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To what extent can an analysis of primary research studies identify the challenges faced by a Down Syndrome singer and contribute to improving vocal training for this demographic

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Part 1: Project Proposal

Context

Within my practice as a freelance vocal coach, I tutor students who are impacted by Down Syndrome ("DS"), a neurogenetic aneuploid disorder (Gardiner et al.,2010) which adversely affects the operation and quality of the voice (Michel and Carney,1964; Montague et al.,1978; Groves and Gray,1985; Moran,1986; Pentz and Moran,1988; Pryce, 1994; Albertini et al.,2010; Krishnamurthy and Ramani,2020), as well as the singing experience, of those students (Pryce, 1994). As a vocal tutor operating a fully inclusive studio which embraces diversity, I am committed to teaching beyond the methods and practices commonly employed by vocal tutors to support neurologically and physically able students, and, to this end, I continue to seek additional and better methods of tutoring singers who face unique challenges, including singers with DS.

Justification

DS is the most frequently occurring autosomal disorder across the Globe (MacLennan, 2019) with an incidence rate of 1 in 1,000-1,1000 births worldwide. (Samuel et al, 2014; Chen et al, 2022). Singing has both physiological and psychological benefits for the general population (Stacy et al, 2002; Keeler et al., 2015) and the proven positive impact of singing, upon matters such as respiratory health (Stacy et al., 2002; Irons et al.,2010;Clift, 2009;Lewis et al.,2016); speech-motor issues and dysphagia (Wan et al, 2010;Stegemoller et al, 2017); immune system activity (Kreutz et al., 2004; Beck et al.,2006); physiological pain management and control (Hopper et al, 2016) mood enhancement and mental wellbeing (Grape et al., 2002; Clark and Harding, 2012) and quality of life (Stegemoller et al, 2017) are very relevant to individuals with DS, who experience these and other issues at least as frequently, and more significantly, than their able-bodied contemporaries (McGuire and Defrin,2015; Rivelli et al.,2022). Thus, singers with DS should be supported with their singing activities and efforts, not least, so that they can share in the positive benefits of singing as proven by primary studies.

Aims, Objectives and Research Method

My earlier research during this Master's study has revealed a dearth of literature specifically dealing with the impact of DS upon a singer's voice. It is submitted that this lack of information negatively impacts upon, and discriminates against, singers with DS, and an important aim of this project is to contribute to research which will improve vocal tuition and equality for DS singers in the teaching studio. One of my objectives in this regard is to present a review which "analyses and synthesizes an existing body of literature by

identifying, challenging, and advancing the building blocks of a theory through an examination of ... prior work” (Post et al., 2020, p.352) and to present a standalone piece of work from which I can enhance my knowledge and understanding and more easily identify gaps in the body of literature that currently exists (Kraus et al, 2022). Despite the lack of directly relevant studies and materials either in the field of vocal pedagogy or in other disciplines, one of my objectives is to conduct this literature review by searching beyond what is apparently not there, move beyond the “box” of safe research (Bannister, 2023, p.2) and attempt to fill some of the gaps in the literature with a comprehensive, multi-disciplinary search of primary research materials. The overall aim of this project is to improve vocal tuition for singers with DS and to this end, one of my objectives is to further refine and adapt my own teaching practices and to share the knowledge I have obtained, over the course of 3 inter-related projects exploring the impact of DS on the voice, through publication.

The success of initiatives I have implemented in my studio, following my previous research on topic, has inspired me to conduct a further, more comprehensive multi-disciplinary literature review, following a tighter methodology and framework (hermeneutics will be applied), utilising tools such as thematic coding, to extend my understanding and search for answers. Accordingly, this current literature review will take the form of a qualitative systematic review, referred to in the Cochrane-Campbell Handbook, Chapter 21, as a Qualitative Evidence Synthesis (QES) (Noyes et al., 2021), which synthesises findings from multiple qualitative studies. This method can identify patterns in data, classify similarities and differences, translate and transfer findings and provide richer, more interpretive data than that found in a single study (Bearman and Dawson, 2013; Booth, 2016; Downe et al. 2019).

More specifically, I intend to carry out this literature review in the following manner:

- identifying as many directly relevant or indirectly relevant primary research studies as possible, by searching across numerous academic databases, including PubMed, ScienceDirect, JSTOR, MEDLINE, Google Scholar, DOAJ, Scopus, ProQuest, Ovid. Literature will be selected depending upon its relevance to my enquiry (including studies undertaken outside of the field of vocal pedagogy which nevertheless explore issues which may impact upon vocal production in DS singers) and the rigor of investigations and data collected in the studies reported. I will use the systematic methods promoted by the Campbell group and the Cochrane collaboration to create a robust inclusion/exclusion criteria.
- reviewing the primary research studies selected, using a Braun and Clarke based thematic analysis to code my data and extract themes, thereby producing a transparent analysis which discloses my selections, decisions and reasons

therefore, enabling evaluation of my research by the reader (Braun and Clarke, 2006). My coding will follow an inductive approach, namely, my descriptors will not be fixed beforehand as is the case with deductive coding, but rather will emerge from my reading and analysis of the literature (Kraus et al., 2022). The procedure adopted will commence with the extraction of important content from titles, abstracts, and keywords of studies and articles, followed by classification of this content to create a shorter list of core descriptors, and subsequent revision of my code book, as appropriate, to combine patterns and develop a shortlist of descriptors/themes (López-Duarte et al., 2016; Dabić et al., 2013 ; Vlacic et al., 2021).

Ethical Considerations

Although my role, experience and intentions as vocal tutor to DS students are declared, so that my position as a researcher is transparent, this work is confined to a comprehensive review and analysis of published literature, to enhance my understanding of why vocal challenges occur within this demographic. I hope to publish my own research as a vocal teacher to DS students in the future (when my extensive and ongoing research on this topic is closer to completion), however, I will not currently be sharing any personal teaching experiences or private student data, and have taken care to preserve student anonymity, even in so far as avoiding disclosure of the number of DS students I teach, so that no student can possibly be identified by any part of this report.

I anticipate that the cross-disciplinary research required for this work will expose me to data and information beyond my field of expertise, and I will endeavour to neutralise any risks to myself, my students, Voice Study Centre and UWTSD, by thorough review of unfamiliar concepts, seeking expert guidance as necessary (e.g. regarding complex medical data) and will not make assumptions where a matter is outside my own experience or knowledge. I will use evaluative frameworks for my research and be transparent regarding my selection of search terms, to enable the reader to assess the depth of my study, as they determine the value of my work. I will check that each study I discuss, or offer as proof of any issue or concept, is accurately cited, so that the reader can review those works independently, and I will ensure the studies I rely upon and cite have not been retracted. (Kara, 2019). I will conduct myself professionally as I research, and will neither plagiarise nor self-plagiarise, showing honesty and respect for other authors and avoiding repetition of any errors which may exist in other works (Kara, 2019). I will take steps to avoid bias by reviewing all studies equally, and will not discuss or rely upon any study, nor part of any study, selectively, to reach a conclusion I might anticipate or desire (Hosseini, 2020). At all times, I will maintain high standards of research so that my research can be used by others (Kara, 2019).

Finally, I will consider the ethical implications of applying knowledge obtained from my literature review to my practice, identifying, evaluating and explaining how I will mitigate any ethical risks that may arise by my future application.

Personal Learning Outcomes

By completing this work I hope to:

- enhance my existing knowledge and understanding of the physiological challenges faced by DS singers;
- acquire sufficient knowledge to create further teaching resources, such as more focussed and appropriate vocal exercises, to support my DS students;
- acquire knowledge and understanding that can be shared professionally, so as to promote greater awareness of the importance and possibilities of singing tuition for DS individuals.
-

Timeline

The Gantt chart at Figure 1 below sets out a timeline for implementation of this project and initiatives post-submission.

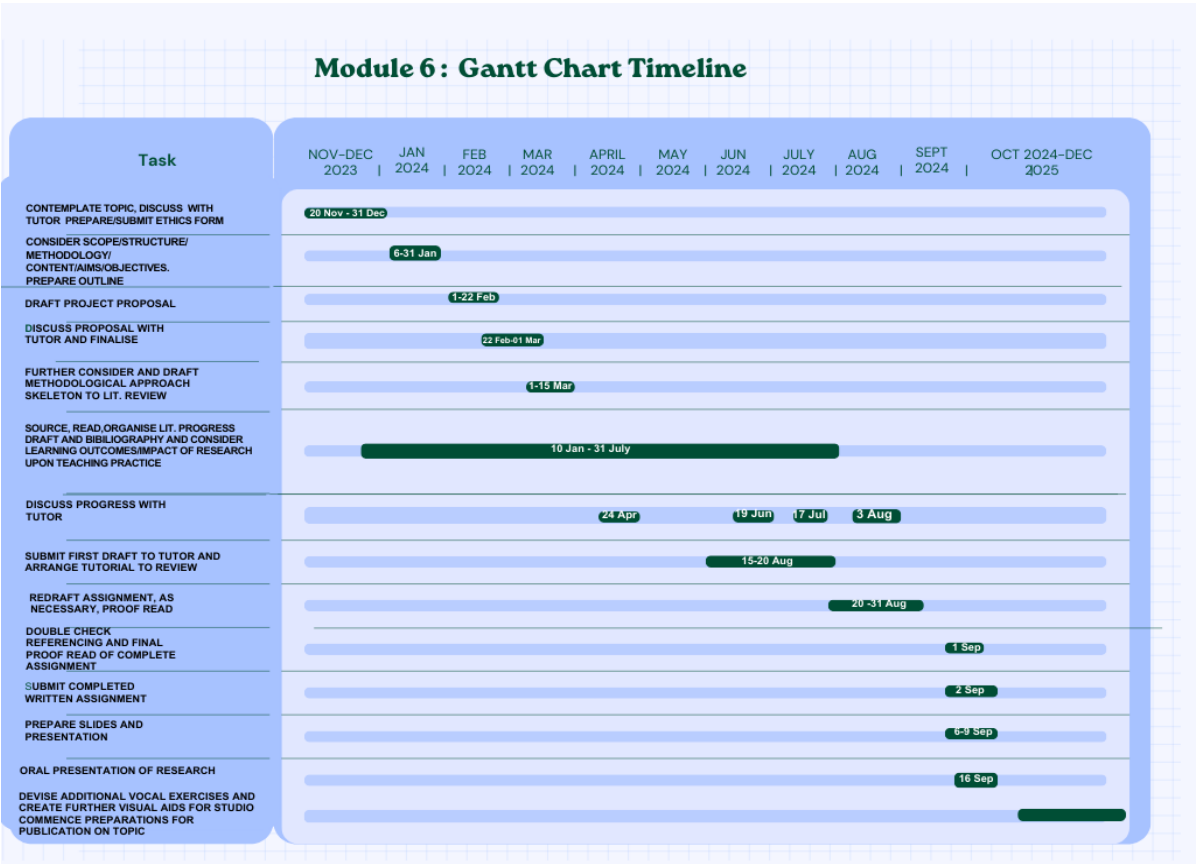


Figure 1: Timeline of Project and Ongoing Implementation Initiatives Post-Completion

(Proposal 1496 words)

Part 2: Project Report

Context and Reasons for This Research

This work enhances and consolidates previous research undertaken on this Master's course to identify and understand the challenges faced by Down Syndrome singers, with a view to improving the vocal training and experiences in the teaching studio of this student demographic. The information acquired from research previously undertaken during this course of study facilitated some understanding of the challenges and physiological issues faced by singers with DS, but also revealed a lack of literature concerning the impact of DS upon the singing voice (in contrast to the literature available on the impact of DS upon speech/spoken voice) and this highlighted the need to carry out a much broader cross-disciplinary review, carried out in a structured and systematic manner, to progress research on this important topic. A systematic and well conducted review as a research method creates a solid base for advancing knowledge and facilitating theory development (Torraco, 2016) and this further literary review is undertaken in the hope that it will reveal further useful information that can be used by vocal teachers to support singers with DS.

Methodology

This literature review will take the form of a qualitative systematic review, referred to in the Cochrane-Campbell Handbook, Chapter 21, as a Qualitative Evidence Synthesis (QES) (Noyes et al., 2021), and will synthesise findings from multiple qualitative studies so as to obtain richer, more interpretive data than might be found in a single study (Bearman and Dawson, 2013; Booth, 2016; Downe et al., 2019). The QES format has been chosen in light of the author's discovery, through previous research on this general topic, of an absence of literature addressing the singing voices of individuals with DS (unlikely to have been filled since previous research was undertaken), indicating the necessity of a comprehensive multi-disciplinary review to progress previous research, synthesising findings from multiple studies across multiple disciplines. This method may facilitate answers to the enquiry raised, if not directly, then potentially via analogy and deduction, and will confirm the current position as regards the existence of literature that has specifically investigated the DS singing voice.

The review is undertaken with a pragmatic lens, considering the practical application of research findings, rather than focussing on purely theoretical issues. The aim of the review is not only to identify and explain vocal challenges of DS singers and the causes, but also to identify strategies or interventions that might effectively address these challenges, so as to enhance vocal tuition of this student demographic. All data contained within any literature, printed, or online, that falls within the inclusion parameters will be considered and assessed

and will form the basis of practical application to the author's teaching practice. Pragmatism is consistent with the hermeneutical methodology that will be applied to this research to guide its collation and presentation. Both pragmatism and hermeneutical philosophy emphasise action as a mode of interpreting experience (Heelan, 1998; Zambas et al., 2015; Breitenwischer and Keiling, 2020) and application of hermeneutics to a QES can enhance research credibility by showing a philosophical depth to the QES process (Winter, 2023).

Thus, a hermeneutic approach was taken to this literature review, along the lines described by Boell and Cecez-Kecmanovic, (2014), where the review was not a linear process (Boell and Cecez-Kecmanovic, 2010; Wolfswinkel, 2013) but rather an interactive and iterative process, sometimes reaching beyond the initial enquiry, aiming for saturation of understanding before closing the literature search (Combs, 2010, p.5). To this end, the two major, intertwining hermeneutic circles proposed by Boell and Cecez-Kecmanovic (2014), the "search and acquisition" circle and the wider "analysis and interpretation" circle were adopted.

Inclusion criteria:

- (1) studies relating to the speaking voice of DS participants, to search for any common issues or challenges potentially also affecting the singing voice;
- (2) studies which appear helpful to the enquiry, regardless of the number of participants or the age range of those participants;
- (3) all relevant literature, without limitation as to publication date;
- (4) studies relating to other disorders or disabilities where there is overlap in the features of these conditions and those of DS, impacting upon vocal production or performance;
- (5) studies reviewed in the author's previous research on this topic, although previously reviewed works will be re-read and reviewed as thoroughly as any new studies, so as to avoid any familiar literature being treated with priority or inappropriate significance or any other form of bias (Schwind and Buder, 2012).

Exclusion Criteria:

- (1) studies identified as potentially relevant upon input of search terms, but not written in English and without an English language translation;
- (2) literature relating to people with DS, but which is focussed on features of DS that do not impact upon vocal production or performance (e.g., common gastro intestinal issues or facial features);

- (3) literature discussing characteristics in healthy singers also encountered by DS singers, but for irrelevant reasons that would not progress the research enquiry, for example the cause of temporary breathy tone (also a characteristic of DS voice) in healthy adolescent singers;
- (4) studies with significant methodological flaws, such as those with weak or no control groups, or those which have little or no evidence based information and/or offer insufficient data to enable critical analysis;
- (5) blogs, articles, websites and other non-peer reviewed literature.

For the purpose of framing the research question and determining inclusion and exclusion criteria, methods proposed by the Cochrane Collaboration and the Campbell group were adopted. The Cochrane Collaboration is an international organisation that aims to support decisions relating to health-care with its library of regulated systematic reviews (Scholten et al., 2005). Inclusion of systematic reviews within the Cochrane library requires reviews to have been undertaken using explicit and systematic methods which minimise bias and lead to the development of high-quality clinical practice guidelines which can safely be used by clinicians and policy makers within the health care system (Korfitsen et al., 2022). The Campbell Collaboration, on the other hand, was established in 2001 to promote positive social and economic change (Li et al., 2023) and it created a library of systematic reviews in alignment with the protocols of the Cochrane Collaboration, additionally highlighting the importance of undertaking searches for unpublished information to avoid publication bias, and considering studies from all around the world, not just from limited jurisdictions.

The author has taken principles from both Campbell and Cochrane into account, such as avoiding bias, transparently reporting and explaining the inclusion and exclusion criteria and considering the principles of the PICO framework, “Patient, Intervention, Comparison, Outcome” (Eriksen, 2019) when determining the research question and developing inclusion and exclusion criteria relevant to the enquiry. Accordingly the research question and the introduction to the work clearly describe the “patient” (singers with DS) and the “intervention” that the author is seeking to facilitate (namely, better vocal sound and training for DS students through intervention, such as targeted vocal exercises, inspired by the research) and also highlight the “comparison” or alternative (by setting out the common limitations faced by singers with DS), and the desired “outcome”, which is clearly stated throughout the work as one which leads to an improvement of the vocal training, vocal sound and learning experience of DS singers.

This literature review has traversed multiple disciplines and, despite the limitations imposed by the inclusion and exclusion criteria, has required an initial perusal of several hundred articles, to establish relevance to the specific enquiry. The review commenced with the input

of a set of author selected search terms into multiple databases, including Google Scholar; Pubmed; Journal of Voice; Frontiers in Human Neuroscience; Elsevier Science; My Bib and Mendeley. The literature that was revealed and reviewed after the first input of search terms, highlighted issues leading to the selection and input of a second round of search terms, and this layered process was repeated several times, as indicated in the table at Figure 2 below.

First Input of Search Terms			
Down Syndrome singers	Down syndrome voices	Down Syndrome +singing	Down Syndrome+ voice
Impact of Down Syndrome on voice	Impact of Down Syndrome on singing voice	Vocal Challenges of Down Syndrome singers	Speech +Down Syndrome
Phonation + Down Syndrome	Genetic disorders +singing	Genetic disorders +voice	Aneuploidy +voice
Second Input of Search Terms			
Respiration/Respiratory function + singing	Breath management +singing	Pulmonary function + singing	Breathing+ singing/singers
Pulmonary/Respiratory function of people with Down Syndrome	Pulmonary/Respiratory issues/complications of people with Down Syndrome	Respiratory function of Down Syndrome singers	
Third Input of Search Terms			
Laryngeal function of a singer	Laryngeal function of people with Down Syndrome	Vibratory system in singers	Vibratory system of people with Down Syndrome
Vibratory cycle of Down Syndrome singers	Vocal cords/folds +Down Syndrome		
Fourth Input of Search Terms			
Resonance + Singing singers	Resonance +voice	Resonance +Down Syndrome	Articulation+Down Syndrome
Fifth Input of Search Terms			
Singers and resonant tuning	velopharyngeal dysfunction +Down Syndrome	hypermobility +impact on voice	hypermobility in Down Syndrome individuals
Sixth Input of Search Terms			
collagen +vocal cords/folds	Collagen +vocal cords/folds+Down Syndrome		
Seventh Input of Search Terms			
Disorders + similar characteristics to Down Syndrome	Disorders that affect the voice		
Eighth Input of Search Terms			
Common features of Down Syndrome and Williams Syndrome			
Ninth Input of Search Terms			
Cerebellum + Down Syndrome	Cerebellum+Basal Ganglia +Down Syndrome		

Figure 2: Layered input of Search Words into Databases

The search of databases was supplemented with a hand-search of reference lists from the most pertinent studies identified. Many of the search terms above revealed literature that was useful by analogy rather than by direct relevance, for example, literature investigating the DS singing voice was not found, but research relating to the features and challenges of DS speech is available, as referenced throughout this review. The adoption of a layered process of inputting search terms broadened the scope of literature identified, but review of this remained manageable by the application of inclusion and exclusion criteria.

In line with the hermeneutic circle concept, the search of databases via keyword searches was supplemented with a “back-chaining” method (Winter, 2023, p.144) which enabled further literature to be sourced from reference lists included in the studies reviewed, and from a consideration of other titles appearing when inputting a citation into the bibliography resource “MyBib”.

As indicated by the Gantt chart at Figure 1 above, the reading undertaken for this literature review was extensive. The first level of systematic screening involved a review of the abstracts of hundreds of articles and studies to identify those eligible for review, before screening those studies to find which were eligible (in accordance with the inclusion and exclusion criteria above) for inclusion within this research. The abstract screening was the

“tedious but vital first step to synthesize the extant literature: winnowing down the overwhelming amalgamation of citations discovered through research databases to the citations that should be “full-text” screened and eventually included in the review” (Polanin et al., 2019, p.338)

As the abstract screening revealed a huge number of potentially relevant studies across multiple professional disciplines, the inclusion and exclusion criteria were crucial to producing a relevant, systematic and manageable body of material, from which patterns and data could be analysed to enable conclusions to be drawn as regards the initial research enquiry posed. After undertaking the abstract screening, full texts of the articles and studies which passed this initial review were obtained and read, and a deeper analysis conducted of those texts. The previous research undertaken on this topic meant that a number of the studies identified as appropriate for the current review were familiar, however these were re-read and reviewed thoroughly, with strict adherence to inclusion and exclusion criteria, in an effort to remove any bias that prior knowledge or familiarity with the studies could otherwise have produced (Hart et al., 2009; Schwind and Buder, 2012) and in accordance with the researcher’s ethical obligation to review all studies equally, and not elevate or prioritise any study, nor any part of it, to validate an existing opinion or belief or to reach a particular conclusion (Hosseini, 2020).

Once the literature to be reviewed had been identified, a Braun and Clarke based thematic analysis, involving a search across the range of texts to find repeated patterns of meaning, was implemented. Data was coded in accordance with patterns perceived and key themes were subsequently generated by the author. The information extrapolated from the review is presented in the second section of this work with an interpretative delivery, forming judgments wherever possible about the information obtained from the literature, and providing insight to the reader via a “dominant authorial voice” (Winter, 2023, p.144) which aligns with the thematic analysis model proposed by Braun and Clarke (Braun and Clarke, 2006).

The thematic analysis promoted by Braun and Clarke (Braun and Clarke, 2006) traverses six distinct stages, commencing with the search for patterns of meaning and issues of interest, and culminating in the reporting of the content and the meaning of themes generated across the data (Braun and Clarke, 2006). The process requires the researcher to (i) familiarise themselves with their qualitative data, (ii) generate initial codes, (iii) generate initial themes, (iv) review those themes, (v) define and name those themes, (vi) produce a written report of the research. Braun and Clark’s process is a reflexive one, meaning that the researcher is always consciously aware of their own preconceptions, bias and position, which impact upon the researcher’s interpretation of the research undertaken. The themes generated and concepts identified in the work below were produced from the pragmatist position of a vocal tutor seeking to improve tuition specifically for singers with DS, in particular, personal students of the author belonging to that demographic. Accordingly themes were developed which related to the impact of issues discussed within the studies in so far as they impacted solely upon vocal production and vocal tuition, and it is acknowledged that some studies may have had a greater impact than others upon the author’s use and interpretation of the data, for example where studies included details of vocal exercises familiar to the author as a vocal tutor.

To find a starting point from which to investigate factors which might impact the singing voice of an individual with DS, the research studies focussing upon the speaking voice of DS individuals were reviewed and coded, before being grouped into central themes, in accordance with the Braun and Clarke method of analysis (Braun and Clarke, 2006), which might also be relevant to the singing voice. At the coding stage the keywords relevant to the research question, which appeared from detailed review of the literature were as follows:

- (i) respiratory weakness
- (ii) hoarse, rough, nasal vocal tone
- (iii) fundamental frequency and pitch perception
- (iv) unstable pitch

- (v) hypermobility
- (vi) hypotonia
- (vii) craniofacial features
- (viii) challenges with articulation
- (ix) methods of speech therapy

These codes were subsequently grouped into the following central themes which became the “key characters” (Crowther, 2016):

- (1) Characteristics of DS Voice
- (2) Respiration issues for DS individuals
- (3) Features affecting DS Voice
- (4) Production of DS Voice
- (5) Improving production of DS Voice

The themes above generated from the data were considered and interpreted within the framework of production of the singing voice, in the four broad stages identified under heading “Vocal Production” below, so as to use the data to address the specific research question posed, and, to facilitate this, create a bridge between the data retrieved in respect of the spoken voice and its potential application to the singing voice. Accordingly, although no literature was found which investigated vocal challenges faced by DS singers, the themes generated to organise the data concerning the DS spoken voice produced patterns which highlighted common speech/singing processes, such as vibration and articulation, and the data was therefore useful, as it enabled the author to use existing professional knowledge to form expert judgments upon the meaning of the works, and adapt some of the data to the singing processes being considered (Sattar et al., 2021). The interpretive nature of the Braun and Clarke thematic analysis model (Braun and Clarke, 2006) is compatible with hermeneutic methodology and its circle of understanding (Boell and Cecez-Kecmanovic, 2010). For the purposes of the present research, the literature was reviewed, digested and understood in a circular movement, involving both subjective and objective interpretation, all the time recognising that the themes could not be isolated from each other, but worked together to create a complete picture (Skehill, 2022).

Literature Review.

Introduction

This literature review seeks to identify, through documentary evidence, the challenges which

may be faced by DS singers during vocal production, and the ways in which their vocal training might be improved. The data below is considered by the practitioner researcher, who is keen to find solutions, through a pragmatic lens (Lester and Costley, 2010), guided by a hermeneutic approach, ensuring continuous engagement with the literature reviewed and developing this to achieve greater understanding and insight. (Boelle and Cecez-Kecmanovic, 2014). An initial consideration of what processes enable vocal production to occur, is followed by a detailed explanation of the contribution of each of those processes and an examination of how each process may be impacted by the physical, physiological or clinical features of DS. Finally the practitioner researcher interprets the data to seek for ways in which vocal challenges identified may be ameliorated, so as to improve the vocal training of singing students with DS.

To date, there is no published data evidencing studies undertaken specifically for the purpose of considering the impact of DS on the singing voice. The studies that exist in relation to DS voice are concerned only with the impact of DS upon speech, and the only study to briefly address the DS singing voice offers the general pronouncement that “very few people with Down syndrome can sing at all” (Pryce, 1994, p.10). Accordingly, the selection of studies considered in this literature review has been necessarily determined by research undertaken outside the field of vocal pedagogy, upon issues which may relate to, but are not directly concerned with, the production of the DS singing voice. The data obtained from the literature has been distilled from a unique perspective, namely through the eyes of the vocal tutor researcher, following a process of thematic analysis, to reach understandings and conclusions based upon analogy, comparison, interpretation and deduction. The researcher benefits from some existing knowledge and experience obtained tutoring singers with DS and this position has impacted the way in which the data retrieved from the literature has been interpreted and used, although the researcher has been mindful of the pitfalls of being an “insider researcher” and taken every step to conduct this research thoroughly and humbly, without assumption or bias.

The Process of Vocal Production

The human voice is produced when a steady airflow from the respiratory system is modulated through the glottis by vocal fold vibration, producing sound which travels through the vocal tract and is then amplified and modified by articulators (Zhang, 2016). Voice production can therefore be considered as travelling through four major processes, namely respiration, vibration, resonance and articulation (Sundberg, 1988; Miller, 2013; Zhang, 2016; Herbst et al., 2023)..

In order to understand the challenges of singers with DS, it is necessary to consider the

importance and operation of each of the singing processes and investigate, through the data, why these processes are more difficult for the DS singer to achieve.

Respiration

A number of features of DS impact upon the respiratory process leading to challenges for the DS singer at this first stage of vocal production. As can be deduced from the data, the challenges to respiration lie not only in the pulmonary capacity of the DS singer, but also in the ability to effectively inhale, exhale and manage the breath using abdominal, laryngeal and pharyngeal muscles.

Respiration is the first stage of the singing process and singing requires specific respiratory demands beyond those required by quiet breathing or breathing for speech (Salomoni et al., 2016). The respiratory system is crucial in generating the airflow that travels between the vocal folds, thus providing the power source of the voice (Tong and Sataloff, 2020).

According to Miller, the simple definition of respiration is “the breath cycle; the exchange of internal and external gases.” (Miller, 2013, p.308). Breathing (otherwise known as pulmonary ventilation) consists of a cycle of two phases, namely inhalation and exhalation. During the inspiratory phase, the diaphragm contracts and descends, increasing the volume of the thorax and allowing the lungs to expand and fill with air, whilst external intercostal muscles assist this expansion of the chest cavity as they contract to pull the ribcage upward and outward (De Troyer et al., 2005; Dimon, 2018). During exhalation the internal intercostal muscles draw the ribs inward as air is released from the lungs (Dimon, 2018). The external intercostals, acting as antagonists to the internal intercostals, hold the rib cage up and out during singing. Upon expiration of air from the lungs, the diaphragm relaxes, and returns to its resting position (Leanderson and Sundberg, 1988). It is helpful to keep in mind this comprehensive summary of the process of respiration, since the challenges of a DS singer may lie not only in achieving an effective exchange of gases during the breath cycle, but also in dealing with weakness of respiratory muscles (including the diaphragm, accessory muscles of respiration, and abdominal muscles) necessary for pulmonary ventilation. Indeed studies have shown that respiratory problems in individuals with DS are due to several contributing factors, such as immune dysfunction, hypotonia, persistent upper respiratory tract symptoms (McDowell and Craven, 2011; Laibsirinon, 2012) and reduction in lung function and chest expansion (Albarelo et al., 2014; Trivedi and Das, 2022). Studies dealing with these and other aspects of challenges faced by DS individuals during respiration are identified and discussed separately below.

(i)The exchange of gases

Studies show that individuals with DS have reduced pulmonary function due to various presentations of restrictive lung disorder (Breuer et al., 2022) and that this condition is present in DS infants (Prayle and Vyas, 2021; Breuer et al., 2022) in older DS children (Mitchell et al., 2003; Verstegen et al., 2014; Danopoulos, 2021; Bull et al., 2022) in young adults (Salgueirinho et al., 2016) and in older adults (Resta et. Al., 2003; Landete et al., 2020).

In a study reported in 2016 (Salgueirinho et al., 2016), the pulmonary function of 17 adults with DS (11 females and 6 males), was compared to the function of 17 adults without the condition (9 females and 8 males). All participants were aged between 20 and 40, and were selected in accordance with rigorous inclusion and exclusion criteria, ensuring that any variances between the two groups were confined to pulmonary function and activity. The participants all undertook supervised lung function testing which measured the volume of air they could expel in one second (FEV1), the total amount of air they could exhale forcefully in one breath after maximum inhalation (FVC), and the maximum flow rate achieved during forceful expiration (PEF). As regards pulmonary function, the participants with DS showed significantly lower values for FEV1, FVC and PEF; indeed they achieved less than 60% of the PEF, FEV1 and FVC values recorded by the control group. These results were consistent with findings of previous studies measuring the pulmonary function of children with DS (Dichter et al., 1993; Pastore et al., 2000; Laibson et al., 2012) and previous studies measuring pulmonary function of adults (Da Silva et al., 2010). This information is significant as it shows that both DS adults and children are severely compromised in their ability to expel air with force or consistent flow compared to healthy controls, which also compromises their vocal production, as breath is the “foundation on which singing is established” (Alderson, 1979, p.28) and breath management is the “essential foundation for all skilful vocalism” (Miller, 2000, p.32).

The cause of the significant difference in pulmonary function and activity of individuals with DS has been investigated and documented at length in the medical field. The mid-face hypoplasia, macroglossia, narrow nasopharynx and shortened palate, typical in individuals with DS, are some of the structural features which predispose them to upper airway obstruction (Mitchell et al., 2003; Prayle and Vyas, 2021), compromising adequate airflow (patent airway) and preventing proper oxygenation and ventilation of the lungs (Mouri and Maani, 2020). Further, as shown in a study using MRI equipment, (Uong et al., 2001) it was found that participants with DS had reduced upper airway size and volume when compared to controls without DS, likely as a result of soft tissue crowding within a smaller mid- and lower-face skeleton. Thus abnormal craniofacial development in DS singers compromises air flow and would have a significant impact upon vocal production for the purpose of singing for the reasons mentioned above. Airflow restriction and its impact upon the singing voice

may be compounded by airway blockages, which are common to DS individuals (Sherry, 1983). In a recent study of DS respiration and oxygen carrying capacity, it was suggested that decreased neural input to the respiratory muscles might limit the efficacy of cough and sigh, which could compromise ability in clearing partially blocked airways (DeRuisseau et al., 2023).

Tracheal anomalies, including tracheal stenosis, tracheal ring deformity, tracheal bronchus, tracheomalacia and laryngomalacia are significantly more common in children with DS (Fockens et al., 2021). Pulmonary parenchymal abnormalities in DS individuals result in an overall diminished lung surface area for gas exchange and reduction in functional reserve of the lungs, which persist throughout life (Primack et al., 1994; Prayle and Vyas, 2021). The compromised gas exchange impacts upon respiration and therefore upon vocal production (Miller, 2013, p.308)

The manifestations of respiratory problems in children with DS include stridor, obstructive sleep apnoea, viral upper and lower respiratory tract infections, lobar pneumonia, pulmonary oedema and hypertension, and asthma (Danopoulos et al., 2021; Prayle and Vyas, 2021). Thus the breath cycle – exchange of gases – as described by Miller (Miller, 2013, p.308) is thought to be compromised in individuals with DS, potentially causing difficulties in adequately supporting the voice in speech (Comblain and Rondal, 1996; Miller and Leddy, 1998; Dodd and Thompson, 2001; Casey and Emes, 2011). The literature confirming the difficulty caused to speech by the flawed exchange of gases during respiration might reasonably lead, it is submitted, to the conclusion that the same difficulties would occur, for the same reason, for the DS singer. Indeed, it is likely that an even greater impact would be felt, considering the more intense breath cycle required to produce the singing voice (Salomoni et al., 2016). The notion that reduced respiratory capacity impacts negatively upon the speech of DS individuals was disputed in a study in 2009 (Lee et al., 2009) where the speech of 9 speakers with DS was analysed acoustically against speech samples provided by 9 healthy, gender and age matched English speakers with the finding that speakers with DS exhibited normal respiratory capacity. However, if it is accepted that the majority of research suggesting a link between compromised respiration and speech in DS individuals is correct, it is reasonable to presume that a singer with DS would also have challenges in supporting and projecting their voice effectively.

(ii) Weakness of respiratory muscles

Respiration also requires effective movement of breath using appropriate muscle groups, namely the inspiratory muscles, the expiratory muscles and the accessory muscles (mainly abdominal) of respiration. The muscles that keep the upper airway open during respiration

may also be considered as muscles of respiration because of their close interaction with the other respiratory muscles. These respiratory muscles are the “mechanical effectors of the breathing system” (Benditt and McCool, 2016, p.169). The literature shows that respiratory muscle strength is compromised in individuals with DS by the generalised hypotonia that affects them (Costa et al., 2015; Mohammed et al., 2021). A review of literature produced by clinical research, has revealed a number of studies confirming this weakness of respiratory muscle and function in individuals with DS. In a study in 2015, (Furlan et al., 2015), the respiratory strength of 20 adults aged between 18 and 35 years (10 with DS, 10 non-syndromic), male and female, was measured by an analog manometer showing maximum inspiratory pressures (MIP) and maximum expiratory pressures (MEP). MIP is a measure of inspiratory muscle strength and is closely connected to diaphragmatic strength, since the diaphragm is the major inspiratory muscle; MEP is generated through abdominal and intercostal muscles (Schoser et al., 2017). The participants with DS had significantly lower MIP and MEP (around 30% and 22% respectively) confirming lower respiratory muscle strength. In a subsequent study (Değer et al., 2017), the strength of the respiratory muscles of 23 children with DS was compared with that of a control group of 30 children and Maximum Inspiratory and Expiratory Pressure (MIP-MEP) was measured. Only 13 children with DS were able to complete the tests due to co-operation issues, but results showed that the MIP and MEP values were significantly lower than the control group ($p < 0.05$), indicating that children with DS had significantly lower respiratory strength. A study undertaken by Pryce (Pryce, 1994) highlighted the significant increase in energy required from DS speakers to initiate voice, as compared to individuals unaffected by DS. Using an electromyographic device which measured the electrical activity occurring when a nerve impulse triggered the contraction of an individual group of fibres in a muscle, the researchers were able, inter alia, to measure the effort required to initiate phonation. The results showed that the average amount of energy required by the 29 adult members of the control group to initiate voice was 72.52 microvolts. Amongst the 30 adult DS participants, the average measurement was 131.57, indicating almost double the amount of effort. This was attributed to the greater energy required to mobilise flaccid muscles weakened by generalised hypotonia, including that of the laryngeal muscles, typical of individuals with DS (Pryce, 1994). It is submitted that this data can be applied to the singing voice of a DS individual, since the same, if not more effort would be required to initiate phonation for this purpose.

Understanding the extent of respiratory challenges of singers with DS is relevant and important to vocal tutors, who will inevitably recommend and teach appropriate breathing techniques to their students. The data shared in the studies uncovered by this literature review may contribute to a greater understanding by tutors of the level of respiratory challenges faced by singers with DS, and encourage appropriate adjustments to tuition, in addition to enhanced awareness and sensitivity of the challenges. A number of studies have

spoken to the positive impact of targeted exercise upon improvement of muscular function during the respiratory process (Sgariboldi et al, 2014; Costa et al., 2015). In 2021, a study was carried out to investigate pulmonary rehabilitation methods in children with DS (Mohamed et al., 2021). More specifically, the study compared the effect of proprioceptive neuromuscular facilitation of respiratory muscles with that of inspiratory muscles training, in relation to respiratory muscle strength, chest expansion, spirometry and functional capacity. Forty-five children of both genders, between the ages of 10 and 13, all with DS were put into 3 groups. Group A undertook proprioceptive neuromuscular facilitation of respiratory muscles (PNF), Group B undertook inspiratory muscle training (RMT) and Group C was the control. PNF is a combination of passive stretching and resistance that can improve pulmonary functions such as expansion, endurance and respiratory muscle strength (Ashwini et al., 2017). RMT is a series of targeted exercises which improve muscle strength and endurance, such as chest resistance exercises to the costal areas to align respiratory muscles with breathing rhythm (Kido et al., 2013) and repetitive breathing resistance exercises (Roth et al., 2010). The treatment programme for groups A and B was carried out 5 times each week for 12 successive weeks and, in addition, all three participant groups undertook 20 mins of aerobic exercise on a bicycle ergometer 5 times each week for 12 successive weeks. MIP and MAP testing was carried out pre and post treatment and showed a significant increase in values for groups A and B, post the treatment programme, leading to the conclusion that both were effective in improving respiratory muscle strength in children with DS, with greatest improvement shown by the group which undertook PNF. Research studies such as these may provide valuable information to a vocal tutor of DS singers, since many of the exercises described in this study and others (Slupska et al., 2019; Baxi et al., 2022) are familiar stretching and breathing exercises which can be safely carried out in the studio.

Vibration

Vibration is another crucial stage in the process of vocal production and it is necessary to consider what comprises and contributes to a successful vibration, to understand why a singer with DS may be unable to achieve this proficiently, leading to compromised vocal production.

Voiced sound is produced when sub-glottal pressure following inspiration, pushes the vocal folds apart, which force is then reversed by the elastic recoil properties of the vocal fold tissue, creating a negative Bernoulli pressure effect which sucks the cords back together and results in a push and pull sequence in every cycle, sustaining the vibration (Titze, 2019) and known as the mucosal wave. The voiced sound, initially just a “buzz” like noise, is then modified and formed into words by the processes of resonance and articulation (Hrelec and

Zhang, 2022), which are discussed separately below.

The successful vibratory cycle depends on the viscoelasticity of the connective tissue of the vocal fold lamina propria which is the major vibratory tissue activated during phonation (Titze, 2000; Prades et al., 2010; Rocha et al., 2021). The vibration of the vocal folds is possible due to the existence of fibrous proteins found in the extracellular space between the cells of the various layers of the vocal fold tissue (Tang et al., 2017). One of these important proteins is elastin which allows tissue to stretch and then recoil and has the ability to extend and contract in repetitive motion when hydrated. (Moore and Thibeault, 2012; Trebacz and Barzyka, 2023). Another of the most abundant proteins in the vocal cords, contributing to the viscoelastic properties and making up approximately 43% of total proteins in the vocal fold lamina propria (Hahn et al., 2006) is collagen. Unlike elastin, collagen does not directly facilitate tissue stretch but provides tensile stiffness and strength to counter elastin's stretch-and-recoil properties, defining the elasticity threshold of the tissue during stretching or loading and ensuring tissue integrity following repeated stress recoil, enabling the vocal folds to meet the demands of the repetitive and high frequency impact of phonation. (Gray et al., 2000). It is known that individuals with DS have an abnormal collagen arrangement, specifically an over-expression of type VI collagen, a collagen crucial for cardiac and skeletal muscles, which is encoded on a gene on the 21st chromosome, and regulates muscle stiffness (Dey et al., 2013). This abnormality in collagen VI is thought to hamper function of the protein, and thereby contribute to muscle weakness (hypotonia), hypermobility and dysautonomia in DS individuals (Dey et al., 2013; Wilson and Tonk, 2022). The predominant collagen types in vocal cords are collagen types I and III, which maintain the structure of the lamina propria and tissue elasticity (Tateya et al., 2007; Buhler et al., 2011) and so an extensive search across multiple disciplinary libraries was undertaken to try to determine whether individuals with DS might also have an abnormal volume or distribution of collagens I and III, directly impacting operation of the vocal folds. That extensive search failed to produce any literature at all concerning the content or distribution of types I or III collagen in DS individuals, and it is submitted that research in this regard would be useful to further explore the specific features of DS which impact upon phonation.

The hypotonia experienced by DS individuals includes hypotonia of their extrinsic laryngeal muscles and pharyngeal wall (Pryce, 1994) and impacts upon laryngeal function, contributing to the typical hoarse, nasal and perceived lower pitched voices of DS individuals. The repeating undulation known as the mucosal wave, which occurs during a normal vibratory cycle is critical to high quality voice production (Krausert et al., 2010). This mucosal wave is dependent on the structure of the vocal tract, such that abnormalities of the vocal folds or vocal tract will cause irregularities in the mucosal wave (Krausert et al., 2010) and therefore, in clinical practice, assessment of the mucosal wave is essential for treating

and diagnosing voice disorders (Jiang et al., 2019). F0 (f₀) is the lowest frequency component of a complex sound wave produced during vibration of the vocal folds and is determined by the rate at which the vocal folds vibrate when voiced sounds are made (Lee and Humes, 2012). Our perception of a fundamental frequency is known as “pitch” (Oxenham, 2012). F0 is measured in cycles per second (Hz); the average male voice typically has an f₀ of between 85 Hz and 155 Hz, whereas a female voice typically has an f₀ of between 165 Hz and 200 and 255 Hz (Fitch and Holbrook, 1970; Baken, 2010; Watson, 2019). The f₀ of vocal fold vibration is impacted by vocal fold tension and sub-glottal pressure, both processes affected by laryngeal muscles. Contraction of cricothyroid muscles elongates the vocal folds and increases tension, whereas contraction of thyroarytenoid muscles decreases the combined tension of the vocal fold (Jiang, 2000). Phonation which shows involuntary instability of fundamental frequency (perceived as pitch to the human ear) or intensity is usually perceived as hoarse voice (Jiang, 2000).

One of the patterns that emerged during coding of the salient research studies, was data regarding examination of the impact of DS upon f₀. Several quantitative studies based on acoustic measurement methods offered mixed results, with most studies finding no f₀ difference between participants with DS and healthy controls. This discrepancy with pitch perception of DS voices promoted enquiry as to whether perception of low pitch was influenced by factors other than the frequency of vocal fold vibration. Prior to a study conducted by Michel and Carney (Michel and Carney, 1964), it was accepted that individuals with DS suffered from voice disorder (Blanchard, 1964; Benda, 1965; Novak et al., 1972) and inevitably spoke with hoarse, monotonous voice and an abnormally low pitch (Benda, 1965; West et al., 1957). Indeed Benda (Benda, 1965) went so far as to say that the “low and raucous” voice of children with DS was so typical, that a diagnosis of the disorder could be made purely on the basis of hearing the voice, without actually meeting the child (Benda, 1965 p. 520). These earlier studies and conclusions as to DS vocal characteristics were based upon clinical perception of factors such as pitch and vocal range. Developments in technology provided researchers with the means to analysis acoustic features of the DS voice with instruments, rather than relying upon clinical judgments, and so perceptions of the DS voice began to be compared alongside acoustic analysis. One of the earliest instrumental studies (Michel and Carney, 1964) challenged the previously accepted perception of the abnormal pitch of DS children, providing empirical evidence via measurement of f₀, that the pitch of DS children was within normal limits for their age, although children with DS were 1.5-2 years behind the control group in terms of physical growth, leading them to conclude that this might in fact be responsible for inaccurate perception of their pitch – the immature physical appearance leading to the perception that the pitch was lower than normal, when in fact it was consistent with chronological age. This study was corroborated by a further study a year later (Hollien and Copeland, 1965) which

reached the same conclusions when it tested the speaking f0 of groups of girls with DS against control groups, and found no major discrepancy in f0 measurement, suggesting that the perception of low pitch might be attributable to the hoarse, rough voice of a child with DS. Some years later, consideration was given to the impact of typical hypotonia upon the DS voice and a hypothesis was developed (Weinberg and Zlatin, 1970) that the hypotonia impacted laryngeal function and lowered the f0 of the individual with DS, leading to the common misperception of the DS voice. A more robust study was conducted in 1973 (Montague et al., 1973) which regulated the rather hap-hazard collection sample of earlier studies, testing the voices of 20 individuals with DS against 20 age-matched typically developing participants. The results of this study corroborated earlier studies (Michel and Carney, 1964; Hollien and Copeland, 1965) and reported no overall difference in the f0 of DS children and their typically developing peers. In 1978 a follow up study was conducted (Montague et al., 1978), comparing perception of pitch against accurate measurement of f0. Once again, the f0 levels measured within normal parameters, yet the perception rated over 60% of the children with DS as having lower pitch. The debate was widened in 1983 by Pentz and Gilbert, (Pentz and Gilbert, 1983) who tested the proposition that f0 level was the only factor responsible for the perception of low pitch amongst DS individuals, and concluded that other acoustic parameters such as jitter, shimmer and noise-to harmonic ratio (which all measured higher in DS participants than in members of the control group) were responsible for the perception of low pitch and hoarse tonal quality. A much more recent study further added to the debate, when it found that across all 5 vowels, DS participants did in fact measure lower values than the control group. This discrepancy with the findings in earlier studies was attributed to language specific differences, as the participants in this study were all Portuguese (Moura et al., 2008). Despite some inconsistencies in results, the review of the literature contrasting actual f0 measurement and perception of pitch is important to vocal tutors of DS singers, as it indicates that perception of lower pitch may not be real, and this in turn could encourage wider exploration of the vocal range of a DS singer (setting aside any assumption that only a lower pitch/vocal range of a DS student is supportable in the studio). Furthermore, focus on other acoustic factors, aside from level and range, which later studies (Moura et al., 2008) have suggested may account for conflict between perception and measured f0, may guide the vocal tutor to better support the DS student, for example with appropriate exercise such as SOVT exercise (Fantani et al., 2017) which, it is submitted, may improve dysphonia caused by jitter (frequency variation of vibratory cycles) and shimmer (amplitude variation of sound waves), thereby improving overall voice quality. Despite investigation as exemplified above, there is no single acoustic correlate of voice quality which consistently emerges in the published literature on point (Kent and Vorperian, 2013).

The dysfunction of collagen type VI in DS individuals mentioned above has also been linked to hypermobility, a universal feature of the DS condition that results in lax joints and ligaments, which, particularly when combined with hypotonia, leads to poor control over body systems and functions (Foley and Killeen, 2019; Jeffery et al., 2021). Singers with hypermobility have difficulty maintaining stable pitch and tone, as the hypermobility of the cricothyroid muscle which elongates and regulates tension of the vocal cords, results in inconsistent tone, unreliable pitch, potential voice breaks and reduced vocal endurance (Jeffery et al., 2021; Menton et al., 2023). Hypermobility may also affect the stability of the rib cage and the diaphragm, leading to inefficient breath support and management, which is particularly problematic for singers. Further, the overall tone and quality of the voice can be affected if hypermobility impacts the alignment and function of features within the vocal tract, such as the jaw and tongue (Bascom et al., 2021). Jaw hypermobility can lead to issues like temporomandibular joint disorder, which can cause pain, inflammation and dysfunction in the jaw and inability to effectively speak or sing (Ghibellini et al., 2015), and hypermobility of the tongue can cause problems with tongue positioning and movement, causing unclear articulation (Baeza-Velasco, 2021).

Anomalies in collagen type VI can also cause dysautonomia, a disorder that disrupts the autonomic nervous system and there is a strong connection between hypermobility and dysautonomia (Gazit et al., 2003). Dysautonomia is common in DS individuals (Dey et al., 2013; Wilson and Tonk, 2022). The autonomic nervous system is relevant to vocal production because of the importance of the vagus nerve for phonation. The vagus nerve has 3 branches, namely the pharyngeal branch which carries nerves to many of the pharyngeal muscles, the superior laryngeal nerve branch which carries nerves to the cricothyroid muscle, and the recurrent nerve branch which innervates the abductor and adductor muscles of the vocal folds. Damage or disruption to these branches of the vagus nerve due to dysautonomia can result in weakness or paralysis of the velum, compromising resonance, difficulties controlling pitch via the cricothyroid muscle and dysfunction of vocal fold alignment, causing breathiness and hoarse voice. (Xue et al., 2014; Cardoso et al., 2019).

DS individuals frequently experience significant voice-related complications which may not be explained by typical syndromic features. A recent study (Hseu et al., 2022) sought to investigate the link between DS and higher risk of subglottic stenosis, vocal fold paralysis, and laryngomalacia. A retrospective review of twenty-three children with DS, all under the age of 18, who had presented to the otolaryngology department of a paediatric hospital between January 2007 and July 2021 with voice related concerns, was undertaken. The data extracted and reviewed included general personal and clinical information and notes of voice assessments, acoustic data, laryngoscopic and/or videostroboscopic exam data, and

medical notes of any relevant surgical procedures performed. The detailed review identified vocal fold immobility as the most common laryngeal pathology. A link was made between the presence of vocal cord immobility and prior cardiothoracic surgery, with over half of the participants having undergone this surgery at the hospital. Although this study was unable to confirm the link conclusively without pre- and post-operative laryngoscopies, the opinion of the senior medical specialists undertaking the review was that the cause of the vocal cord immobility was due to laryngeal nerve damage during cardiac surgery. The higher incidence of cardiac surgery in DS children predisposes them to such nerve damage (Mudd and Noelke, 2018), as the left recurrent laryngeal nerve is in close proximity to the aortic arch (the top part of the main artery carrying blood away from the heart) and iatrogenic trauma to the recurrent laryngeal nerve during cardiac surgery is cited as the most common cause of vocal cord immobility in infants and older children (Khariwala et al., 2005; Zbar and Smith, 1996; Lee et al., 2018; Barr et al., 2021; Kenny et al., 2022) although spontaneous recovery was possible in infants (Barr et al., 2021). This information, acquired via this multi-disciplinary literature review, is useful knowledge for teachers of younger DS singers, as not only can it be an indication that a breathy voice in a DS student, particularly a child, may not respond to attempts to improve breathiness with vocal exercise, but the presence of a very breathy voice, easily detected by an experienced vocal tutor, may be good reason to suggest medical follow up to the student, to investigate the extent and possibility of treatment for any laryngeal nerve injury that a particularly breathy voice might suggest. Indeed, although beyond the scope of this work, the research undertaken in relation to this link between laryngeal nerve damage and vocal cord immobility has revealed the existence of minimally invasive surgical intervention, namely, endoscopic arytenoid lateral abduction, which fixes the arytenoid cartilage in the maximum abducted position in order to straighten the vocal cord and improve phonation (for speech and singing) (Trozzi et al., 2022).

Resonation

Resonation is another crucial element of vocal production and although a full discussion of the intricacies of resonance and acoustics is beyond the scope of this work, in order to fully understand why singers with DS face challenges with achieving resonant voice, it is first necessary to consider how vocal tract resonance occurs in healthy singers..

Vocal resonance has been defined by McKinney as “the process by which the basic product of phonation is enhanced in timbre and/or intensity by the air-filled cavities through which it passes on its way to the outside air” (McKinney, 2005, p.27). A resonant voice has been described as a voice that projects well (Myers et al., 2015) is tension free and easy to produce (Verdolini et al., 1998; Yiu et al, 2016), gives a sense of vibration at the front of the

face or “forward tone” (Titze, 2001; Rakerd et al, 2019) and is harmonic (Titze,2001; Wolfe et al.,2009).

The vocal tract is the airspace which extends from the vocal folds to the lips (Story, 2016). The oscillating airflow generated by vibration of the vocal folds creates a wave which travels through the airspace of the vocal tract formed by the relative positions of the lips, tongue, jaw and soft palate, and then leaves the lips as a shaped voiced sound containing a number of unique acoustic features (Story, 2016). Vowel identity and sound quality (timbre) are impacted by shape and size of the resonating cavities (pharynx, oral and nasal) in the vocal tract (Wolfe et al., 2009). The strength, tone, pitch and resonance of the voice are dependent on the structure and neuro-muscular control of the laryngeal and respiratory systems (Espir and Rose, 1983). Singing (and speech) require appropriate anatomical structure and control, to enable positioning of the tongue, lips, jaw and soft palate in such a way as to adjust acoustic resonances to specific frequencies and produce broad peaks or bands of resonance-enhanced power in the voice known as formants, which are characteristic of vowels and some consonants (Wolfe et al., 2009). In singing, vocal tract resonance can be used to boost one or more harmonics of the voice, as, for example, when soprano singers, whether trained or not (Garnier et al., 2010), tune the first resonance R1 (which falls, approximately, in the frequency range D4-C6) to the pitch frequency f_0 , in a process known as R1: f_0 tuning, or when alto singers using belt technique tune R1 to the second harmonic $2 f_0$, in a process known as R1: $2 f_0$ tuning (Smith et al., 2013). Vocal tract resonance can be purposefully adjusted by both male and female singers to deliver certain singing styles or qualities or techniques such as “twang”, “forward” or “open” (Bloothoof and Pomp, 1986; Hertegard et al.,1990;Titze et al., 2003) in an effective and safe manner. This process of resonant tuning can enhance the tone and quality of the voice (Rakerd 2019), increase volume and projection (Titze, 2021), reduce fatigue and avoid strain on the vocal folds (Verdolini et al., 1998), improve the efficiency of voice production (Koberlein et al., 2021)and ensure clarity of diction by maintaining distinct vowels sounds (Wolfe et al., 2009). As is evident from the literature referenced throughout this section, the range of complex adjustments to the vocal tract, utilised by healthy singers to achieve an effective and typically pleasing vocal output, are unavailable to the DS singer, due to anatomical incumbrances upon the DS vocal system.

Resonance requires a singer to have control of their vocal system, including management of breath (as discussed in the above section on “Respiration”), control of laryngeal and pharyngeal muscles, and control of the tongue, lips, jaw and soft palate. Structurally, resonance is optimised by the shape and size of the vocal tract (Ahn et al., 2015). DS individuals have a smaller mouth and jaw, flat nasal bone and narrow nasopharynx, prominent lips, shortened hard palate and reduced palatal volume, reduced facial and

pharyngeal height, weak laryngeal and pharyngeal muscles, a variety of dental anomalies, mid-face hypoplasia and low muscle tone in the tongue causing protrusion (Jacobs et al., 1996; Dellavia et al., 2007; Carolina et al., 2008; Moura et al., 2008; Sforza et al., 2012; Antonarakis et al., 2020). Each of these features interferes with resonance and creates an abnormal vocal tone (Kummer, 2016). The size of the oral and pharyngeal cavities influence how sound travels and produces certain acoustic characteristics, impacting resonance and vocal quality (Ahn et al., 2015). The size of the pharynx is adjusted primarily by constrictor muscles and is at its optimum size when these muscles are at rest. The size of the pharynx is directly related to the production of formant 1, which is the lowest, originating frequency of a vowel (Kent and Vorperian, 2018). Individuals with DS have notably smaller oropharynx volumes (Uong et al., 2001; Khurana et al., 2024) and generalised hypotonia of muscles throughout the body, including pharyngeal muscles (Mitchell et al., 2003; Madhavan et al., 2023). The smaller size of the oropharynx and the weakness of constrictor muscles which limit adjustment within the pharynx, impact upon production of formants, affecting resonance of the DS voice (Rakerd, 2019). Further, the shortened vocal tract of DS individuals (Vorperian et al., 2023) and the abnormally small/narrow oral/nasal cavities in DS individuals, influence which overtones are emphasised (Pruthi and Espy-Wilson, 2004), thus affecting the voice timbre and contributing to the common perception that DS voices are low in pitch and hoarse/gruff in quality. Tongue position (which may have limited movement in DS individuals due to its placement within an abnormally small oral cavity and hypotonic muscles) and compromised embouchure adjustment (affected by hypotonia of the facial and laryngeal muscles in DS individuals) both impact upon resonant frequencies, by impeding the size and shape of resonating cavities, and the position of the soft palate (lower in DS individuals due to a compressed craniofacial structure) also impacts negatively upon the balance of oral and nasal resonance. (Wolfe, 2009; Dholvan and Anitha, 2014). The reduced bone growth in the head and face of DS individuals impacts upon attachment locations and support of muscles used during speech (and, therefore, singing) (Kent and Vorperian, 2013), creating further challenges with resonance. Neuroanatomical differences may also play a role in the functioning of voice (Madhavan et al., 2023). In 1988 a review of neurology literature concluded that individuals with Down syndrome had fewer cortical neurons and a decreased neuronal density compared to typically developing individuals which disrupted the accuracy, speed, and consistency of voiced sounds in DS individuals (Kemper, 1988). Recent neuroanatomical studies, including MRI studies of adolescents with DS, have reported smaller cerebellar volumes, compared to age-matched neurotypical peers (Wilson et al., 2019), an anomaly that creates challenges with timing and clarity (Person et al., 2008), as referenced in a study carried out on songbirds to test whether cerebellum could influence activity of the basal ganglia and therefore be instrumental in song learning (Pidoux et al., 2018).

The physical and physiological features common to DS individuals and discussed in the literature reviewed and referenced above have been shown to impact upon their speech production and, it is submitted, the information in this data is useful also for the purposes of understanding incumbrance to the singing voice. If resonance is hindered or interrupted by anatomical abnormalities in the mouth, nose, palate and jaw and weakness of laryngeal and pharyngeal muscles impacting control of articulators and intra-oral spaces, it is reasonable to deduce from the data showing detrimental impact upon DS speech, that the singing voice will be similarly affected.

One of the patterns seen in the literature was the tonal quality of DS voices. DS voices are perceived as being hoarse, monotonous, breathy and nasal (Michel and Carney, 1964; Pryce, 1994; Montague and Hollien, 1973; Moran et al., 1988; Moura et al., 2006; Lee et al., 2009). Research which discussed breathy voice purely in the context of healthy adolescent voices was added to the exclusion list, but some of the studies that remained revealed that this tonal quality, common in individuals with DS, was shared by individuals with another genetic disorder, Williams Syndrome or Williams-Beuren Syndrome (WS). Upon conducting searches enquiring as to the impact upon the voice of WS, literature was discovered indicating that the hoarse, breathy, rough tonal quality of the voices of individuals with WS is caused by a deficiency in vocal cord elastin (Vaux et al., 2003; Watts et al., 2008; Hildago et al., 2018) and low muscle tone (Mervis and Velleman, 2011). The data confirming typical weakness of laryngeal muscles in WS individuals (Pober, 2010; Copes et al., 2016; Shalev et al., 2019), comparable to the weakness of laryngeal muscles in DS individuals (Novak, 1972; Pryce, 1994; Moura et al., 2008) provoked further searches as to therapeutic interventions which might exist to improve WS vocal function. A range of vocal function exercises (VFE's) (Stemple et al., 1994; Angadi et al., 2019) commonly utilised for WS individuals, including breath management, warm up, stretch, contract and power exercises, was discovered and these exercises, all familiar to vocal tutors, could perhaps also assist vocal function of DS individuals.

Another relevant piece of information revealed, when reviewing studies potentially relevant to the rough, hoarse tonal quality of DS individuals, was the correlation between strained voice and dysphonia in healthy singers. Although, in accordance with the exclusion criteria, a comprehensive analysis of that literature was inappropriate, a study by Mckenna and Stepp (Mckenna and Stepp, 2018) which considered the relationship between acoustical and perceptual measures of vocal effort, provided a rich source of references dealing with the correlation between excessive vocal effort and voice disorder, and highlighted other useful studies where hoarse and rough vocal tones were linked to strain. This information is of interest, particularly as it aligns with the findings of Pryce (Pryce, 1994), mentioned above, that significantly greater energy and effort is required by DS individuals to initiate voice,

which it is submitted, could lead to strain, contributing to the rough tonal quality of DS singers and speakers.

Hypernasality (abnormal nasal resonance during the production of non-nasal sounds) has been cited as another common characteristic of the DS voice (Rolfe et al., 1979; Kline and Hutchison, 1980). In speech, what would be regarded as normal resonance, is achieved through an appropriate balance of oral and nasal processing, depending on whether vowels, voiced oral consonants or nasal consonants are being spoken. When a nasal vowel is produced, the soft palate is lowered and air flows out through the nose and mouth simultaneously. By contrast, oral vowels are produced without nasalisation and air flows only through the mouth. In nasal consonants (“m”/”n”/”ng”) the palate is again lowered but air flows out only through the nose and the oral cavity is fully occluded (Kowovi et al., 2019; Gwizdzinski et al., 2023). Therefore, nasal sounds are generated by a coupling of the nasal and oral cavities, so as to open the velopharyngeal port and allow sounds to resonate in the nasal cavity (Cler et al., 2021). Oral sounds which, in the English language, include most vowels and vocalic consonants (Kent and Rountrey, 2020) are produced when the velopharyngeal port is closed (Shadi et al., 2022). An inability to uncouple the nasopharynx and oropharynx, such that acoustic energy is shared, is typical in DS individuals and is caused by hypotonia of the pharyngeal and laryngeal muscles, (Kummer, 2006; Kummer, 2011(a), Kummer, 2016; Kent et al., 2021). This failure to uncouple the cavities results in velopharyngeal dysfunction, causing hypernasality (Kummer, 2006; Kummer, 2011 (a)). Hypernasality should not be confused with nasal air emission; although these terms are often used interchangeably in the literature, they are distinct symptoms of velopharyngeal dysfunction that typically occur on different speech sounds. Hypernasality is a resonance phenomenon that occurs primarily on vowels, whereas nasal air emission is an aerodynamic phenomenon that occurs primarily on oral pressure consonants, especially voiceless consonants (Kummer, 2011(b)). When Velopharyngeal dysfunction is present, both hypernasality and nasal air emission can occur in the same speaker, but they are different entities.

Although hypernasality is frequently mentioned in the literature as a feature of DS, no published studies which discuss this in the context of the DS singer and/or provide direction for improving this quality in DS singers have been identified. As the literature review undertaken for this work was necessarily multi-disciplinary due to the lack of literature regarding the DS voice in the field of vocal pedagogy, data from studies undertaken by researchers in the field of speech therapy was reviewed and these studies contain information and direction that may be explored by a singing tutor of DS students. For example searches on “velopharyngeal dysfunction” which includes such symptoms as the shortened palate typical of DS individuals (Dellavia et al., 2007; Klingel et al., 2017) revealed

papers containing very detailed exercises created by Dr Ann Kummer, a much published researcher and senior director of the Division of Speech-Language Pathology at Cincinnati Children's Hospital, for improving hypernasality due to velopharyngeal dysfunction caused by cleft palate. Further searches identified other literature and exercise plans created by Dr Kummer, addressing symptoms commonly experienced by DS individuals, affecting phonation (Kummer, 2006; Kummer 2011(a);Kummer , 2011(b)). Notably, Dr Kummer emphasises that any vocal therapy cannot correct structural dysfunction, only improve errors of vocal placement or vocal technique affecting voice quality and function (Kummer, 2006;Kummer, 2011(a).

Articulation

Articulation may be considered as the final stage in successful vocal production and is as important as the previous stages discussed. The way in which effective articulation is achieved in healthy singers needs to be considered to understand why this process might be challenging for the singer with DS.

Articulation in the narrower sense of the word refers to the movement of articulators in the oral tract (lips, tongue, velum, cheeks, jaw, larynx) to produce a specific articulatory movement (e.g. vowel or consonant formation), and in the wider sense includes vocal fold activity and phonation (Zhu, 2015). Each of these meanings will be dealt with separately below.

It has been written that the performer who cannot articulate the words of a song so that they are understood has defeated the whole purpose of his art (Bunch, 1997). What's more, since movements of articulatory structures like the tongue, lower jaw, soft palate and lips directly influence the shape of the resonator, faulty diction and inefficient articulation inevitably impoverish tonal quality (Dayme, 2009). The skilful separation of vowels and consonants when singing and the shaping of words with articulators requires motor control of velopharyngeal structures (Moon and Jones, 1991; Myers and Finnegan, 2015) which studies show may be challenging for DS singers to achieve (Roberts et al., 2005; Timmins et al., 2009; van Bysterveldt, 2010). Down syndrome speech typically shows phonological immaturity such as final consonant deletion, use of stops for fricatives and inappropriate stresses at the end of words rather than on the initial syllable or the head of the tone group (Bray, 1998). Syllable deletion is also common in the speech of people with DS and it has been suggested that this may be a strategy, subconsciously employed to facilitate simplification of the rhythmic structure of the speech sample, thereby making neurophysiological process of speaking easier (Heselwood et al., 1995). As indicated in the section on "Resonation" above, reduced size of the oral cavity and a high palatal arch

(Bhagyalakshmi et al., 2007) may influence tongue placement which may impact upon articulation (Rosin et al, 1988) and the relative macroglossia which is a typical feature of DS may interfere with articulatory placement, as the tongue does not have sufficient space in which to move fluently (Miller and Leddy, 1998;Bunton and Leddy, 2010). Notably, the historical and common perception of macroglossia (oversized tongue) in DS individuals has been discredited, with researchers now agreeing that tongue size in DS individuals is in fact comparable to healthy contemporaries, but that the smaller oral cavity gives the misleading impression that the tongue is larger than normal (Guimaraes et al., 2008; Kent and Vorperian, 2013). A study in 2008 (Guimaraes et al., 2008) investigated whether adults with DS suffered from true macroglossia or relative macroglossia. Sixteen participants with DS and sixteen age/gender matched members of a control group underwent sagittal and axial MRI examinations to compare the distance between the tongue and various intra-oral points, as well as the size of the tongue relative to several craniofacial parameters features. The results of those examinations showed that in fact the tongues of all the DS participants (2,432 mm²) were significantly smaller than those of the control group (2,767 mm²; $P=0.02$). The craniofacial bony parameters were also smaller in all the DS participants as compared to the control group, however the size of the tongue relative to the craniofacial parameters was larger in the patients with Down syndrome (0.54) than in the controls (0.46; $P<0.001$). Nevertheless the limited space within the mouth causes the tongue to fill more of this cavity, and this, together with weakness of muscles operating the tongue, results in restricted movement of the tongue which compromises resonance and intelligibility (Wilson et al., 2019). A study in 2010 (Bunton and Leddy, 2010) aimed to investigate and identify vowel production characteristics for the four English corner vowels produced in monosyllabic words by two adult speakers with Down syndrome, and to compare the data obtained against 2 age and gender matched controls. This was achieved by measuring the “articulatory working space area” to determine the range of motion and positioning that could be achieved by the articulators (tongue, lips, jaw) during vowel production, and by also assessing the acoustic vowel space and the speed of articulatory movement/mobility of the participants. The participants with DS exhibited significantly reduced articulatory working space area and smaller acoustic vowel space area as well as slower speeds of articulation. The authors of the study concluded that these factors contributed to the impaired speech intelligibility of the DS participants and that the reduced articulatory working space was likely related to the anatomical and functional differences associated with DS, namely a smaller oral cavity and the limited range of articulators affected by syndromic hypotonia (Bunton and Leddy, 2010).

It would appear from the literature that many of the same features impacting resonance also impact articulation, in the broader sense, in DS singers. The organs which effect articulation are dependent on muscle function and the generalised hypotonia, which is typical of DS individuals, is well documented and discussed above in the section on “Resonation”. The

weakness of muscles in the vocal tract and anatomical differences affecting phonation, such as mid-face hypoplasia and relative macroglossia in DS individuals, prevent precise articulation and compromise intelligibility (Miller and Leddy, 1998) reducing the quality of both speech (Bunton and Leddy, 2010) which would also impact articulation when singing. The smaller oral and pharyngeal spaces in DS individuals impact upon how sound travels through the vocal tract, determining the acoustic characteristics of the output, and the consequent nasalisation of sounds, as discussed above, may also affect intelligibility.

The impact that smaller cavity size can have on vocal output was demonstrated in a study of participants (without DS or any other any form of disorder) to investigate the impact of body position upon vocal tract acoustics (Vorperian et al., 2015), including any impact upon the configuration or function of the articulators, either when speaking or at rest. This study confirmed other earlier research which found that nasopharyngeal and oropharyngeal cavities are smaller when the body is in the supine position (Jan et al., 1994; Sutthipraporn et al., 2008; Traser et al., 2013; Bae et al., 2014). This was attributed in the Vorperian study (Vorperian et al., 2015) to the effect of gravity pulling back the tongue root when the subject was lying down, such that the size of the pharyngeal cavity was reduced. Additional research has shown that respiration is also affected by body position, with a supine position reducing the cross sectional area of the upper airway, particularly in the region between the hard palate and bottom of the uvula (Van Holsbeke et al., 2014). Gravitational effects did not however extend to lingual function, so tongue movement, for example, was not affected by body position (Dietsch et al., 2013). The Vorperian study (Vorperian, 2015) investigated the impact of a supined body position on vocal tract acoustics, by means of an acoustic pharyngometry device (which measures oral and pharyngeal dimensions by means of sound wave reflection). It is submitted that the data from this study is of relevance, since singers with DS have a similarly small pharyngeal cavity (Pryce, 1994; Guimaraes et al., 2008; Kent and Vorperian, 2013). The Vorperian study (Vorperian, 2015) found increased f_0 and, most notably, significantly raised values in the f_3 formant. The latter finding is of interest to a singing tutor of DS students, since raised f_3 values have been linked to changes in voice quality, such as a more breathy or rough voice (Keating et al., 2015; Gonzalez et al., 2024), typical characteristics of the DS vocal tone, as discussed above.

Conclusion and Reflection on Practice

This comprehensive literature review has shown that analysis of primary research studies is a credible and useful research method for identifying challenges faced by singers with Down Syndrome. Further, in the hands of a reader who, like the author, has knowledge and experience in the field of vocal pedagogy, it is submitted that the literature may also assist to identify safe and effective ways, based upon familiar vocal exercise and technique, to tackle

some of the challenges faced by singers with DS, arising from syndromic features and physiological complications of this disorder.

This conclusion is reached in spite of the fact that, over the course of several extensive literature reviews undertaken by the author in recent years, culminating in this particularly comprehensive and systematic review, not a single piece of literature exploring the impact of DS upon the singing voice has been found. This is surprising considering the large volume of data available in relation to the impact of DS upon speech and the spoken voice, and the widely accepted view that sufferers of DS have a particular attraction towards music and singing, and benefit medically, physically, psychologically and socially from being exposed to music in all its forms (Flowers, 1984; Stratford and Ching, 1989; Buckley, 2006; Wylie, 2006; Pienaar, 2012; Moreno-Garcia et al., 2020). The only study identified that even acknowledged the singing voices of individuals with DS, was that undertaken by Pryce (Pryce, 1994) where once again, the focus was on exploring the impact of DS upon speech, but brief mention was made of the fact that most sufferers of Down syndrome “could not sing at all” (Pryce, 1994, p. 10).

Although the literature as regards the impact of DS upon vocal production relates only to the spoken voice, there are common elements as regards production of spoken voice and singing voice, including common features of vibration, resonance and articulation. (Lindblom and Sundberg, 2007; Rakerd, 2019; Marczyk et al., 2022). This has enabled the author, as an experienced vocal tutor, familiar with the mechanics of vocal production, to consider the nature of challenges affecting DS speech explored by the literature, and to make reasonable deductions and develop opinions as to how this information might be transferred and applied to challenges encountered by DS singers.

For example, the information arising out of the studies showing that DS individuals do not have lower voices than their healthy contemporaries but that their “lower” pitch is in fact explained by the inaccurate perception of their pitch by the listener who is misled by the typical pitch instability and rough tonal quality of DS individuals (Pentz and Gilbert, 1983; Pryce, 1994) gives rise to interesting points of consideration in the teaching studio. Is it possible that vocal tutors are also guilty of assuming that the perceived low speaking voice of the DS student, necessarily assigns them to a certain limited vocal range when singing? Could exercises which vocal tutors regularly use to improve intonation in healthy singers – for example slides to help accurately perceive the distance between notes, or pivot note exercises (finding a pivot note from which the singer practises accurately moving up and down consecutive semi tones) to develop muscle memory for notes and intervals – be successfully utilised by DS singers to improve their pitch instability and accuracy and thereby reveal a greater vocal range, perceptible to the listener? The evidence of negative

perception of the DS voice (Michel and Carney, 1964; Hollien and Copeland, 1965), is perhaps indicative of prejudice, conscious or not, towards singers with DS, and acknowledgement of this possibility may encourage a tutor to be more open minded about the possibilities of the DS singer. As is the case with all healthy singers, every DS voice is unique, in spite of typical challenges (O'Leary et al.,2019) and, it is submitted that it is desirable for vocal tutors to refrain from assumption and prejudice when tutoring a singer with DS and instead seek a tailored approach to their tuition, using their knowledge, expertise and experience to assist DS students navigate the individual issues affecting their singing voice. To this end the author has benefitted from the literature review undertaken, by enhancing existing knowledge which can now be used in the teaching studio.

In a similar vein, if the abnormally small oral cavities and shorter jawbone and palate of the DS singer make resonance and articulation challenging, can vocal tutors utilise or adapt common exercises to focus on these features when singing, for example, encouraging repetition of vowel practice and observance of increasing intra-oral spaces, supported by gentle oral stretching exercises, to improve clarity and tone? Implementation, directed by the vocal tutor, of common respiratory exercises, undertaken at an appropriate pace and intensity, may assist DS singers to achieve connected and supported breath management.

The author's deliberations regarding the possibilities of DS vocal training have arisen from information revealed in the literature reviewed, which again leads to the conclusion that comprehensive and structured analysis of primary research studies - even if not directly relevant to the enquiry posed - can assist to inform and inspire the vocal tutor of DS singers, understanding their challenges and providing more focussed and sensitive tuition. The author has already adjusted her teaching approach towards DS students, based upon research previously undertaken on this topic. The creation of simple but effective exercises, fun to do, and demonstrated upon the walls of the teaching studio by large scale, brightly coloured posters to engage DS students who might suffer from other incumbrances such as weak vision or compromised ability to understand or concentrate, has proven effective. It is anticipated that further adjustments will be made to the author's teaching methods of DS students, following the completion of this latest, more comprehensive research, with focus directed, for example, upon exercises for improving pitch stability and articulation, and other exercises targeted at strengthening pharyngeal muscles - yawns, effortful swallows, resistive tongue exercises and the like. The aim will be to incorporate more formality and routine into a section of the lessons taught to DS students, encouraging regular vocal exercise and monitoring, over time, the impact of this upon their vocal production and singing technique. The implementation of these initiatives will be carefully considered, with emphasis on light hearted delivery and demonstration, to maintain engagement and cooperation during lessons.

Finally another important matter, brought to prominence by this research, is the continuing lack of literature dealing directly with challenges faced by DS individuals when singing. The lack of information and data is most striking in the field of vocal pedagogy and, in view of the many proven benefits of singing for all individuals (Stacy et al, 2002; Keeler et al., 2015; Stegemoller et al, 2017), the known connection between music/singing and improvement of certain symptoms of DS (Wylie, 2006; Pienaar, 2012; Moreno-Garcia et al., 2020) and the fact that singing has long been used a tool in DS speech therapy (Cohen, 1992; Schlaug et al., 2010; Wan et al., 2010; Morena-Garcia et al., 2020), this lack of data is regrettable and should be addressed, to avoid discrimination and lack of professional assistance for DS singers, who would benefit from being better supported in this art form, as information collected from this review indicates.

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APPLICATION FOR ETHICAL APPROVAL

In order for research to result in benefit and minimise risk of harm, it must be conducted ethically. A researcher may not be covered by the University's insurance if ethical approval has not been obtained prior to commencement.

The University follows the OECD Frascati manual definition of **research activity**: "creative work undertaken on a systematic basis in order to increase the stock of knowledge, including knowledge of man, culture and society, and the use of this stock of knowledge to devise new applications". As such this covers activities undertaken by members of staff, postgraduate research students, and both taught postgraduate and undergraduate students working on dissertations/projects.

The individual undertaking the research activity is known as the "principal researcher".

Ethical approval is not required for routine audits, performance reviews, quality assurance studies, testing within normal educational requirements, and literary or artistic criticism.

Please read the notes for guidance before completing ALL sections of the form.

This form must be completed and approved prior to undertaking any research activity. Please see Checklist for details of process for different categories of application.

Delete the Guidance Notes at the end of the form BEFORE submitting your application

SECTION A: About You (Principal Researcher)

1	Full Name:	Karen Lesley Linker			
2	Tick all boxes which apply:	Member of staff:	☐	Honorary research fellow:	☐
3	Undergraduate Student	☐	Taught Postgraduate Student	☒	Postgraduate Research Student
4	Institute/Academic Discipline/Centre:	WAPPAR			
5	Campus:	Carmarthen			
6	E-mail address:	karenlinkervocalist@outlook.com			
7	Contact Telephone Number:	+852 6606 8043			
For students:					
8	Student Number:	211960			
9	Programme of Study:	MA in Professional Practice (Voice Pedagogy)			
10	Director of Studies/Supervisor:	Debbie Winter			

SECTION B: Approval for Research Activity

1	Has the research activity received approval in principle? (please check the Guidance Notes as to the appropriate approval process for different levels of research by different categories of individual)	YES	☒	NO	☐
					Date
2	If Yes, please indicate source of approval (and date where known):	Research Degrees Committee	☐		
3	<i>Approval in principle must be obtained from the relevant source prior to seeking ethical approval</i>	Institute Research Committee	☐		
4		Other (write in) WAPPAR Ethics Board Representative	☒		

SECTION C: Internal and External Ethical Guidance Materials

Please list the core ethical guidance documents that have been referred to during the completion of this form (including any discipline-specific codes of research ethics, and also any specific ethical guidance relating to the proposed methodology). Please tick to confirm that your research proposal adheres to these codes and guidelines.		
1	UWTSD Research Ethics & Integrity Code of Practice	☒
2	UWTSD Research Data Management Policy	☒
3		☐

SECTION D: External Collaborative Research Activity

1	Does the research activity involve collaborators outside of the University?	YES	☐	NO	☒	
2	If Yes, please provide the name of the external organisation and name and contact details for the main contact person and confirmation this person has consented to their personal data being shared as part of this collaboration.					
3	Institution					
4	Contact person name					
5	Contact person e-mail address					
6	Has this individual consented to sharing their details on this form?	YES	☐	NO	☐	
7	Are you in receipt of a KESS scholarship?	YES	☐	NO	☐	
8	Is your research externally funded	YES	☐	NO	☐	
9	Are you specifically employed to undertake this research in either a paid or voluntary capacity?	Voluntary	YES	☐	NO	☐
10		Employed	YES	☐	NO	☐
11	Is the research being undertaken within an existing UWTSD Athrofa Professional Learning Partnership (APLP)	If YES then the permission question below does not need to be answered.	YES	☐	NO	☐
12	Permission to undertake the research has been provided by the partner organisation	(If YES attach copy) (If NO the application cannot continue)	YES	☐	NO	☐

Where research activity is carried out in collaboration with an external organisation

13	Does this organisation have its own ethics approval system?	YES	☐	NO	☐
14	If Yes, please attach a copy of any final approval (or interim approval) from the organisation				

SECTION E: Details of Research Activity

1	Indicative title:	To what extent can a document analysis of primary research studies contribute to enhancing vocal tuition for singers with Down Syndrome, Parkinson's Disease or severe Speech Disorder.		
2	Proposed start date:	06 January 2024	Proposed end date:	02 Sept 2024
3	Introduction to the Research (maximum 300 words) Ensure that you write for a Non-Specialist Audience when outlining your response to the three points below: • Purpose of Research Activity • Proposed Research Question • Aims of Research Activity • Objectives of Research Activity Demonstrate, briefly, how Existing Research has informed the proposed activity and explain • What the research activity will add to the body of knowledge • How it addresses an area of importance.			
4	Purpose of Research Activity The purpose of this research is to: 1) Conduct a reflective review of the impact of the peer-reviewed material collated in previous modules on my practice with students who experience Down Syndrome, Parkinson's disease and Stammering. 2) Identify gaps in my knowledge and peer-reviewed literature that could further develop my understanding and aid the creation of teaching tools. 3) Use document analysis and thematic analysis to: a) Carefully select relevant new literature by creating a methodical inclusion/exclusion criterion. b) Rigorously coding the selected literature to thoroughly evaluate the peer-reviewed material. 4) To evaluate how the literature could impact my professional practice and consider the ethical implications of applying the knowledge to practice. The onus of the project is on the evaluation of literature only. Data on my students will not be collected. (this box should expand as you type)			
5	Research Question To what extent can a document analysis of primary research studies contribute to an evidence-based vocal training framework for singers with Down Syndrome, Parkinson's Disease or severe Speech Disorder. (this box should expand as you type)			
6	Aims of Research Activity The overarching aim of the research is to build on further research and inform my practice with new knowledge acquired by conducting a desktop review. The long-term aim is to build a teaching framework and create teaching tools.			

	(this box should expand as you type)
7	Objectives of Research Activity 1) Conduct a reflective review to evaluate achievements, gaps in my knowledge and further areas of desktop research. 2) Explore methodology to create a robust inclusion/exclusion criterion. 3) Use both the Document Analysis framework to ensure quality literature is explored. 4) Use Thematic Analysis to code the literature and engage in a thorough evaluation. 5) Evaluate the ethical implications for practice application. 6) Evaluate the practical implications for the teaching studio. It may allow for the design of a teaching framework word count permitting. (this box should expand as you type)
8	Proposed methods (maximum 600 words) Provide a brief summary of all the methods that may be used in the research activity, making it clear what specific techniques may be used. If methods other than those listed in this section are deemed appropriate later, additional ethical approval for those methods will be needed.
9	- Conduct a critical review by drawing on reflective theory. Theories such as Kolb, Gibbs and Brookfield's reflective cycle will provide the scaffolding for me to synthesise previous modules, evaluate the implications for practice and consider the ethical implications for teachers working with sensitive client groups. - I may include my case notes and other reflective journaling data here. - Draw on methodologies used by the Campbell group and the Cochrane collaboration when conducting a qualitative evidence synthesis to create a robust inclusion/exclusion criteria. For example, the CASP and other similar frameworks could be adapted to suit the needs of the project. - Interrogate the literature through the application of Braun and Clarke's thematic analysis. I will use the framework to code the data and organise it into themes.

10	Location of research activity Identify all locations where research activity will take place.		
11	Private studio in Hong Kong (this box should expand as you type)		
12	Research activity outside of the UK If research activity will take place overseas, you are responsible for ensuring that local ethical considerations are complied with and that the relevant permissions are sought. Specify any local guidelines (e.g. from local professional associations/learned societies/universities) that exist and whether these involve any ethical stipulations beyond those usual in the UK (provide details of any licenses or permissions required). Also specify whether there are any specific ethical issues raised by the local context in which the research activity is taking place, for example, particular cultural and/or legal sensitivities or vulnerabilities of participants.		
13	I am using research methodology to conduct a rigorous desktop review rather than collect participant data. (this box should expand as you type)		
14	Use of documentation not in the public domain: Are any documents NOT publicly available?	NO ☐	☒
		YES ☐	☐
15	If Yes, please provide details here of how you will gain access to specific documentation that is not in the public domain and that this is in accordance with prevailing data protection law of the country in question and England and Wales. (this box should expand as you type)		

SECTION F: Scope of Research Activity

1	Will the research activity include:	YES	NO
2	Use of a questionnaire or similar research instrument?	☐	☒
3	Use of interviews?	☐	☒
4	Use of diaries?	☐	☒
5	Participant observation with their knowledge?	☐	☒
6	Participant observation without their knowledge?	☐	☒
7	Use of video or audio recording?	☐	☒
8	Access to personal or confidential information without the participants' specific consent?	☐	☒
9	Administration of any questions, test stimuli, presentation that may be experienced as physically, mentally or emotionally harmful / offensive?	☐	☒
10	Performance of any acts which may cause embarrassment or affect self-esteem?	☐	☒
11	Investigation of participants involved in illegal activities?	☐	☒
12	Use of procedures that involve deception?	☐	☒
13	Administration of any substance, agent or placebo?	☐	☒
14	Working with live vertebrate animals?	☐	☒

15	Other primary data collection methods, please explain in this box For example, 'focus groups'. Please indicate the type of data collection method(s) in this box and tick the accompany box.		
16	Details of any other primary data collection method: My own teaching notes and reflective observations on my teaching journey. These are part of my everyday teaching practice. (this box should expand as you type)	☒	☐

If NO to every question, then the research activity is (ethically) low risk and **may** be exempt from **some** of the following sections (please refer to Guidance Notes).

If YES to any question, then no research activity should be undertaken until full ethical approval has been obtained.

SECTION G: Intended Participants

1	Who are the intended participants:	YES	NO
2	Students or staff at the University?	☐	☒
3	Adults (over the age of 18 and competent to give consent)?	☐	☒
4	Vulnerable adults?	☐	☒
5	Children and Young People under the age of 18? (Consent from Parent, Carer or Guardian will be required)	☐	☒
6	Prisoners?	☐	☒
7	Young offenders?	☐	☒
8	Those who could be considered to have a particularly dependent relationship with the investigator or a gatekeeper?	☐	☒
9	People engaged in illegal activities?	☐	☒
10	Others (please identify specifically any group who may be unable to give consent) please indicate here and tick the appropriate box.	☐	☐
11	Other – please indicate here: (this box should expand as you type)		

12	Participant numbers and source Provide an estimate of the expected number of participants. How will you identify participants and how will they be recruited?	
13	How many participants are expected?	N/A (this box should expand as you type)
14	Who will the participants be?	N/A (this box should expand as you type)
15	How will you identify the participants?	N/A (this box should expand as you type)

16	Information for participants:	YES	NO	N/A
17	Will you describe the main research procedures to participants in advance, so that they are informed about what to expect?	☐	☐	☒
18	Will you tell participants that their participation is voluntary?	☐	☐	☒
19	Will you obtain written consent for participation?	☐	☐	☒
20	Will you explain to participants that refusal to participate in the research will not affect their treatment or education (if relevant)?	☐	☐	☒
21	If the research is observational, will you ask participants for their consent to being observed?	☐	☐	☒
22	Will you tell participants that they may withdraw from the research at any time and for any reason?	☐	☐	☒
23	With questionnaires, will you give participants the option of omitting questions they do not want to answer?	☐	☐	☒
24	Will you tell participants that their data will be treated with full confidentiality and that, if published, it will not be identifiable as theirs?	☐	☐	☒
25	Will you debrief participants at the end of their participation, in a way appropriate to the type of research undertaken?	☐	☐	☒
26	If NO to any of above questions, please give an explanation			
27	(this box should expand as you type)			

28	Information for participants:	YES	NO	N/A
29	Will participants be paid?	☐	☒	☒
30	Is specialist electrical or other equipment to be used with participants?	☐	☐	☒
31	Are there any financial or other interests to the investigator or University arising from this study?	☐	☒	☐
32	Will the research activity involve deliberately misleading participants in any way, or the partial or full concealment of the specific study aims?	☐	☐	☒
33	If YES to any question, please provide full details			
34	(this box should expand as you type)			

SECTION H: Anticipated Risks

1	Outline any anticipated risks that may adversely affect any of the participants, the researchers and/or the University, and the steps that will be taken to address them. If you have completed a full risk assessment (for example as required by a laboratory, or external research collaborator) you may append that to this form.		
2	Full risk assessment completed and appended?	Yes ☐ No ☒	
3	Risks to participants For example: emotional distress, financial disclosure, physical harm, transfer of personal data, sensitive organisational information		
4	Risk to Participant: In relation to the reflective review, I may draw on my existing teaching records and notes. These notes are personal and could reveal the identify my students if mishandled. (this box should expand as you type)	How will you mitigate the Risk to Participant My teaching records and observations will be anonymised. (this box should expand as you type)	
5	If research activity may include sensitive, embarrassing or upsetting topics (e.g. sexual activity, drug use) or issues likely to disclose information requiring further action (e.g. criminal activity), give		

	details of the procedures to deal with these issues, including any support/advice (e.g. helpline numbers) to be offered to participants. Note that where applicable, consent procedures should make it clear that if something potentially or actually illegal is discovered in the course of a project, it may need to be disclosed to the proper authorities			
	N/A (this box should expand as you type)			
6	Risks to investigator For example: personal safety, physical harm, emotional distress, risk of accusation of harm/impropriety, conflict of interest			
	Risk to Investigator: Problems caused by the challenges of cross-disciplinary research. It includes: a) the lack of relevance to my field of enquiry. b) Complexity caused by the medical nature of the literature c) Ethical problems that could arise for future application (this box should expand as you type)	How will you mitigate the Risk to Investigator: - Use evaluative frameworks to consider the implications of future application both ethical and practical. - Engage in an unbiased evaluation of the literature including its limitations and relevance to the teaching studio - Be transparent about future potential and limitations. (this box should expand as you type)		
7	University/institutional risks For example: adverse publicity, financial loss, data protection			
	Risk to University: Adverse publicity/damage to reputation (this box should expand as you type)	How will you mitigate the Risk to University: Professional conduct at all times Diligent review of literature and sensitive treatment of details of how theory was applied to practice. Ensuring complete anonymity of students referred to in my teaching notes and observations. (this box should expand as you type)		
8	Disclosure and Barring Service			
9	If the research activity involves children or vulnerable adults, a Disclosure and Barring Service (DBS) certificate must be obtained before any contact with such participants.	YES	NO	N/A
10	Does your research require you to hold a current DBS Certificate?	☐	☒	☐

SECTION I: Feedback, Consent and Confidentiality

1	Feedback What de-briefing and feedback will be provided to participants, how will this be done and when?
	N/A (this box should expand as you type)
2	Informed consent Describe the arrangements to inform potential participants, before providing consent, of what is involved in participating. Describe the arrangements for participants to provide full consent before data collection begins. If gaining consent in this way is inappropriate, explain how consent will be obtained and recorded in accordance with prevailing data protection legislation.
	N/A (this box should expand as you type)
3	Confidentiality / Anonymity

	Set out how anonymity of participants and confidentiality will be ensured in any outputs. If anonymity is not being offered, explain why this is the case.
	Review of previous application of theory to practice will remain strictly anonymous and only personal notes will be used as evidence. (this box should expand as you type)

SECTION J: Data Protection and Storage

In completing this section refer to the University's Research Data Management Policy and the extensive resources on the University's Research Data Management web pages (<http://uwtsd.ac.uk/library/research-data-management/>).

1	Does the research activity involve personal data (as defined by the General Data Protection Regulation 2016 "GDPR" and the Data Protection Act 2018 "DPA")?	YES	NO
	<i>"Personal data" means any information relating to an identified or identifiable natural person ('data subject'). An identifiable natural person is one who can be identified, directly or indirectly, in particular by reference to an identifier such as a name, an identification number, location data, an online identifier or to one or more factors specific to the physical, physiological, genetic, mental, economic, cultural or social identity of that natural person.</i>	☐ x	☐
2	If YES, provide a description of the data and explain why this data needs to be collected:		
	I use an identification code for my teaching notes to avoid using personal names. (this box should expand as you type)		
3	Does it involve special category data (as defined by the GDPR)?	YES	NO
	<i>"Special category data" means sensitive personal data consisting of information as to the data subjects' – (a) racial or ethnic origin, (b) political opinions, (c) religious beliefs or other beliefs of a similar nature, (d) membership of a trade union (within the meaning of the Trade Union and Labour Relations (Consolidation) Act 1992), (e) physical or mental health or condition, (f) sexual life, (g) genetics, (h) biometric data (as used for ID purposes).</i>	☐ x	☐
4	If YES, provide a description of the special category data and explain why this data needs to be collected:		
	My teaching notes contain sensitive information relating to Parkinson's, Down Syndrome and Stammering. These need to be drawn upon to conduct a thorough reflective analysis of my practice. It is important I use them so that the project is appropriately situated within my professional practice. (this box should expand as you type)		
5	Will the research activity involve storing personal data and/or special category data on one of the following:	YES	NO
6	Manual files (i.e. in paper form)?	☐	☒
7	University computers?	☐	☒
8	Private company computers?	☐	☒
9	Home or other personal computers?	☒	☒
10	Laptop computers/ CDs/ Portable disk-drives/ memory sticks?	☐	☒

11	"Cloud" storage or websites?	☒	☐
12	Other – specify:	☐	☒
13	For all stored data, explain the measures in place to ensure the security of the data collected, data confidentiality, including details of password protection, encryption, anonymisation and pseudonymisation:		
	My personal teaching notes will be anonymised and stored on computer with an encrypted password. I use secure cloud-based storage for all my notes that is password protected. I will use a unique identifier rather than naming my students. <i>(this box should expand as you type)</i>		
14	All Data Storage		
15	Will the research activity involve any of the following activities:	YES	NO
16	Electronic transfer of data in any form?	☐	☒
17	Sharing of data with others at the University?	☐	☒
18	Sharing of data with other organisations?	☐	☒
19	Export of data outside the European Union or importing of data from outside the UK?	☐	☒
20	Use of personal addresses, postcodes, faxes, emails or telephone numbers?	☐	☒
21	Publication of data that might allow identification of individuals?	☐	☒
22	Use of data management system?	☐	☒
23	Data archiving?	☐	☒
24	If YES to any question, please provide full details, explaining how this will be conducted in accordance with the GDPR and DPA (and/or any international equivalent):		
	n/a <i>(this box should expand as you type)</i>		
25	List all who will have access to the data generated by the research activity:		
	Principal researcher <i>(this box should expand as you type)</i>		
26	List who will have control of, and act as custodian(s) for, data generated by the research activity:		
	Principal researcher Tutor as regards drafts of the principal researcher's work during the module <i>(this box should expand as you type)</i>		
27	Give details of data storage arrangements, including security measures in place to protect the data, where data will be stored, how long for, and in what form. Will data be archived – if so how and if not why not.		
	Only my teaching notes will be used and these are all stored on my laptop that is password protected and encrypted with a password. Once I no longer teach the student all data is removed. <i>(this box should expand as you type)</i>		
28	Please indicate if your data will be stored in the UWTSD Research Data Repository (see https://researchdata.uwtسد.ac.uk/). If so please explain. <i>(Most relevant to academic staff)</i>		
	n/a		

	<i>(this box should expand as you type)</i>		
29	Confirm that you have read the UWTSD guidance on data management (see https://www.uwtsd.ac.uk/library/research-data-management/)	YES	☒
		NO	☐
30	Confirm that you are aware that you need to keep all data until after your research has completed or the end of your funding	YES	☒
		NO	☐

SECTION K: Declaration

31	The information which I have provided is correct and complete to the best of my knowledge. I have attempted to identify any risks and issues related to the research activity and acknowledge my obligations and the rights of the participants. In submitting this application I hereby confirm that I undertake to ensure that the above named research activity will meet the University's Research Ethics and Integrity Code of Practice which is published on the website: https://www.uwtsd.ac.uk/research/research-ethics/		
	Signature of applicant:	<i>Karen Lister</i>	Date: 22 Nov 2023

For STUDENT Submissions:

32	Director of Studies/Supervisor:	Laura James	Date: 15.1.24
33	Signature:	<i>Laura James</i>	

For STAFF Submissions:

34	Academic Director/ Assistant Dean:		Date:
35	Signature:		

Checklist: Please complete the checklist below to ensure that you have completed the form according to the guidelines and attached any required documentation:

☒	I have read the guidance notes supplied before completing the form.
☒	I have completed ALL RELEVANT sections of the form in full.
☒	I confirm that the research activity has received approval in principle
☐	I have attached a copy of final/interim approval from external organisation (where appropriate)
☐	I have attached a full risk assessment (and have NOT completed Section H of this form) (where appropriate) ONLY TICK IF YOU HAVE ATTACHED A FULL RISK ASSESSMENT
☒	I understand that it is my responsibility to ensure that the above named research activity will meet the University's Research Ethics and Integrity Code of Practice.
☒	I understand that before commencing data collection all documents aimed at respondents (including information sheets, consent forms, questionnaires, interview schedules etc.) must be confirmed by the DoS/Supervisor, module tutor or Academic Director.
☒	I have deleted the guidance notes before submitting the PG2 for consideration

RESEARCH STUDENTS AND STAFF ONLY

All communications relating to this application during its processing must be in writing and emailed to pgresearch@uwtsd.ac.uk, with the title 'Ethical Approval' followed by your name. You will be informed of the outcome of your claim by email; therefore it is important that you check your University and personal email accounts regularly.

STUDENTS ON UNDERGRADUATE OR TAUGHT MASTERS PROGRAMMES should submit this form (and receive the outcome) via systems explained to you by the supervisor/module leader.

To what extent can an analysis of primary research studies identify the challenges faced by a Down Syndrome singer and contribute to improving vocal training for this demographic

Appendix 2

Glossary Of Medical Terms

Term	Meaning		
Aneuploidy/Anaeuploid disorder	A genetic disorder where the total number of chromosomes doesn't equal 46		
Collagen type I	The most abundant fibrillar collagen found in almost all connective tissues		
Collagen type III	Another fibrillar collagen producing elastic network		
Collagen type VI	a type of collagen primarily associated with the extracellular matrix of skeletal muscle.		
Craniofacial	Relating to the skull and the face		
Dysautonomia	A nervous system disorder that disrupts autonomic body processes		
Hypermobility	Movement of joints beyond normal range		
Hypotonia	Decreased muscle tone and muscle weakness		
Laryngomalacia	Soft larynx caused by floppiness of the laryngeal tissues above the vocal cords		
Lobar pneumonia	A type of pneumonia characterized by the infection and inflammation of one or more lobes of the lung		
Macroglossia	Enlarged tongue		
Mid-face hypoplasia	Where the middle region of the face is underdeveloped or recessed, including the upper jaw, cheekbones and eye sockets.		

To what extent can an analysis of primary research studies identify the challenges faced by a Down Syndrome singer and contribute to improving vocal training for this demographic

Appendix 2

Glossary Of Medical Terms

Nasopharynx	The top part of the throat		
Oropharynx	The middle part of the throat behind the mouth		
Proprioceptive neuromuscular facilitation	A stretching technique to improve range of motion		
Pulmonary hypertension	High blood pressure in the blood vessels that supply the lungs		
Pulmonary odema	A condition in which too much fluid accumulates in the lungs, interfering with a person's ability to breathe normally		
Pulmonary parenchymal abnormality	Decreased vessel diameter or vascular compression		
Restrictive lung disorder	Any disorder which leads to a decrease in the total volume of air that the lungs are able to hold		
Stridor	An abnormal, high-pitched respiratory sound produced by irregular airflow in a narrowed airway		
Subglottic stenosis	A narrowing of the airway below the vocal cords (subglottis) and above the trachea		
Temporomandibular joint disorder	A group of more than 30 conditions that cause pain and dysfunction in the jaw joint and muscles that control jaw movement		
Tracheal bronchus	A congenital anomaly causing a bronchus outgrowth from the right tracheal wall to the the upper lung lobe		

To what extent can an analysis of primary research studies identify the challenges faced by a Down Syndrome singer and contribute to improving vocal training for this demographic

Appendix 2

Glossary Of Medical Terms

Tracheal ring deformity	A birth defect that causes the tracheal ring to be O-shaped instead of C-shaped		
Tracheal stenosis	A narrowing of the trachea that interferes with normal breathing		
Tracheomalacia	Collapse of the airway when breathing		
Vagus nerve	The longest nerve of the autonomic nervous system in the human body		
Velum	The soft palate at the top of the mouth		
Williams Syndrome	A genetic disorder caused by the deletion of genetic material on chromosome 7		