in nature; this figure compares with 50% in individuals with generalised intellectual disability (ID), 27.7% with functional dysphonia, and only 10% in TD controls, indicating that there is "a high level of perceived introversion amongst people with learning disabilities of all sorts" (Pryce, 1994, p. 109); a finding which further questions the stereotypical view of high social confidence in DS and ID generally.

The most negative rating for the DS group, and that with the biggest disparity between the DS and TD speakers was for the parameter 'intelligent-unintelligent' with the DS group being rated as sounding particularly unintelligent (40%) in comparison to the TD group whose ratings fell firmly towards the 'intelligent' end of the scale (72%). As all the individuals within the DS group have some degree of ID this finding suggests that this feature is perceptually salient within voices; however as ID would be likely to also impact negatively across other speech and language domains (e.g. articulation and language skills) it is difficult to be certain of the extent that listeners were able to disassociate these poorer parallel abilities in order to focus on voice features alone when judging this parameter in particular. Intelligence was not one of the parameters investigated by Moran, Labarge and Haynes (1988) thus there is no basis for comparison with other DS studies, however Lass, Ruscello, Bradshaw and Blankenship (1991) and Lass, Ruscello, Harshaw and Blankenship (1993) in studies of the judgements by adolescents of children with voice disorder characterised by ‘hoarseness’ and ‘breathiness’ and of children with dysarthria respectively found that both groups were rated as sounding significantly less ‘smart’ and significantly more ‘foolish’ than their TD peers. Similarly in adults, Turcotte, Wilson, Harris, Seikaly and Rieger (2009) identified that speakers with disordered voice quality as a consequence of surgical and non-surgical treatment for laryngeal cancers were rated as sounding significantly less ‘clever’ than a control group of non-dysphonic adults (interestingly a significant age effect was also found, whereby older speakers were rated less positively than younger speakers). These findings indicate that even where intellectual disability is not present atypical vocal features contribute towards a significantly more negative perception of cognitive skills in comparison to those without voice and/or speech disorders. Moreover, all of the above studies identified intelligence ratings of speakers with voice disorders to be more negative than controls to a statistically significant level, whilst Lass et al. (1991) found that although ratings of ‘happiness’ and ‘confidence’ by adolescents were lower for voice-disordered children than their peers they did not differ significantly; thus it would appear that intelligence is a characteristic which is particularly sensitive to negative perception in speakers with atypical vocal quality.

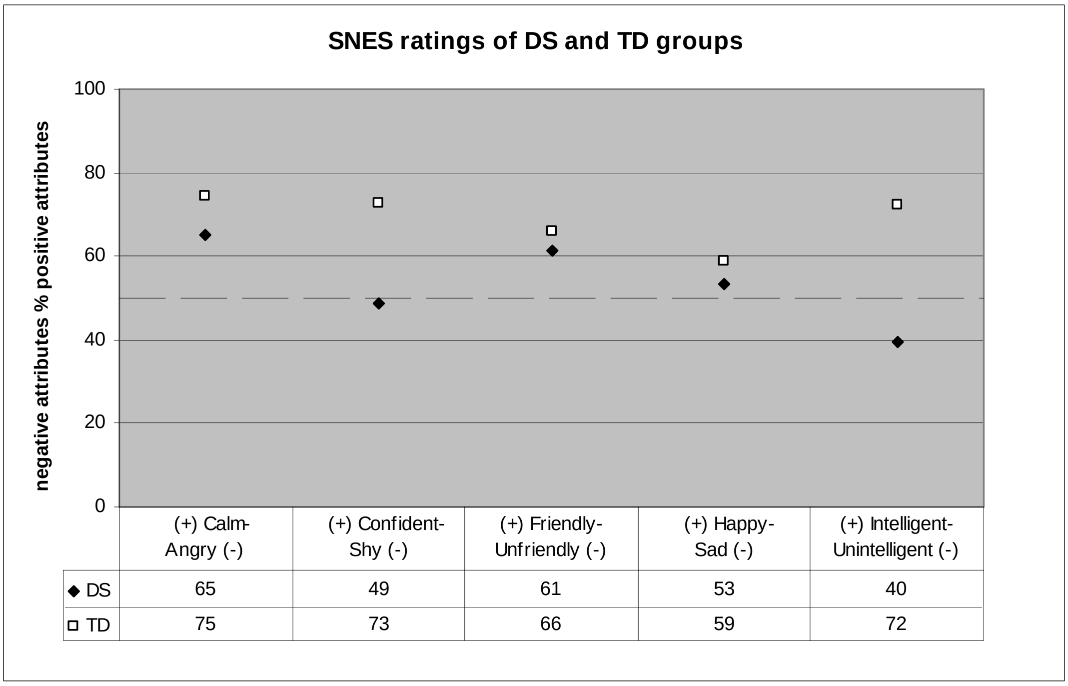


Figure 5.1: Diagram illustrating the relative positions of overall ratings (percentage) for 'calm-angry', 'confident-shy', 'friendly-unfriendly', 'happy-sad' and 'intelligent-unintelligent' parameters for DS and TD groups by SNES raters
---- neutral rating (50%)

Another significant difference between the DS and TD groups emerged for judgements of 'gender' by the SNES listeners. As raters had a third option in this parameter ('unsure') over and above basic judgements of 'male' or 'female', the very large disparity between correct judgements made cannot be taken in isolation, as it might have been the case that listeners opted for the 'not sure' option more frequently for the DS voices rather than simply being incorrect in their judgements. However, the greater percentage of both 'unsure' ratings and 'incorrect' judgements for the DS group confirms that these listeners did have some difficulty in identifying the gender of DS speakers from audio recordings alone. This finding supports earlier research by Montague (1976) which identified that listeners found judgements of the gender of children with DS on the basis of their voices to be problematic.

In the final parameter of 'age', again there was a significant difference between the judgements for the age-matched DS and TD groups. Although the median values for both groups are identical (14.00 years) the difference can be seen to be in the wider range of age judgements associated with the DS group. This variance indicates that there is less agreement between raters about how old the voices of children and young people with DS sound than within the same judgements of typical peers. It is notable that outlying judgements for the DS group all fall within the older rather than younger range, and that no such outliers exist within the age judgements of the TD group. Both the TD and DS groups were rated as sounding older than their actual median ages (DS, 11.42 years & TD, 12.17 years); for the DS group this equates to a difference of just over two and a half years which is a marked contrast to the findings of Montague (1976) where children with DS were perceived to be more than two years younger than their actual chronological age.

The results of the SNES raters support the a priori hypotheses that listeners will rate the abilities and personality characteristics of the DS group more negatively than the TD group, and that judgements of gender will be less accurate for the children with DS. However it does not support the hypothesis that children with DS will be judged as sounding younger than their chronological age and younger than their age-matched TD peers.

5.2.2.2 <u>MES raters</u>

Within the MES group all five semantic differential parameters (summarised in figure 5.2) were again rated significantly more negatively for the DS group, meaning that the children with DS were judged to sound less calm, less confident, less friendly, less happy and less intelligent than their peers. The biggest disparities between the ratings of the two groups were for the parameters of ‘confident-shy’ and ‘intelligent-unintelligent’, the DS group being rated just below neutral (49%) in terms of confidence (i.e. not confident but not shy) compared to high ratings of confidence in the TD group, whilst DS intelligence ratings fell well below neutral into the ‘unintelligent’ end of the scale (37%), again compared to much higher ratings of intelligence in the TD group (69%).

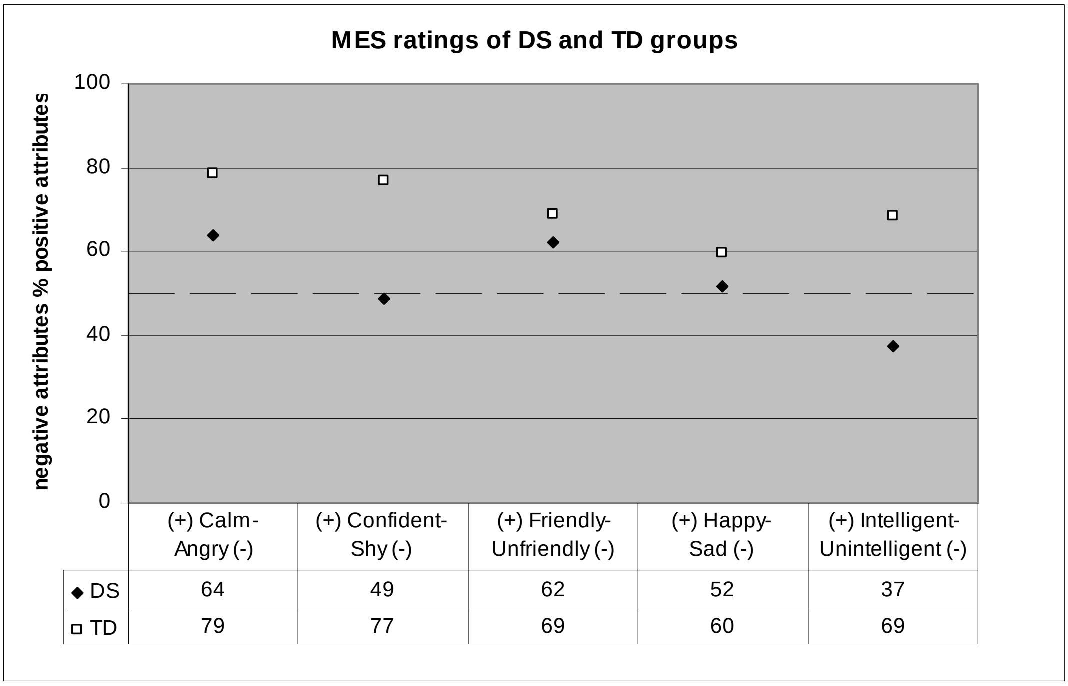


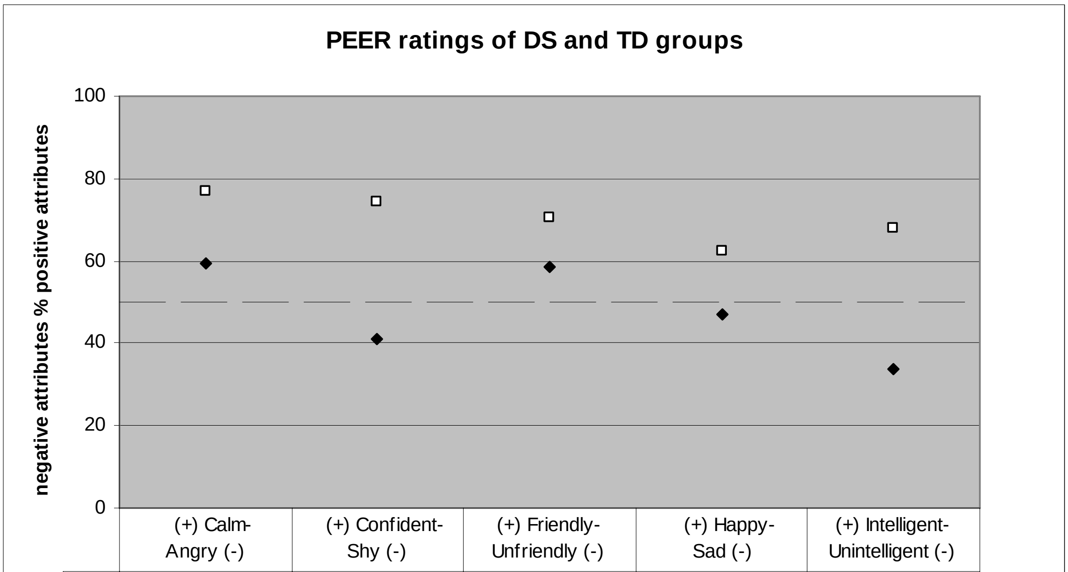
Figure 5.2: Diagram illustrating the relative positions of overall ratings (percentage) for 'calm-angry', 'confident-shy', 'friendly-unfriendly', 'happy-sad' and 'intelligent-unintelligent' parameters for DS and TD groups by MES raters
---- neutral rating (50%)

Judgements of 'gender' were significantly different for the DS and TD groups, with a considerably higher percentage of 'incorrect' and 'unsure' judgements. These findings, in common with those of the SNES raters, indicate some degree of ambiguity in the perception of gender for DS speakers.

A significant difference was also found between the judgements of 'age' for the DS and TD speakers, which in common with the SNES raters reflected the greater variation in the range of age judgements for the DS group (DS IQR, 6.00; TD IQR, 3.00) rather than a difference in the median value (both 14.00 years). Again both groups were judged to sound older than their actual median age; the DS group by approximately two and a half years and the TD group by just under two years.

Only judgements of age by the MES raters failed to support the given hypotheses.

5.2.2.3 PEER raters



	(+) Calm-Angry (-)	(+) Confident-Shy (-)	(+) Friendly-Unfriendly (-)	(+) Happy-Sad (-)	(+) Intelligent-Unintelligent (-)
◆ DS	60	41	59	47	34
□ TD	77	74	70	63	68

Figure 5.3: Diagram illustrating the relative positions of overall ratings (percentage) for 'calm-angry', 'confident-shy', 'friendly-unfriendly', 'happy-sad' and 'intelligent-unintelligent' parameters for DS and TD groups by PEER rater
---- neutral rating (50%)

The PEER listeners also rated the children with DS significantly more negatively than the TD group across all of the semantic differential parameters of 'calm-angry', 'friendly-unfriendly', 'happy-sad', 'confident-shy' and 'intelligent-unintelligent' (figure 5.3). Again, particularly low ratings were found for the DS group for confidence and intelligence, both being well below the neutral rating of 50% (indicating they were perceived as sounding shy and unintelligent in comparison to the high ratings of confidence and intelligence found for their TD peers).

For the additional question posed to the PEER listeners regarding if they would they like to 'spend time with' the individuals that they listened to, the group with DS were again rated significantly more negatively than the TD speakers (overall scores 34% versus 69%). This finding indicates very strongly that TD PEER listeners have a significant preference for forming friendships with other children who share similar voice features to their own; although again it is difficult to be sure that the PEER listeners followed the instruction to not base judgements on features other than the way the voices sounded. The PEER raters' preference for vocal features which are similar in character to their own supports the research of Saxton (2006) who found that children rated male voices with higher pitch more positively than males with lower pitch whilst the opposite pattern was observed in adult raters (higher mean pitch level being a characteristic of younger speakers). Saxton also makes the point that vocal (and facial) preferences are likely to occur as a result of the habitual environment, thus the typical features to which individuals are exposed to frequently are likely to become those which they consider to be more attractive, and consequently individuals will prefer to interact with others who share those familiar, attractive features.

The judgements of 'gender' by the PEER listeners also differed significantly between the DS and TD groups, with a lower overall percentage of correct judgements and higher numbers of incorrect judgements and 'not sure' judgements for the DS group, indicating difficulty in judging gender within DS speakers.

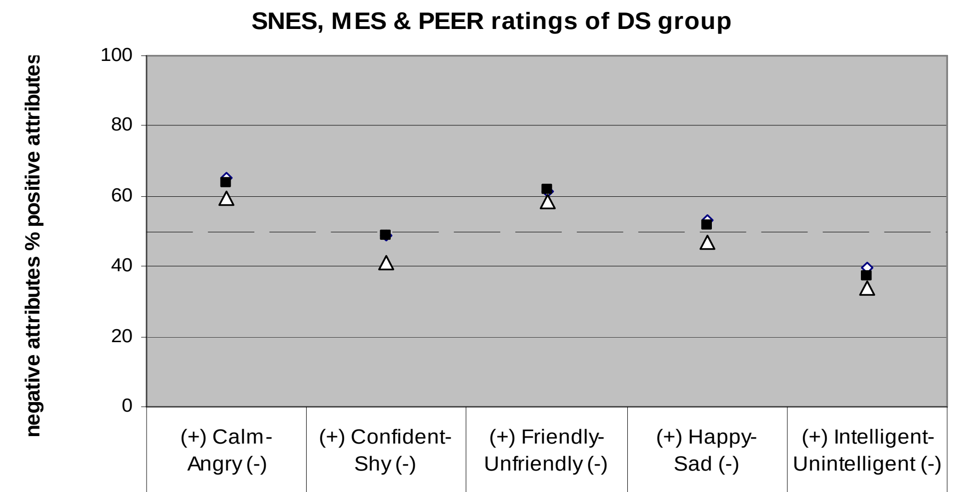
For the final parameter of 'age' the statistically significant difference between the DS and TD groups again reflects the larger variation in age judgements for the DS group. Both groups were judged to have a median age of 13.00 years, but the IQR for the DS group was much greater (7.00 years) than that for the TD group (3.00 years). Although both groups have been perceived as being older than their actual age (DS, 11.42 years; TD, 12.17 years) the difference for the DS group is just over one and a half years (1.58 years) and less than a year (0.83 years) for the TD group, again suggesting that children and young people are more accurate in their judgement of the voices of their peers than are adult raters. Only the judgements of age failed to support the a priori hypotheses.

5.2.3 Analysis of differences between DS and TD ratings between listener groups

A surprisingly consistent pattern of judgements can be seen between the SNES, MES and PEER listener groups for both the DS and TD groups.

5.2.3.1 <u>DS ratings by SNES, MES and PEER raters</u>

As can be seen in figure 5.4, the ratings for the five semantic differential parameters for the DS group, by all three groups of raters, follow a very similar pattern of positive to negative judgements: ‘calm-angry’ being the most positively judged, followed by ‘friendly-unfriendly’, ‘happy-sad’, ‘confident-shy’, with ‘intelligent-unintelligent’ being scored the lowest.



	(+) Calm-Angry (-)	(+) Confident-Shy (-)	(+) Friendly-Unfriendly (-)	(+) Happy-Sad (-)	(+) Intelligent-Unintelligent (-)
◇ SNES	65	49	61	53	40
■ MES	64	49	62	52	37
△ PEER	60	41	59	47	34

Figure 5.4: Diagram illustrating the overall percentage ratings for ‘calm-angry’, ‘confident-shy’, ‘friendly-unfriendly’ ‘happy-sad’ and ‘intelligent-unintelligent’ by the SNES, MES and PEER listeners for the DS group
---- neutral rating (50%)

There are no significant differences between the ratings of the SNES and MES groups across any of the questionnaire parameters (table 5.2). It might have been hypothesised that the greater experience of, and exposure to, people with unusual vocal characteristics (as would be likely in a special school environment) might have contributed to a less judgemental perception of voice disorder by the SNES raters, thus resulting in more positive judgements of the children with DS than by the MES group. Conversely, that greater exposure might have been suggested to cause the SNES raters to recognise and correctly associate any unusual voice patterns within the DS group with children with special educational needs, potentially resulting in more negative judgements of ability. Given the current drive for educational inclusion within mainstream schools for children with ID, the lack of differences between these two groups of education staff can be seen as a very positive and reassuring factor for the parents of children with DS, who may have been concerned that a lack of experience of disability might have meant that teachers within mainstream schools were less positive about the skills and potential of children with DS, consequently having lower educational and social expectations than might be appropriate.

Statistically significant differences between raters (DS ratings)			
	SNES versus MES	**SNES versus PEER**	**MES versus PEER**
calm-angry	none	$p < 0.001$	$p = 0.001$
confident-shy	none	$p < 0.001$	$p < 0.001$
friendly-unfriendly	none	none	none
happy-sad	none	$p < 0.001$	$p = 0.001$
intelligent-unintelligent	none	$p < 0.001$	none
gender	none	none	none
age	none	$p = 0.001$	$p = 0.008$

Table 5.2: Summary of statistically significant ratings of the DS group between SNES, MES and PEER raters

Although following the same pattern of judgements as the SNES and MES raters, the PEER listeners can be seen to have rated more negatively than both the education staff groups across all the semantic differential questionnaire parameters (figure 4). Statistically significant differences were found between the PEER and MES ratings for ‘calm-angry’, ‘confident-shy’ and ‘happy-sad’, and between the SNES and PEER raters for those and the ‘intelligent-unintelligent’ parameter also (table 5.2). These findings are in contrast to earlier research examining listener judgements of non-speech abilities made about children with voice disorders characterised by 'hoarseness' and 'breathiness'. Although the judgements made by children (Lass, Ruscello, Stout & Hoffman, 1991), adolescents (Lass, Ruscello, Bradshaw & Blankenship, 1991) and adults (Ruscello, Lass & Podbesek, 1988) in these voice studies were all more negative about those children with voice disorder than the TD controls, Lass, Ruscello, Bradshaw and Blankenship (1991) observed that the child and adolescent groups rated less negatively than the adults. This finding was suggested to be a consequence of younger people having less rigid attitudes towards voice disorders; however in this current study, children from the Edinburgh area appear to be more judgemental of their voice disordered peers than do adult raters. It may be the case that the nature of the adult raters had an impact on this finding. Both groups of adults consisted of education staff, and as professionals working with children on a regular basis (both with and without disabilities) it is possible that that experience has lead them to be somewhat less judgemental about the personality and ability traits of children than adult raters used in previous studies.

From these findings it is evident that despite the positives found in the similar ratings of the two education staff groups, great concern must be expressed regarding the negative perception of DS speakers by their TD peers. This concern is compounded by the results of the 'spend time with' parameter posed only to PEER raters, where a desire to form some level of friendship with the DS speakers (i.e. 'yes' judgements) was expressed in only 13% of ratings, compared to 50% for the TD speakers, with an overall percentage calculation taking into account the 'no' and 'not sure' judgements being almost twice as high for the TD group than the DS group (DS, 35%; TD, 69%). These ratings indicate that children with DS are deemed considerably less desirable as potential friends for TD children than other TD speakers, and serve to highlight a very real risk of social exclusion within the educationally inclusive setting. The education of students about communication disorders and the need for them to encounter individuals with such disorders within the classroom is noted to be an effective way of bringing about a change in attitude towards those with communication difficulties (Lass, Ruscello, Bradshaw & Blankenship, 1991) and as such it may be the case that as educational inclusion continues more acceptance of those with vocal differences may gradually occur.

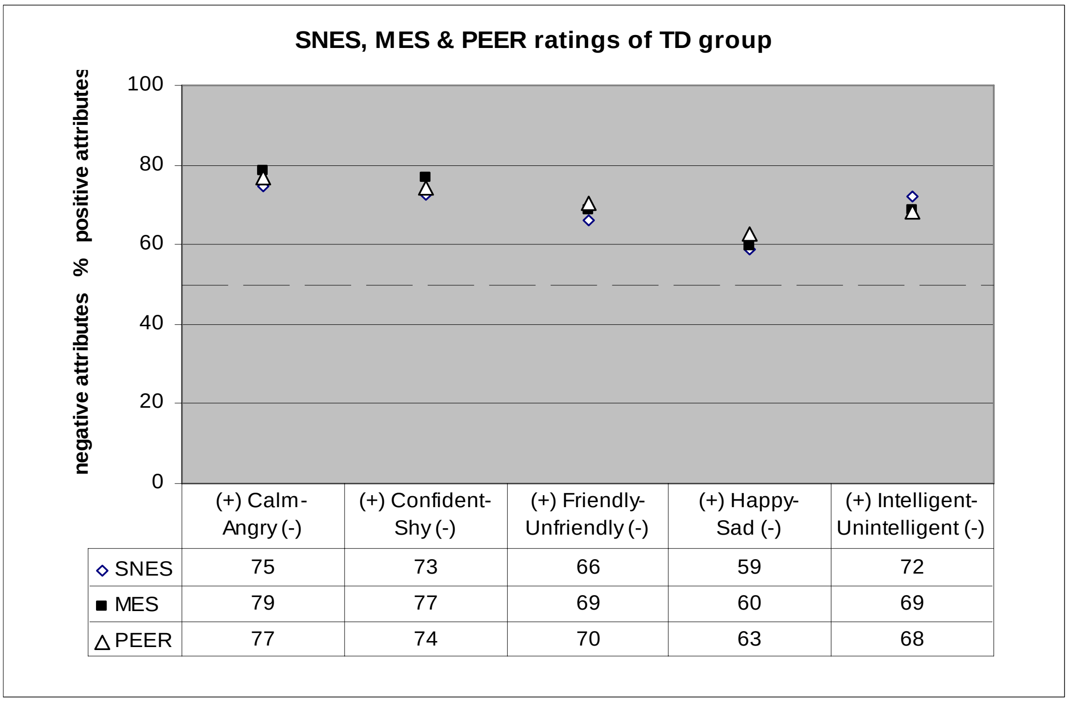
No significant differences were found between any of the listener groups for the parameter of ‘gender’ for the DS children (table 5.2).

For judgements of ‘age’ significant differences were found between the PEER listeners and the SNES and MES raters, whilst no significant difference was found between the two education staff groups (table 5.2). Although all three listener groups rated the DS speakers as sounding older than their actual median age, the PEER listeners were more accurate in their judgements than either of the education staff groups by one full year. All three groups were similar in that the range of their age judgements for the DS children was much wider than for the TD children, demonstrating less agreement within listener groups about the age of the DS speakers, indicating that all three groups experienced some difficulty in estimating the ages of the DS group accurately.

The striking similarities between the SNES and MES ratings of the DS group in relation to the number of significant differences between the education staff and the PEER ratings suggests a fundamental difference between adult and child perception of voice. The more negative judgements across all five semantic differential parameters by the PEER raters indicates a more judgemental perspective, which appears to become less apparent into adulthood, perhaps as a consequence of increasing social awareness and acceptance of differences.

5.2.3.2 TD ratings by SNES, MES and PEER raters

For the TD speakers, all three groups of listeners again demonstrated a similar pattern of ratings for the semantic differential parameters (figure 5.5) although much more overlap in judgements was evident, with no single group of listeners rating more negatively or positively than the others across all the parameters. This greater parity in judgements compared to the DS ratings would indicate a greater acceptance by the PEER raters of voices which sound similar to their own than of those with DS. The only significant difference between the two education staff groups was for the ‘calm-angry’ ratings, with the MES group rating the TD speakers more positively. Between the SNES and PEER raters significant differences existed for the ‘friendly-unfriendly’, ‘happy-sad’ and ‘intelligent-unintelligent’ parameters; the PEER group rating more positively for the first two and more negatively for the latter. No significant differences were found between the MES and PEER groups for any of the 5 semantic differential parameters for the TD speakers (table 5.3).



	(+) Calm-Angry (-)	(+) Confident-Shy (-)	(+) Friendly-Unfriendly (-)	(+) Happy-Sad (-)	(+) Intelligent-Unintelligent (-)
◇ SNES	75	73	66	59	72
■ MES	79	77	69	60	69
△ PEER	77	74	70	63	68

Figure 5.5: Diagram illustrating the overall percentage ratings for ‘calm-angry’, ‘confident-shy’, ‘friendly-unfriendly’, ‘happy-sad’ and ‘intelligent-unintelligent’ by the SNES, MES and PEER listeners for the TD group
---- neutral rating (50%)

As with the DS speakers no differences were found between any of the listener groups for the parameter of ‘gender’ for the TD speakers.

Significant differences were found to exist between both the education staff listeners and the PEER raters for judgements of age. All groups judged the TD speakers to be older than their actual group median age, but with the PEER group being significantly closer to the actual age than either of the education staff raters, suggesting that the PEER raters may have a more instinctive recognition of voices that are similar in age and vocal features to their own than adult listeners. It is possible that the adult listeners may also have been at a disadvantage in judging age as the vocal profile of Edinburgh-speakers would differ from that of children and young people in their own locality, making them less aware of vocal features that are typical within different stages of maturation in south-east Scotland than the TD PEER raters.

This lack of familiarity with Edinburgh-speakers might have been expected to lead the education staff to make less positive ratings of the TD group than the PEER raters, however the mixed pattern of ratings across all three listeners groups, with PEER raters on occasion rating most negatively, indicates that familiarity was not a key factor in the more positive perception of the TD speakers.

Statistically significant differences between raters (TD ratings)			
	SNES versus MES	SNES versus PEER	MES versus PEER
calm-angry	$p = 0.002$	none	none
confident-shy	none	none	none
friendly-unfriendly	none	$p < 0.001$	none
happy-sad	none	$p = 0.007$	none
intelligent-unintelligent	none	$p = 0.002$	none
gender	none	none	none
age	none	$p < 0.001$	$p < 0.001$

Table 5.3: Summary of statistically significant ratings of the TD group between SNES, MES and PEER raters

5.2.4 Analysis of the effect of gender and age on PEER ratings

When divided on the basis of gender and age, some interesting effects were noted within the PEER listener group's judgements of the DS and TD speakers (table 5.4).

Effects of gender and age on peer ratings of DS and TD groups				
	gender effects		age effects	
	DS	TD	DS	TD
calm-angry	none	none	none	none
confident-shy	$p < 0.001$	none	none	none
friendly-unfriendly	none	none	none	none
happy-sad	none	none	none	none
intelligent-unintelligent	$p < 0.001$	none	$p < 0.001$	none
spend time with	none	none	none	none
gender	none	none	none	none
age	none	$p = 0.03$	none	$p < 0.001$

Table 5.4: Statistically significant differences within PEER ratings for DS and TD groups when divided on the basis of gender and age

5.2.4.1 Gender effects

For the judgements of the DS speakers, gender effects within the PEER listener group were identified within only two of the eight questionnaire parameters ('confident-shy' & 'intelligent-unintelligent'; the two parameters judged most negatively by all listener groups). In both cases the female listeners rated more positively than the males, indicating a somewhat less judgemental perception of voice features which differ from their own. No significant differences were found between genders in the DS ratings of 'calm-angry', 'friendly-unfriendly', 'happy-sad', 'spend time with', 'gender' or 'age', making it difficult to draw any hard and fast conclusions regarding the existence of gender differences in voice perception of DS speakers.

Within the TD ratings the only parameter to differ significantly between male and female listeners was that of 'age'. Although no difference existed between the median and IQR values, and both males and females judged the TD children to be older than their actual age, the range of male judgements was found to be spread across a younger age band which was closer to the actual median age of the TD children than the judgements by the female PEER raters. As the majority of the TD speakers were males (7 out of 8) of a similar age, this finding may suggest that it is easier for listeners to judge age when the vocal characteristics presented reflect those of their own voices. Again the general lack of gender differences within the ratings of the TD speakers makes it impossible to draw any clear conclusions.

5.2.4.2 Age effects

Again, very few differences in ratings were found when the PEER group was split on the basis of age, making it difficult to identify any particular pattern that could be associated with increasing age.

It might have been hypothesised that the inherent differences previously discussed between adult and child raters for the DS speakers would have resulted in a clear distinction between younger and older PEER raters, with the older raters making more positive (i.e. more adult-like) judgements, however this was not in fact the case. For the judgements of the DS group, only a single difference was found between the younger and older PEER raters. In this instance the younger raters judged the DS speakers to be significantly more intelligent than the older PEER raters. The presence of more female raters within the younger PEER group (younger, 47% female; older, 43% female) might have meant there was more of a gender effect on differences than a genuine age effect (group numbers were too small to evaluate this possibility further). Alternatively, as the median age of the DS group (11.42 years) is closer to the age of the younger PEER raters (12.42 years) than the older PEER raters (15.57 years) then it may be the case that the older raters naturally perceived these younger speakers to sound less intelligent than themselves. As it has been noted that "age differences in attitudes within a gender may be greater than differences in attitudes between genders" (Nowicki, 2006, p. 345) perhaps the latter proposition is most likely.

For the TD group the only difference existed in judgements of 'age' with the younger group of PEER raters estimating the average age of the TD speakers more accurately than the older PEER raters. Again, as the average median age of the TD group (12.17) was closer to that of the young raters (12.42) it could be argued that these younger raters were able to recognise the speakers' voices as sounding similar to their own making them better placed to estimate their age than the older PEER raters.

Thus it can be seen from the data when divided by both gender and age that no particular patterns of ratings can be defined between male and female raters and younger and older raters. These findings are in common with those of Turcotte et al. (2009) who identified that the age, gender (and occupation) of listeners did not appear to influence judgements made about the voices of speakers who had been treated for laryngeal cancers, and of Gannon and McGilloway (2007) who noted no difference between genders in a questionnaire about social and educational inclusion of their peers with DS. Findings also offer support to the observation of Saxton (2006) who in a study of vocal attractiveness traits found that that child judgments shift gradually to match those of adults during the course of development.

5.2.5 Summary

The high overall 'consistency of group judgements' percentage values indicate that the system devised to exclude raters who showed poor consistency between repeated judgments of the same voices was effective. Consequently it is reasonable to attach a high degree of confidence to the reliability of the data of those raters included in the final analysis of each parameter.

As predicted, and in common with the findings of Moran, LaBarge and Haynes (1988) judgements reflected significantly more negative ratings for the children with DS than the TD children across all the semantic-differential parameters judged by all three groups of listeners. Ratings of confidence and intelligence were consistently seen to be the most negatively judged parameters for the DS group and those with the greatest disparity between the DS and TD ratings, whilst degree of calmness and friendliness were rated less negatively by all the listener groups. No significant differences were found between any of the five semantic differential parameters for the SNES and MES groups for the DS speakers, indicating that increased experience of children with speech and language difficulties within special-schools does not lead to more positive or negative perception of DS speakers based on voice features alone; suggesting that such perception is more instinctive or intuitive than experiential.

The higher percentage of 'incorrect' and 'unsure' judgements and the fewer 'correct' judgements made about the DS speakers indicates that all three listener groups had more difficulty in judging the gender of the DS speakers than the TD speakers. Findings are in line with those of Montague (1976) who notes that "…sexual identification of the Down's syndrome child through perceived voice may be exceedingly difficult…".

Significant differences were found between the judgements of age of the DS and TD children by all three groups of listeners, however, as the median values for the groups did not differ, these differences reflect the considerably wider range of age judgements for the DS speakers, indicating a lack of agreement within the listener groups. In contrast to the earlier finding of perceived immaturity in children with DS, where they were judged to be approximately two years younger than their actual age (Montague, 1976) both the DS and TD speakers in this study were judged by all of the listener groups as sounding older than their chronological age. PEER listeners were more accurate in their judgements of the ages of the DS and TD speakers than either of the adult rater groups; this suggests that judgements of age might be easier when the speakers' ages are similar to those of the listeners.

Although females rated more positively than males in some of the parameters, no particular pattern could be discerned in the judgements of PEER listeners when they were grouped on the basis of gender or age; a finding which supports recent research by Turcotte et al. (2009) in judgements made about people with laryngeal cancers and by Gannon and McGilloway (2007) concerning judgements about the social and educational inclusion of children with DS, whilst also confirming the gradual change in vocal judgements as a consequence of maturation recognised by Saxton (2006).

The lack of any significant differences between the judgements of the SNES and MES raters may be of comfort to parents of children with DS who are facing the prospect of their child being enrolled in a mainstream school as it is evident that mainstream teachers would be no more or less judgemental of ability and character and thus likely to have similar levels of expectation of children with DS as teachers in special-schools. However, this positive finding is tempered by the observation that PEER listeners rated the DS speakers more negatively than adult listeners across all the semantic differential parameters, and in particular showed an overwhelming preference to want to 'spend time with' other TD children rather than children with DS, highlighting a very great potential for children with DS to be socially excluded within the mainstream setting on the basis of how they sound. It may be the case that over time educational inclusion may lead TD students to become more familiar with children with atypical vocal features and thus more accepting of them within their social circles. Increased experience of, and education about communication disorders have been found to be effective in changing attitudes towards disability (Lass, Ruscello, Bradshaw & Blankenship, 1991); however as it was observed in recent research that familiarity with a person with DS, either as a friend or relative, did not result in more favourable responses from TD peers concerning their attitudes towards social interaction with children with DS in mainstream schools (Gannon & McGilloway, 2007), perhaps there is still some way to go in achieving genuine educational and social inclusion for children with DS.

5.3 Study 2: Acoustic Analysis of Voice

The acoustic analysis findings of the overall DS and TD groups, the groups split by gender and the DS and TD speakers presented to listeners in study 1 will be discussed in the following sections.

5.3.1 Overall DS and TD groups

For the overall DS (22) and TD (52) groups a rather surprising set of acoustic analysis results were found. When compared using independent samples t-tests for F0 mean, F0 mean (stdev), jitter (rap & ppq5), shimmer (apq3 & apq5), harmonic-to-noise ratio (HNR), and spectral tilt (SPT; 1-5kHz & 2-5kHz) the only parameter to differ statistically significantly was that of SPT (both 1-5kHz & 2-5kHz). In both cases the DS groups demonstrated significantly higher mean levels of SPT than the TD groups (1-5kHz: DS, 16.53 (3.08) & TD, 13.89 (2.56) $p < 0.001$; 2-5kHz: DS, 22.61 (2.63) & TD, 19.56 (2.88), $p < 0.001$) in contrast to the a priori hypothesis.

5.3.1.1 F0 mean and F0 mean (stdev)

The lack of identified differences between the DS and TD groups in F0 mean and F0 mean (stdev) supported the proposed hypothesis. Thus this research confirms the findings of earlier F0 research in English-speaking school-aged children with DS, indicating that levels are comparable to their TD peers (Michel & Carney, 1964; Hollien & Copeland, 1965; Montague, Brown & Hollien, 1974; Pentz & Gilbert, 1983). Interestingly, this study is at odds with the findings of Moura et al., (2008) of lower F0 compared to TD peers in young Portuguese-speaking children (aged 3-8 years), suggesting that differences may be to some extent language-specific. Although the children in the Moura et al. study are younger than those in the current study, differences cannot be attributed to this factor as pre-school aged English-speaking children with DS have been found to have the opposite pattern of significantly higher F0 in comparison to TD pre-school peers (Weinberg & Zlatin, 1970), thus adding weight to the proposal of language-specific differences.

Although no significant difference was found in the F0 (stdev) values between the DS and TD groups, the IQR of the group's F0 stdev values were found to be greater for the DS speakers (6.55) than the TD speakers (3.97). This finding indicates that there is a considerable difference in the range of F0 standard deviation values between individual speakers in this group, serving as a reminder that DS speakers must be considered to be a heterogeneous group rather than a group of speakers exhibiting a standard DS vocal profile. This finding of variability may also lend some support to the proposal of Montague, Hollien, Hollien and Wold (1978) that variance in F0 rather than a deviance in the actual F0 level contributes to the frequently-cited perception of low pitch in DS speakers.

5.3.1.2 Jitter (rap & ppq5), shimmer (apq3 & apq5) and harmonic-to-noise ratio (HNR)

Based on earlier research it had been hypothesised that significant differences would exist between the DS group and their TD peers across the parameters of jitter (frequency perturbation), shimmer (amplitude perturbation) and HNR. Jitter and shimmer levels have been found to be increased in English-speaking (Pentz & Gilbert, 1983) and Portuguese-speaking (Moura et al., 2008) children with DS whilst HNR has been identified as being significantly lower (indicating increased levels of noise within the acoustic signal). The lack of identified differences within these parameters between the DS and TD groups in this study is therefore a stark contrast to previous research in children with DS. Lee, Thorpe and Verhoeven (2009) did find reduced jitter values in young adults with DS in comparison to TD peers, however they had attributed this unexpected result to increased vocal awareness as all the DS subjects in their study belonged to a theatre youth group. As the children and young adults in this study had higher jitter results than their peers (just not significantly so) it does seem likely that the reduced jitter seen by Lee, Thorpe and Verhoeven is not typical of all young people with DS.

It is clear that children with DS from the Edinburgh area have similar jitter, shimmer and HNR profiles to those of their age-matched TD peers from the same geographical area, however what is not clear is whether the lack of differences is a consequence of vocal differences within the DS speakers from this area in comparison to DS speakers in previous studies or within the vocal profiles of the control subjects in comparison to controls from other areas. Although some degree of natural variation is expected in speakers from different geographical locations, as speakers with DS are recognised to be somewhat constrained in their voice production by their atypical vocal tract physiology, it would be considered unlikely that such factors could be overcome to the extent that a significantly different DS vocal profile could be found in the SE of Scotland than to other parts of the UK. Thus differences in the vocal quality of the TD control subjects from this area in relation to controls used in previous DS studies must be considered to be the most likely factor in the lack of identified differences in jitter, shimmer and HNR. Unfortunately, due to differing methodologies (the Pentz & Gilbert (1983) study analysing perturbation and HNR in isolated vowels only) it is not possible to directly compare the TD values found in that study against those identified in this acoustic analysis, making it difficult to establish conclusively whether the TD values of Edinburgh speakers differ fundamentally to those of controls in previous research. The Moura et al. (2008) research also uses isolated vowel samples, but moreover as language-specific differences in F0 findings have already been indicated between this study and the Portuguese DS research findings it seems unwise to directly compare norms across other acoustic parameters. Presently, there is no normative acoustic analysis data for young Edinburgh speakers to compare against these TD findings and as such no way of determining whether the perturbation and HNR values found in the TD controls of this study are higher than expected.

A further point to consider is the finding by Yui (1999), that although increased jitter and shimmer have been found to be able to support differentiation between dysphonic and non-dysphonic voices (values being increased in dysphonic speakers) some dysphonic speakers have been found to have acoustic measurements which fell within normal limits. More specifically, jitter and shimmer have been found to fail to differentiate between male speakers with and without identified voice disorders, whilst HNR has been similarly unsuccessful for both genders, causing Estella, Maa and Yui (2005) to question the sensitivity of these measures in clearly distinguishing between typical and disordered voices in all instances.

Despite the absence of supporting data from previous research, this study does indicate that there are surprising similarities in the acoustic vocal profiles of DS and TD children within the Edinburgh area. As the control subjects have by definition typically-developing vocal tract anatomy and function, it seems logical to suggest that other factors, such as sociolinguistic norms for this area, may give rise to the similar acoustic properties of voice observed. Increased jitter and shimmer have been found to be associated with harsh phonation type (Farrús, Hernando & Ejarque, 2007), whilst lower HNR correlates with increased whisper or breathy voice (Mathieson, 2001); as both these phonation settings are closely associated with DS speakers (Montague & Hollien, 1973) it may be the case that harsh and whispered phonatory settings are also typical of TD Edinburgh speakers resulting in the similar acoustic profile. In study 3 expert judgements of the perceptual quality that the DS and TD acoustic profile gives rise to will enable examination of the nature of how these features are produced in both groups in terms of their habitual vocal tract, muscular and phonation settings. From the acoustic analysis findings it is anticipated that the TD group will reflect similar phonatory settings to the DS group (increased harsh and whispered phonation as a consequence of the similar jitter, shimmer and HNR findings) but that differences in the way these phonation features are produced will be identified in the degree of muscular tension and habitual vocal tract settings used. This proposal will be investigated further in the discussion of the results of study 3.

5.3.1.3 Spectral tilt (1-5kHz & 2-5kHz)

The only parameter to differ significantly between the DS and TD groups was that of SPT, with the DS speakers having significantly higher SPT values within analysis of both the 1-5kHz and 2-5kHz frequency ranges. According to Lofqvist and Mandersson (1987) SPT or spectral slope is the rate at which the strength of the harmonics in the glottal source decrease as their frequency increases. As the frequencies of the various harmonics reflect the number of the harmonic multiplied by the F0, the F0 level is related to the degree of SPT. Also significant is the rate of change of airflow during the phonatory cycle, meaning that SPT will vary considerably depending on the phonatory setting of the speaker at any time (Clark & Yallop, 1995). More specifically, during the cyclical phases of vocal fold opening and closure, the closure phase occurs more rapidly than the opening phase and it is this abrupt vocal fold closure that is responsible for the generation of the majority of the energy in an acoustic wave, particularly the higher frequency harmonics (Baken & Orlikoff, 2000), thus with increasing effort the vocal fold mechanism becomes more efficient, and there will be a less rapid reduction in the strength of harmonics, equating to a lower SPT value. In typical voice production increased effort will result in increased volume; as a louder voice is associated with more high frequency harmonics there is a strong relationship between the degree of SPT and vocal amplitude (essentially a louder voice will have a lower SPT and the level of SPT will increase as the amplitude decreases). Thus SPT is expressed in decibels (dBs) per octave, the value indicating the degree of attenuation (weakening) associated with each doubling of harmonic frequency. A typical SPT value is recognised as approximately 12dB per octave, whilst a much lower slope of 6dB per octave would be indicative of the high frequency energy not tailing off so quickly, resulting in a more strident sound.

SPT has received comparatively little attention in comparison to other acoustic features of voice and presently the only other research to examine the SPT profile of individuals with DS is that of Moura et al. (2008) in young Portuguese-speaking children, aged 3-8 years of age. Intriguingly, Moura et al. found the opposite pattern of SPT in their DS speakers, finding them to demonstrate significantly lower SPT values than the TD control subjects. The Moura et al. study examined values in sustained vowel samples, whilst the current study analysed across all voiced segments of speech; Hillenbrand and Houde (1996) note that differing methodologies can impact on findings, stating that SPT measures used within a reading passage were found to have substantially greater predictive power compared to measures used in isolated vowels, indicating that findings from sustained vowels may not necessarily translate to more complex speech, and possibly explaining the differences between the two studies.

Moura et al. related their lower finding of SPT to a more breathy and forced voice in DS speakers than TD controls. In contrast Baken and Orlikoff (2000) associate breathy phonation with a steeper SPT of approximately 18dB per octave, as the glottal closure is more sluggish in this phonation type (i.e. having a less efficient/rapid closure mechanism) resulting in the production of less powerful higher harmonics which tail off more quickly. This view is shared by Klatt and Klatt (1990) who note that alongside an increase in noise in the higher harmonics, breathiness is seen to increase the amplitude of the first harmonic which may lead to a greater SPT. In study 3, it is discussed whether a greater incidence of the perceptual quality of whispered or breathy phonation (which could be associated with the higher SPT values observed) was found in the DS group.

The degree of muscular tension within the walls of the vocal tract is also considered to be a factor in SPT values. Greater muscular tension within the pharynx would cause less acoustic damping, resulting in reduced absorption of acoustic energy as the strength of the harmonics decline less rapidly than would be seen in a more lax muscular setting (Guion, Post & Payne, 2004). Similarly, within adult populations hypoadduction of the vocal folds has been found to correspond to higher SPT values, whilst the reverse pattern of hyperadduction has been found to be directly related to lower SPT (Mendoza, Munoz, & Naranjo, 1996). The findings of higher SPT within the DS speakers in this study would support a hypotonia-related hypothesis, whereby the low muscle tone associated with DS affects negatively both the muscular tension of the vocal tract and the efficiency of vocal fold closure.

5.3.2 DS and TD groups split by gender

Mann Whitney U-tests were used to identify statistically significant differences across all the acoustic parameters studied both between and within DS and TD speakers when grouped by gender.

5.3.2.1 Comparison of DS and TD male speakers and DS and TD female speakers

When the male DS speakers (13) were compared to the male TD speakers (34) and the female DS speakers (9) compared to the female TD speakers (18) the same pattern as was found for the overall groups was identified, with the only significant differences between groups being found for the parameter of SPT. In the case of the males, SPT values for both 1-5kHz and 2-5kHz were significantly greater in the DS males than the TD males (1-5kHz: DS males, 16.91 (4.66); TD males, 13.47 (2.54), $p = 0.001$, & 2-5kHz: DS males, 23.33 (3.13); TD males, 19.68 (4.22), $p = 0.003$) whilst only the higher frequency SPT values differed significantly between the DS and TD females; again the DS females evidencing higher SPT values than the TD females (2-5kHz: DS females, 22.67 (3.32); TD females, 19.59 (4.59), $p = 0.011$). Despite not being statistically significantly different, the females with DS also had a greater SPT at 1-5kHz than the TD females, indicating the same direction of differences as found at higher frequencies, if not the same extent; it is possible that the smaller sample size of the female groups had an impact on statistical analysis. These findings indicate that differences are not gender-specific.

5.3.2.2 Comparison of female and male speakers within both DS and TD groups

Within groups, significant differences were found between female and male speakers for both the DS and TD groups.

For the DS speakers significant differences in F0 mean, F0 mean (stdev) and HNR were identified, with female DS speakers demonstrating significantly higher values for each than male DS speakers (F0 mean: DS female, 236.71 (72,32); DS males, 136.63 (75.06), $p = 0.006$; F0 mean (stdev): DS females, 18.28 (10.49); DS males, 9.48 (7.14), $p = 0.025$; HNR: DS females, 14.94 (2.59); DS males, 12.91 (1.66), $p = 0.025$).

A very similar pattern between females and males was also found for the TD group, with statistically significant differences being identified in F0 mean (TD females, 226.4 (33.66); TD males, 174.73 (104.66), $p < 0.001$); F0 mean (stdev) (TD females, 12.6 (2.93); TD males, 9.14 (6.52), $p = 0.001$) and HNR (TD females, 14.08 (2.7); TD males, 12.21 (3.47), $p = 0.003$); in each case, like the DS females, the TD females were found to have higher scores than the males. However the TD group differed from the DS group in that a significant difference was also found between females and males for the parameter of shimmer (apq5), with females having a lower shimmer value (TD females, 5.15 (1.38); TD males, 6.25 (1.84), $p = 0.006$).

F0 is a well-recognised marker between male and female voices across languages; the average F0 of adult American English-speaking females being reported to be approximately 1.7 times higher than is found in American males (Klatt, 1987) and similarly within Spanish speakers, females have been found to have an F0 level approximately 1.8 times higher than that of their male counterparts (Trittin & de Santos y Lleó, 1995). In adult speakers differences in F0 between genders are recognised to be principally a consequence of physiological differences between the vocal tracts of males and females. The vocal folds of adult males have been found to be up to 50% larger than those of females (Henton, 1992) which during phonation results in a longer oscillation period of the periodic signal, producing a lower F0

(Heffernan, 2004). Other non-physical factors, such as social environment, education and the influence of the geographical area that a speaker lives in can also influence male to female vocal differences (Carlson & Granström, 1997) and furthermore Whiteside and Marshall (2000) argue the case for a sociophonetic factor, in that women tend to articulate more carefully.

According to Raymond and Kent (2003) there are fundamental differences in the size and composition of the larynx between young children and adults. In infants the vocal folds are between 3 and 5mm in length, which by five years of age will have reached approximately 7.5mm. The distinctive layering of the folds, which is termed 'lamination', is absent in infants, and will gradually develop over time, with the lamina propria (the outermost layer of the folds) not being complete until at least twelve years of age. Even up to the age of twelve, the dimensions of the larynx in pre-pubescent children have been found to be approximately twice as small as those of adults (Wysocki, Kielska, Orszulak & Reymond, 2008). There is some disagreement regarding whether differences exist in the physiology and size of the larynx between young boys and girls, however it is reported that any slight distinctions do not contribute towards significant differences in F0, both having a mean of approximately 250Hz until puberty (Raymond & Kent, 2003). Some small differences in the course of F0 development between genders have been noted prior to puberty; Hacki and Heitmuller (1999) finding that habitual pitch and speaking pitch range lowered between the ages of eight and nine years for boys and between seven and eight years for girls. Glaze, Bless, Milenkovic and Susser (1988) propose that differences pre-puberty may be correlated with increasing age, height and weight, however, Kent (1976) notes that changes in the F0 of children may not necessarily reflect age related development, but rather methodological differences between studies and variability in vocalisations by subjects. The onset of puberty gives rise to a range of anatomical changes in the human vocal tract which have considerable implications for the development of adult-like voice and signal the onset of emerging differences in F0 between boys and girls. Alongside growth of the various structures, a change in their relative positions within the vocal tract also occurs. Primarily, this is seen in the larynx, which is found to lower (de la Bretèque,

1990) and increase in circumference, with males exhibiting a greater overall increase than females (Wysocki, et al., 2008) which correspondingly, causes a greater drop in F0 in boys than girls (Pedersen, Møller, Krabbe, Bennet & Svenstrup, 1990); early studies showing the decrease in females to be about half that of males (Linke, 1953; McGlone & Hollien, 1963). Further to this, growth is evident in the tip of the epiglottis, the hyoid, the glottis, and the inferior margin of the cricoid cartilage, which all lower significantly in relation to the cervical spine (Westhorpe, 1987). The gradual nature of voice change, together with individual differences makes it difficult to identify a clear point of change between child and adult voice features (Andrews and Summers, 2002; Fuchs, Fröehlich, Hentschel, Stuermer, Eberhard Kruse & Knauft, 2007). However, according to Raymond and Kent (2003) the greatest downward shift in F0 in boys can be seen between the ages of twelve and fifteen years. Lee, Potamianos and Narayanan (1999) reported a 78% decrease in F0 for males between these ages and no significant change was observed after the age of fifteen years, indicating that voice change is more or less complete by that age. Similarly in females early research showed that no significant difference was found between the F0 of girls aged fifteen, sixteen and seventeen years and young adult females (Michel, Hollien & Moore, 1965) indicating that voice change must occur at an earlier age. Hollien and Paul (1969) later confirmed that changes in F0 occur in most girls before the age of fifteen years, becoming reasonably stable during the immediate post-pubescent period. Based on this evidence, the significant differences found in F0 between the male and female DS speakers and the male and female TD speakers in this study is likely to be indicative of the presence of adult-like voices in some of the older subjects in both groups as a consequence of the developmental growth associated with the onset of puberty.

As well as the significant differences found in the median values of the F0 mean and F0 mean (stdev) parameters it was also notable that the females with DS demonstrated a much greater IQR value for F0 mean (stdev) than the males with DS. This higher value indicates a wider variation between the range of F0 mean (stdev) values of individual speakers within the female group (i.e. some speakers have high variability in F0 whilst others have much less variation around the mean F0 value).

Those individuals with higher F0 mean (stdev) values would be perceived by listeners as having a wider pitch range and conversely those with lower values would be perceived as having a smaller pitch range; a particularly narrow range being perceived as monotonous. Interestingly the opposite pattern was observed in the TD group, where the IQR of the male TD speakers for F0 mean (stdev) was considerably wider than for TD females, again showing increased variability in the F0 mean (stdev) values between individual male speakers. These findings are confirmed perceptually in both the DS and TD groups on listening to the recordings, where there is a notable difference between some of the younger speakers who have a marked 'sing-song' quality, evidencing a wide pitch range, compared to some of the older speakers whose speech is somewhat lacking in inflection.

The male TD speakers also had a wider IQR for the F0 mean parameter, which indicates a wide variation in the F0 mean values across the group as a whole (i.e. some speakers have low average F0 and others higher average F0) and is likely to be indicative of the spread of pre-pubertal voices through to adult-like voices; again confirmed perceptually when listening to recordings. As adult-like male voices have a lower F0 than female voices and pre-pubertal male and female children have roughly the same F0 level, the difference between the F0 of the youngest and oldest speakers is therefore going to be greater for males than for female speakers.

HNR is a measure of the amount of additive noise in the voice signal which is measured in dB. Two principle factors are related to HNR. Firstly, greater levels of noise in the acoustic signal are suggested to be a consequence of the production of turbulent airflow at the vocal folds (Hillenbrand, 1987) occurring as a result of excessive airflow due to inadequate or ineffective closure of the folds (Ferrand, 2002). Secondly, aperiodic vocal fold vibration can also result in increased noise levels, in contrast to periodic vibrations which give rise to the harmonics of the speech waveform (Ferrand, 2002). Both jitter and HNR are predictors of additive noise; although Ferrand (2002) observed that substantial changes in HNR within the speech signal corresponded to little or no changes in jitter, prompting the suggestion that HNR is a more sensitive measure of additive noise than jitter. It should be noted that some studies report noise-to-harmonic ratio (NHR) rather than harmonic-to-noise ratio (HNR) thus in the former increased values represent increased noise, whilst conversely in the latter increased values indicate reduced levels of additive noise in the speech signal.

A range of HNRs have been reported for adults without vocal pathologies, from an average of 7.23dB (Bertino, Bellomo, Miani, Ferrero & Staffieri, 1996) up to 18 dB in healthy adult Spanish speakers (Fernández Liesa et al., 1999) with other research finding a more middle-ground, indicating values of between 11dB to 13 dB (de Krom, 1993; Murry, Brown & Rothman, 1987) thus making it difficult to identify a specific, typical HNR value. This disparity may reflect differences in measurement techniques, differences in the type of recording used and differences between subjects (Ferrand, 2002) making comparison across studies problematic. Values appear to be relatively stable in young adults and through middle-age, with a tendency to decrease in later years (Ferrand, 2002). Less data is available on paediatric norms, but Ferrand (2000) does report the HNR of children aged four, five, eight and ten years to be lower than adult levels with girls having a significantly higher HNR (i.e. lower additive noise) than boys in some vowel contexts but not others. Fundamental differences between child and adult HNR values suggest that comparison of acoustic values would not be valid (Ferrand 2000).

Significant differences have been identified between genders, with some conflicting results between studies. When measuring NHR, Rodrigues, Behlau, Pontes (1994) found levels of 8.63dB and 10.17dB for males and females respectively, whilst Naufel de Felippe, Grillo and Grechi (2006) reported similar values of 9.56dB in males compared to 10.98dB in females (also NHR); both indicating that females have higher levels of additive noise than males. In contrast, Heffernan (2004) reported the opposite pattern of significantly lower levels of noise (higher HNR values) in both Japanese speaking and English speaking females compared to their male counterparts when measuring the phoneme /s/ (HNR: Japanese females, 13.8dB versus males, 11.95dB; and English speaking females, 15.22dB, versus males, 12.35dB).

Within this study both the DS females and the TD females had higher HNR values than the DS and TD males respectively (DS females, 14.94dB; DS males, 12.91dB & TD females, 14.08dB; TD males, 12.21dB); these results are in line with the direction and values of those of Heffernan (2004) and show that the speech signals of both DS and TD females from the Edinburgh area have significantly less additive noise than those of male DS and TD speakers respectively. Both DS and TD speakers evidenced similar HNR values indicating that additive noise is not a feature which separates these two groups.

The degree of noise in the acoustic signal is regarded as a useful quantitative measure in the confirmation of a perceptual diagnosis of dysphonia (Jotz, Cervantes, Abrahã, Settanni & Carrara de Angelis, 2002); lower HNR (i.e. increased noise) is reported as being perceived as 'rough' (Eskenazi, Childers & Hicks, 1990; Martin, Fitch & Wolfe, 1995), 'hoarse, breathy and rough' (McAllister, Sundberg & Hibi, 1996) and 'rough, gravelly and breathy' phonation (Mathieson, 2001). Female voices have a tendency to be more breathy or whispered than male voices, which according to Henton and Bladon (1985) is a consequence of having a significantly higher first than second harmonic, as well as increased aspiration noise in the vowel spectrum (Klatt & Klatt, 1990); although significant variation between speakers is reported within genders. Increased breathiness in females may not be a universal finding, as Trittin and de Santos y Lleó (1995) report that breathiness in Spanish females is less apparent than in American females and that it does not differ significantly between Spanish females and males, which they suggest as evidence towards the assumption that breathiness may be a 'learned, cultural behaviour'. This is supported by findings of differences in degree of breathiness between different speech communities; significant differences in HNR being found between Italian and Polish male speakers (Wagner & Braun 2003), African American English and Standard American English speakers (Purnell, Idsardi & Baugh, 1999) and between received pronunciation (RP) speakers and an accent group termed Modified Northern speakers in the UK (where both males and females in the latter were more breathy than the RP speakers) in a study by Henton and Bladon (1985). Heffernan (2004) states that such differences strongly support the claim that breathiness is not solely a consequence of physiological differences within the vocal folds of female and male speakers.

The females in this study (both DS and TD) did not have increased noise in relation to the males, and thus these findings do not seem to support the presence of more breathy phonation being associated with female speakers. On face value the contrary (increased breathiness in males) would seem to be the case, however, as rough or harsh phonation is also associated with increased noise (lower HNR) it may possibly be the case that the higher noise levels found in both the DS and TD males is associated with increased perturbation. The perceptual analysis in study 3 discusses further the particular phonation type that is associated with the relative HNR values of the male and female DS and TD speakers.

As breathiness is a consequence of additive noise in the higher frequencies of the speech signal, due to incomplete adduction of the vocal folds as a result of low muscular effort (Laver 1980) it would have been expected that the hypotonia associated with DS speakers would have resulted in significantly lower HNR values, reflecting increased noise, in both males and females. The lack of differences in HNR values across the groups as a whole suggests that other factors, perhaps sociolinguistic, are at play, whereby the TD speakers make voluntary adjustments to their vocal tract settings which result in a similar breathy/harsh quality to that of the DS speakers whose settings are constrained by their altered physiology. Again, this proposal is evaluated within the perceptual analysis of study 3.

Shimmer or amplitude perturbation is defined as short-term instability in the intensity of the vocal signal, the measurement of which quantifies the degree of variability in intensity of the fundamental vocal note (Mathieson, 2001), which in this study is expressed in percentage form. Increased shimmer values are linked to a range of vocal pathologies such as vocal fold lesions, spasmodic dysphonia and unilateral laryngeal nerve paralysis, although inconsistency in the airflow through the glottis during phonation can also lead to this type of irregularity (Hirano & Bless, 1993). Typically it is the imbalance in mass and tension between the vocal folds which produces irregular vocal fold function (Titze, 1994) however, although increased perturbation values are associated with dysphonic voice, even within speakers without vocal pathology some irregularity in the glottal cycle is to be expected (Bonilha & Deliyski, 2008). Higher shimmer has been found to correlate with breathiness ratings (McAllister, Sundberg & Hibi, 1996), but in contrast it has also been reported that shimmer appears to be more related to the irregular vocal fold function associated with rough phonation (Kreiman, Gerratt & Percoda, 1990; Wolfe & Martin, 1997) rather than "the unmodulated airflow accompanying phonation in the breathy voice type" (Awan & Roy, 2005, p 277).

In adult populations average shimmer values have been reported to be 2.25% in males and 1.98% in females (Robinson, Mandel & Sataloff, 2005) whilst within paediatric populations Nicollas, Garrel, Ouaknine, Giovanni, Nazarian, and Triglia (2008) reported that shimmer values did not differ between genders or with increasing age in children between the ages of six and twelve years. It has been found recently that both jitter and shimmer increase with decreasing vocal loudness and that gender differences may be related to differences in habitual loudness levels adopted by male and female speakers (Brockmann, Storck, Carding & Drinnan, 2008).

In this current study, differences were found between genders within the TD group only (shimmer (apq5): TD females, 5.15 (1.38); TD males, 6.25 (1.84), $p = 0.006$). Some degree of difference in shimmer between gender is unsurprising given that the ages of the TD females (median age, 14.67) and males (median age, 14.08 years) in this study are older than those examined by Nicollas et al. (2008) and thus may be beginning to show adult-like values. Although these shimmer values are higher than the findings reported in adults by Robinson, Mandel and Sataloff (2005) they do conform to the expected profile of greater values being present in male speakers than female speakers. As this study analysed connected speech rather than isolated vowels, higher perturbation values are not completely unexpected.

Interestingly an area that did not differ between genders, which would have been expected to from previous research findings was that of SPT. Research has shown that SPT values tend to be higher in females than males principally as a direct consequence of anatomical differences in the thickness and size of the vocal folds and in the increased length of the vocal tract; these physical attributes producing lower formant frequencies in male speakers (Ho, 2001). The fact that both DS and TD females failed to conform to this predicted SPT profile suggests that there is something in common with both groups that over-rides the effects of their smaller vocal tract physiology. As Ho (2001) notes that speaking habits also consistently affect vocal patterns, again it seems reasonable to suggest that there are some sociolinguistic effects at work which either cause the SPT of young Edinburgh females to be slightly lower than expected or conversely cause the values of young Edinburgh males to be higher, resulting in some degree of overlap between the genders in both DS and TD populations.

5.3.3 Comparison of DS and TD speakers presented to listeners in study 1

Within the analysis of differences between the overall DS and TD groups significant differences were found only in SPT; levels being higher in the DS group than the TD group. These differences were reflected when the overall groups were divided on the basis of gender, with male DS speakers having greater SPT values at both 1-5kHz and 2-5kHz than TD male speakers, and female DS speakers demonstrating a steeper SPT at higher frequencies (2-5kHz) than TD female speakers. Thus it would have been expected that the same pattern of increased SPT would have been apparent in the DS speakers presented to listeners in study 1; in fact this was not the case, with no significant differences being identified by Mann Whitney U-tests between the eight DS and eight TD speakers across F0 mean, F0 mean (stdev), jitter (ppq5), shimmer (apq3 & apq5), HNR or either of the SPT measures. However, on closer examination of the data it is evident that the trend in SPT is in the expected direction (greater in DS speakers) therefore it is likely that the failure to find significant differences is a consequence of small group size.

Also surprising is the finding of a significant difference between the groups for the parameter jitter (rap) (DS, 1.01 (0.35), TD, 0.83 (0.26), $p = 0.05$). No differences in jitter measures were found in the overall groups or when the groups were divided for gender. As the DS and TD speakers from study 1 were predominantly male (7 out of 8 speakers in each group) they would have been expected to echo the acoustic analysis profile shown between overall DS males and TD males.

In adults a range of average jitter values have been reported, not always with good agreement. Jitter values of 0.69% were found in young adults, 0.57% in middle-aged adults and 0.66% in the elderly (Ferrand, 2002), while in contrast, Wilcox and Horii (1980) found that older subjects obtained higher levels of jitter than younger subjects (0.73% and 0.55%, respectively). Several studies have found jitter levels in females to be lower than values for males, although not finding those differences to be significant (Horri, 1980; Behlau & Tosi, 1985; Morente et al., 2001). In contrast other research has identified the opposite pattern of higher jitter levels in females. Mean values of 0.54% in healthy males and 0.63% in healthy females were reported by Robinson, Mandel and Sataloff (2005). Similarly in an analysis of vowels /a/ and /é/ also in healthy adults between twenty and forty-five years, females were found to have average levels of 0.62% and 0.59%, with males having slightly lower values of 0.49% and 0.5% respectively (Naufel de Felippe, Grillo & Grechi, 2006). Higher female values were also found by Araujo, Grellet and Pereira (2002); females speakers having an average level of 0.85% compared to 0.37% in males.

In an analysis of two hundred and twelve children, jitter, like shimmer, was found not to differ significantly with age or gender (Nicollas, Garrel, Ouaknine, Giovanni, Nazarian & Triglia, 2008). This concurs with earlier research of sustained vowels of women, men, and six to nine year old children, where significant gender differences were found only for the adult speakers (Sussman & Sapienza, 1994).

Differences in findings are likely to be a consequence of the variability in methods between different studies and the acoustic analysis packages used (Naufel de Felippe, Grillo & Grechi, 2006) which makes it difficult to compare findings directly. In illustration of this, it has been reported that perturbation measures appear to be more reliable when applied to sustained vowels rather than across connected speech samples (McAllister, Sundberg & Hibi, 1996).

Perceptually, jitter, along with HNR has been found to correlate at a reasonable level with hoarseness, breathiness and roughness, whilst jitter, HNR and shimmer correlated with the perception of breathiness (McAllister, Sundberg & Hibi, 1996).

In this study, it may be the case that the editing process used for the recordings of the speakers in study 1, whereby the longer and more complex utterances of the TD speakers were cropped to be closer in nature to those of the DS speakers (who had lesser language skills as a consequence of intellectual disability) had some effect in producing results which differ to overall findings. Although this finding does support the a priori hypothesis of increased jitter in DS speakers, as the finding of increased jitter (rap) is only just significant ($p = 0.05$) and the second jitter (apq5) measure is not significant, and taking into account the small group size and most significantly the failure to reflect the pattern of the larger, overall DS and TD groups, it is unlikely to be truly representative and thus should be interpreted with caution.

5.3.4 Summary

In common with the earlier research findings of Michel and Carney (1964), Hollien and Copeland (1965), Montague, Brown and Hollien (1974) and Pentz and Gilbert (1983) acoustic analysis of recordings of the overall DS and TD speakers has shown that no significant differences in mean F0 level exist between the two. Findings are at odds with the increased F0 in young Portuguese-speaking children with DS identified by Moura et al. (2008), suggesting that there may be some language-specific component. Although no difference was found in F0 mean (stdev), DS speakers were observed to have a wider IQR, demonstrating increased variation within the DS group, reinforcing the heterogeneous nature of DS speakers.

No significant differences were found in measures of jitter, shimmer and HNR, which is a stark contrast to earlier findings of increased jitter and shimmer and decreased HNR in English and Portuguese-speaking children with DS respectively (Pentz & Gilbert, 1983; Moura et al., 2008); thus failing to support the hypotheses of increased jitter and shimmer and decreased HNR in the DS speakers in this study. The lack of identified differences may be related to sociolinguistic features of TD Edinburgh speakers resulting in vocal settings which have similar acoustic properties to those of speakers with DS. It should be noted that research has shown that jitter, shimmer and HNR can be unreliable measures in distinguishing dysphonic and non-dysphonic voices (Estella, Maa & Yui, 2005).

The only significant difference found between the overall DS and TD groups was that of SPT, with DS speakers having greater values at both 1-5kHz and 2-5kHz than TD speakers. Again this failed to support the hypothesis of lower SPT in DS speakers and is in direct opposition to the findings of lower SPT in Portuguese-speaking children with DS by Moura et al. (2008). Higher SPT would be consistent with the finding of increased values associated with hypoadduction of the vocal folds (Mendoza, Munoz & Naranjo, 1996) which would be a likely consequence of the hypotonia that is regarded as being associated with DS speakers.

When divided by gender, DS males and DS females (in comparison to TD males and females respectively) were found to reflect the same pattern as the overall groups, with differences being identified only in the parameter of SPT (at both 1-5kHz & 2-5kHz for males, and at the higher frequencies only for females); thus indicating that differences and similarities between the acoustic profile of DS and TD speakers are not gender-specific.

Within groups, differences between genders again followed a very similar pattern, with both DS and TD females having significantly higher levels of F0 mean, F0 mean (stdev) and HNR than their male counterparts. As F0 tends to be very similar in prepubescent children (Raymond & Kent, 2003) differences are likely to be an indication that developmental changes in anatomy which accompany the onset of puberty have taken place in some speakers in both the DS and TD groups. Increased HNR levels in DS and TD females indicate the presence of less additive noise in the acoustic signal than was found in males in both groups; earlier conflicting findings between genders might suggest that differences are to some extent sociolinguistic in nature.

In addition TD females were found to have lower levels of shimmer (apq5) than TD males, which is a finding in common with the adult study of Robinson, Mandel and Sataloff (2005); although the shimmer values are higher (in both DS and TD groups) which may reflect the analysis of all voiced segments of connected speech rather than of isolated, sustained vowels. As Nicollas et al. (2008) found no difference in the shimmer values of children under the age of twelve years this is a further indication of developmental change occurring within this group.

Unlike earlier research no significant difference was found between genders within either group for SPT, which may again reflect sociolinguistic constraints.

Finally within the eight DS and eight TD speakers presented to listeners in study 1, the findings of the overall DS and TD groups were not replicated; the only significant difference being found for jitter (rap) with the DS speakers having a higher value. The modest significance level ($p = 0.05$) taken into account with the small group size and the failure to reflect the profile of the larger sample (of which these speakers were a subset) indicates the need to exercise caution extrapolating this finding to the wider DS population.

The relationship between these acoustic findings and the perceptual quality of DS and TD speakers from the Edinburgh area are examined in the following study 3.

5.4 STUDY 3: PERCEPTUAL ANALYSIS OF VOICE

The perceptual analysis findings from the VPAS for the overall DS and TD groups, the groups split by gender and the DS and TD speakers presented to listeners in study 1 will be discussed in relation to the twelve individual sections of the VPAS (labial settings, mandibular settings, lingual settings, pharyngeal settings, velopharyngeal settings, larynx height settings, muscular tension settings, phonation features, pitch, loudness, temporal organization and other features).

5.4.1 Consistency of rater judgements

Agreement between the two VPAS raters was extremely high across the twelve sections; the lowest percentage agreement figure being 85.71% for judgements of 'tongue body raising' for the DS speakers. This high inter-rater reliability suggests good validity of results.

5.4.2 Labial settings

5.4.2.1 Spreading – rounding/protrusion

The overall DS group were found to have a significantly more rounded/protruded lip pattern than their TD peers, supporting the a priori hypothesis. This finding is unsurprising given the physical appearance of the lips of people with DS which have a strong tendency to be thick and everted (Mackenzie Beck, 1997). The same finding of increased rounding/protrusion in the DS speakers was also found in the analysis of the speakers from study 1.

When the overall groups were divided on the basis of gender some interesting results were found. As would be suggested by the similarities in physiology between females and males with DS no significant differences were observed between the two (although there was evidence of more rounding protrusion within the females with DS). This finding is in sharp contrast to the finding of significantly greater levels of lip spreading in the TD females compared to their TD male counterparts who were associated with more neutral lip settings. A spread lip position is consistent with a more ‘smiley’ lip pattern which may be indicative of a difference in attitude towards the task (females possibly being more relaxed in their participation or more eager to please than the male participants). Holmes (2001) states that within society females typically use more standard forms of language and pronunciation than males and that there is a greater expectation on them to not cause offence, and as such they tend to be more sensitive to their communication partners. Indeed, in TD populations research has shown that women smile more in situations that are tense or strained than males (as might be the case when meeting the researcher for the first time), which is suggested to be a consequence of females feeling the desire to try to make the situation more relaxed (Woodzicka & LaFrance, 2004). From this it seems reasonable to suggest that the spread lip setting might be an expression of this compliant or amiable female behaviour. Although the males with DS had slightly greater rounding/protrusion values than the TD males, the difference was not found to be statistically significant. In contrast, marked (and statistically significant) differences were evident between the DS and TD female speakers. The females with DS had very high levels of lip rounding/protrusion compared to the spread setting observed in the TD females, thus it would seem that the females with DS were not conforming to the gender-specific lip setting differences associated with typical-development. From a sociolinguistic perspective it might be the case that the habitual rounded/protruded lip pattern of female DS speakers may have the effect of making them appear less socially accommodating or friendly than their TD female peers. As research has shown that not only do females smile more than males but that females who do not conform to this gender-specific trait are perceived as being less happy than their male counterparts (Deutsch, Lebaron & Fryer, 1987) then this lack of conformity may have particular negative consequences for how females with DS are perceived. However, the isolated feature of less spread lip setting may be counterbalanced in DS by the general appearance of what Zebrowitz (1997) terms ‘babyfaceness’, which is characterised by features associated with immature cranial development such as a small nose, larger forehead with a sunken bridge, fuller cheeks and a rounder chin which are suggested to elicit more protective and nurturing reactions from communication partners.

Aside from the gender differences, early research into the voice and social status (based on social index scores) of thirty-two adult speakers from Edinburgh found lip setting to be a differentiating factor between high and low status. A spread lip setting was more associated with speakers with higher social standing, whilst those with lower social status were described as having a 'pursed' lip setting (Esling, 1978). Thus the habitual rounded/protruded lip pattern of people with DS may also be associated with lower social groups.

Rounded or protruded lip posture also has the effect of altering the dimensions of the vocal tract, lengthening it slightly which can cause a reduction in the level of pitch (Rammage, Morrison, Nichol, 2001) most notably within the higher formants (Laver, 1980). The consequences of altered pitch features will be considered in the 'Pitch' section later in the discussion.

Also apparent was a tendency for a much wider range of spread-rounded/protruded lip values being associated with the DS speakers than the TD speakers, a finding which indicates the heterogeneity of people with DS. As such, features like protruded lip setting may be generally associated with the condition but not necessarily typical of the habitual pattern of all speakers with DS.

5.4.2.2 Labiodentalisation

No differences in labiodentalisation were found between any of the groups and this feature was not found to be typical of either the DS or TD groups studied.

5.4.2.3 Minimised – extensive lip range

Minimised range of lip movements was found to be associated with the DS speakers in the overall analysis and that of the speakers in study 1, both being significantly reduced in comparison to the TD groups; this finding is in keeping with the a priori hypothesis.

Again, when analysed on the basis of gender some interesting differences were evident. As in the spread-rounded/protruded findings there was no significant difference between the range of the male and female DS speakers, although the males had a slightly higher median (more minimised range) than the females. In contrast a significant difference was found between the TD females and males, where although the median value was the same (neutral) the males had a wider IQR indicating more variability between male subjects which extended further into the minimised range than the females. As previously discussed, females tend to use more standard pronunciation which could be argued to require a wider range of articulatory movements. Unlike the spread-rounded/protruded settings both the male and female DS groups were significantly more minimised in their lip movements in comparison to the male and female TD groups respectively.

Low muscle tone has been shown to cause lip function to be reduced in conditions such as the flaccid dysarthria seen after some strokes (Murdoch, 1990) thus as hypotonia is known to affect the muscles of the lips of individuals with DS (Limbrock, Fischer-Brandies & Avalle, 1991; Mizuno & Ueda, 2001) then it seems reasonable to suggest that the reduction in the range of lip movement seen in DS in this study is related to reduced lip muscular tone and strength.

In natural speech the lips move between spread and rounded positions according to the articulatory demands of individual segments. As lip setting impacts on pitch production then this limited range of movement is likely to impact on the variability of pitch. This hypothesis will be examined later in the 'Pitch' analysis section.

5.4.3 Mandibular settings

5.4.3.1 Close – open jaw

Clear differences were found between the overall DS group and the TD group for close – open jaw setting, with the DS speakers having a significantly more open jaw. This pattern was also found in the analysis of the speakers from study 1. Both results support the given hypothesis.

Between genders it was found that there was no significant difference between the females and males with DS. The musculature of the jaw requires more effort to close the jaw rather than open it (Van Riper & Irwin, 1958) therefore this finding would again be consistent with the presence of hypotonia which reduces the ability of both genders to achieve and sustain a typical jaw setting. Interestingly, within the TD groups, although the median value was the same (both neutral) the TD males had a wider range of jaw positions which extended further into the close jaw range than the females, causing a significant difference between the two. As in the labial settings it may be the case that the females adopted more standard forms which used a more open or relaxed jaw, or perhaps were slightly less anxious about the task than the TD males.

Esling (1978) found a higher incidence of close jaw in Edinburgh speakers with a higher social index than those with lower social indices (although in a second analysis of the same subjects he concluded that jaw closeness levels were not as high as previously identified). Similarly, in his study of eight boys from the same Edinburgh areas he found close jaw position to be only associated with those boys with higher social standing (female speakers were not analysed). These findings are confirmed by later research in Glasgow, finding that working-class speakers are distinguished from middle-class speakers by having (amongst other features) a high degree of open jaw (Stuart-Smith, 1999). In light of these findings, the more open jaw which is a feature of DS in this study might be at risk of being considered to be associated with lower social status.

5.4.3.2 Protruded jaw

Unsurprisingly, due to the underdevelopment of the maxilla which is associated with DS (Mackenzie Beck, 1988) and which causes the mandible to sit in a relatively more forward position, significantly higher levels of jaw protrusion were seen in the overall DS group and the speakers with DS from study 1. This again supports the a priori hypothesis.

As seen in the close-open jaw analysis, no significant difference was found between males and females with DS for protruded jaw, although the males did have a greater amount of protrusion (median level 3, which is on the border of an extreme degree for this setting, compared to a median rating of 2 for females). Similarly, no difference was found between the TD females and males, both having a neutral median rating indicating that protrusion is not characteristic of either gender within typical Edinburgh speakers. In contrast Esling (1978) found protruded jaw to be typical of the adults and male boys with lowest social status in his Edinburgh research; a finding which again associates the typical DS profile with that of those with lower social index scores. Again, jaw position, like lip position, appears to be a physiological constraint which might contribute towards negative sociolinguistic interpretation or misinterpretation of voice in DS.

5.4.3.3 Minimised – extensive range

Although the median values were identical for the range of jaw movements in the overall DS and TD groups, the group with DS had a wider IQR which included speakers with more minimised movements which created a significant difference between the groups; a finding consistent with the given hypothesis. The same values were found for the speakers from study 1 with DS, however no significant difference was identified in statistical testing; this is likely to be a consequence of the considerably smaller sample size.

No significant difference existed between the male and female DS speakers, which is likely to indicate that low jaw muscle tone and strength affects both genders equally. Within the TD groups there was a significant difference in jaw range (males having a wider IQR extending into the minimised range) however this difference seems to be a consequence of outliers in the male group rather than a typical male pattern (as the median and IQR were neutral for both groups, indicating that minimised jaw is not a typical Edinburgh feature), and when this analysis was repeated excluding the outliers no significant difference was found.

5.4.4 Lingual settings

5.4.4.1 Retracted – advanced tongue tip

Both the overall DS group and the speakers from study 1 had significantly more advanced tongue tip ratings than their TD peers. This is unsurprising given the relative macroglossia which is typical in DS as a consequence of the smaller oral cavity resulting from reduced growth of the maxilla and is in line with the given hypothesis.

When divided on the basis of gender no significant difference was found between the female and male speakers with DS. In contrast a significant difference was found between the TD gender groups with the females having a more fronted tongue tip setting. As the female TD group is actually slightly older than the male group, this disparity cannot be accounted for by developmental differences (younger children tending to have a more fronted tongue position) and thus is likely to be another example of sociolinguistic differences between genders. There is a general tendency for females to use a more fronted tongue setting (Mackenzie Beck, 2005a) which can bring about the perception of immaturity and vulnerability, and according to Mathieson (2001) may be used as strategy to appeal to the protective nature of the opposite sex; thus this finding would appear to be a typical gender difference. Although females are expected to have an advanced tongue position relative to males, the females with DS were found to have a significantly greater degree of fronting than their TD female peers. This difference was even more marked between the DS males and TD males, where the TD males favoured a neutral setting (not advanced and not retracted) whilst the median rating for the males with DS (in common with the DS females) was on the border of a severe presentation of advanced tongue tip and the IQR extended further into this severe range. This profile might have been expected to lead the DS speakers to produce speech sounds which were more typical of much younger children and consequently to sound a great deal younger than their chronological age, however, as shown in study 1 the DS speakers were actually judged as sounding older than their actual mean age; a finding which might reflect other vocal features rather than tongue position, such as phonation type.

5.4.4.2 Backed – fronted tongue body

In common with the advanced tongue tip and in keeping with the a priori hypothesis, the tongue body was also found to be significantly more fronted in the overall DS group. This finding was not replicated in the speakers from study 1, although the small group size may have been a factor as the results for this group were in the expected direction.

Given the lack of difference between the female and male DS speakers for tongue tip setting and the organic constraints associated with this condition, it is perhaps surprising that a significant difference was found between the two for tongue body setting, with the females having a significantly more fronted position. This difference was echoed between the TD females and males, demonstrating again that tongue fronting is more consistent with female habitual settings than male. No significant difference was found between the two male groups, although the TD males demonstrated a slightly backed tongue body whilst the males with DS had a neutral setting. The females with DS had the most fronted tongue body position (on the border of severe fronting) compared to the slight fronting (median rating of 1) observed in the TD females. In this instance the DS group appear to conform to the expected gender differences.

In Edinburgh adult speakers a backed tongue body was found to be typical of those with lower scores in social indices but not of those with a higher social status (Esling, 1978) and this setting was later replicated in working-class Glasgow speakers but not those judged to be middle class (Stuart-Smith, 1999). However, Esling noted that differences between social groups were not so marked within his study of boys in Edinburgh which he suggested was due to the relative reduction in the size of the vocal tract. Thus it is possible that the younger average age of the children and young people in this study compared to Esling's adult study may to some extent account for the minimal finding of tongue body backing in TD males. Another factor may be that despite canvassing for participants across the city of Edinburgh, this study may have failed to recruit speakers who had the markedly backed tongue position that Esling noted to be associated with areas of high social deprivation. As social index scores were not used in this study it is impossible to determine if this is the case.

According to Lin, Jiang, Noon and Hanson (2000) a more protruded tongue position is associated with increased jitter and shimmer values, and indeed the females with DS (the only group to have fronted tongue tip setting and tongue body setting on the boundary of the severe range) were found to have higher levels of perturbation than their male DS counterparts in the acoustic analysis.

5.4.4.3 <u>Lowered – raised tongue body</u>

The overall DS group were found to have a significantly raised tongue body setting in relation to their TD peers, a finding which supports the given hypothesis. Again, this finding was not observed in the DS speakers from study 1 who had identical median and IQR values to the TD speakers. It is possible that the younger age of the speakers in study 1 may have resulted in more raised findings in the DS group, although this was not the case in the TD group who had identical median (IQR) values to the overall group, thus it may be that small sample size has confounded findings.

When analysed by gender there was a clear division between the DS and TD groups with both males and females with DS having significantly more raised tongue settings than both the TD groups (DS females having a slightly higher tongue position than the DS males, although not significantly so). In contrast there was a significant difference between the TD females and males, females again having a slightly more raised tongue from the neutral ratings identified in the TD males. From these findings it appears that although raised tongue setting is associated with DS generally it is more so in females with DS, which echoes the gender pattern seen in the TD Edinburgh speakers.

These findings are intriguing as the more open jaw position which is typical of DS speakers would suggest a lowered tongue body setting, as the mandible is sitting further away from the roof of the mouth. However, it is likely that the relative macroglossia in DS, as a consequence of the smaller oral cavity volume from reduced growth of the maxilla, causes the tongue to sit in a higher position and thus overrides the open jaw posture.

5.4.4.4 Minimised – extensive range

In analysis of both the overall groups and the speakers from study 1, and in keeping with the a priori hypothesis, the speakers with DS were found to have significantly more minimised tongue movements than their TD peers.

A significant difference was observed between the male and female speakers with DS, with the males have a more minimised median rating and a wider IQR than the females with DS. The TD males and TD females were also found to have significantly different results however this was a consequence of outliers in the male group and when removed there was no statistical difference between the genders. As both groups had a neutral median value and an IQR of zero, it is clear that neither minimised nor extensive tongue body range is a characteristic of typical Edinburgh speakers and thus this is an area of significant difference between typical and DS speakers. Although the males with DS had the most minimised tongue body range of all the gender groups (the median rating being on the border of the severe range and the IQR extending into this area) the females with DS were also more minimised than both the TD males and females. As this feature is common to both genders with DS (as seen in the labial and mandibular results) this is most likely to be a consequence of a combination of the reduced oral cavity volume limiting tongue movement and the hypotonia associated with the syndrome.

5.4.5 Pharyngeal settings

5.4.5.1 Constriction - expansion

A clear distinction was found between the overall DS and TD groups for pharyngeal constriction-expansion, where the DS group has significantly higher levels of pharyngeal constriction; again supporting the given hypothesis. This finding was replicated in the eight DS speakers from study 1. In both cases the TD median rating and IQR were neutral indicating that neither pharyngeal constriction nor expansion are characteristic of typical Edinburgh speakers.

Females and males with DS had identical median (1) and IQR (1) values compared to the neutral median and IQR values in both the TD males and females, which suggests that the constriction is a consequence of organic differences within the vocal tract of people with DS (namely a more congested oral space as a consequence of lack of growth of the maxilla and the relative macroglossia which this causes).

A pharyngeal quality was found to be typical of both Edinburgh and Glasgow working-class speakers (Esling, 1978; Stewart-Smith, 1999) which in the case of the Glasgow speakers Stewart-Smith attributed to some retraction of the tongue root (i.e. a backed tongue body position) indicating the inter-relationship between some vocal variables. As tongue backing was not seen to any great extent in the TD children and young people in this study, then it makes sense that no evidence of pharyngeal constriction was found in these typical speakers also. Again the habitual setting of the DS group has been shown to be similar to that of Edinburgh and Glasgow speakers with lower social index scores.

5.4.6 Velopharyngeal settings

5.4.6.1 Audible nasal escape

Audible nasal escape is one of the few settings which is only judged as present in the severe range (i.e. it is not possible to have a moderate presentation of this setting).

A significant difference was found between the overall DS and TD groups, with the DS speakers having a higher degree of audible nasal escape; a finding which supports the given hypothesis. This setting was not associated with any of the TD speakers, confirming that it is not a feature associated with the typical Edinburgh population. In the analysis of the speakers from study 1 a rather anomalous result was observed where no significant difference was found between the eight DS and eight TD speakers, however as three of the eight DS speakers had marked nasal escape whilst none of the TD speakers presented with this feature then it is likely that the smaller sample size was a factor.

Interestingly, a significant difference was found between the DS males and females, whereby only one of the nine overall females with DS had any evidence of audible nasal escape in comparison to five of the thirteen males. It is not possible to say for certain whether this is a feature which is truly representative of male speakers with DS or rather a finding which highlights the variability of features within individual speakers. However, as hypotonia, which is known to contribute to velopharyngeal incompetence and therefore audible nasal escape (Biavati, Sie, Wiet & Rocha-Worley, 2009) affects both males and females with DS then it seems most likely that the finding of more nasal escape in males with DS is a consequence of individual differences rather than a genuine gender-specific trait. As previously indicated this setting was not evident in either of the TD gender groups.

5.4.6.2 Denasality – nasality

In line with the a priori hypothesis both the overall DS group and the DS speakers from study 1 were shown to have significantly higher levels of nasality then their TD peers. For the overall group the degree of nasality was on the boundary of severe presentation (median rating of 3) and the IQR extended further in this direction, whilst in the TD group the nasality rating was lower (median of 2) with an IQR which extended down towards neutral from the median setting. Similar results were found in the analysis of the speakers from study 1, but in this instance the DS results had slightly more extreme nasality settings than seen in the overall DS group.

Between the females and males with DS it was found that males had statistically significantly higher levels of nasality than the females (the females being on the boundary of extreme nasality with a median of 3 and the males being judged to sound even more extreme with a median rating of 4). Both genders with DS had higher levels of nasality than either the male or female TD groups. A similar pattern was found in the TD gender groups (although to a lesser degree, both being within the moderate range) where the TD males had higher levels of nasality than their female counterparts. Thus it appears that although the degree of nasality is greater across the board in DS, this setting adheres to the same gender profile as found in TD Edinburgh speakers.

Although nasality is higher in speakers with DS these findings suggest that some degree of nasality is also typical of Edinburgh speakers, but more marked in males than females; a finding also observed in Glasgow speakers (Stuart-Smith, 1999). The gender differences observed in this study and in Glasgow speakers are at odds with the claim that “women's voices are normally more nasal than men's” (Bloom, Zajac & Titus, 1999, p. 278) suggesting that nasality needs to be considered according to norms for the target population.

Esling (1978) found nasality to be associated with Edinburgh speakers with higher social status and Stuart-Smith echoes this sentiment stating that working-class speakers in Glasgow often use increased nasality when mimicking those from the middle-classes. In contrast highly nasal voices have been found to be judged as being of lower social status in terms of perceived occupation, education, ambition and intelligence and to sound less friendly, sociable, trustworthy, sympathetic and likeable than those with less strong nasality (Pittam, 1987, 1989) and to be associated with less positive gender stereotypes (Bloom, Zajac & Titus, 1999) making it difficult to evaluate whether the increased nasality associated with DS is in fact a positive social feature.

Increased nasality is frequently discussed in relation to inadequate velopharyngeal closure, and as such increased levels of nasality would be expected in people with DS where the velum is likely to be negatively affected by the presence of hypotonia. However, it is known that the velum does in fact sit in a slightly open position during the course of typical speech without producing audible nasality, leading Laver (1980) to contend that nasality is an auditory concept which is concerned with more than just the position of the velum, rather it incorporates the effect of velic activity on the pharynx and its connected structures, such as the tongue and larynx. Laver also notes that nasality may not be only a consequence of resonance within the nasal cavity (although he does concede that it is the most significant chamber in the production of nasal resonance) as increased muscular tension within secondary resonating sites such as the pharyngeal and laryngeal cavities have been suggested to produce what is termed 'cul-de-sac resonance'. Significantly, where muscle tension is relaxed there is a tendency for the 'cul-de-sac resonance to dissipate (West, Ansberry & Carr, 1957). The muscular tension settings of people with DS will be examined in a later section of the discussion.

5.4.7 Larynx height settings

5.4.7.1 Lowered – raised larynx

As people with DS have been perceived as having lower than average pitch it was hypothesised that the larynx position in these speakers would be lower than typical, however no significant difference was found between the DS and TD speakers within the overall analysis or within the analysis of the speakers from study 1.

When split by gender the only significant difference to be found was between the TD females and the TD males, where although the median values were the same (neutral) the female IQR extended up to a rating of 1 (slight raised larynx) and the male IQR extended down to -1 (slight lowering); as a lower larynx is generally associated with male speakers the direction of this result was to be expected. The lack of a gender difference between the DS females and DS males may be a product of the vocal tract configuration which is associated with this syndrome and/or a lack of conscious manipulation of voice by the females with DS to conform to the expected gender pattern. Functionally, this lack of difference might account in part for the difficulties experienced by listeners in judging the gender and age of the speakers with DS in study 1.

Raised larynx has been found to be a distinctive feature of all Edinburgh speakers, however, a significantly higher incidence of this setting was observed in adults and young male speakers of lower social status, twice as many being seen in boys of the lowest social group compared to boys of he highest social group (Esling, 1978).

Pitch and F0 are intrinsically linked to laryngeal position and function; how these findings relate to the ratings of pitch and F0 findings will be considered later in the pitch section of the discussion.

5.4.8 Muscular tension settings

5.4.8.1 Lax – tense vocal tract

As hypotonia is a predominant feature of DS it was hypothesised that laxness of the vocal tract would be a clear differentiating feature between people with DS and their TD peers. In fact, although the overall DS group were found to have significantly laxer vocal tract ratings, the difference was actually quite slight (both groups having a neutral median of zero, but the TD group also having an IQR of zero whilst the DS group's IQR extended down to -1, just into the lax vocal tract range). A more lax muscular setting is associated with increased absorption of acoustic energy, which would support the higher SPT values observed in the DS speakers in study 2. The vocal tract values were identical in the analysis of the speakers from study 1 but as no significant difference was found this may be as a consequence of reduced sample size. Given the previous findings which have appeared to confirm the reduced muscular function of the vocal articulators of speakers with DS as a consequence of hypotonia, it is unusual that the tension features do not conform to this profile more strongly.

When analysed for gender differences it was found that the male speakers with DS (neutral median with an IQR extending down into the lax end of the scale) echoed exactly the pattern of the overall DS group. In contrast, the DS females and the TD females and males all had neutral median and IQR values, indicating that vocal tract laxness or tension is not an habitual feature of any of these groups. As suggested before, hypotonia is likely to affect people with DS of both genders equally, thus the significant difference in the male DS speakers may be due to individual differences in speakers within that group rather than indicative of an habitual male setting.

5.4.8.2 <u>Lax – tense larynx</u>

Again, due to hypotonia, it was hypothesised that the speakers with DS would have a laxer laryngeal setting than their TD peers, but as in the above vocal tract tension settings results were not as anticipated. A significant difference was found between the overall DS and TD groups but it was the DS speakers who were found to have increased laryngeal tension. This result echoes that of Moran and Gilbert (1982) where more than 70% of judges perceived laryngeal tension in nine of sixteen adults with DS. Laryngeal tension would be consistent with the more 'forced voice' described by Moura et al. (2008) in young Portuguese-speaking children with DS. It is also consistent with the proposition that people with DS require considerably more effort to initiate movements of the musculature of the larynx to initiate phonation (Pryce, 1994); that effort potentially resulting in increased laryngeal tension. The TD speakers from study 1 had a much lower median value than the overall DS group (zero compared to 1.5) indicating that those speakers with increased laryngeal tension with DS were not represented in study 1. In both cases the median (IQR) values for the TD groups were 1 (1), indicating a slight degree of tense larynx.

Between genders, although the females with DS had higher levels of laryngeal tension than the males with DS (median 2 versus median 0.5) this difference was not statistically significant. The only significant difference was found between the DS and TD females, where again the females with DS had higher levels of tension than their TD counterparts; the TD females having identical ratings to the TD males (median 1, IQR 1) indicating that only slight tension is a feature of typical Edinburgh speakers.

Esling (1978) found that in Edinburgh adults tense phonation was most prevalent amongst those with the lowest social index scores whilst the opposite pattern of lax phonation was associated with higher social standing. Lax vocal tract tension in conjunction with increased laryngeal tension has been shown to be associated with the 'stereotypical male Glasgow voice', which according to Stuart-Smith (1999) will facilitate harsh voice production. Phonation type will be evaluated in the following section of the discussion.

5.4.9 Phonation features

5.4.9.1 Neutral – non-neutral voice

All speakers (DS and TD) were judged as having non-neutral voice. The terms 'neutral' and 'non-neutral' should not be confused with 'normal' or 'non-normal', as the VPAS takes into account the significant variation seen in speakers from different language or speech communities (Mackenzie Beck, 2005b). Neutral phonation is defined as the result of regular and efficient vocal fold vibration (also termed 'modal voice') thus 'non-neutral' phonation should be considered "as representing voices which deviate from neutral by an alteration in the basic type of vibration (the voicing type), by the addition of audible fricative airflow through the glottis (laryngeal frication) or by irregularity/perturbation of the basic voicing type (laryngeal irregularity)" (Mackenzie Beck, 2005b, p. 22).

According to Mackenzie Beck (2005b) neutral voice is relatively rare, therefore these findings are unsurprising and in line with the given hypothesis.

5.4.9.2 Falsetto

None of the DS or TD speakers in this study were judged to use falsetto, demonstrating that this voicing type is not characteristic of either DS or typical Edinburgh speakers.

5.4.9.3 Creak

Creak or 'glottal fry' is the presence of "discrete audible pulses of voice" (Mackenzie Beck, 2005b, p. 23). This label is used where there is no voice component (i.e. only creak). In this study no speakers were judged to fit this criterion.

5.4.9.4 Creaky voice

The term creaky voice is used where some creakiness is observed alongside other phonatory features.

A significant difference in creakiness was found between the overall DS and TD groups. However, it is notable that the median rating of both groups was neutral (rated 0) and that the significance came from only a slight difference in the IQR of the groups (DS IQR, 0, compared to the TD IQR, 1). These values indicate that creakiness is not typical of speakers with DS from the Edinburgh area, but that it is present in some TD speakers from the same area. In the analysis of the speakers from study 1 no significant difference was found, but on closer observation this was due to the fact that all of the three speakers with DS (from the overall 22) who were judged to use creaky voice were present in the DS group in study 1.

Analysis by gender identified some interesting findings. All groups had a neutral median rating, and all with the exception of the TD males had an IQR which was also scored at zero. A wider IQR of ‘2’ was found for the TD males and this was sufficient to create a significant difference between the TD males and females, suggesting that creakiness is more characteristic of TD males than females in the Edinburgh area. This finding is echoed in Glasgow speakers (Stuart-Smith, 1999) and in speakers of RP and an accent group termed ‘Modified Northern English’ (Henton & Bladon, 1988). The general lack of creakiness in both males and females with DS is therefore more critical for the males, as low levels of creaky voice would be anticipated in females.

A clear social division was observed in Edinburgh adults and young boys by Esling (1978) for creakiness, where speakers with higher social status were found to use this feature significantly more than those with lower social index scores, indeed creakiness was not present in any of the boys from the lower social group. Interestingly Stuart-Smith (1999) found that creakiness did not correlate with social class in Glasgow speakers, highlighting the variety in voice production across even close geographical areas and the subtleties associated with those differences. If there is a social class division regarding the use of creakiness in Edinburgh and if people with DS do find creak difficult to produce then there may be strong implications for those children and young people with DS who belong to families who habitually use creakiness as an indicator of their social group.

Early evaluation of vocal fold function identified that the production of creaky voice involves strong adductive tension and medial compression alongside weaker longitudinal tension, with some involvement of the false (ventricular) folds also (Hollien, Moore, Wendahl & Michel, 1966). This is a complex configuration which requires considerable articulatory effort and fine motor control, thus it is possible that creaky voice is particularly difficult for people with DS due to the reduced muscle tone and control, and the increased muscular effort required to initiate phonation in DS.

When accompanied by a drop in pitch and amplitude, creakiness also serves a communicative function of voice, being used to signal the end of an utterance and indicating to the listener that the speaker has finished his or her communicative turn (Cutler & Pearson, 1986). The lack of creaky voice in DS may be indicative of the physical difficulties inherent in producing this feature or alternatively may be linked to lack of awareness of the prosodic function of creak, or perhaps more likely, a mixture of the two.

5.4.9.5 Whisper

As with creak, whisper is the label used to describe phonation where there is whisper with no voice component. No speakers were judged to have this phonatory feature.

5.4.9.6 Whispery voice

Whispery phonation was found to be significantly greater in both the overall DS group and the speakers with DS from study 1 in comparison to their TD peers. These findings are in accord with the a priori hypothesis of increased whisperiness in speakers with DS due to the likelihood of inefficient vocal fold closure allowing air to escape during phonation. Whispery values were high for both groups (rated at 3 which is on the border of a severe presentation) which is in line with earlier research showing that breathiness was the most commonly perceived vocal characteristic in adults with DS (Moran & Gilbert, 1982).

Increased whispered or breathy phonation is associated with higher SPT values (Baken & Orlikoff, 2000; Klatt & Klatt, 1990) thus the findings of significantly greater whispery phonation ratings for the DS groups supports the significantly higher SPT values observed in the overall DS group in the acoustic analysis. Although a significant difference in SPT was not also found between the DS and TD speakers from study 1 the direction of the results (increased SPT values in the DS speakers) suggests that this was a likely consequence of small sample size. As lower HNR (increased noise) has also been shown to correlate with increased whisper or breathy voice (Mathieson, 2001) the non-significant HNR results between the overall DS and TD groups and the DS and TD speakers from study 1 are somewhat surprising. Despite the finding of a significant difference in whispery voice, as ratings were relatively high in both the DS and TD groups it is evident that whispered phonation is a general phonatory characteristic of DS and TD speakers from the Edinburgh area. For these TD speakers this setting reflects accent-based modification of voice whilst for the DS speakers this quality is a likely consequence of their atypical vocal tract structure and function. Thus, it could be seen as a positive for the speakers with DS from this particular locality that their phonation type is consistent with that of their peers, even if it is to a greater extent, as in populations where whisperiness is not habitual it could be seen as a further differentiating factor.

When split by gender, although the median values were the same (both rated at 2) the higher IQR of the TD females in relation to the TD males resulted in a significant difference being found between the two. This might point to possible social differences in the use of whispered voice between genders. Breathy or whispered voice is much more common in female speakers than in males (Ishi, Ishiguro & Hagita, 2008) and in particular at higher pitches and in younger women (Abberton, 2008) although there will be considerable variation between individual speakers and between different accent groups. Whispered or breathy phonation type has long been believed to be associated with “more self-effacing, submissive, meek personalities”

(Laver, 1968, p. 50) which is in keeping with the more socially-accommodating female traits described by Holmes (2001) earlier in the discussion.

This finding was not replicated between the females and males with DS, both having identical median and IQR values (median, 3 IQR, 1) seeming to confirm the equal effects of vocal tract differences on phonation between genders. No significant difference was found between the TD and DS females, although the median was slightly higher for the females with DS, whilst in contrast the males with DS had significantly greater whisperiness compared to the TD males. These finding suggest that males with DS have a higher degree of whisperiness than is typical for males in Edinburgh but that females with DS who produce equivalent levels of whisperiness find themselves in line with expected female margins.

Increased noise (lower HNR level) tends to be seen more in females than males however the opposite pattern of significantly more noise in male speakers was found in both the DS and TD groups in this study. Increased noise suggests more whispered/breathy phonation but in contrast the VPAS results indicated significantly greater whisperiness in the TD females in comparison to their male counterparts, whilst the males with DS were found to have exactly the same median and IQR ratings as the females with DS; this would suggest that whispered phonation on its own does not account for differences in HNR. Similarly, it had been expected that due to generally smaller vocal tract anatomy female speakers would have higher SPT values than males (Ho, 2001) however no significant differences were identified in this acoustic parameter between the genders in either the DS or TD groups. This finding backs up the absence of identified differences in perceived whisperiness between the DS males and females, but does not support the significant difference found between the TD gender groups. As females are typically associated with higher breathiness/whisperiness, and all the gender groups have relatively high levels of whispered phonation ratings, it seems most likely that it is the Edinburgh males who have high whisperiness for their gender (rather than the females having particularly low levels).

In terms of relevance to local voice production, whispery phonation was found to be more associated with groups of lower social status than those with higher social index scores in Edinburgh adults and boys (Esling, 1978) and to be an indicator of working-class speakers in Glasgow of both genders, although (in common with this study) it featured significantly more in female voices than in male voices (Stuart-Smith, 1999).

5.4.9.7 Harsh voice

Although the median values were identical (scored at 1) the slightly wider IQR of the overall DS group resulted in a significantly higher rating for harsh voice in comparison to the overall TD group; which again supports the a priori hypothesis. No significant difference was found for the subset of DS speakers from study 1, but as in previous analyses small sample size is likely to have compromised results to some extent. The low median rating for harshness is rather surprising given the plethora of descriptive labels which represent this quality used in reference to people with DS within the published literature, and serves to indicate that the whisperiness described previously seems to be a more salient perceptual phonation feature than harshness in these speakers with DS from the Edinburgh area, although harshness does play a part in voice of both these DS and TD groups.

Despite the significant difference between the overall groups the equal median ratings give credence to the lack of identified differences in the acoustic parameters of jitter, shimmer and HNR, all of which have been found to be associated with harsh phonation type. There was a finding of increased jitter in the DS speakers from study 1 but again this has to be considered against the small numbers and also the slight degree of statistical significance found ($p = 0.005$).

Between genders the only analysis to evidence a significant difference was between the DS and TD females, both having an identical median of 1 but the DS group having a slightly wider IQR indicating more variation in the presence of harsh phonation within the DS females. Generally harsh voice is less associated with females than males but in this study that proved not to be the case with no significant difference being found between the TD males and females, and in fact the females had a higher median value for harshness than the males. However, it should be taken into consideration that the TD males did produce significantly more creaky voice than their female TD counterparts, suggesting phonatory settings reflecting ‘whisperness+harsh voice’ may be more associated with TD females compared to more ‘whisperiness+creaky voice’ in TD males. As was expected from the similar vocal tract structure and function, no significant difference was found between the harshness ratings of the males and females with DS.

No differences were found in the jitter and shimmer values between the DS males and females which corresponds with the absence of significant differences between the two in harshness ratings. Between the TD gender groups only one of the shimmer variables differed (shimmer apq3, being higher in males) and no difference was found in jitter values, again agreeing with the non-significant VPAS harshness ratings. As harsh voice is also associated with increased noise (lower HNR) it might have been the case that the significantly lower HNR values seen in both the TD and DS males may have related to this phonation setting. However, as harsh phonation did not differ significantly between the male and female DS or TD speakers, and in fact the female DS speakers had a wider IQR for this setting and the female TD speakers had a higher median value compared to the male speakers this seems not to be the case.

Harsh phonation type tends to be perceived rather negatively, having been found to correlate with more dominant and authoritative characteristics (Laver, 1968) and in both genders to indicate impatience and aggressiveness (Tanner, 2007). This setting is also associated with both adults and young boys of lower social status in Edinburgh (Esling, 1978). As in whispered phonation, the presence of this shared phonatory feature between the DS and TD speakers from this Edinburgh study might be seen as positive in terms of people with DS being perceived as being less different than might be the case in areas where whisper and harshness are not generally observed in the TD population.

5.4.9.8 Tremor

This feature was found not to be typical of either the DS or TD speakers, all groups having neutral median and IQR values.

5.4.10 Pitch

5.4.10.1 Low – high mean

A significant difference was found between the overall DS and TD groups with the DS group having significantly lower mean pitch ratings than their peers (DS median 1, TD median 0). As lip rounding and protrusion is associated with reduced pitch (Rammage, Morrison, Nichol, 2001) this finding is in line with the significantly more rounded/protruded lip posture ratings for the DS group. A much wider IQR was also noted for the overall DS group indicating the wide variance in pitch levels between the individual speakers who made up this group and again confirming the heterogeneity of people with this syndrome. This result was not replicated in the DS speakers from study 1, however as the median pitch rating (-2) was actually lower in this DS subgroup than the overall DS group and the IQR the same, the lack of a statistical difference would seem to come from the wider IQR seen in the TD speakers in this analysis compared to the overall TD group (both having a neutral median).

Interestingly, despite the significant perception of lower pitch in DS there were no significant differences in the level of F0 (the acoustic correlate of what listeners perceive as pitch) or larynx height ratings between either the DS and TD overall groups or those speakers from study 1. The non-significant F0 results are in common with those of early research into F0 in children with DS which identified that despite the perception of low pitch F0 level did not differ significantly from that of TD children (Michel & Carney, 1964; Hollien & Copeland, 1965; Montague, Brown & Hollien, 1974; Montague, Hollien, Hollien & Wold, 1978; Pentz & Gilbert, 1983). It is possible that, as suggested by Hollien and Copeland (1965) the perception of low pitch is a consequence of the effects of interaction between various other vocal characteristics which combine in DS to give the illusion of reduced pitch in the presence of typical F0 values. More specifically, the wider IQR observed in the DS group might offer support to the proposition of Montague, Hollien, Hollien and Wold (1978) that it may be the greater variance in F0 values which is responsible for the perceptual rating of low pitch in DS.

Between genders some notable differences were identified. Despite both the TD females and males having a neutral median (indicative of not low and not high pitch) a significant difference was found as a result of the IQR of the females group extending up into the higher pitch scale whilst the male IQR extended down into the lower pitch scale. This finding is unsurprising given that higher pitch is associated with female speakers and lower pitch with males and is in agreement with the finding of higher F0 in the TD females in the acoustic study and the significantly higher larynx height ratings of the TD females compared to the TD males.

The pitch gender differences did not extend to the speakers with DS, where no significant difference was found between the males and females, but surprisingly this is at odds with the finding of significantly higher F0 in the females with DS (although the females with DS did have a higher median perceptual pitch rating than their male counterparts which is likely to have contributed to this disparity). Interestingly, the perceptual pitch ratings do correspond with the lack of significant difference in larynx height ratings between the DS males and females.

When the male and female DS groups were compared against their respective TD gender groups significant differences were found, with both the male and female DS speakers having significantly lower pitch ratings than their peers, illustrating that the perception of low pitch in DS is not gender specific. As in the overall DS group analysis, these significant perceptual pitch differences between the TD and DS females and the TD and DS males were not in keeping with the non-significant F0 results observed in the instrumental analysis or the non-significant differences in larynx height ratings. Within both gender groups the speakers with DS were found to have considerably wider IQR values, again demonstrating significant variability between speakers.

Interestingly, low pitch has been found to be associated with dominance whilst high pitch signals submissiveness (Frick, 1985). Given the tendency for people with ID generally, and DS specifically, to be regarded as somewhat passive and vulnerable, this interpretation of low pitch would appear to be very much at odds with how these individuals are viewed within society.

5.4.10.2 Minimised – extensive range

The range of pitch movement between high and low was found only to differ significantly between the TD females and the TD males, and even then the median values were both neutral and the difference lay in a slightly wider (and lower) IQR rating for the TD males (female IQR, 0 and male IQR, -1). Although there was not a similar finding between the DS males and females, the males with DS did have a more minimised median rating indicating that speakers with DS from Edinburgh do echo the pitch range gender pattern observed in TD speakers, if not to the same extent.

The solitary significant finding of gender differences taken in combination with no overall difference between the DS and TD speakers suggests that range of pitch is not particularly problematic in DS in comparison to typical speakers from the Edinburgh area; a finding which is rather surprising given the reputation for reduced pitch movement in DS. Thus it could be the case that this finding reflects a slightly reduced range of pitch in the TD Edinburgh speakers which brings them closer to the values of speakers with DS than controls in studies from other parts of the UK.

5.4.10.3 Low – high variability

The perceptual results for pitch variance are the same as for minimised – extensive pitch range, with the only statistically significant result being between the TD males and females. Again both median values are equal (neutral) but the male IQR is slightly lower (-1, indicative of lower variability).

5.4.11 Loudness

5.4.11.1 Low – high mean

Between the overall DS and TD groups, despite the identical neutral median rating for both groups, the wider IQR of the DS group extending into the low volume range (DS, 2 compared to TD, 0) has resulted in a statistically significant difference. This would indicate that generally volume is at a level comparable to TD peers for the majority of speakers with DS, but that for some this feature is reduced. The overall DS group had significantly steeper SPT values than the TD group (which is associated with a more rapid tailing off of acoustic energy and reduced volume) but not significantly different shimmer values. According to Brockmann, Storck, Carding and Drinnan (2008) shimmer has been found to increase with decreasing vocal loudness. However, in this study it was the case that despite the absence of a significant difference, the DS group actually had lower shimmer values then their TD peers, which would be expected to indicate higher loudness levels. It was notable also that the DS group had significantly reduced respiratory support ratings, as poor breath support is a strong factor in reduced volume and length of utterances (Mackenzie & Laver, 2004).

No significant difference was found between the speakers from study 1; however in this analysis the IQR for the TD speakers was wider than in the overall group as one speaker in this sub-group had relatively low mean volume in comparison to the rest of his group. The SPT and shimmer values in this group did not match the pattern of the overall groups (neither being significantly different from the TD values and one of the shimmer values being higher in the DS group) however there was agreement with the degree of respiratory support, the speakers with DS having reduced capacity ratings.

Within genders there were no differences between the mean volume ratings of the TD males and females, the median and IQR for both being neutral. This is in keeping with the lack of differences in SPT and respiratory support. At odds to this is the finding that the females actually had significantly lower values in one of the shimmer measures, which might have been expected to point to higher loudness levels; this suggests that SPT and ratings of respiratory support are more reliable measures of loudness production than shimmer.

Similarly for the DS speakers there were no identified gender differences which corresponded with the absence of differences in SPT, shimmer or respiration values. This finding indicates that the overall differences in loudness seen in the DS speakers are not gender specific.

When males and females within the two groups were compared directly, an interesting result was seen. A significant difference was found between the DS males and TD males with the DS speakers having the same neutral median but a much wider IQR (a rating of 2 compared to a neutral value for the TD speakers) indicating some speakers had reduced loudness levels. Again, the SPT values of the DS males were significantly higher than those of the TD males, the respiratory support significantly lower and no difference found in shimmer. Exactly the same values were found between the DS females and TD females however this did not result in a significant difference in loudness, which may be due to the smaller sample size of this group. As before no significant difference was found in SPT and respiratory support was significantly reduced. The shimmer values are somewhat ambiguous as only one was significant, although both were higher for the DS females.

It has been reported that loud voices are associated with anger and more dominant personalities whilst quieter voices can signal sadness (Williams, 1997). Thus it is possible that the less loud voices of people with DS may contribute to the stereotypical perception of being less dominant and more passive.

5.4.11.2 Minimised – extensive range

The only significant difference in loudness range was found between the DS males and the TD males, and was as a consequence of the larger IQR of the DS males (both medians were neutral). In fact all the median values for all the groups were rated as neutral thus reduced range of volume does not appear to be a parameter which is common in either the TD or DS speakers in this study.

5.4.11.3 Low – high variability

Amplitude variability did not differ significantly between the overall DS and TD groups or between the DS and TD speakers from study 1. Some differences were observed between gender groups, but notably not between the TD males and females (median 0 and IQR 0 in both cases). Significant differences did exist between the males with DS and their TD male counterparts and between the females with DS and the TD females (the DS groups demonstrating less variability in both cases). Further comparison of the males and females with DS showed that the males with DS were rated as having lower variability than the females. However, as all groups had a neutral median it appears that low or high variability of loudness is not generally characteristic in either gender of TD or DS Edinburgh speakers.

5.4.12 Temporal organization

5.4.12.1 Continuity

Mackenzie Beck (2005b, p. 18) describes continuity as "…the incidence of pauses within a speech sample. Marking a speaker as having an interrupted setting indicates the presence of inappropriate silent pauses between words or syllables".

A significant difference in continuity was found between the overall DS and TD groups and the DS and TD speakers from study 1. In both cases the degree of continuity was significantly more interrupted in the speakers with DS. A wide IQR (3) was noted for the overall DS group in particular, indicating that there is considerable variability between individual speakers.

There was no difference in the continuity ratings of the TD females and the TD males (both rated as neutral) or between the DS females and the DS males. Similarly, when females were compared against females and males with males it was apparent that both the females and males with DS had significantly higher interrupted continuity ratings than their TD gender counterparts, indicating that differences are not gender-specific.

These findings highlight that speakers with DS in this study have significant difficulties producing words and utterances in a fluid manner. Aside from conventional dysfluency (which was present in only a few speakers with DS) a range of factors, including the presence of ID, reduced speed and accuracy of the articulators as a consequence of hypotonia, apraxia of speech or expressive language difficulties may all contribute to this interrupted vocal profile.

5.4.12.2 Rate

Although the median value for both the overall DS and TD group was neutral a significant difference in the rate of speech was found between the overall DS and TD groups and between the DS and TD speakers from study 1 (the DS groups both having a wider, lower, IQR).

As in the continuity measures there were no significant differences between the TD males and females or between the DS males and females (although the females with DS had a lower median rating). The males with DS were found to have significantly slower rate than the TD males and similarly the females with DS were significantly slower than their TD female counterparts. Although there are clear statistical differences between the DS and TD speakers (both overall and within the subgroups) the maximum level of reduced rate in the DS speakers is only -1 (DS females) indicating only slight slowing of speech rate.

As with continuity, processing and production difficulties as a consequence of ID, apraxia, hypotonia and reduced language skills are all likely to impact on the speed of production. Slower rate tends to be associated with sadness, and where overly slow can disrupt the natural rhythm of speech and potentially cause the listener to lose interest (Williams, 1997).

5.4.13 Other features

5.4.13.1 Respiratory support

Significant differences were identified in the respiratory support of the overall DS and TD speakers and between the DS and TD speakers from study 1. In both instances the DS speakers were judged to have reduced respiratory capacity. "The generation of sound in the larynx depends on the coordination of the laryngeal and respiratory systems, with appropriate levels of air pressure, air volume and airflow being fundamental to phonation and articulation (Mathieson, 2001, p. 51) thus voice production in DS can be considered at serious risk of being compromised as a consequence of poor breath support. A series of inspiratory and expiratory muscles coordinate to enable effective lung function, and thus in DS it may be the case that hypotonia impairs muscle function to some degree.

No significant differences were found between the TD males and females or the DS males and females, indicating that low respiratory function is not typical of Edinburgh speakers and that the reduced function observed in the overall DS speakers is not gender specific. As would be expected from the pattern of these and the overall results, significant differences were found between the males with DS and the TD males and between the females with DS and the TD females, the DS speakers having significantly reduced respiratory support ratings compared to their TD peers.

5.4.13.2 Diplophonia

This feature was not found in any of the DS or TD speakers which clearly identifies diplophonia as an atypical vocal feature for both groups.

5.4.14 Summary

Marked differences were observed in the perceptual analysis of voice between the DS and TD speakers in this study. Some ratings corresponded with the acoustic profiles identified in study 2 whilst others were not backed-up by expected instrumental findings.

The speakers with DS were found to have significantly more protruded lip pattern than their TD peers and significantly less range of lip movement. No differences were found between males and females with DS suggesting this is a consequence of the shared characteristics of their anatomy. TD females were observed to have a more spread lip pattern than any of the other speakers which may reflect a more ‘smiley’, socially accommodating persona; hence females with DS appear to be particularly at odds with the typical gender lip setting.

Jaw position was significantly more open and protruded in the DS speakers compared to their TD peers and the range of movement minimised. Again the absence of differences between males and females with DS suggests that this feature is organic in origin.

Significantly more advanced tongue tip, fronted and raised tongue body and minimised range of tongue movements were all associated with speakers with DS and are likely to be related to the relative macroglossia which is a feature of this syndrome. Females with DS, like their TD female counterparts, had a more fronted tongue body setting than the males, whilst the males with DS demonstrated a more minimised range of tongue movement.

A significantly higher degree of pharyngeal constriction was found in both the males and females with DS, which again is a likely consequence of structural differences.

Although some nasality appeared to be the norm in the TD speakers a significantly higher degree was observed in the DS speakers, falling in the extreme range of this setting for the males with DS and borderline extreme for the females with DS. Significantly more audible nasal escape was also associated with the DS groups. Both findings indicate that velopharyngeal function is compromised in this syndrome, most likely as muscular strength and tone is diminished.

The lack of difference in the perceptual rating of larynx height was surprising, given the perception of low pitch. The TD females had significantly higher larynx position ratings than their male counterparts (a finding which is expected and relates to the higher pitch and higher F0 levels usually found in females) but this was not replicated in the females with DS. Whether it is the case that females with DS are constrained by their vocal tract structure and function or that they do not adjust their larynx height to fit the female norms of this geographical area is unclear; a mix of the two seems likely.

The wider IQR of the DS group compared to their TD peers for vocal tract tension created a significant difference despite identical neutral median values; again this finding was surprising given the hypotonia associated with DS which would have been expected to result in greater differences in the medians between groups. Moreover, a laxer larynx had been hypothesised but in fact the DS speakers were found to have significantly increased laryngeal tension compared to their TD peers. This suggests increased vocal effort and is consistent with the higher degree of energy required to initiate phonation within speakers with DS (Pryce, 1994).

In terms of phonation, significant differences were identified in creaky voice, whispery voice and harsh voice, the former being significantly higher in the TD speakers and the latter two significantly higher in the DS speakers. Creaky voice, although significantly greater in the TD group did not appear to be particularly common in either group (DS & TD both rated as neutral), and as TD males had wider creaky ratings than their female counterparts, it appears that this setting is associated with Edinburgh males more than females. It is possible that reduced muscular control is a factor in the low creaky ratings of the DS group. Whispery voice and harsh voice were seen to be features of both the DS and TD groups, although a greater degree was observed in the DS speakers. In contrast to the TD males and females, no significant differences were found between the males and females with DS for any of the phonation settings indicating that their organic structure constrains voice production across both genders equally.

Despite the absence of differences between the overall DS and TD groups in F0 the DS speakers were perceived as having significantly lower mean pitch; a finding in common with earlier voice research in DS by Michel and Carney (1964), Hollien and Copeland (1965), Montague, Brown and Hollien (1974), Montague, Hollien, Hollien and Wold (1978) and Pentz and Gilbert (1983). No differences were found in the variability of pitch or the pitch range. All three pitch measures differed significantly between the TD males and females but tellingly no such differences were observed between the genders in the DS speakers, again suggesting structural constraints or possibly a failure to adapt to typical pitch gender norms.

A significant difference was found in mean loudness between the overall DS and TD speakers, with the DS speakers having reduced loudness ratings, but no differences found in variability or range of loudness. Between genders no differences were found in any of the measures between the TD males and females and only variability differed between the DS genders (males having a slightly wider IQR). As the median values for all the groups were neutral it appears that extreme or minimised loudness features are not typical of either DS or TD speakers from the Edinburgh area.

Both continuity and rate of speech were found to be significantly different, the DS group having more interrupted speech and a slower rate than the TD group. Between genders both the TD males and females had neutral ratings indicating that neither setting is typical of Edinburgh speakers, whilst again there was an absence of difference between the DS genders.

Finally, respiratory support was found to be significantly reduced in the DS speakers, which is likely to be related to lower muscle function as a consequence of hypotonia. Diplophonia was not found to be a feature of either the DS or TD speakers.

Many of the habitual vocal features associated with the DS groups in this VPAS study, such as protruded lip setting, open and protruded jaw, constricted pharynx and whispery and harsh phonation have been associated with Edinburgh speakers who score more lowly on social index scales (Esling, 1978) and as such might contribute towards a more prejudiced, negative perception of speakers with DS.

5.5 CORRELATIONS BETWEEN STUDIES 1, 2 AND 3

In this section correlations for the combined data of the sixteen DS and TD speakers from study 1 are examined in relation to each of the questionnaire parameters. Due to the high number of variables only strong correlations (+/-0.7) are discussed.

5.5.1 Calm – angry ratings

The negative correlation for lip spreading – rounding/protrusion indicates that more calm ratings are associated with more spread lip setting; as spread lip setting is indicative of a smiling configuration then this result is unsurprising. VPAS ratings of nasality also indicated a negative correlation, meaning that more calm ratings correlated with more denasality, which in turn means that higher angry ratings are associated with more nasal resonance. This finding is in keeping with the negative listener perceptions of nasality identified by Pittam (1987, 1989) and Bloom, Zajac and Titus (1999). As lip protrusion and higher nasality were found to be associated with the DS speakers then these findings indicate that children with DS are at a higher risk of being judged as less calm and more angry than their TD peers.

5.5.2 Confident – shy ratings

High ratings of confidence were found to correlate with high ratings of friendliness, happiness and intelligence and in turn with more positive responses to the 'spend time with' parameter of study 1; children with DS were rated less positively than their TD peers in these parameters which is reflected in the particularly low confidence ratings they received. In study 2 high confidence correlated with reduced SPT measures; as lower SPT indicates less tailing-off of acoustic energy and is associated with more forceful or strident voice then it is unsurprising that this is the case given that a stronger voice is likely to be characteristic of higher confidence. This is confirmed by the finding in study 3 that decreased whisperiness (associated with more efficient vocal fold closure) correlates with high confidence ratings. High pitch ratings also correlated with high confidence, and this was in keeping with the correlation between raised larynx and high confidence. Also in study 3, spread lip pattern correlated with higher confidence, as did extensive lip and tongue range and close jaw, whilst negative correlations were found for protruded jaw and interrupted continuity; both of which are typical of the DS speakers.

5.5.3 Friendly – unfriendly ratings

High friendly ratings correlated positively with high confidence, happiness, intelligence and more positive ‘spend time with’ ratings. This indicates that speakers who are deemed to sound shy, sad and unintelligent are perceived as sounding less friendly; this is of particular concern for individuals with DS as in study 1 speakers with this syndrome were judged as sounding significantly less confident, happy and intelligent than their TD peers.

Increased F0 values in study 2 also correlate with higher friendly ratings, which is consistent with the correlation between higher perceived pitch level and friendliness in study 3. As increased lip protrusion lengthens the vocal tract and thus reduces pitch, it is notable that increased protrusion correlates negatively with high friendliness (confirming the relationship between pitch and friendliness ratings). Visually this finding is unsurprising as the opposite setting (lip spreading) is consistent with smiling, which is generally considered a friendly gesture. Also in study 3 increased lip and tongue range of movement were found to correlate with high friendly values; both of which were rated more negatively in the DS group.

5.5.4 Happy – sad ratings

Higher happiness ratings were found to correlate positively with higher confidence, friendliness and intelligence ratings, and also with higher judgements of speakers sounding female in the gender parameter. As TD females have been shown to have a more spread lip pattern (high spreading also correlated with high happiness ratings) then this finding suggests that the spread lip pattern is perceived by listeners as sounding happier. As in the confidence ratings both the SPT measures in study 2 were found to correlate negatively (i.e. higher happiness ratings were associated with lower SPT values). F0 mean, raised larynx, high perceived pitch and extensive pitch range all correlated positively with higher happiness ratings which is in keeping with the findings of Scherer (1981) that happiness correlates with higher pitch measures. Extensive lip and tongue range also correlated positively whilst interrupted continuity correlated negatively, indicating that the less fluid speech associated with the DS speakers is perceived more negatively.

5.5.5 Intelligent – unintelligent ratings

Increased ratings of confidence, friendliness and happiness all correlated positively with higher intelligence ratings in study 1, as did a more positive response to the 'spend time with' parameter. In line with the high happiness ratings a more spread lip pattern in study 3 also correlated with high intelligence judgements. Also in study 3 close jaw, decreased jaw protrusion, increased tongue range, pharyngeal expansion, faster speech rate and less interrupted speech correlated positively with high ratings of intelligence; all of which are polar opposites of the profile identified in speakers with DS in study 3.

5.5.6 'Spend time with' ratings

High confidence, friendliness and intelligence ratings all correlated positively with more 'yes' responses to the parameter 'spend time with', indicating that it was these features, over the other parameters of calmness, happiness, gender and age, that were most important for peers when judging who they wanted to build friendships with. As some degree of ID is a typical consequence of DS this result demonstrates the difficulties faced by these children in the formation of meaningful relationships with their TD peer group. Higher levels of pharyngeal expansion also correlated positively with higher spend time with ratings; again as constriction in the pharynx is a direct consequence of organic differences in DS this is an aspect which cannot be overcome by children with DS. Finally, interrupted continuity correlated negatively (less interruptions being perceived more positively) confirming its significant role in vocal social judgements.

5.5.7 Gender ratings

As described in the happy-sad correlations, higher ratings of happiness correlated positively with more female judgements of speakers in study 1. The majority of the correlations for gender centred around pitch values. In study 2 higher F0 mean and standard deviation both correlated positively with more female judgements, whilst in study 3 increased larynx height, higher mean perceptual pitch and range of pitch all correlated positively. All indicate the relationship between higher pitch measures and gender. The only negative correlation was for both the SPT measures (where lower values were associated with more female ratings). As a lower SPT indicates reduced attenuation of the harmonics of speech which is associated with a less lax larynx, which in turn is associated with higher pitch production, then this finding is not surprising.

5.5.8 Age ratings

The final parameter of age judgements also concerns measures relating to pitch. Low F0 mean, low F0 mean standard deviation, lowered larynx height and lower mean perceptual pitch all correlate negatively with age (i.e. they are associated with speakers who are rated as sounding older). Conversely, higher F0 mean and standard deviation, higher position of the larynx in the vocal tract and higher pitch are all associated with younger speakers. These correlations are entirely expected as they conform to typical norms for age and pitch.

5.6 CASE STUDIES OF SPEAKERS FROM STUDY 1

Relationships between the acoustic and perceptual profiles of individual speakers and the ratings received in the listener questionnaire will be examined for some of the individual speakers from study 1 in the following sections.

5.6.1 Speaker DS14

The median (in bold), IQR (in brackets) and the minimum to maximum age judgements of the three listener groups for speaker DS14 are shown in table 5.5. This male is 16.08 years old yet in the SNES, MES and PEER groups the maximum judgements of age are considerably older (70, 80 & 70 years respectively). Although some variation around the actual age would be expected, this margin is clearly unusual and as such the acoustic and perceptual features of this speaker are of interest.

AGE: Individual median (IQR) & min-max ratings for DS14 and DS6				
	actual age	**SNES ratings (30)**	**MES ratings (30)**	**PEER ratings (59)**
DS14	16.08	**16** (4) 8-70	**16.5** (5) 10-80	**15** (3) 5-70
DS6	16.50	**16.5 (4)** 11-25	**15.5 (5)** 12-40	**15 (2)** 11-20

Table 5.5: Summary of median (IQR) and min-max range of judgements of age made about speakers DS14 and DS6 by the SNES, MES and PEER listeners in the 'age' parameter of study 1

For DS14 protruded lip and jaw pattern, advanced tongue tip and body and minimised lip range are rated in the severe range by at least one of the two expert raters, whilst nasality and minimised tongue range ratings are in the extreme range by both raters. High whisperiness, harshness and slow rate are prevalent, and are characteristics which might point to the misperception of an older voice. DS6 is a male of approximately the same age who does not have such an extensive range of age judgements. The VPAS ratings for many of the parameters are very similar for these two speakers but some key differences can be seen. Notably DS14 has a more lax vocal tract, lower mean perceptual pitch, more harshness and a much slower speaking rate than DS6. Acoustically both are again similar, with the only obvious differences being in shimmer values. DS14 has higher shimmer values which are typically associated with lower loudness levels however perceptually loudness does not appear to be a distinguishing feature between the two speakers. The lower perceived pitch for speaker DS14 is also not backed-up by the acoustic F0 value which is actually higher for DS14 than DS6. From these observations it would seem that it may be the phonatory features of increased harshness together with the slow speech rate of DS14 which have been key to these excessive age judgements. Indeed, in a study of acoustic cues and their relevance to perceptions of age Harnsberger, Shrivastav, Brown, Rothman and Hollien (2008) found that when the speaking rate of middle aged persons was artificially decreased by 20% then naive listeners judged them to sound older.

Although there were extremely high age judgments for DS14 there were other raters in study 1 who judged him as sounding much younger than his chronological age. Thus it may be the case that listeners find his particular constellation of vocal characteristics unusual and as such difficult to estimate age accurately. Poor levels of attention during the listening task also cannot be ruled out.

It is notable that DS14 also received the lowest overall ratings for the ‘spend time with’ parameter (23%, taking into account the ‘yes’, ‘no’ and ‘not sure’ ratings combined) and the most negative calm-angry ratings (i.e. judged to sound most angry) for the SNES and PEER raters and the second most negative ratings for the MES raters; which based on vocal features alone puts this speaker at a considerable disadvantage in forming appropriate relationships with his peers.

5.6.2 Speaker DS13

By a considerable margin DS13, who is a 14 year old male, was judged to have the most negative confident-shy and happy-sad ratings (i.e. to sound to be the least happy and confident speaker) by all three of the listener groups in study 1. In contrast TD7 (who is also a male of 14 years of age) scored highly in both parameters.

Although differences were found between the two speakers, for example in lip pattern (DS13 being protruded whilst TD7 was slightly spread), jaw setting (DS13 having a more open and protruded jaw) and tongue setting (DS13 having more fronting and a minimised range) similarities were seen in pharynx configuration (slight constriction), nasal resonance (both having moderately high nasality ratings) and phonation features (a mixture of whispery, harsh and creaky voice; albeit that DS13 had higher creaky and whispery ratings and TD7 higher harshness ratings). However it is in the ratings of pitch, loudness and temporal features that strong differences become evident. DS13 was shown to have a much lower mean, range and variability of pitch (in keeping with his lowered larynx ratings, more protruded lip position and lower F0 mean and standard deviation values and in contrast to the raised and tense larynx which was associated with the relatively higher pitch seen in TD7). Lower loudness mean and range, slower rate and more interrupted speech were also seen in DS13, compared to the more neutral values of TD7. These findings support those of Scherer (1981) who found low mean and range of pitch and loudness together with slow tempo to be associated with sadness and low volume to be associated with low ratings of confidence.

The lower pitch of DS13 is in keeping with his lower F0 mean and standard deviation values from study 2, and the lower perceived volume supports the higher shimmer values. Interestingly, given the greater harshness ratings of TD7, his jitter values were less than half of those of DS13; however DS13 did have higher creakiness ratings which are likely to have contributed to this high perturbation. The higher SPT (which would cause steeper attenuation of harmonics) and lower HNR (associated with increased noise) seen in DS13 fit with his lower levels of laryngeal tension and reduced vocal tract tension ratings compared to TD7.

As with DS14 these very low ratings within two of the eight parameters of study 1 indicate that over and above the other physical, communication and intellectual difficulties associated with DS, this speaker will also have to overcome negative perceptions based on his voice quality.

5.6.3 Speaker TD5

The only typically developing speaker to receive the greatest number of inaccurate judgements in any of the study 1 parameters was TD5 (a 10 year old male) for judgements of gender. The second most inaccurately judged speaker, with ratings close to those of TD5, was DS26, also a young male (10.58 years). Similarities in the vocal profiles of these two speakers may point to possible causes of this misperception.

Both speakers had a similar high spread lip pattern but TD5 had more labiodentalisation whilst DS26 had a more extensive lip range. Larger differences were observed in jaw pattern as DS26 had a strongly protruded jaw, more open jaw and a very slightly more minimised range of movement compared to TD5's neutral values. Tongue values were similar (although DS26 had a greater degree of fronting of the tip and body and body raising) except for range which was diminished in DS26 and neutral in TD5. More pharyngeal constriction and a higher degree of nasality were also found in DS26 (although nasality was high for both speakers). Both had raised larynx but DS26 had higher ratings and DS26 also had a slightly lax vocal tract with a tense larynx compared to the neutral ratings of TD5. High whispery voice with low harshness levels were seen in both speakers but the whisper ratings were slightly higher for DS26. Just above neutral pitch values were seen for TD5 (which were surprisingly low given his high F0 value in the acoustic analysis) whilst much higher mean, range and variation of pitch were attributed to DS26.

Loudness, temporal organisation and respiration measures were very similar for both speakers, all being around neutral. Acoustically the spread lip pattern in both is consistent with the high F0 values (both being higher than the older TD and DS female speakers) and the relatively low jitter corresponds to low levels of perceptual harshness. The high whispery component of phonation might have been expected to yield high SPT and low HNR values, but in relation to the other speakers this doesn't seem to be a clear pattern for these two boys.

According to the more fronted tongue, higher and more tense larynx, more whispered voice and higher pitch values for DS26 it would have been expected that this male speaker would have been attributed more inaccurate 'female' gender judgements than TD5, but this was not the case. Perhaps the very different protruded and open jaw ratings and the higher nasality ratings of DS26 had the effect of reducing the impact of the other more female-typical settings. Regardless of some vocal differences, both are young males and as such have a number of child-like qualities, most notably fronted tongue position and low perturbation in phonation. Perceptually both also appeared rather excited or animated in their speech recordings, findings reflected in the high degree of spread lip posture ratings and the positive pitch and high F0 values. According to Ho (2001) pitch level and vocal tract length are the two most significant correlates to judging a speaker's gender; as both boys were of small stature and would thus have smaller, shorter vocal tracts than the older speakers, and given that there are no particularly strong differences in vocal tract morphology prior to the onset of puberty (Mackenzie Beck, 1997) then it is likely that these youthful vocal characteristics have had the effect of making the voices of these two speakers sound more ambiguous in terms of their gender than their older peers. In both boys this appears to be a typical developmental pattern which, if typical development continues, should alter with the onset of puberty when significant growth of the laryngeal structures in particular will bring about more masculine vocal features such as reduced pitch.

6
CONCLUDING COMMENTS

In the preceding chapter the findings of study 1 (ratings of the voices of TD children and children with DS by education staff and TD peers), study 2 (the instrumental analysis) and study 3 (perceptual analysis by expert SLT judges) were discussed in relation to evidence from previous research about the impact of vocal differences and how various features might interact. In this final chapter the key points will be summarised and consideration given to the social and clinical implications of the findings for children and young people with DS and their families. A short critical evaluation of the study will follow, identifying strengths and weaknesses within the research process, which will lead on to suggestions as to the direction that future research in this area might take.

6.1 SUMMARY OF FINDINGS AND THEIR IMPACT ON CHILDREN AND YOUNG PEOPLE WITH DS

A wide range of differences in the perceptual characteristics of the DS and TD speakers were found in study 3, with the children with DS having more protruded lips and jaw, more open jaw, more fronted and raised tongue, more minimised lip, jaw and tongue range, more constricted pharynx, higher nasality and nasal escape, less vocal tract tension, higher laryngeal tension, more whispered and harsh but less creaky phonation, lower pitch and loudness, more interrupted speech with slower rate and a higher degree of poor breath support; many of which have previously been found to be features associated with lower social status in Edinburgh speakers (Esling, 1978). These findings support earlier research indicating that vocal differences are frequently associated with people with DS, being likely to be borne out of structural and functional differences of the vocal apparatus and associated conditions such as hearing impairment. However, vocal features within the speakers with DS were found to vary widely, as did the degree of severity, which serves as a reminder that this is a heterogeneous group and thus that any therapeutic interventions need to be tailored to the needs of the individual.

Despite these significant differences it was apparent that the TD speakers also had relatively high levels of whisper, perturbation and nasality (albeit to a lower level than the speakers with DS) as well as equivalent larynx height ratings, which are likely to have contributed towards the fewer than expected differences between the two groups in the instrumental analysis findings of study 2. In that study there were no significant differences in jitter and shimmer (which were expected to be higher in the speakers with DS), or in HNR (which was expected to be lower), although as anticipated from previous research F0 levels were also not significantly different. Only SPT was found to differ between the TD and DS speakers, and even that was in the opposite direction to the a priori hypothesis. However, the higher level of SPT observed in the speakers with DS in this study would be consistent with the high levels of hypotonia associated with this syndrome, which would cause the laxer vocal tract to absorb more acoustic energy and the vocal folds to work less efficiently. The increased SPT in this group of English-speaking children with DS is in contrast to the only other SPT finding in people with DS, which showed lower SPT in young Portuguese-speaking children with DS in relation to TD peers (Moura et al., 2008); as that study also identified a different pattern of F0 compared to this and other English-speaking studies of children with DS (values being significantly higher in the Portuguese DS group) then it may indicate language-specific differences in the voice features of typically-developing children.

The large number of non-significant instrumental findings indicates some degree of similarity in the vocal characteristics of children with DS compared to those of the TD control subjects in this particular part of Scotland, in contrast to the significant acoustic analysis differences found between DS subjects and controls from other parts of the UK. For example, the similarities in measures of perturbation and noise (jitter, shimmer and HNR) are reflected in the pattern of phonation settings observed in both groups in the perceptual VPAS ratings (although featuring to a higher degree in the DS speakers). Thus both groups reflected a whispery voice with harshness, or (most predominantly in the TD males) whispery voice with creakiness.

It might have been expected then that these phonatory similarities between the DS and TD speakers from the Edinburgh area would lead to a less negative perception of voice for the children with DS. However, in study 1, across all the questionnaire parameters the children and young people with DS were rated significantly more negatively than the TD control subjects by both the groups of education staff raters (SNES & MES) and the TD PEER raters. Thus they were judged as sounding less calm, less confident, less friendly, less happy, and less intelligent than those without voice differences. Moreover they received more inaccurate judgements about gender and age than their TD peers.

Although no hypothesis was made concerning potential differences between the ratings of the different rater groups, it might have been predicted that the SNES raters would have judged the DS speakers either more positively than the MES raters (due to increased experience of people with voice disorders within special schools) or conversely more negatively (due to increased recognition that the unusual vocal features belonged to children with ID). However, ratings between the SNES and MES raters for the DS speakers did not differ significantly in any of the parameters, which suggests that judgements of character and ability based on voice are somewhat instinctive rather than being based on greater or lesser exposure to unusual voice features. Thus, even though both groups rated the speakers with DS significantly more negatively than the TD speakers, it can be seen that children and young people with DS aren't perceived any more negatively by education staff in mainstream schools than those in special school environments. As more and more children with DS are accessing mainstream education, this finding may be of particular interest and relief to parents and carers of young children with DS facing inclusive education, who may quite naturally fear that the lesser experience of and training around the needs of people with ID associated with mainstream might result in more negative perceptions of ability and thus lower levels of expectation; both of which could significantly affect educational and social potential.

Despite the positives identified above between the adult raters, the PEER raters were shown to be particularly negative in their attitudes towards the children with DS. This was most clear in their response to being asked if they would like to 'spend time with' the speakers that they were rating, where a huge gulf was found in the positive ratings between the groups (DS 'yes', 13% compared to TD 'yes', 50%) which highlights the considerable difficulty that children with DS face in forming meaningful friendships with their TD peers. This, coupled with the fact that TD children have been shown to prefer the company of other TD children over children with DS (Hamilton, 2005) presents a genuine barrier to social inclusion within mainstream education. Hamilton proposes that more direct strategies to promote peer interaction and thus further develop social skills may be required, pointing out that, according to Hanline (1993), in situations where relatively intrusive strategies have been employed (e.g. positioning children, prompting and reinforcing peer interaction and modelling appropriate social behaviours) children with disabilities have been found to demonstrate more interaction, which has then been reciprocated by TD peers. Clearly these are measures which would need to be implemented in the younger years of education in order for more social inclusion to be seen through the secondary years. Whether schools feel that they have the resources to effectively manage such programmes, particularly with the increasing numbers of children with special educational needs accessing mainstream education, remains to be seen.

6.2 CLINICAL IMPLICATIONS

The findings of this research indicate that there are vocal features associated with children and young people with DS from the Edinburgh area which disassociate them from their TD peers, such as the habitual lip, mandible and tongue settings, pharyngeal constriction, increased nasal resonance, altered muscular tension, phonation and prosodic differences, together with slower, more interrupted speech and poorer breath support. However there are also some features which, although not as extreme in the TD children, are common to both groups, particularly relating to the increased nasality ratings and a high degree of whispered voice in combination with some perturbation (harshness and/or creaky voice).

SLTs routinely prioritise caseloads according to clinical risks and the impact of those risks. In the same way clinicians invariably have to weigh-up the severity and impact of the different communication difficulties observed in individual clients to determine which merit treatment. Voice very often receives a lower priority in therapy, which may not be unreasonable where a client has severe intelligibility issues pertaining to poor articulation, phonological awareness or language skills. However, as this study and previous research has shown that there is considerable potential for negative judgements of ability and character based on voice, which have significant implications for social inclusion and thus psychological well-being then perhaps clinicians need to consider placing more importance on the remediation of voice disorder.

A key stumbling block in treating voice disorder in DS is the question of potential for change: people with DS are recognised to have a range of structural differences in their vocal anatomy which bring about altered voice and which are therefore going to be impervious to change. Furthermore there is frequently some concern about how accessible vocal therapies are to people with ID in terms of being able to understand instructions, comply with interventions and generalise skills into their everyday communication. Added to this is the dearth of knowledge of the TD voice norms for children from the Edinburgh area which has made it difficult to establish appropriate goals; this study has gone some way in identifying typical patterns in this population.

Traditional voice techniques have been relatively unexplored in DS. However, it may be the case that some functional improvements can be brought about in some areas of voice.

Poor breath support is a significant factor in DS. There are a range of vocal exercises based on improving the control of breath for speech. In a voice therapy programme for children with DS, Van Vuren (2009) suggests practising slow steady breaths versus short quick ones by making one big soapy blowing bubble followed by lots of little small ones or by blowing tissues pieces across a table in the same manner, whilst coordination of breathing may be improved through counting to three during inspiration and exhaling with a long slow 'ssssss' or 'aaaaaah' until the breath is gone. An element of competition can be brought into activities by seeing if children can count higher as they breathe in or out, which may help with motivation.

Limited range in the movements of the lips, tongue and jaw have all been identified in DS and contribute significantly to decreased intelligibility. Therapy exercises to increase these ranges may be able to bring about voice benefits as well as crossover improvements in articulation, such as increased vowel distinction.

Nasality may be reduced by simply opening the mouth wider during speech, which can help to improve the oral-nasal balance (Pentz & Moran, 1988), although it needs to be kept in mind that some degree of nasality is typical of Edinburgh speakers.

The increased laryngeal tension in DS may be tackled by indirect strategies, such as encouraging awareness of head and neck posture and relaxation of the neck and shoulders. More direct strategies might include the yawn/sigh technique, which according to Mathieson (2001) can greatly improve voice by prolonging the inspiration phase, causing the musculature of the pharynx to relax, thereby allowing maximum opening of the airway. The high tension of the larynx is likely to reflect the increased muscular effort required to initiate vocal fold movement in DS, and therefore clinicians need to consider that fatigue and low motivation might be factors which influence therapy.

Finally, the prosodic aspects of pitch and volume may also benefit from therapeutic intervention. Anecdotal reports suggest that identification of stressed words and syllables within utterances might help to make the cadence of speech more appropriate and increase intelligibility. More specifically pitch can be targeted through exercises designed to expand intonation range and level. Van Vuren (2009) suggests discrimination activities such as listening to voices and deciding if they are high or low pitched or thinking about how animals sound (giving the example of a squeaky mouse versus a low-voiced elephant) as well as production activities like counting at a low then high pitch or getting progressively higher in pitch. However, as a note of caution, although pitch tends to be perceived as being significantly lower, instrumental evaluation of F0 hasn't supported this finding, therefore clinicians need to be certain that pitch is problematic before tackling this feature. Awareness of volume can be encouraged, not only to increase intelligibility (where the voice is too quiet) but also to minimise the risk of damage where the voice is too loud. Much the same as for pitch a series of exercises involving moving from low to high volume can be used.

As hypotonia is understood to lessen as the child with DS matures then it is possible that some spontaneous improvements in voice quality may be brought about with this increasing muscle tone and strength, or perhaps that children may be better able to reap the benefits of therapy as muscular tone stabilises.

The mode of intervention may be a prime factor in the success of therapy. Music and singing have been identified as particular interests of people with DS (Rosner, Hodapp, Fidler, Sagun & Dykens, 2004) and according to Barker (1999) over a period of two years singing therapy was observed to bring about significant improvements in the conversational speech intelligibility of a young adult female with DS. Singing is proposed to stimulate auditory discrimination and memory skills, encourage repetition and therefore practise of words and sounds and to develop phonological awareness through the identification of onset and rime (Barker, 1999) and as such may be a particularly useful tool for voice therapy in this population who are recognised as having low levels of attention and motivation. As the visual channel is stronger than the auditory channel in DS (Buckley, 1993) then it may be the case that clinicians can increase motivation and the chance of successful therapy by utilising imaging techniques which provide visual feedback. Pryce (1994) has suggested that the use of electromyographic biofeedback would be an excellent way of helping people with DS to see the degree of tension in their vocal fold musculature. Similarly, a very recent study of the benefits of electropalatography in articulation therapy in DS found that the visual feedback associated with this technique had significant long-term benefits in helping the children to correct their tongue positions and thus contributed towards increased intelligibility (Wood, Wishart, Hardcastle, Cleland & Timmins, 2009).

More generally clinicians can encourage good vocal hygiene, including minimisation of abusive behaviours such as shouting or straining, emphasise the importance of drinking plenty of fluids (an aspect that is often a problem for people with ID) and highlight to parents and carers the danger of potentially damaging environments such as smoke-filled rooms which may cause irritation to the vocal apparatus and exacerbate breathing difficulties. As hearing impairment is so very common in DS, audiological referral to evaluate hearing function is key. Finally due to the considerable differences in vocal features and the severity of those features, together with the acknowledged attention deficits and low motivation, interventions must be individualised rather than syndrome specific.

6.3 Critical evaluation of research and future direction of research

This study's aims were to investigate the voice quality of children and young people with DS in comparison to TD age-matched peers, using both instrumental and perceptual measures, to establish the judgements of character, ability, age, gender and social desirability made by listeners about the voices of those children and to determine whether specific instrumental and perceptual ratings of voice quality correlate with specific listener judgements. A thorough perceptual and instrumental analysis has identified significant vocal differences between the DS and TD speakers, but with some overlap in features, which are likely to have contributed to the number of unexpected similarities in instrumental findings. Despite these similarities a questionnaire-based analysis of listener judgements revealed that the speakers with DS were rated significantly more negatively than their TD peers by all groups of raters across all the questionnaire parameters. A series of statistical correlations identified how the results of these three studies interacted, allowing for the beginnings of increased understanding of the impact of specific vocal features on specific listener judgements. Therefore it is the opinion of the researcher that these three key aims have been achieved in the course of studies 1, 2 and 3 of this research.

This study has been a hugely challenging but enjoyable process. It has necessitated a considerable amount of background reading in both voice and DS in order to plan and implement the method of data collection and to analyse the findings appropriately. As with any research there are strengths and weaknesses associated with this study, which will be discussed in the following sections. Some of these points lead on naturally to aspects of voice in DS which this study did not assess and as such indicate areas that would be appropriate for future research in this field.

6.3.1 Positive aspects of this research project

6.3.1.1 Sample size

It is notoriously difficult to recruit large numbers of people with disorders such as DS for research purposes. As this study was aligned to the much larger MRC speech motor control in DS study at QMU it was possible to approach participants with DS and their parents and carers involved in that study with a view to their participation in this study of voice. In order to make this proposition a desirable one the decision was taken (between the studies) that the voice recording necessary for this study could be done during one of the assessment or therapy sessions attached to the MRC speech motor control study. By doing so no additional effort (in terms of commitment or attendance at the university) was required, which consequently led to the relatively high number of DS subjects (n = 22). A considerable amount of effort was put into the recruitment of the TD peer raters/control subjects and the two groups of education staff. In total thirty schools and the Scouting organisation were approached, which despite only a small number of those agreeing to take part, still yielded high participant numbers (TD subjects, 102, SNES, 52 and MES, 45) which contributes significantly to the validity of the findings.

6.3.1.2 Breadth of the research

This study is, to the researcher's knowledge, the first in the field of DS to establish perceptual and instrumental features of voice quality, their impact on listeners' judgements of ability, character, age, gender and social desirability and to explore the relationship between the various findings. This study design has therefore been able to add to the current knowledge base but also to extend that knowledge further, offering more insight into the social impact of disordered voice. Educational inclusion for children with special educational needs is the current trend in the UK, meaning that more and more children with complex and multiple needs are accessing mainstream schools. This study has been able to establish that despite all the listener groups rating the speakers with DS more negatively than their TD peers, there is no difference in vocal perception between teachers in special and mainstream schools, which may be of particular reassurance to parents and carers of children with DS. However this research has also been able to show that these children are at considerable risk of being isolated socially within mainstream as their TD peers demonstrated a particularly negative response to their voices. These findings may help those involved in the care and education of children and young people with DS to be aware of the negative perception caused by differences in voice and its impact on social acceptance by peers, and to guard against instinctive, erroneous judgements based on atypical features.

6.3.2 Negative aspects of this research project

6.3.2.1 Absence of direct involvement in the DS data collection by the researcher

The recordings of the speakers with DS were completed during the MRC speech motor control assessment or therapy sessions by the SLT researcher for that study which meant that the voice researcher had little direct contact with the children with

DS and their data collection. However there were sound reasons for this being the case. This decision was taken after discussion with that researcher about how to cause the least disruption to her sessions. It was felt that as these sessions were already very busy and were completed in a small room with the researcher, the child and a parent or carer that to have another person (the voice researcher) involved would be too much of a distraction for the child and would also be likely to cause the activities of the session (including the recording) to be rushed in order to keep within appointment times. As high numbers had been achieved precisely because parents and carers were not being asked to keep additional appointments this eventuality was to be avoided if at all possible. Another significant factor was that the SLT researcher was well-known to the children, whilst the voice researcher was not, and as such it was considered that a familiar person offered the best chance of gaining a recording from these children which reflected their habitual vocal patterns. The voice researcher prepared all the resources required to elicit the voice sample and ensured that the SLT researcher understood clearly what was required. The voice researcher observed a recording session (with permission from the participants) via a clinic camera to ensure that the process was adequate, and was reassured that this was the case. However, despite these measures the quality of a small number of the recordings was unsuitable for voice analysis (due to low recording level and high background noise) and these subjects had to be excluded from the study.

6.3.2.2 Depth of the research

As this study looked to cover a broad range of voice analysis involving perceptual and instrumental techniques, the impact of voice, and the interactions between studies, in retrospect it feels that this was perhaps at the expense of the level of detail that the researcher was able to explore about each. For example, some aspects of previous voice research in DS were not addressed, such as instrumental evaluation of the formant amplitude levels of vowels. Also it would have been nice to be able to explore the relationship between the voice measures and the intelligibility measures of the MRC speech motor control study; this could be a possible source of future research in DS. However, as this is a piece of research that was for the most part completed by a single person, it is acknowledged that there are limits on what can reasonably be achieved.

6.3.2.3 Isolation of the effects of voice

This study purports to be a study of voice and its impact on listener judgements. However, due to the constellation of disability associated with DS it is difficult to isolate the role of voice in the judgements made by raters. The effects of poor articulation and phonological awareness, accompanied by reduced grammatical and vocabulary skills and lack of story-telling coherence were controlled to some extent by presenting to raters the recordings of the DS speakers with the highest expressive language scores (based on the CELF expressive language component completed as part of the MRC speech motor control study assessment battery). This was enhanced by editing out overly long and complex structures used by the TD speakers and presenting those individual utterances in a random order, and also by direct instruction of the listeners to focus not on the clarity of words or sentences or the sense of those utterances but on the way the voices sound. Previous studies played samples backwards but this method was rejected as it was felt that it would distort the natural prosodic voice features which are necessary for judgements about voice.

6.3.2.4 DS perception of voice

Although this study was valuable in gaining an insight into the perception of TD and DS voices by education staff and a group of TD peers, it would have been extremely interesting to evaluate the perception of a group of people with DS about these same recordings. This would have been difficult in this study as the method of evaluation (the listener questionnaire) would have had to be changed significantly to make it accessible to young raters with DS. Poor auditory short-term memory, literacy and comprehension problems and sensory deficits, taken together with the possible effect of a bias in results due to unintentional prosodic prompts (e.g. changes in word stress or volume if reading out the questionnaire options) by the person supporting the raters with DS to complete the questionnaire would make this task a challenging one. Additionally, it would have been difficult to recruit enough people with DS (over and above those already recorded) of the appropriate age, geographical background and with sufficient hearing and visual acuity and adequate comprehension skills to complete the task. For these reasons it was decided to concentrate on groups of raters who could all access the same unmodified listener questionnaire, thus making their results directly comparable. It may be that a more qualitative approach needs to be taken where people with DS describe what they think in a less controlled manner and key themes are extrapolated from the findings. DS vocal self-perception and the perception of the voices of peers would be an aspect of research which would be extremely informative, giving an insight into awareness of atypical features as well as an understanding of how this group feels about these vocal differences.

6.4 CONCLUSION

Voice disorder has long been recognised as a feature of DS (Strazzulla, 1953; Schlanger, 1962; Tredgold & Soddy, 1963; Blanchard, 1964; Benda, 1965; Novak, Sedlackova, Klajman & Betlycwski, 1967). This study confirms a range of perceptual vocal differences between children and young people with DS and their age-matched TD peers, which are not always borne out by instrumental analysis of voice; perhaps indicating that there is a combined effect of vocal differences which causes greater perception of atypical voice features. In contrast to the given hypothesis significantly greater SPT was observed; a finding which would support the negative effects of hypotonia on the vocal folds and vocal tract. As predicted, significant differences were not found in measures of F0, but unexpectedly jitter, shimmer and HNR values failed to differ also. In line with hypotheses, special-needs and mainstream education staff and TD peers were all found to rate the voices of the children and young people with DS significantly more negatively than those of the TD controls, and in particular the TD children indicated very strongly that the voices of the speakers with DS were not socially desirable, demonstrating clear difficulties in the potential for the formation of meaningful friendships between children and young people with DS and their TD peers.

Thus it is clear from the findings of this research that voice plays a significant role in how speakers are perceived by listeners and as such highlights the need for clinicians to consider whether improvements to voice quality can be made for individual speakers with DS. Where it is judged that direct therapy would not prove to be successful then it may be the case that indirect interventions, in the form of helping to make those involved in the education and care of people with DS aware of the potential for negative judgements as well as encouragement of improved vocal hygiene, could help to maximise vocal potential.

Significant improvements in healthcare mean that there are now more people with DS and these numbers are expected to rise (Fonseca, Amaral, Ribeiro, Beserra & Guimaraes, 2005). If it is recognised that this growing population of people with DS have the right to have their voices heard and accepted in our society, then those professionals with the skills and training to treat or advise about voice disorder need to be working towards making that right a reality.

APPENDICES

Appendix I: Information sheet for parents of children with DS

Queen Margaret University

EDINBURGH

Information for Parents of Children with Down's Syndrome

The Down's Syndrome Association is funding a three year PhD research project at Queen Margaret University, Edinburgh, into the **voice quality of children and young people with Down's syndrome and the judgements that listeners make about these children based on their voices.**

Why investigate voice quality?

Our voice features play an important part in how we are viewed by others, allowing us to express who we are through our own individual characteristics as well as helping to convey different emotions such as anger, happiness or frustration.

As many children with Down's syndrome now attend mainstream schools rather than special schools it is likely that they spend more time with other children of their own age without disabilities. Therefore it is important to understand how both teachers and peers respond to, and make judgements about, the voices of children with Down's syndrome and in particular how this might impact on the development of friendships with peers.

The Research

This project will record the speech of a number of children with Down's syndrome as well as children without Down's syndrome, in order to compare the two.

Three groups of listeners will be asked to make judgements about how they think each voice sounds (e.g. *happy*, *sad, angry, clever, confident, shy).* The groups of listeners will be:

- Special-needs education staff.
- Mainstream education staff.
- Children without developmental delay/disorder

The researcher will then analyse the voices at the university using computer software and a perceptual voice assessment.

What will this involve for your child?

This project is running alongside the Medical Research Council (MRC) study of speech control, with which your child is already involved.

During one of the therapy or assessment sessions for the articulation study your child would be asked to look at a series of pictures and describe what they see and this activity would be recorded. This means that **your child would not be required to attend any additional sessions at the university.**

Risks of Taking Part

There are no health or safety risks associated with participation. All personal information will be held securely at the university and accessed only by the researcher and supervisory team.

Thank you for taking the time to read this information. Should you agree to allow this study access to your child's voice recordings, please sign the attached consent form. If at any point you or your child wishes to withdraw consent, this can be done without giving a reason and will not affect the therapy received as part of the MRC project.

If you would like any further information, please contact:

Rebecca Rodger
PhD Research Student
Speech Science Research Centre
Queen Margaret University
Edinburgh
EH21 6UU

Tel 0131 474 0000
e-mail: rrodger@qmuc.ac.uk

If you have any concerns and wish to speak to someone who is independent from this research please contact:

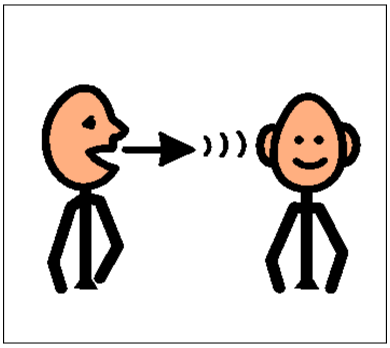
Prof Fiona Gibbon (Head of Subject) 0131 474 0000

Appendix II: Information sheet for children with DS

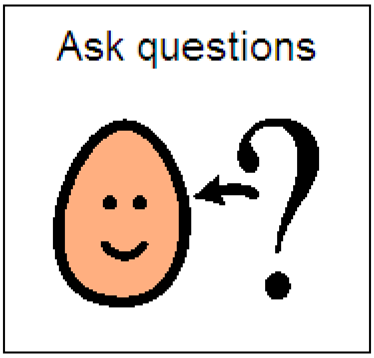
Queen Margaret University
EDINBURGH

Information for children with Down's syndrome

Would you like to take part in a study about how your voice sounds?



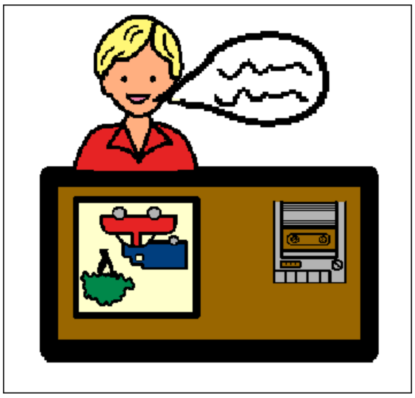
- Please read this information with an adult and ask questions about anything you don't understand.



What do you have to do?

- When you come to Queen Margaret University to work with the speech and language therapist she will ask you to look at 5 pictures and tell her what you see.

- Your voice will be recorded so that some adults and children can listen to it later and tell us how they think it sounds.



Appendix III: Consent form for child participants

Queen Margaret University
EDINBURGH

Participant Identification Number:

CONSENT FORM – CHILD PARTICIPANTS

PhD Research Project: Voice Quality and Associated Listener Judgements.

Please tick box

1. I confirm that I have read and understood the information sheet for the above study. ☐

2. I understand that my/my child's participation is voluntary and that I/he/she is free to withdraw at any time, without giving reason. ☐

3. I agree to being/that my child may be **audio-recorded** for this study. ☐

4. I agree that my/my child's audio-recordings may be kept for **research/teaching** purposes. ☐

5. I have/my child has read the consent form and agree to take part in the above study. ☐

______________________	__________	________________
Child's name	Date	Signature of child
______________________	__________	________________
Parent's / guardian's name	Date	Signature of parent / guardian
______________________	__________	________________
Researcher	Date	Signature

Appendix IV: Information sheet for parents of TD children

Queen Margaret University

EDINBURGH

Information for Parents

A PhD research project at Queen Margaret University, Edinburgh is assessing **voice quality and the judgements which listeners make about voices based on how speakers sound.** Part of this study will look at the voices of **children who have no voice problems**.

Why investigate voice quality?

Our voice features play an important part in how we are viewed by others, allowing us to express who we are through our own individual characteristics as well as helping to convey different emotions such as anger, happiness or frustration. People with unusual voices have been found to be judged more negatively, which may affect self-esteem and the development of friendships or relationships.

The Research

This project will record the speech of a number of children with and without voice problems which will then be rated by three groups of listeners. The listeners will make judgements on how they think each voice sounds (e.g. *happy, sad, angry, clever, confident, shy).* The groups of listeners will be:

- Special-needs education staff.
- Mainstream education staff.
- Children without developmental delay/disorder.

The researcher will then analyse the voices at the university using computer software and a perceptual voice assessment.

What will this involve for your child?

Your child would meet with the researcher (who is a qualified speech and language therapist) and be asked to describe 5 pictures; this activity would be recorded. Your child would then listen to the voices of some other people and fill in a questionnaire about how he/she thinks they sound. **If your child takes part he/she will receive a £10 gift voucher for Virgin Megastore.**

Further Information

Thank you for taking the time to read this information. If you agree to your child taking part, **please fill in the attached consent form**. If you would like any further information, please contact:

Rebecca Rodger
Speech Science Research Centre
Queen Margaret University, Edinburgh
EH21 6UU
Tel: 0131 474 0000
e-mail: rrodger@qmuc.ac.uk

If you have any concerns and wish to speak to someone who is independent from this research please contact:

Prof Fiona Gibbon (Head of Speech and Language Therapy) 0131 474 0000

Appendix V: Information sheet for TD children

Queen Margaret University

EDINBURGH

Information for Students Taking Part in Voice Study

Would you like to take part in a study about how voices are judged?

What do you have to do?

You will listen to the voices of 20 people telling a story and complete a simple questionnaire about how each voice sounds (by ticking or circling the answers which best describe the voice you are listening to). This will take about 25 minutes.

E.g. Does voice number 1 sound:

- male - female - not sure
- very happy - quite happy - in the middle - quite sad - very sad
- Would you like to spend time with this person? yes - not sure - no
- How old is the person speaking?

You will then be asked to look at 5 pictures and talk about what you see, this will be recorded. Later, the researcher will analyse the all the recorded voices at the university using computer software and a speech therapy rating scale. The recording will take about 10 minutes.

If you complete the questionnaire and the recording you will receive a £10 gift voucher for Virgin Megastore.

Appendix VI: Information sheet for education staff

Queen Margaret University

EDINBURGH

Information for Education Staff Participants

A three year PhD research project at Queen Margaret University, Edinburgh proposes to examine **voice quality, and the judgements about ability and character which listeners associate with a range of speakers.**

Why investigate voice quality?

Voice quality plays a significant role in how we are perceived by others, allowing expression of our individuality through habitual vocal characteristics, which can be adapted according to our mood and environment. Unusual vocal features have been found to be associated with negative judgements about the speaker which may have considerable impact on self-esteem and social interaction.

The Research

Recorded speech samples will be rated by three groups of listeners who will make judgements about what they think are the character traits and abilities which best match each voice-recording. The groups of listeners will be:

- Special-needs education staff
- Education staff working in mainstream schools
- A group of children

What will this involve?

Participation in this project will involve you listening to a series of recorded voice samples. For each voice you would be asked to complete a short questionnaire to rate how you think that voice sounds. The process is estimated to take about 25 minutes.

Risks of Taking Part

There are no anticipated risks in participation in this voice study. Your anonymity will be preserved as all questionnaires will be coded rather than named and only the research team will have access to your responses.

Thank you for taking the time to read this information. Should you agree to participate, please complete the attached consent form. If at any point you wish to withdraw consent, this can be done without giving a reason.

Further Information

If you would like any further information, please contact:

Rebecca Rodger - PhD Research Student
Speech Science Research Centre
Queen Margaret University
Edinburgh
EH21 6UU
Tel: 0131 474 0000
e-mail: rrodger@qmuc.ac.uk

If you have any concerns and wish to speak to someone who is independent from this research please contact:

Prof Fiona Gibbon (Head of Subject) Tel 0131 474 0000

Appendix VII: Consent form for adult participants

Queen Margaret University

EDINBURGH

Participant Identification Number:

CONSENT FORM – ADULT PARTICIPANTS

PhD Research Project: Voice Quality and Associated Listener Judgements.

Please initial box

1. I confirm that I have read and understood the information sheet for the above study and have had the opportunity to ask questions. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason. ☐

3. I have read the consent form and agree to take part in the above study. ☐

Name of participant	Date	Signature of participant
Name of person taking consent (if different from researcher)	Date	Signature
Researcher	Date	Signature

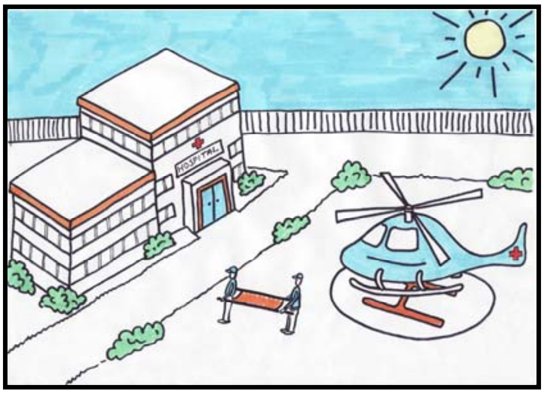
Appendix VIII : Stimulus pictures used to elicit language samples in study 1

1.

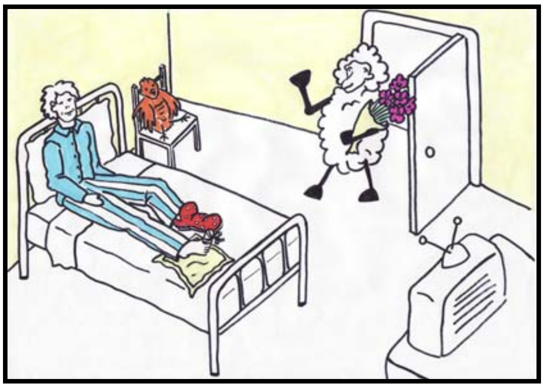
2.

3.

4.



5.



Appendix IX : Demographic information and results of expressive language test scores for the speakers from study 1

Summary of demographic information and expressive language test scores for the speakers from study 1				
	Gender	**Age**	**School type**	**CELF-P UK Expressive subtest age-equivalent scores**
DS13	M	14.00	mainstream	4.00
DS26	M	10.58	mainstream	4.00
DS24	M	11.58	mainstream	3.67
DS14	M	16.08	mainstream	4.00
DS7	M	11.00	mainstream	3.50
DS30F	F	10.08	mainstream	2.92
DS6	M	16.50	mainstream	3.25
DS8	M	11.25	mainstream	4.42
TD2	M	12.00	mainstream	n/a
TD1	M	12.25	mainstream	n/a
TD5	M	10.00	mainstream	n/a
TD4F	F	11.83	mainstream	n/a
TD7	M	14.00	mainstream	n/a
TD3	M	12.08	mainstream	n/a
TD8	M	14.50	mainstream	n/a
TD6	M	15.00	mainstream	n/a

Appendix X: Samples of speech rated by listeners in study 1

DS24: (49 seconds)

The boy hurt his toe, it was bleeding.

It was raining.

The boy, and, uh… a chicken.

Umbrella moved into the car.

Men took the boy to the hospital.

Boy frightened, the chicken flying.

Sheep came, gave…took some flowers to the boy.

Chicken called for help.

Car bashed into a tree

TD5: (42 seconds)

It's got black wheels.

There's a sheep with a red car.

And the person has a…is knocked over, and has a bleeding toe.

With a yellow umbrella in their hand.

And the sheep is come with flowers.

There's a tree that's losing its leaves.

And the chicken is going after it.

A person wearing red clothes, a scarf and a hat.

And next to the person is a chicken.

The person is in the hospital with his chicken.

Appendix XI: Voice rating questionnaire for TD peer raters

CODE: _________

VOICE QUESTIONNAIRE

How old are you? ☐

Are you a boy ? ☐ Or a girl? ☐ (tick box)

Draw a circle around the words that best describe how the voices sound.
Circle **ONE** answer only for questions 1-7 and write the age of the person speaking in the box in question 8.

VOICE 1 sounds:

1. male female not sure
2. very happy quite happy in the middle quite sad very sad
3. very calm quite calm in the middle quite angry very angry
4. very confident quite confident in the middle quite shy very shy
5. very friendly quite friendly in the middle quite unfriendly very unfriendly
6. very intelligent quite intelligent in the middle quite unintelligent very unintelligent
7. Would you like to spend time with this person? yes no not sure
8. How old is the person speaking? ☐

Appendix XII: Voice rating questionnaire for education staff raters

ADMINISTRATION CODE:______

VOICE QUESTIONNAIRE

Please tick the appropriate box

OCCUPATION: Special Needs Education Staff ☐ Mainstream Education Staff ☐

ROLE ____

GENDER: Male☐ Female☐

AGE: < 20 ☐ 20-29☐ 30-39☐ 40-49☐ 50-59☐ 60+☐

INSTRUCTIONS: Listen to the first voice and complete the questions for **VOICE 1** in the following questionnaire. Repeat for each voice played.

Circle **ONE** answer only for questions 1-6 and write the age of the speaker in the box in question 7.

VOICE 1 sounds:

1. male female not sure
2. very happy quite happy neutral quite sad very sad
3. very calm quite calm neutral quite angry very angry
4. very confident quite confident neutral quite shy very shy
5. very friendly quite friendly neutral quite unfriendly very unfriendly
6. very intelligent quite intelligent neutral quite unintelligent very unintelligent
7. How old is the person speaking? ☐

Appendix XIII: Samples of speech analysed in study 2 (acoustic analysis) and study 3 (perceptual analysis)

DS24: (1 minute 23 seconds)

It was raining. The boy, and, uh… a chicken. (long pause) Wet, boy put (up) a umbrella. Umbrella moved into the car. Boy frightened, the chicken flying. Chicken called for help. The boy hurt his toe, it was bleeding. The sheep put his hands on his ears. Car bashed into a tree. Umbrella blowed into ground. Men took the boy to the hospital. Sheep came, gave…took some flowers to the boy. And a little bit in the (unintelligible) boy is lying in bed. And plaster his toe. (long pause) And (long pause) an oh, television.

TD5: (1 minute 41 seconds)

There's a sheep with a red car. It's got black wheels. Erm…some… A person wearing red clothes, a scarf and a hat. (pause) With a yellow umbrella in their hand. And next to the person is a chicken. I think (laughs). Erm… There's a tree that's losing its leaves. (pause) Grass and clouds. The person has dropped his umbrella (pause) in front of the sheep's car (pause). And the chicken is going after it (laughs). The chicken cries for help. (pause) And the person has a…is knocked over, and has a bleeding toe. And the umbrella is flying away. And there's a helicopter (pause) and people carrying something (laughs). And they're going into the hospital. Eh… The person is in the hospital with his chicken (laughs). He's very hurt (pause) his toe's been wrapped in bandages. And the sheep is come with flowers. Eh… and there's a TV in front of the guy (laughs). A blue shirt (pause) and blue and white trousers, or pyjamas or whatever they are.

Appendix XIV: Praat script for instrumental analysis of F0, jitter, shimmer, HNR and SPT created by Dr Felix Schaeffler, SHS, QMU, Edinburgh

```
form Please enter pitch minimum and maximum for analysis
	positive minimum 100
	positive maximum 500
endform
clearinfo
pbot = minimum
pceil = maximum
pt$ = selected$("Sound")
labid = 0
firsthstar = 1
#select Sound 'pt$'
#To Pitch (cc)...  0 100 15 no  0.03 0.45 0.01 0.35 0.14 500
select Sound 'pt$'
plus Pitch 'pt$'
To PointProcess (cc)
select TextGrid 'pt$'
nofint = Get number of intervals... 1
print label'tab$'interval_nr'tab$'duration'tab$'mean_pitch'tab$'mean_sdev'tab$'
print nof_periods'tab$'jit_rap'tab$'jit_ppq5'tab$'shim_apq3'tab$'
print shim_apq5'tab$'HNR'tab$'autocorr'tab$'energy_diff15'tab$'energy_diff25
printline
for i from 1 to nofint
	select TextGrid 'pt$'
	label$ = Get label of interval... 1 i

	if label$ != "" & label$ != "U"
		start = Get starting point... 1 i
		end = Get end point... 1 i
		select Sound 'pt$'
		plus Pitch 'pt$'
		plus PointProcess 'pt$'

		voirep$ = Voice report... start end pbot pceil 1.3 1.6 0.03 0.45
		dur1 = end-start
		dur2 = extractNumber(voirep$, "duration: ")
		meanpitch = extractNumber(voirep$, "Mean pitch: ")
		sdev = extractNumber(voirep$, "Standard deviation: ")
		nofp = extractNumber(voirep$, "Number of periods: ")
		rap = extractNumber(voirep$, "Jitter (rap): ")
		rap = rap*100
		ppq5 = extractNumber(voirep$, "Jitter (ppq5): ")
		ppq5 = ppq5*100
```

continued

```
			apq3 = extractNumber(voirep$, "Shimmer (apq3): ")
			apq3 = apq3 * 100
			apq5 = extractNumber(voirep$, "Shimmer (apq5): ")
			apq5 = apq5 * 100
			hn = extractNumber(voirep$, "Mean harmonics-to-noise ratio: ")
			acorr = extractNumber(voirep$, "Mean autocorrelation: ")
			select Sound 'pt$'
			Extract part... start end rectangular 1 no
			To Spectrum... yes
			ediff15 = Get band energy difference... 1000 5000 0 1000
			ediff25 = Get band energy difference... 2000 5000 0 2000
			tmpname$ = pt$ + "_part"
			select Spectrum 'tmpname$'
			plus Sound 'tmpname$'
			Remove
			if nofp > 14
				print
'label$"tab$"i"tab$"dur1:3"tab$"meanpitch:3"tab$"sdev:3"tab$'
				print
'nofp"tab$"rap:3"tab$"ppq5:3"tab$"apq3:3"tab$"apq5:3"tab$'
				print 'hn:3"tab$"acorr:3"tab$"ediff15:3"tab$"ediff25:3'
				printline
			endif
		endif

endfor
```

Appendix XV: Vocal Profile Analysis Scheme form (Laver, Wirz, Mackenzie & Hiller, 1991; revised 2007, Laver & Mackenzie Beck)

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: …………………. Date of recording: ……… Judge: ………… Recording ID: ………

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range						
2. Mandibular			Close jaw						
			Open jaw						
			Protruded jaw						
			Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade						
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body						
			Raised tongue body						
			Lowered tongue body						
			Extensive range						
			Minimised range						
5. Pharyngeal			Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal						
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx						
			Lax larynx						

C. PHONATION FEATURES

		Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
	SETTING			1	2	3	4	5	6
10. Voicing type	Voice								
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery								
12. Laryngeal irregularity	Harsh								
	Tremor								

D. PROSODIC FEATURES

13. Pitch	Mean		High						
			Low						
	Range		Extensive range						
			Minimised range						
	Variability		High						
			Low						
14. Loudness	Mean		High						
			Low						
	Range		Extensive range						
			Minimised range						
	Variability		High						
			Low						

E. TEMPORAL ORGANIZATION

15. Continuity		Interrupted						
16. Rate		Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	
	Inadequate	
18. Diplophonia	Absent	
	Present	

Appendix XVI: Completed VPAS form for DS13

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS13** Date of recording: **11/05/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion	x	✓				
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range		x	✓			
2. Mandibular			Close jaw						
			Open jaw	✓	x				
			Protruded jaw	x		✓			
			Extensive range						
			Minimised range	x					
3. Lingual tip/blade			Advanced tip/blade				x ✓		
			Retracted tip/blade						
4. Lingual body			Fronted tongue body		x ✓				
			Backed tongue body						
			Raised tongue body	x					
			Lowered tongue body						
			Extensive range						
			Minimised range				x ✓		
5. Pharyngeal			Pharyngeal constriction	x					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx		x	✓			
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract	x	✓				
9. Laryngeal tension	x✓		Tense larynx						
			Lax larynx						

C. PHONATION FEATURES									
	SETTING	Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
				1	2	3	4	5	6
10. Voicing type	Voice		x ✓						
	Falsetto								
	Creak								
	Creaky				x	✓			
11. Laryngeal frication	Whisper								
	Whispery					x ✓			
12. Laryngeal irregularity	Harsh			✓					
	Tremor								

D. PROSODIC FEATURES									
13. Pitch	Mean		High						
			Low		x	✓			
	Range		Extensive range						
			Minimised range				x ✓		
	Variability		High						
			Low				x ✓		
14. Loudness	Mean		High						
			Low		x ✓				
	Range		Extensive range						
			Minimised range		x ✓				
	Variability		High						
			Low						

E. TEMPORAL ORGANIZATION								
15. Continuity		Interrupted	x	✓				
16. Rate		Fast						
		Slow	x ✓					

F. OTHER FEATURES		
17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XVII: Completed VPAS form for DS26

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS26** Date of recording: **06/05/2008** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
1. Labial			Lip spreading	✓	x				
1. Labial			Labiodentalization	✓					
1. Labial			Extensive range	x	✓				
1. Labial			Minimised range						
2. Mandibular			Close jaw						
2. Mandibular			Open jaw	x ✓					
2. Mandibular			Protruded jaw			x ✓			
2. Mandibular			Extensive range						
2. Mandibular			Minimised range	x					
3. Lingual tip/blade			Advanced tip/blade			x	✓		
3. Lingual tip/blade			Retracted tip/blade						
4. Lingual body			Fronted tongue body		x ✓				
4. Lingual body			Backed tongue body						
4. Lingual body			Raised tongue body	✓	x				
4. Lingual body			Lowered tongue body						
4. Lingual body			Extensive range						
4. Lingual body			Minimised range	x	✓				
5. Pharyngeal			Pharyngeal constriction		x				
5. Pharyngeal			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
6. Velopharyngeal			Nasal			x		✓	
6. Velopharyngeal			Denasal						
7. Larynx height			Raised larynx		✓	x			
7. Larynx height			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
8. Vocal tract tension			Lax vocal tract	✓					
9. Laryngeal tension			Tense larynx		x				
9. Laryngeal tension			Lax larynx						

C. PHONATION FEATURES									
		Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
	SETTING			1	2	3	4	5	6
10. Voicing type	Voice		x ✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery					✓	x		
12. Laryngeal irregularity	Harsh			x					
	Tremor								

D. PROSODIC FEATURES									
13. Pitch	Mean		High				x ✓		
			Low						
	Range		Extensive range	x	✓				
			Minimised range						
	Variability		High		✓	x			
			Low						
14. Loudness	Mean		High						
			Low	✓					
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION								
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES		
17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XVIII: Completed VPAS form for DS24

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS24** Date of recording: **09/05/2008** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion		x ✓				
			Lip spreading						
			Labiodentalization		x	✓			
			Extensive range						
			Minimised range		✓	x			
2. Mandibular			Close jaw						
			Open jaw	✓	x				
			Protruded jaw		x	✓			
			Extensive range						
			Minimised range	x					
3. Lingual tip/blade			Advanced tip/blade		x	✓			
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body		x ✓				
			Raised tongue body	✓	x				
			Lowered tongue body						
			Extensive range						
			Minimised range		✓	x			
5. Pharyngeal			Pharyngeal constriction	✓		x			
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape				✓	x	
			Nasal				✓	x	
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx	✓					
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx		x				
			Lax larynx	✓					

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x ✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery					✓	x		
12. Laryngeal irregularity	Harsh			x ✓					
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean		High						
			Low	✓	x				
	Range		Extensive range						
			Minimised range	✓	x				
	Variability		High						
			Low		x ✓				
14. Loudness	Mean		High						
			Low	✓	x				
	Range		Extensive range						
			Minimised range	x					
	Variability		High						
			Low		x				

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity		Interrupted			x	✓		
16. Rate		Fast						
		Slow	x					

F. OTHER FEATURES

17. Respiratory Support	Adequate	
	Inadequate	x✓
18. Diplophonia	Absent	x✓
	Present	

Appendix XIX: Completed VPAS form for DS14

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS14** Date of recording: **19/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion		x	✓			
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range		x	✓			
2. Mandibular			Close jaw						
			Open jaw	✓	x				
			Protruded jaw			x	✓		
			Extensive range	✓					
			Minimised range	x					
3. Lingual tip/blade			Advanced tip/blade			x	✓		
			Retracted tip/blade						
4. Lingual body			Fronted tongue body			x ✓			
			Backed tongue body						
			Raised tongue body	x					
			Lowered tongue body						
			Extensive range						
			Minimised range				x ✓		
5. Pharyngeal			Pharyngeal constriction	✓	x				
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal					x ✓	
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx		x ✓				
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract		x ✓				
9. Laryngeal tension	x✓		Tense larynx						
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
				1	2	3	4	5	6
10. Voicing type	Voice		x ✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery					x ✓			
12. Laryngeal irregularity	Harsh				x ✓				
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean		High						
			Low		✓		x		
	Range		Extensive range	✓					
			Minimised range						
	Variability		High	x	✓				
			Low						
14. Loudness	Mean		High	✓					
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate		Fast						
		Slow			x ✓			

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XX: Completed VPAS form for DS7

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS7** Date of recording: **05/06/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion	x					
			Lip spreading		✓				
			Labiodentalization		✓				
			Extensive range						
			Minimised range	x					
2. Mandibular			Close jaw						
			Open jaw	✓	x				
			Protruded jaw	✓					
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade	x ✓					
			Retracted tip/blade						
4. Lingual body			Fronted tongue body		x ✓				
			Backed tongue body						
			Raised tongue body		x ✓				
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal			Pharyngeal constriction	x					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape				x ✓		
			Nasal			✓	x		
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx		x ✓				
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract	✓					
9. Laryngeal tension			Tense larynx		x				
			Lax larynx						

		Present		Scalar Degree					
				Moderate			Extreme		
C. PHONATION FEATURES	SETTING	Neutral	Non-neutral	1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery					x ✓			
12. Laryngeal irregularity	Harsh			x					
	Tremor								
D. PROSODIC FEATURES									
13. Pitch	Mean		High						
			Low	✓	x				
	Range		Extensive range						
			Minimised range	x ✓					
	Variability	x✓	High						
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						
E. TEMPORAL ORGANIZATION									
15. Continuity			Interrupted	x	✓				
16. Rate		x✓	Fast						
			Slow						
F. OTHER FEATURES									
17. Respiratory Support			Adequate						
			Inadequate	x✓					
18. Diplophonia			Absent	x✓					
			Present						

Appendix XXI: Completed VPAS form for DS30F

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS30F** Date of recording: **13/12/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion		x ✓				
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range		x	✓			
2. Mandibular			Close jaw						
			Open jaw		x ✓				
			Protruded jaw		✓				
			Extensive range						
			Minimised range	x					
3. Lingual tip/blade			Advanced tip/blade			x ✓			
			Retracted tip/blade						
4. Lingual body			Fronted tongue body			x ✓			
			Backed tongue body						
			Raised tongue body	x					
			Lowered tongue body						
			Extensive range						
			Minimised range		x	✓			
5. Pharyngeal			Pharyngeal constriction	x ✓					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx	x ✓					
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx						
			Lax larynx	✓					

C. PHONATION FEATURES									
	SETTING	Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
				1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				✓	x			
12. Laryngeal irregularity	Harsh				x				
	Tremor								

D. PROSODIC FEATURES									
13. Pitch	Mean		High						
			Low		x				
	Range	x✓	Extensive range						
			Minimised range						
	Variability		High						
			Low	x ✓					
14. Loudness	Mean		High	x ✓					
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability		High	x ✓					
			Low						

E. TEMPORAL ORGANIZATION								
15. Continuity		Interrupted		x	✓			
16. Rate		Fast						
		Slow	x	✓				

F. OTHER FEATURES		
17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXII: Completed VPAS form for DS6

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS6** Date of recording: **14/06/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion		✓	x			
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range	x		✓			
2. Mandibular			Close jaw						
			Open jaw	x	✓				
			Protruded jaw			x	✓		
			Extensive range						
			Minimised range		x				
3. Lingual tip/blade			Advanced tip/blade			x	✓		
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body	x	✓				
			Raised tongue body		✓	x			
			Lowered tongue body						
			Extensive range						
			Minimised range			x ✓			
5. Pharyngeal			Pharyngeal constriction	✓	x				
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape				x ✓		
			Nasal					x ✓	
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx		x ✓				
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract	✓					
9. Laryngeal tension			Tense larynx	x					
			Lax larynx						

C. PHONATION FEATURES									
		Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
	SETTING			1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky			✓					
11. Laryngeal frication	Whisper								
	Whispery					x	✓		
12. Laryngeal irregularity	Harsh			x					
	Tremor								

D. PROSODIC FEATURES									
13. Pitch	Mean		High						
			Low		x ✓				
	Range		Extensive range						
			Minimised range	x	✓				
	Variability	x✓	High						
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION								
15. Continuity		Interrupted	✓					
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES		
17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXIII: Completed VPAS form for DS8

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **DS8** Date of recording: **15/11/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading	x	✓				
			Labiodentalization	x	✓				
	x✓		Extensive range						
			Minimised range						
2. Mandibular			Close jaw						
			Open jaw						
			Protruded jaw		x	✓			
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade		x ✓				
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body	x	✓				
			Raised tongue body	x	✓				
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal			Pharyngeal constriction		x ✓				
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal						
			Denasal	✓					
7. Larynx height			Raised larynx		x ✓				
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract	x					
			Lax vocal tract						
9. Laryngeal tension			Tense larynx			x ✓			
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky				x	✓			
11. Laryngeal frication	Whisper								
	Whispery			✓	x				
12. Laryngeal irregularity	Harsh			x					
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean		High		x ✓				
			Low						
	Range		Extensive range	✓					
			Minimised range						
	Variability		High		x ✓				
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range		Extensive range		x ✓				
			Minimised range						
	Variability		High		x ✓				
			Low						

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXIV: Completed VPAS form for TD2

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD2** Date of recording: **10/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading	x ✓					
			Labiodentalization	✓					
			Extensive range						
			Minimised range	✓					
2. Mandibular			Close jaw		✓				
			Open jaw						
			Protruded jaw	✓					
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade	x					
			Retracted tip/blade	✓					
4. Lingual body			Fronted tongue body						
			Backed tongue body	x	✓				
			Raised tongue body						
			Lowered tongue body	✓					
			Extensive range						
			Minimised range	✓					
5. Pharyngeal	x✓		Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal	x	✓				
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx	x ✓					
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx	✓					
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
				1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x ✓				
12. Laryngeal irregularity	Harsh			x ✓					
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean	x✓	High						
			Low						
	Range		Extensive range						
			Minimised range	x ✓					
	Variability		High						
			Low	x ✓					
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXV: Completed VPAS form for TD1

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD1** Date of recording: **10/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion	✓					
			Lip spreading						
			Labiodentalization	✓					
			Extensive range						
			Minimised range	✓					
2. Mandibular			Close jaw	✓					
			Open jaw						
			Protruded jaw	✓					
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade	x					
			Retracted tip/blade	✓					
4. Lingual body			Fronted tongue body						
			Backed tongue body	✓					
			Raised tongue body	✓					
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal	x✓		Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal	x ✓					
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx	x ✓					
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx	✓					
			Lax larynx						

C. PHONATION FEATURES

		Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
	SETTING			1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x	✓			
12. Laryngeal irregularity	Harsh								
	Tremor								

D. PROSODIC FEATURES

13. Pitch	Mean		High						
			Low	x ✓					
	Range		Extensive range						
			Minimised range	x ✓					
	Variability		High						
			Low		x	✓			
14. Loudness	Mean	x✓	High						
			Low						
	Range		Extensive range						
			Minimised range	✓					
	Variability		High						
			Low	x		✓			

E. TEMPORAL ORGANIZATION

15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXVI: Completed VPAS form for TD5

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD5** Date of recording: **28/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading		x ✓				
			Labiodentalization	x		✓			
	x✓		Extensive range						
			Minimised range						
2. Mandibular	x✓		Close jaw						
			Open jaw						
			Protruded jaw						
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade		x ✓				
			Retracted tip/blade						
4. Lingual body			Fronted tongue body	x ✓					
			Backed tongue body						
			Raised tongue body	x ✓					
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal	x✓		Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx	x ✓					
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension	x✓		Tense larynx						
			Lax larynx						

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
C. PHONATION FEATURES									
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x	✓			
12. Laryngeal irregularity	Harsh			x					
	Tremor								
D. PROSODIC FEATURES									
13. Pitch	Mean		High	x ✓					
			Low						
	Range		Extensive range	x ✓					
			Minimised range						
	Variability		High	x ✓					
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						
E. TEMPORAL ORGANIZATION									
15. Continuity		x✓	Interrupted						
16. Rate		x✓	Fast						
			Slow						
F. OTHER FEATURES									
17. Respiratory Support			Adequate	x✓					
			Inadequate						
18. Diplophonia			Absent	x✓					
			Present						

Appendix XXVII: Completed VPAS form for TD4F

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD4F** Date of recording: **28/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading		x	✓			
			Labiodentalization	✓					
	x✓		Extensive range						
			Minimised range						
2. Mandibular	x✓		Close jaw						
			Open jaw						
			Protruded jaw						
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade	x	✓				
			Retracted tip/blade						
4. Lingual body			Fronted tongue body	x	✓				
			Backed tongue body						
			Raised tongue body	x	✓				
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal	x✓		Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal	x	✓				
			Denasal						
7. Larynx height			Raised larynx	x					
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx	x					
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x ✓				
12. Laryngeal irregularity	Harsh								
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean		High	x ✓					
			Low						
	Range		Extensive range	✓					
			Minimised range						
	Variability		High	✓					
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXVIII: Completed VPAS form for TD7

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD7** Date of recording: **22/02/2008** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS	moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion						
			Lip spreading	✓					
			Labiodentalization						
			Extensive range						
			Minimised range	✓					
2. Mandibular			Close jaw		x✓				
			Open jaw						
			Protruded jaw						
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade		✓				
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body						
			Raised tongue body	x	✓				
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal			Pharyngeal constriction	✓					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx		✓				
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx		x	✓			
			Lax larynx						

C. PHONATION FEATURES									
	SETTING	Present		Scalar Degree					
		Neutral	Non-neutral	Moderate			Extreme		
				1	2	3	4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky			x	✓				
11. Laryngeal frication	Whisper								
	Whispery				x ✓				
12. Laryngeal irregularity	Harsh			✓	x				
	Tremor								

D. PROSODIC FEATURES									
13. Pitch	Mean		High	✓					
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION								
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES		
17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXIX: Completed VPAS form for TD3

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD3** Date of recording: **09/07/2007** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial	x✓		Lip rounding/protrusion						
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range	✓					
2. Mandibular	x✓		Close jaw						
			Open jaw						
			Protruded jaw						
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade			Advanced tip/blade	✓					
			Retracted tip/blade						
4. Lingual body			Fronted tongue body	✓					
			Backed tongue body						
			Raised tongue body	✓					
			Lowered tongue body						
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal	x✓		Pharyngeal constriction						
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx	x					
			Lowered larynx						
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension	x✓		Tense vocal tract						
			Lax vocal tract						
9. Laryngeal tension			Tense larynx	x ✓					
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x ✓				
12. Laryngeal irregularity	Harsh			x ✓					
	Tremor								

D. PROSODIC FEATURES

		Neutral		1	2	3	4	5	6
13. Pitch	Mean	x✓	High						
			Low						
	Range		Extensive range						
			Minimised range	x ✓					
	Variability		High						
			Low	x ✓					
14. Loudness	Mean	x✓	High						
			Low						
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION

	Neutral		1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXX: Completed VPAS form for TD8

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD8** Date of recording: **14/03/2008** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial			Lip rounding/protrusion	x	✓				
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range	x	✓				
2. Mandibular			Close jaw						
			Open jaw	x					
			Protruded jaw	✓					
	x✓		Extensive range						
			Minimised range						
3. Lingual tip/blade	x✓		Advanced tip/blade						
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body	x	✓				
			Raised tongue body	x					
			Lowered tongue body						
			Extensive range						
			Minimised range	x	✓				
5. Pharyngeal			Pharyngeal constriction	✓					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal	x	✓				
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx			x ✓			
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract	x	✓				
9. Laryngeal tension			Tense larynx	x ✓					
			Lax larynx						

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky			x	✓				
11. Laryngeal frication	Whisper								
	Whispery					x	✓		
12. Laryngeal irregularity	Harsh					x ✓			
	Tremor								

D. PROSODIC FEATURES

				1	2	3	4	5	6
13. Pitch	Mean		High						
			Low			x ✓			
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						
14. Loudness	Mean		High						
			Low	x	✓				
	Range	x✓	Extensive range						
			Minimised range						
	Variability	x✓	High						
			Low						

E. TEMPORAL ORGANIZATION

			1	2	3	4	5	6
15. Continuity		Interrupted						
16. Rate		Fast						
		Slow	✓					

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXXI: Completed VPAS form for TD6

VOCAL PROFILE ANALYSIS PROTOCOL

Speaker: **TD6** Date of recording: **01/02/2008** Judge: **Rater 1 (x) & rater 2 (✓)**

	FIRST PASS		SECOND PASS						
				moderate			extreme		
	Neutral	Non-neutral	SETTING	1	2	3	4	5	6
A. VOCAL TRACT FEATURES									
1. Labial	x✓		Lip rounding/protrusion						
			Lip spreading						
			Labiodentalization						
			Extensive range						
			Minimised range	x	✓				
2. Mandibular			Close jaw						
			Open jaw	x ✓					
			Protruded jaw						
			Extensive range						
			Minimised range	x ✓					
3. Lingual tip/blade	x✓		Advanced tip/blade						
			Retracted tip/blade						
4. Lingual body			Fronted tongue body						
			Backed tongue body	✓					
			Raised tongue body	x					
			Lowered tongue body	✓					
	x✓		Extensive range						
			Minimised range						
5. Pharyngeal			Pharyngeal constriction	✓					
			Pharyngeal expansion						
6. Velopharyngeal			Audible nasal escape						
			Nasal		x	✓			
			Denasal						
7. Larynx height			Raised larynx						
			Lowered larynx	x					
B. OVERALL MUSCULAR TENSION									
8. Vocal tract tension			Tense vocal tract						
			Lax vocal tract	x					
9. Laryngeal tension			Tense larynx						
			Lax larynx	✓					

C. PHONATION FEATURES

	SETTING	Present: Neutral	Present: Non-neutral	Scalar Degree: Moderate 1	2	3	Extreme 4	5	6
10. Voicing type	Voice		x✓						
	Falsetto								
	Creak								
	Creaky								
11. Laryngeal frication	Whisper								
	Whispery				x ✓				
12. Laryngeal irregularity	Harsh			✓		x			
	Tremor								

D. PROSODIC FEATURES

		Neutral	Non-neutral	1	2	3	4	5	6
13. Pitch	Mean		High						
			Low	x					
	Range		Extensive range						
			Minimised range				x ✓		
	Variability		High						
			Low		✓	x			
14. Loudness	Mean	x✓	High						
			Low						
	Range		Extensive range						
			Minimised range		✓				
	Variability		High						
			Low	x	✓				

E. TEMPORAL ORGANIZATION

	Neutral	Non-neutral	1	2	3	4	5	6
15. Continuity	x✓	Interrupted						
16. Rate	x✓	Fast						
		Slow						

F. OTHER FEATURES

17. Respiratory Support	Adequate	x✓
	Inadequate	
18. Diplophonia	Absent	x✓
	Present	

Appendix XXXII: Published article: 'Speech production in Down syndrome'

Timmins, C., Cleland, J., Rodger, R., Wishart, J., Wood, S. and Hardcastle, W. (2009) Speech production in Down syndrome
Down Syndrome Quarterly 11 pp 16-22

SPEECH PRODUCTION IN DOWN SYNDROME

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Keywords: Down syndrome, speech development, intelligibility, delay/disorder, phonology, phonetics

Running head: Speech in Down syndrome

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SPEECH PRODUCTION IN DOWN SYNDROME

INTRODUCTION

In comparison to their non-verbal ability levels, people with Down syndrome present with relative deficits in expressive speech and language, and relative strengths in vocabulary comprehension (Chapman, 2006). As part of the difficulty with expressive language, speech is particularly impaired. Speech has been found to be considerably less well developed than levels of cognitive ability would predict, suggesting a specific difficulty in this area.

The speech disorder in Down syndrome is thought to result from impairments in almost all of the systems required for successful speech. In addition to a specific behavioural phenotype (see e.g. Fidler, 2005), people with Down syndrome present with a specific anatomical profile that may affect speech production (Miller, Leddy & Leavitt, 1999; Spender et al. 1995). The oral cavity is smaller than in typically developing speakers (Vorperian, Kent & Gentry, 2004), with a narrow palatal vault and reduced palatal length (Pilcher, 1997; Westerman, Johnson & Cohen, 1974). These features affect the movement of the tongue which is similar in size to typically developing speakers but seems considerably larger in relation to the oral cavity size (Vorperian et al., 2004). It has been widely suggested that these differences may contribute to the reduced intelligibility in speech production (Dodd & Thompson, 2001; Smith & Stoel-Gammon, 1983, Stoel-Gammon, 1997, 2001), although one should be cautious in attributing specific speech disorders to structural abnormalities, due to the well-known ability of the speech production system to accommodate to anatomical changes in the oral region (Bloomer, 1957).

In addition to these anatomical differences, people with Down syndrome also perform poorly in most areas of motor functioning (Frith & Frith, 1974; Spano et al., 1999; Spender et al., 1995). Motor speech difficulties are also common, with hypotonia a possible important factor in reduced intelligibility. Alternatively, problems with motor planning and with the co-ordination of speech movements may suggest an apraxic element to the speech disorder seen in Down syndrome (Kumin, 2006). For example, Barnes, Roberts, Mirret, Sideris, and Misenheimer (2006) found that boys with Down syndrome showed significantly lower levels of lip, tongue, velopharynx, larynx and coordinated speech function than typically developing boys matched for nonverbal mental age, as well as lower levels of coordinated speech movements than boys with fragile X (another common cause of intellectual disability) who were matched for both chronological and nonverbal mental age.

Kumin (2006) found that the majority of children with Down syndrome show signs of childhood apraxia of speech but this disorder is rarely diagnosed in Down syndrome. One way of assessing motor control is using maximum performance tasks, for example repetition of syllables, or sequences of syllables at maximum rate (diadochokenetic rates, DDK). McCann and Wrench (2007) found that DDK rates were not reduced in people with DS but that accuracy, especially of sequences, was greatly reduced. A slow rate of DDK usually indicates dysarthric (neuromuscular difficulty) features, whereas difficulty with accuracy, especially in sequencing, may indicate apraxic-like difficulties (motor planning problems).

Although motor control is problematic in Down syndrome, it is the development of the systematic use of sounds in language, phonological development, that has received the most attention in the literature, with specific attention directed at whether this is delayed (i.e. following the same pattern of development as in typical speakers but at a slower rate and with a lower developmental ceiling) or is disordered (i.e. following an idiosyncratic developmental pattern, at odds with typical speakers). Research findings are still equivocal on this issue (e.g. Stoel-Gammon, 1980; Van Borsel, 1996, Roberts et al., 2005). This distinction is not only of theoretical relevance. It is also important in relation to the planning of intervention as the course and content of therapy for a given individual may be determined depending on whether speech appears to be delayed or disordered. A disordered profile may also suggest that spontaneous improvements are less likely and that specific interventions may have to be designed.

Considering the anatomical and motor planning difficulties faced by this population, the actual articulation (phonetics) of speech would also be expected to be affected. Studies of phonology provide valuable information on speech development but much more detailed phonetic analysis of speech production is required in order to assess and treat the speech and voice problems experienced by many children with Down syndrome more effectively. Research findings in both of these keys areas are presented below, starting from pre-linguistic development and first word stages through to adult speech production.

PRE-LINGUISTIC DEVELOPMENT

Speech production is preceded by an important period of pre-linguistic development. According to some researchers (Dodd, 1972; Smith & Oller, 1981), aspects of pre-linguistic development, such as babbling or acquisition of segments (speech sounds) are neither atypical nor delayed. For example, Dodd found no differences in number of utterances, length of utterances, variety of segments produced in the babbling stages by infants with and without Down syndrome aged 9 to 13 months. Similarly, Smith and Oller also reported "substantial similarities" (p.46) in the production of reduplicated babbling (repetition of syllables) in infants with and without Down syndrome. However, Lynch, et al. (1995), who analysed the age of babbling onset in 13 infants with Down syndrome over a wider age range (0;4 to 1;6 yrs) and in 27 age-matched typically developing infants, found that the infants with Down syndrome showed no signs of babbling until 9 months of age; in contrast, the mean onset of babbling in the typically developing infants was around 6 months of age. They also found that the production of babbling was unstable in their infants with Down syndrome, suggesting motor control difficulties at even this early stage of vocal development. In a longitudinal study, Steffens, Oller, Lynch and Urbano (1992) reported similar developmental patterns in age-matched infants with and without Down syndrome aged 4 to 18 months, although they acknowledged that the spacing between their recording intervals may have been too long and that larger data samples should have been recorded in order to allow a more in-depth analysis of potential developmental differences in the two groups of infants.

More detailed work in this area by Smith and Stoel-Gammon (1996), however, found that the development of particular types of pre-linguistic babbling, reduplicated (e.g. 'bababa') versus variegated (e.g. 'bada') babbling, among infants with Down syndrome between 6 months and 2 years of age was similar to that previously reported among typically developing age-matched infants. They also found no age related differences in phonetic complexity in their infants with Down syndrome, a developmental pattern similar to that reported in typically developing infants in one of their earlier studies (Smith, Brown-Sweeney & Stoel-Gammon 1989).

In sum, research on pre-linguistic development in infants with Down syndrome currently includes contrasting findings. There is some evidence that, in both form and content, babbling emerges as in typical development and is neither delayed nor disordered. There is also, however, some conflicting findings suggestive of a delay in the emergence of early babbling in infants with Down syndrome, along with some evidence of disorder in the underlying motor control system.

FROM FIRST WORDS ONWARDS

Phonological development: evidence for delay

Lenneberg (1967) interpreted the speech patterns of people with Down syndrome as indicating a delay in phonological development. This was supported in subsequent studies. For example, Stoel-Gammon (1980) analysed speech production from the spontaneous speech of 4 young children with Down syndrome (3;10, 5;6, 6;1 and 6;3 yrs) and found that they were able to produce all of the consonants of English but not necessarily in the correct word position. Production patterns were similar to those seen in normal phonological development, leading Stoel-Gammon to conclude that development was similar to that of typically developing children, but at a slower rate. Bleile and Schwarz (1984) also assessed phonological aspects of the speech production of three children with Down syndrome, aged 3;4, 3,6 and 4;6, and concurred with Stoel-Gammon's view that the children followed a delayed but similar pattern of development to typically developing children.

Smith and Stoel-Gammon (1983) likewise found parallels with typical developmental processes when comparing the development of consonants in 5 children with Down syndrome (followed from 3 to 6 years) and 4 typically developing children (followed from birth to 3 years). They found similar patterns of production between the two groups in the development of the consonants /p t k b d g/. For example, at the beginning of words these consonants were produced with more accuracy and frequency than when in word-final position, although the children with Down syndrome produced more errors in both cases.

Smith and Stoel-Gammon (1983) also looked at four developmentally important phonological processes (final stop devoicing, initial stop de-aspiration, final stop deletion and initial stop cluster reduction), all of which are integral to successful speech production and effective communication. They found that by the age of 3 years, typically developing children showed adult-like abilities (very low frequency use) of final consonant deletion and initial stop de-aspiration, but initial cluster reduction and final stop devoicing were less adult-like in both the children with Down syndrome and their typically developing peers at this same stage of their early linguistic development. However, there was a large difference between the groups in the length of time it took for the next developmental step to occur, that is, the children with Down syndrome showed some evidence of delayed, but not, disordered development. The typically developing children gained more adult-like productions at a rate of 38% per year, whereas the children with Down syndrome became adult-like in only 6% increments, showing an overall 4 year difference in achieving this same level of proficiency. On the basis of these findings, Smith and Stoel-Gammon concluded that phonological acquisition is delayed for children with

Down syndrome, although they pointed out that their sample was small, both in respect to the linguistic variables examined and the number of children studied.

Van Borsel (1996) directly addressed the question of delayed versus disordered development in Down syndrome, aiming to analyse areas of phonological development which had hitherto been neglected (such as vowel production and consonant distortions). This study involved 20 adolescents and young adults with Down syndrome (15;4 to 28;3 yrs) and at typically developing children (2;6 to 3;4 yrs), selected on the basis that they had not yet fully acquired all aspects of speech production. Many errors were found to be similar in the two groups, with consonant and vowel errors especially similar, but with more errors being made by the speakers with Down syndrome.

Phonological development: evidence for disorder

There are a number of additional studies that suggest that phonological development in children with Down syndrome is not simply a case of delay, especially with regard to speech. For example, Kumin, Councill and Goodman (1994), analysed the emergence of consonants in the speech production of 60 children with Down syndrome, aged 9 months to 9 years, and found that the order of acquisition of phonemes did not follow the same pattern as in typical development, with, for example, /sh/ (the initial sound in 'sheep') appearing earlier than /f/ (the initial sound in 'feet').

There is further evidence of possible disorder in terms of increased variability and inconsistency in the speech of children with Down syndrome. For example, Dodd and Thompson (2001) compared speech production in children and adolescents with Down syndrome, aged 5 to 16 years, and in a group of children with inconsistent phonological disorder, aged 3 to 6 years, who were matched for severity of speech disorder on the basis of the percentage of consonants produced correctly within single words. They found that all of the children with Down syndrome were inconsistent in their productions of the target words, with a mean inconsistency score which was very similar to that found in the children with inconsistent phonological disorder (67% in the group with Down syndrome and 62% in the group with inconsistent phonological disorder). Previous evidence (Dodd, 1976 and 1995 Burt, Holm and Dodd, 1999) indicated that a third group of children with a straightforward delay in phonology had inconsistency ratings of less than 20%, suggesting that the inconsistency in DS is not due to delay. Moreover, inconsistency ratings in typically developing speakers were only 10%. This led them to suggest that the inconsistency in speakers with Down syndrome was not simply due to a delay in development but that it might stem from different causes, such as underspecified phonological representations or differences in language learning environments.

Further evidence for disorder in speech development comes from a large-scale study in which Roberts et al. (2005) analysed speech production in detail, including the accuracy of consonant production and the use of differing phonological processes (see above) among boys with Down syndrome aged 3 to 14 years, typically developing boys of the same age, and boys with fragile X syndrome similar to the Down syndrome group in chronological and cognitive age. For percentage consonants correct, they found a significant difference between boys with Down syndrome and the typically developing boys. While some developmentally appropriate substitution processes were used at similar rates across the two groups (e.g. velar fronting, fricative simplification), other processes were being used much more frequently by the boys with Down syndrome (e.g. final consonant deletion, consonant cluster reduction). A higher number of atypical phonological processes were also found, including lateralization of sibilants, de-affrication and deletion of nasals. These findings are evidence for both a delay and a disorder in speech development in Down syndrome.

A recent study by Cleland et al. (in press) concurs with the idea of a picture of mixed delay and disorder. In their study of 15 children and adolescents with Down syndrome, 66% of phonological processes were developmental in nature, suggestion mostly delayed acquisition. However, all of the children presented with at least one atypical or non-developmental speech error. When this occurs in other groups of children with speech impairments the diagnosis is usually one of disorder rather than delay.

Overall then, the evidence on phonological development indicates that speakers with Down syndrome display slower and more variable speech, with a higher incidence of atypical speech errors (disordered) than is found in typically developing speakers. Consistent with an earlier review of this field by Stoel-Gammon (1997), the findings described above suggest patterns of both delay and disorder in the phonological development of children with Down syndrome.

Phonetic aspects of speech development: evidence for delay

While phonological analysis of speech development provides important information regarding the onset of speech sounds in Down syndrome (and their order of acquisition), phonetic analysis can provide a more detailed picture of the actual production of these speech sounds. Phonetic analysis of speech development in children with Down syndrome is still a relatively small area of research but one that has been gathering momentum in recent years. Most of the research finds atypical features of speech articulation that would suggest a disordered pattern but there is some work that finds evidence of typical patterns of speech production.

In a study of phonetic and phonological aspects of speech production, Van Borsel (1988) found that 5 Dutch girls with Down syndrome ranging in age from 16;5 to 19;9 years were able to produce all of the speech sounds that would be expected in their native language. Although he did not examine the actual articulation of the speech sounds that were produced, he provided useful information on the phonetic inventory of the participants. He reported similar phonetic distortions in both the participants with Down syndrome and a group of very young typical children (aged 2;6-3;4) suggesting delay. However, additional distortions were found in speakers with Down syndrome, including ‘denasalisation’, ‘dentalisation’ and ‘wet’.

Phonetic aspects of speech development: evidence for disorder

Segmental aspects of speech production: consonants and vowels

Many phonetic studies of speech in Down syndrome include acoustic analysis techniques as they allow a much more detailed look at speech production and can be useful in assessing whether speech is delayed or disordered. For example, Callahan Mandaluk, Zajac, Harris, Roberts and Cox (2006) used acoustic analysis to investigate duration and spectral qualities of fricative sounds (e.g. /s/ as in ‘sun’ and /z/ as in ‘zoo’) produced by children with Down syndrome aged 6;3 to 15;11 years, chronologically age-matched typically developing children, and children with fragile X syndrome. They found that the distinctions in duration between /s/ and /z/ patterned differently for children with Down syndrome. For example, /z/ was longer in duration than /s/, but this pattern was reversed in both the typically developing children and in the children with fragile X syndrome. In contrast, the acoustic analysis of /s/ and /sh/ revealed that the production of these two sounds was similar in the children with Down syndrome and the children with fragile X syndrome but was different in the typically developing children, emphasising the need to use a range of measures when comparing speech outputs across different child populations.

Technological advances in the analysis of speech are proving useful for providing more fine grained insights into the nature and range of speech difficulties in children with Down syndrome. For example, the technique of electropalatography (EPG) has been used to record the timing and location of tongue-palate contact with great precision. It utilises a custom-made artificial palate contacting 62 electrodes and allows observations to be made of any fine phonetic differences that may exist in the speech output of people with a range of speech disorders. To date, EPG has not yet been widely used in studies of speech development in Down syndrome but it could prove to be highly useful in identifying profiles of speech productions in this population and, by providing more detailed information, particularly helpful in assessing whether the speech difficulties shown are delayed or disordered in their characteristics. EPG enables the detection of subtle differences in speech production, some of which may not be apparent from perceptual and acoustic analyses alone.

In one EPG study, the first carried out with speakers with Down syndrome, Hamilton (1993) found greatly increased tongue/palate contact for oral and nasal sounds in three young adults with Down syndrome (aged 17, 17 and 20 years) when compared to a typical adult speaker. She suggested that this increased tongue contact was possibly due to the size of the tongue in relation to a small oral cavity, implicating anatomical difference in the speech production deficits of this population. Timmins et al. (2007) also found high levels of variability in the production of /s/ and /sh/ in 6 children with Down syndrome aged 10 to 18 years. They concluded that this increased variability could result from an immature motor system, suggesting a delay in motor control. Together these studies suggest that the speech disorder in Down syndrome goes beyond a difficulty in phonological acquisition, with differences in anatomy and motor control leading to subtle phonetic differences that can be measured using instrumentation such as EPG.

In a study using EPG therapeutically, Gibbon et al. (2003) presented a case study in which EPG was successfully used to treat velar fronting in a 10-year-old child with Down syndrome. Velar fronting is common in younger typically developing children, suggesting that the child in this case study had delayed phonology. However, during EPG-based treatment, double alveolar/velar articulations were observed in the productions of targets /t/ and /k/, indicating that the child was aware of two distinct sounds, and had attempted to produce them differently, but had done so in an atypical way. These errors would not have been identified without EPG analysis of her changing speech patterns.

Further evidence of the potential utility of EPG in the assessment and treatment of intelligibility problems in Down syndrome is reported by Wood et al.

(2009) in their case study report of two children with Down syndrome (11;7 and 14;11 years) who had participated in an ongoing, longitudinal study of speech motor difficulties in children with Down syndrome aged 8 to 18 years. Following 12 weeks of EPG-based therapy, despite having very different phonetic and phonological profiles, both children had made objectively-measurable advances in their speech, although some intelligibility issues still remained (e.g. in speech rate and voice - see below). The visual supplementation of auditory feedback provided by EPG seemed especially effective with these two children, with gains maintained post-therapy and in the absence of EPG feedback.

Non-segmental aspects of speech production: Voice quality

A high incidence of voice disorders has long been reported in Down syndrome (Strazzulla, 1953; Schlanger, 1962; Tredgold & Soddy, 1963; Blanchard, 1964; Benda, 1965; Novak, Sedlackova, Klajman & Betlycwski, 1967) yet voice quality has not received the same level of investigation as other speech and language domains.

Voice quality in Down syndrome is affected by both structural and functional vocal tract anomalies which vary between individuals. Structural differences, such as a constricted pharynx (Jacobs, Gray & Todd, 1996), a smaller larynx (Venail, Gardiner & Mondain, 2004), and under-developed maxilla contributing to relative macroglossia and a tendency to constriction of the tongue body at the palate (Beck, 1997), are clearly life-long constraints contributing towards disordered voice production. However improvements in the functioning of the vocal apparatus, for example, through decreasing hypotonia with age (Penrose & Smith, 1966), might allow for a degree of change with development. Equally, early attention to hearing impairment, which is considered to affect ability to self-monitor vocal output (Montague & Hollien, 1973; Montague, 1976), and to the development of appropriate strategies to encourage vocal efficiency and minimise vocal abuse

(Pryce, 1994), may help to encourage the development of more typical voice patterns.

Studies of voice quality in Down syndrome have primarily been focused on phonation type, nasal resonance and pitch characteristics.

With regard to phonation type a variety of terms reflecting the perceptual features of harsh and whispery voice are frequently reported, including ‘harsh with a wheezy admixture’ (Novak, 1972), ‘rough and breathy’(Montague & Hollien, 1973), and ‘deep, with harsh and hoarse quality’ (Bolfan-Stosic& Hedever, 1999). These perceptual features correlate with more objective acoustic analysis findings of vocal fold irregularities: increased frequency and intensity perturbation (jitter & shimmer) and increased noise in the acoustic signal (represented by lower harmonic-to-noise ratio) in comparison to the general population (Moura et al., 2008).

There is some evidence of perceived differences in nasal resonance, but the findings are not entirely consistent. Although hypernasality is identified in some subjects by experienced raters of voice, these features are not always reported in perceptual studies using inexperienced listeners (Montague & Hollien, 1973; Pentz & Gilbert, 1983; Rolfe, Montague, Tirman, & Vandergrift, 1979), suggesting that nasal resonance disorder is less salient perceptually in Down syndrome than phonation type.

Perceptually, pitch has long been reported as being perceived to be lower in persons with Down syndrome than in typically developing persons (Benda, 1949; Strazzula, 1953). Despite this, acoustic analysis has revealed no significant differences in the level of fundamental frequency (F0) of English-speaking, school-aged children with Down syndrome and their typically-developing peers (Hollien & Copeland, 1965; Michel & Carney, 1964; Montague, Brown & Hollien, 1974; Pentz & Gilbert, 1983). Higher F0 levels have been found in pre-school children with Down syndrome (Weinberg & Zlatin, 1970). It has been suggested that that F0 levels are naturally higher in very young children with Down syndrome, and these levels tend to decrease at around five to six years of age, bringing the F0 into line with typically developing peers (Montague, Brown & Hollien, 1974). However, the opposite pattern of lower F0 was found in young Portuguese-speaking children with Down syndrome (Moura et al., 2008), suggesting that some differences may be language-specific. Higher than typical F0 levels have also been identified in adults with Down syndrome (Lee, Thorpe, & Verhoeven, in press; Moran & Gilbert, 1978); reduced laryngeal growth during puberty in comparison to typically developing populations and improvements in muscle tone are considered to be key factors (Moran & Gilbert, 1978).

Rhythmic and timing aspects of speech production

Heselwood, Bray and Crookston (1995) analysed the timing of speech production in a conversation sample of an adult male with Down syndrome in order to establish whether there was a relationship between production errors and prosodic elements of speech (e.g. rhythm, timing, stress, intonation, etc). They noted a relationship between speech articulation and prosodic structure, with the least errors found in the pre-pausal groups with the stressed syllable. Heselwood et al. also found that the same words were produced both successfully and unsuccessfully throughout the conversation. They discount the suggestion that this may be a case of intra-speaker variability as the words produced correctly always appeared near the end of a prosodic unit (where there was no stressed syllable between the end of the word and the next pause). The number of syllables in a phrase also effected the disordered production of speech (with less syllables equating to better speech production).

The Heselwood et al. study is a good example of the potential advantages of phonetic analysis in developing understanding of the precise nature of speech articulation in Down syndrome. A subsequent study with six adult speakers with Down syndrome (Flipsen, 1999) usefully built on these findings, demonstrating that intelligibility (as assessed from an intelligibility index involving more than segmental accuracy), was increased in the pre-pausal position but that consonant and vowel production was unaffected by prosodic structure.

Brown-Sweeney and Smith (1996) also used acoustic techniques to analyse speech timing and speech precision in two groups of 8 children with Down syndrome (aged 6;8 to 7;9 years and 11;10 to 12;10 years) and in a group of age-matched, typically developing children. They noted significant differences between the children with Down syndrome and the typical developing children, in variability, articulatory accuracy and syllable repetition rate, but not in segment duration.

The phonetic studies described briefly above have been able to provide much more detailed information on the nature of speech production in Down syndrome than was previously available. Phonetic approaches also highlight the differences in the speech disorder present in Down syndrome with most studies providing evidence for disorder in this area. In combination with the findings from the many phonological studies of speech production, a clearer picture is forming about the speech difficulties that exist in this population.

CONCLUSION

The speech difficulties associated with Down syndrome are thought to result from impairments in almost all of the systems required for successful speech production. Diagnoses of dysarthria, apraxia and phonological delay/disorder can all be argued for, but it is not clear which should be the primary diagnosis. This makes it extremely difficult to design interventions to improve speech in persons with Down syndrome.

In this review, we examined whether, speech in Down syndrome is predominantly delayed in its development or whether it is better described as being more fundamentally disordered in nature. Speakers with Down syndrome vary considerably with regard to their phonetic and phonological abilities and speech profiles, with this likely to be a direct result of the wide range of structural, anatomical and motoric difficulties inherent to Down syndrome. As Stoel-Gammon (1980) highlighted, any analysis of speech development in Down syndrome always needs to consider the very wide individual differences that are found when analysing at group level.

Findings from studies to date are broadly in agreement that phonology in Down syndrome follows a pattern consistent with delayed speech development, but also that some aspects of this development can be disordered. Detailed work into phonological processes has shown that children with Down syndrome use many phonological processes which are common in normal development but that these co-exist alongside atypical processes, such as de-affrication and lateralization of sibilants (Roberts et al, 2005).

The phonetic abilities of people with Down syndrome are characterized by atypical patterns of articulation (Hamilton, 1993), high levels of articulation variability (Timmins et al., 2007), rhythmic and timing differences (Brown-Sweeney and Smith, 1996; Heselwood et al., 1995) and pitch differences (Moura et al., 2008). All of these findings suggest that the speech difficulties associated with Down syndrome may require much more detailed analysis if the more disordered aspects of speech output are to be detected. Instrumental approaches may also help to pinpoint the cause (or causes) of the low levels of speech intelligibility found in this population.

Speech development in Down syndrome is clearly a complex area. Future research should be focused on more detailed analyses of the speech production of speakers with Down syndrome, making greater use of the more objective speech analysis techniques which are currently available. Only then will it be possible to identify the origins and precise nature of the speech difficulties experienced by most children with Down syndrome and to treat these with interventions that are appropriately, and if necessary, individually designed.

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