

TITLE PAGE

Assignment Submission Form

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Assignment Title: Independent Study Module (Extended) Module Code: BMPF7013

Submission Date: 22nd May 2024

You work will not be marked unless it is accompanied by this statement.

I confirm that this is my original work and that I have adhered to the university policy on plagiarism.

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INDEX

Portfolio

Introduction and rationale.....	p.2
Planned programme of study.....	p.3
Aims and objectives	p.3
Research objectives.....	p.3
Literature Review Progress.....	p.4
Eligibility Criteria.....	p.6
Search Terms.....	p.8
Critical Reflection.....	p.9

Written assignment

Recognising Joint Hypermobility.....	p.13
Recognising Hypermobility Spectrum Disorder.....	p.15
Genetic Basis of Joint Hypermobility.....	p.16
The Role of Collagen in Connective Tissues.....	p.16
Connective Tissue Structures of the Larynx and Vocal Tract.....	p.18
Cricoarytenoid joint.....	p.19
Cricothyroid joint.....	p.21
Extrinsic muscles of the larynx.....	p.21
The Extracellular Matrix of the Vocal Folds.....	p.23
Further effects of Hypermobility on muscles.....	p.23
Further Research Questions.....	p.24

Appendices.....

Bibliography.....

Portfolio

An Introduction and Rationale

Most of my practice lies in training musical theatre practitioners in higher education. Within the institution I teach in, 84% of students are female-born; these percentages are similar across the musical theatre training industry (Clifford, 2021). Hypermobility is more prominent in women than men (Tinkle, 2020; Janssin et al., 2004; Clinch et al., 2011). Not only this, but hypermobility has also been found to be more prevalent in performers, including dancers, gymnasts, and musicians. (HMSA, 1992; NHS, 2020; Jeffery et al., 2022) Therefore, it is no coincidence that as a Vocal Coach specialising in Musical Theatre training in Higher Education, I have experienced a trend in students presenting with hypermobility.

In my previous research, I aimed to explore if and how hypermobility affects the voice and from this research, I developed some interventions that may help and support students who experience hypermobility (see Appendix 1). My literary review revealed that the literature concerning hypermobility and the voice is very limited. The most relevant search criteria to my research question brought up only three relevant papers in the Journal of Voice and none in Pubmed, SAGE, Medline, the Cochrane Library and Google Scholar. During the process, my research intention changed from looking at the direct effect of hypermobility on voice production to the indirect effects caused by the impact of hypermobility on posture. It became prevalent that the literature was lacking in the field of vocology, and instead, it leaned heavily on the fields of medicine and sports science and focused on the effect of hypermobility on the joints. This literature was mainly directed towards muscular imbalances that affect posture and alignment (Clarke, Holmes, Feeley, et al., 2013), which has been recognised as significantly impacting voice production in singing pedagogy for many years. Many vocologists have emphasised the importance of good posture in voice production, such as Rubin, Frederick, Schneider and Titze, to name a few (Rubin et. al, 2004; Arboleda and Frederick, 2008; Schneider et.al, 1997). From this research, I generated a compendium of exercises (see appendix 1) that may help strengthen the proprioception of joints affected by hypermobility that are considered crucial for good posture (Titze, 1982) such as the knees, shoulders, neck (Rubin et al., 2004) and core.

My previous research gave me a foundation of knowledge of Hypermobility Spectrum Disorder (HSD), which is the term used to comprise the various extents of people who present with hypermobility (Lowth and Tidy, 2019). This knowledge has been invaluable to

my teaching studio, allowing me to recognise hypermobile tendencies in students and support them with targeted exercises. However, I have merely touched the surface of the literature relating to hypermobility. Grahame (2013) describes HSD as ‘misunderstood, overlooked, misdiagnosed and mistreated’ in clinical settings. I strive not to fall victim to this shortfall whilst maintaining best practice, using my knowledge to support students and help direct them to appropriate practitioners and not take on the role of a healthcare advisor. I wish to look further into how HSD directly affects the vocal mechanism. This will also give me the opportunity to create teaching frameworks for teachers of hypermobile singers in my future studies.

The rationale for this study is to obtain a higher knowledge of the extrinsic and intrinsic muscles of the vocal mechanism and their roles within vocal production. I have a sufficient understanding of the larynx, its cartilaginous formations and muscular constructs. Still, I need to have a higher depth of comprehension of the connective muscles to begin to explore whether a singer can experience hypermobility in the larynx. This will ensure I can implement best practice by applying evidence-based exercises and advice in my teaching.

Planned programme of study

Aims and Objectives

- To analyse new research in hypermobility and singers
- To understand how hypermobility affects muscle tissue.
- To better understand the connective intrinsic muscles of the larynx.
- To better understand the connective extrinsic muscles of the larynx.
- To critically evaluate if the larynx is at risk from Hypermobility spectrum disorder.
- To identify if hypermobility in the larynx can alter voice production.
- To extensively collate and identify hypermobility literature.

Research objectives

- By the end of the project, I will be able to articulate the complex muscular structures of the larynx confidently.
- I will have strengthened my ability to look through a critical lens and apply this to my research.

- I will have explained various types of hypermobility clearly and concisely, allowing vocal teachers to use this information in their observations.

Here is the chart for my proposed timeline for my research.

Task	Dec				Jan				Feb				March				April				May			
Topic Agreed with Tutor																								
Aims and Objective to be identified																								
Book, journal and website search																								
Book, journal and website review																								
Draft study																								
Rewrites																								
Submission																								

Literature Review Process

In this literature review, I will critically evaluate the documents that arise from my search criteria to deepen my understanding whilst gaining new knowledge (Shulman, 1999).

Rigorous literature review plays a vital role in the continuous teaching and learning process and gives access to best practice through evidence-informed teaching (Boyer 1990).

Shulman (2000) emphasised that research is “an essential facet of good teaching” and should be an “expected repertoire of scholarly practice” and is supported by the work of Huber and Morreale (2002), who agree that this, in turn, will allow for a higher “quality of student learning”(p2).

I have chosen to begin this literature review using my previous research as a springboard and continuing along the journey of Bloom's taxonomy of learning. My review will be centred around research questions that seek to analyse and examine the questions that arose during my initial research on hypermobility and the singer based on practice-based observations.

- 1.) What causes hypermobility in joints? Are there any trends in the genetic makeup of people who suffer from joint hypermobility?
- 2.) Can the effects of hypermobility affect the larynx and vocal folds?
- 3.) What are the intrinsic connective structures of the larynx?
- 4.) What are the extrinsic connective structures of the larynx?

In this literature review, I will be looking principally in the field of medicine. Though medicine is not my field, I believe it will provide rigorous and trustworthy data that will allow me to understand the genetic structure of hypermobility and anatomy to actualise this research and both my research and practice moving forward. Previously, the literature concerning hypermobility in the world of vocal pedagogy was lacking. I feel it is essential for me to revisit the previous search criteria (see Appendix 2) to ensure I still maintain the most up-to-date information possible in these areas.

I thrive in a fluid research setting, and so I find the iterative research process most fruitful for my undertaking. This freedom to move interchangeably between data collection and analysis gives an opportunity for new insights rather than rigid linear steps (Twohig, 2024). I will ensure that I comprehensively document all changes in the research process to allow for full transparency. (Tuval-Mashiach, Rivka, 2016)

For this research I have chosen to undertake a systematic literature review, as this method is commonly used to synthesise scientific evidence in medical research (Lame, 2019) in a way that is unequivocal and replicable. Systematic literature reviews minimise bias by following a four-step protocol that begins with the aims and objectives of the review. Then, it sets out clearly defined inclusion and exclusion criteria for the studies to be reviewed. Followed by defining how these studies will be identified and, lastly, the plan of analysis. Following these steps will minimise bias, support the rationale of the study, and help me structure my research, which I can find challenging at times.

Nightingale (2009) states that to avoid bias within a systematic literature review, these protocols should not be altered within the process unless absolutely necessary. Though this may hinder an iterative research process, I believe it will still aid in creating a robust research question developed through rigorous, trustworthy, evidence-based research. Borry et al. (2006) rightly state that healthcare providers and those who apply skills within their own practice acquired from these providers' expertise have a moral obligation to work from the most reliable and up-to-date research. This protocol was birthed from the work of Cochrane and the Cochrane Collaboration (Chandler & Hopewell, 2013) and will influence my inclusion criteria.

Eligibility Criteria

Establishing thorough inclusion and exclusion criteria heightens the study's validity by minimising the risk of biased conclusions (Patino, 2018). However, these criteria must be evaluated carefully to determine how they will impact the study's external validity. For this study, I will use the following electronic databases: PubMed, National Library of Medicine, ScienceDirect, and Cochrane Library.

Inclusion criteria

My preliminary research has shown that the nosology for hypermobility was updated in 2017 (Caroll, 2023; Atwell et al., 2021). Due to this, I will be looking at the literature concerning hypermobility published from 2017 onwards. This is important to facilitate heightened reliability in the study by avoiding out-of-date research (Barroso, Sandelowski, Voils, 2006). However, I do believe that there may be useful insight existing before this time, and limiting myself to the past seven years of research may exclude these from my study. If I find papers that precisely match my search criteria for hypermobility, I will approach them through a historical lens. While researching genetics, anatomy, microbiology and vocal pedagogy, I will use publications from the past 30 years and cross-check between them to ensure I am reviewing the most relevant studies and using the most recent terminology used (Borrt et al., 2006) in medicine and science.

I will use only peer-reviewed literature in this study due to the need for evidence-based research in science, medicine and voice pedagogy. I will use primary sources, such as scholarly books and peer-reviewed journals, to achieve this. These sources are subjected to a meticulous review process by specialists within their discipline to guarantee rigour and transparency before publication (Prager et al., 2018). Preliminary searches have shown that I will need to take material from multiple fields to explore the possible risks of hypermobility on the voice fully. The foremost fields I must explore include medicine, rheumatology, microbiology, Otolaryngology, and vocal pedagogy. The journals I will review from these fields include, but are not limited to;

- Microbiology
 - Open Biology
 - Biomolecules
- Medicine
 - American Journal of Medical Genetics

- British Medical Bulletin
- Journal of the American Board of Family Medicine
- Scientific Reports
- BMC Sports Science, Medicine and Rehabilitation
- Rheumatology
 - Rheumatology
 - Rheumatology and Immunology Research
 - Dermatology and Rheumatology
- Vocal Pedagogy
 - Journal of Voice
 - The Voice and Speech Review
 - Journal of Singing
- Otolaryngology
 - Indian Journal Otolaryngology, Head, Neck, Surgery
 - Clinical Otolaryngology
 - The Journal of Laryngology and Otology

Hypermobility Spectrum Disorder and Ehlers-Danlors Syndrome underwent reclassification and nomenclature in 2017 (Carroll, 2017), to further distinguish between the two syndromes. However, joint hypermobility remains a central symptomatic characteristic of both syndromes. Due to this, I will be including EDS or hypermobile EDS in my search criteria.

Studies examining human body characteristics must identify a participant's full demographic (Prager et al., 2018). Failing to do so will diminish the replicability of a study, which will have a negative impact on its reliability. I will not be limiting my review process by means of sex or gender. Though this may seem unnecessary to state, failing to do so by my inclusion criteria would compromise this study's rigour (Clayton, 2016;2018). Though I have observed sexual dimorphism in favour of hypermobility in female-born individuals, omitting studies with male participants would lead to bias in my research. Within my academic institution, there are six female students, for every one male. It wouldn't be wrong to assume that the reason I am observing more hypermobility in females than males is due to the higher ratio of females within the institution. These observations are supported by findings from the preliminary search that show hypermobility is more prominent in women than men (Tinkle, 2020; Janssin et al., 2004; Clinch et.al, 2011). Further investigation has found impactful studies with male participants that can provide exciting insights and aid in rigorous research.

Following Prager et al.'s (2018) justification for providing a full demographic, I must consider the age range of the participants I wish to examine in my literature review. I will be looking at

vocal anatomy and the microbiology of the human body, so the studies I review must contain participants with fully developed musculoskeletal systems. To achieve this, I will review studies with only post-pubescent participants; this is often categorised as adults or 18+ in search criteria, but I will include studies with females of the age 16 and above, and this is the age females are considered to reach full physical maturity (Breehl, Caban, 2023). Carroll (2023) states that an individual's Beighton score can be influenced by ethnicity; this is supported by research from Morlino and Castori (2023) and Castori et al. (2017). I will not limit my search to specific ethnic study groups. Doing this would create bias and exclude possible insights in my study.

Exclusion Criteria

In a population study, Remvig, Jensen and Ward (2007) and Seow, Chow and Khong (1999) found that joint hypermobility was more common in Inuit, African and Asian people. Findings also showed intermedial numbers of hypermobility in North, Central and South American populations and smaller numbers in European and Australian populations. These findings show a strong influence of ethnicity on joint hypermobility (Carroll, 2023; Morlino and Castori, 2023; Castori et al., 2017) but also prove that hypermobility is not exclusive to any ethnical population. In this desktop study, I will be excluding any literature that focuses on one singular ethnical demographic to avoid population bias. I will be excluding any research that has a sexual bias, as hypermobility is not exclusive to any singular sex.

The overlap of characteristics of Hypermobile Spectrum Disorder and Ehlers-Danlos Syndrome relates to hypermobile-Ehlers-Danlos Syndrome or classic type Ehlers-Danlos syndrome, two of the thirteen sub-categorisations of EDS. The eleven other categories of Ehlers-Danlos Syndrome differ more significantly from Hypermobility Spectrum Disorder and are not appropriate for cross-examination; therefore, they will not be included in the search criteria.

Search Criteria

The initial search criteria have been developed from a preliminary search into the risks of hypermobility of the larynx. I believe this criteria may grow during the research process as I will be looking into fields that may reveal new insights that need further research. This may also include new terminology that may reveal new pertinent research when added to the search criteria. Initially, my search terms revealed a large number of results in my chosen databases. I was unable to minimise this with advanced filters so I used the addition of a

Boolean NOT operator to ensure my exclusion criteria were removed from the results. This helped to minimise the irrelevant articles in my search results and lowered the time it took to manually filter the final results (Dinet, Favart, Passerault, 2004).

Search terminology included;

- Joint hypermobility
 - AND diagnosis, symptoms
- Hypermobility Spectrum Disorder
 - AND diagnosis, symptoms
- Hypermobility
 - AND genetic traits, genetic mutation, molecular structures
- Connective tissue
 - AND structure, function, genetic
- Larynx
 - AND anatomy, intrinsic muscles, extrinsic muscles
- Hypermobility
 - AND voice, singing

In addition to each of these terms, exclusion operators were used to negate unwanted themes in the literature;

- NOT adolescent
- NOT Arthrochalasia EDS(aEDS), Brittle Cornew Syndrome (BCS), Cordia Vascular EDS(cvEDS), Classic-like EDS (clEDS), Dermatosparaxis EDS (dEDS), Kyphoscoliotic EDS (kEDS), Musculocontractural EDS (mcEDS), Myopathic EDS (mEDS), Periodontal EDS (pEDS), Spondylodysplastic EDS (spEDS), Vascular EDS (vEDS)

Critical reflection

This study arose as a direct continuation of my previous research. I found myself reflecting upon where my previous research left me, with defined knowledge of the effects of hypermobility on the alignment and proprioception of the body but with little further knowledge of how hypermobility directly affects the larynx. I believe this set me on the path of Schöns's (1991) four stages of reflection, as I desire to support students with any form of hypermobility and provide resources to give vocal coaches the knowledge to do the same.

Reflecting on my work, I became aware that I was missing crucial knowledge that could help me justify creating this pedagogical tool. To arrive at the 'result' I must consider all the possible 'problems' and continually reflect on how I have actively resolved these problems. Only then can I begin to undertake the challenge of creating my final result.

There is a distinct lack of literature on the effect of hypermobility on the larynx. Tracy (2021) conducted an experiment that explored the experiences of singers diagnosed with Hypermobility Spectrum Disorder. This is the only study of its kind connecting the direct effects of diagnosed Hypermobility Spectrum Disorder on the singer's experience within vocal pedagogy literature. This resulted in a monumental amount of background reading from multiple fields, which, as a dyslexic, I found completely overwhelming. Initially, the task was manageable, using the citation tool Papership to annotate and make notes as I read. But the more articles I read, it became clear that I would need to cover such varying information to achieve my research goals whilst cross-referencing to ensure the rigour and validity of studies. I believe I fell behind whilst using Papership as I was unable to create folders in this programme, which led to disorganisation and time-consuming information searches. On reflection, I will use a different programme in further research and return to using a data management plan, which may help to minimise overwhelm. I will return to Briney's strategy, which I found particularly helpful in past (Briney, 2015).

I believe that after taking a break from my studies and having a rather turbulent personal life on my return to studies that in this time my research skills have weakened. I did not perceive how much material I would need to revisit before I began my research, and as a result, I did not include this in my planned programme of study. Once this issue had arisen, I was too far along my timeframe to correct this mistake. I intend to learn from this mistake and include 'revision' time in my next programme of study.

My timeline was also affected by my preconceived assumptions about where the effects of hypermobility would be most likely in the larynx. Through this research, I found that though my knowledge of the intrinsic musculature of the larynx was very strong, my understanding of extrinsic muscular function needed to be improved. I had assumed that vocal issues arising from hypermobility were far more likely to occur intrinsically, but the literature I found connecting vocal dysphonia to hypermobility was more common in the extrinsic muscles. Due to this assumption, I had planned to approach extrinsic musculature lastly in my research process. Therefore, this realisation that I would need to spend more time on this area of research came late in the process leaving me feeling rushed on an area I wished to

spend more time on. To right this in the future, I will add a preliminary reading stage to my research, and this will allow me to regulate my time better.

I also had an unexpected return to search criteria when I began looking into the genetic basis of joint hypermobility and found new terminology I believed needed to be researched further. At this point, I developed further search criteria to include the new development in my research. These included;

- Extracellular Matrix
 - AND structure, function
- Collagen
 - AND structure, function
- Vocal folds AND larynx
 - AND extracellular matrix, collagen

I regularly found my search criteria giving matching results in the thousands, and manually checking each result would have been impossible. I overcame this problem by using Boolean NOT functions to eliminate recurring unwanted themes, which created a far more refined search that gave more realistic search result numbers.

I believe that I have fallen short on multiple occasions during this research process, which I may have found possible to correct had I managed my time better to allow for further rewrites. Reflecting on this has enlightened me about the specifics of these shortfalls and given me an opportunity to create strategies to resolve them in the future. This has furthered my research skills for future projects.

An in-depth study into whether hypermobility poses a risk to the structures used for phonation in the post-pubescent human body.

Jeffery (2021) conducted an experiment that explored the experiences of 156 professionally trained singers diagnosed with Hypermobility Spectrum Disorder or hypermobile type Ehlers-Danlos Syndrome. Though many reported an advantageous wide vocal range, 65% of participants had experienced vocal issues including; dysphonia, vocal fatigue, breaks in phonation and the inability to achieve volume and maintain modal phonation. This study is currently the only one of its kind and provided crucial insights into hypermobile singers' experiences but left the underlying causes of their experiences to be questioned, therefore leaving the possibilities for interventions by vocal coaches yet to be explored.

The study of Joint Hypermobility (JH) was first documented in rheumatological literature in 1967 by Kirk, Ansell and Bywaters (Malfait et al. 2006) and is described as the ability of a patient to move their limbs beyond the normal parameters along the physiological axes. At this time, it was believed to be purely a musculoskeletal disorder (Grahame, 2013) and was named Joint Hypermobility Syndrome (JHS). Many terms have been used to describe the phenomenon of a joint moving passively and actively beyond the perceived norm, such as joint laxity, double jointedness, or joint hyperlaxity. In this paper, I will be using the term joint hypermobility to accumulate all of these terms. In the last decade, more attention has been brought to the study and classification of hypermobility disorders. During this time, an overlap of symptoms became prevalent between JHS and other hereditary connective tissue disorders (HCTD), such as hypermobility type Ehlers-Danlos Syndrome (historically named type III) (Atwell et al. 2021). This prompted an updated nomenclature of this disorder created from expert opinion. Carroll (2023) argues that this confusion is due to the clinical unfamiliarity, owing to the vague historical classification surrounding the disorder, which has resulted in a multitude of nosology with overlapping symptoms (Castori, Tinkle, Levy, Grahame, Malfait and Hakim. 2017). In 2017, JHS underwent reclassification and was renamed Hypermobility Spectrum Disorder, which contained four subtypes (Carroll, 2023, Atwell et al., 2021). At the same time, type III Ehlers-Danlos Syndrome underwent a change in nosology to help distinguish it from generalised HSD, and it was renamed hypermobile EDS (hEDS). As a result of this new nomenclature, the previously used terms are now considered outdated within the medical community (Castori et al, 2017; Carroll, 2023).

Recognising Joint Hypermobility

It is important to understand that a person can have symptomatic joint hypermobility without having HSD. When working in a close environment such as a vocal studio, having the knowledge to recognise this differentiation could be highly beneficial to a coach's pedagogical toolkit and to students' understanding of their body's parameters. The most recent identification tool comes from Hakim and Graham (2003) in the form of a 5-point questionnaire:

1. Can you now [or could you ever] place your hands flat on the floor without bending your knees?
2. Can you now [or could you ever] bend your thumb to touch your forearm?
3. As a child, did you amuse your friends by contorting your body into strange shapes, or could you do the splits?
4. As a child or teenager, did your kneecap or shoulder dislocate on more than one occasion?
5. Do you consider yourself “double-jointed”?

If a student is able to answer yes or two or more of these questions this would suggest that a teacher must consider the presence of hypermobility (Folci and Capsoni, 2016). In my previous research, I developed the five-point questionnaire to become more specific to the voice studio (see Appendix 3) which is intended to be used during initial consultations with students. This tool is then partnered with the Beighton hypermobility score (Fikree, 2013), which is a 9-point physical test for joint laxity (see Figure 1). It must be noted that this is an initial assessment tool to identify how many joints are affected in a person and not a measure of the degree of laxity in the joint (Atwell, 2021).

The Beighton scoring system has been deemed the most recognised tool for the identification of joint hypermobility in an individual (Castori et al, 2017). If an adult (18 years or older) presents with a score of 4 or more, they are considered hypermobile by the Beighton Score (Carroll, 2023; Morlino and Castori, 2023; Atwell et al, 2021), with 0 considered as the perceived normal range of motion. However, the validity of this scoring system has been questioned in recent years due to a possible influence of ethnicity, age, gender or physical fitness on joint mobility. It also does not show if hypermobility is more

pronounced in the upper or lower limbs or between the dominant or nondominant limbs of a person (Jindal et al. 2016).

Despite this criticism the Beighton scale remains the most routinely used tool and has been reported with good to excellent reliability (Jindal 2016). It is considered a quick and easy tool that can be used with no training (Morlino and Castori, 2023) without the need for physical contact (Carroll, 2023). This makes it the ideal tool for use within the vocal studio and validates its use by vocal coaches who are not necessarily medically or clinically trained.

<u>Specific Joint Laxity</u>	<u>Yes</u>		<u>No</u>
1. Can you touch your forearm with your thumb?	Left	Right	
2. Can you stretch your little finger back more than 90 degrees?	Left	Right	
3. Can you bend your elbows backwards?	Left	Right	
4. Can you bend your knees backwards?	Left	Right	
5. Can you place both hands on the floor without bending your knees?	Yes		No

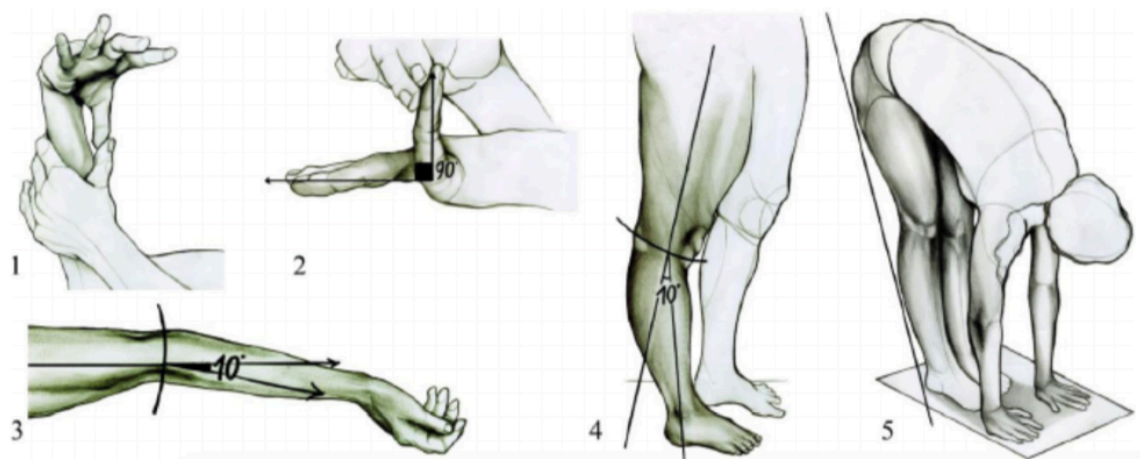


Figure. 1 - Beighton Score for Joint Hypermobility (Beighton et al., 2007)

Recognising Hypermobility Spectrum Disorder

Hypermobility spectrum disorder (HSD) is determined by a score of 4 or more in a person aged 50 or above, 5 or more in a post-pubescent to a 50-year-old person or 6 or above in prepubertal adolescents on the Beighton Score. They must also present with at least one secondary musculoskeletal (MCS) expression, such as altered proprioception, pain, joint realignment, Kyphosis or Scoliosis (Atwell, 2021). In my five years of teaching voice within musical theatre training in higher education, I have taught students who present with each of these musculoskeletal issues. I believe this is not an uncommon observation in this environment. This may be due to the higher percentage of females to males training in this field at this level (Clifford, 2012) and the fact that hypermobility is more prevalent in women than men (Janssin et al., 2004; Clinch et al., 2011; Tinkle, 2020; Jindal 2016) and even more so in performers (HMSA, 1992; NHS, 2020; Jeffery et al., 2022). I believe this also highlights the importance of a thorough initial consultation within the vocal studio. I may have been unaware of some of these MCS complaints had I not opened up a dialogue with students, as many of these manifestations are not visible.

Hypermobility Spectrum Disorder has four subcategories, Historical, Peripheral, Localised and Generalised, all of which must present with at least one secondary MCS manifestation; however, each presents differently on the Beighton Score (Atwell, 2021). Generalised Hypermobility Spectrum Disorder (G-HSD) which is the presentation of a high Beighton score (dependent on age as above), whereas Localised Hypermobility Spectrum Disorder (L-HSD) differs in that a person may only present with hypermobility in a single joint or joint group. Historical hypermobility spectrum disorder (H-HSD) is when a person experiences a high score of joint hypermobility in their life, which decreases with age. Lastly is Peripheral Hypermobility Spectrum Disorder. This differs from the other HSDs as JH is only present in the hands (which can be determined using the Beighton Score) and feet, which can be determined using Hakim and Graham's (2003) 5-point questionnaire.

I believe that having knowledge of all forms of HSD will allow vocal coaches greater awareness of the possible causes of maladaptive postural alignments. Pliego-Arreaga et al (2024) stated that the 2017 reclassification of HSD has highlighted the need for comprehension of the spectrum to allow professionals the knowledge to fully understand the impact HSD can have on an individual.

Genetic basis of hypermobility

The literature concerning the effects of hypermobility on phonation is lacking, with only one related paper published in recent years. Jeffery (2021) found that research on voice function in people presenting with joint hypermobility was limited to clinical studies and two cohort studies, which focused on people with Ehlers-Danlos Syndrome (EDS). Though EDS, particularly hypermobile-EDS (hEDS), has many overlapping features of HSD (Coussens, 2022; Carroll, 2023; Morlino and Castori, 2023), it has very different and more invasive clinical diagnostic tools as unlike HSD, EDS has a genetic marker. Therefore it can be argued a vocal coach should not intervene without first having a student seek medical intervention for diagnosis. Jefferys' 2021 study of the 'Experiences of voice, ability, and loss in singers with hypermobility spectrum disorder' was prompted by the lack of literature and highlighted that only Meizel's (2020) account of their personal experiences of singing with HSD was documented at the time. Due to this lack of literature I feel it is important for there to be an understanding of what causes a joint to become hypermobile to understand if hypermobility poses a risk to the structures of the Larynx.

Hypermobility Spectrum Disorder is not caused by a specific genetic molecular defect and therefore, cannot be identified by any precise laboratory tool (Malfait et al. 2006). Despite this, research has found that joint hypermobility is caused by a mutation in the production and assembly of collagen in the body causing abnormalities in the proteins of the extracellular matrix (Pliego-Arreaga et al., 2024; Fikree, 2013; Aubry-Rozier; 2021). Studies have also found reduced levels of serum tenascin-X (Yamada et al., 2019) in 45% of GHSD sufferers with a disproportionate amount of women, which is extensively documented (Janssin et al., 2004, Clinch et al., 2011, Tinkle, 2020 and Jindal 2016,) showing that joint hypermobility is more prevalent in women than men.

The Role of Collagen in Connective Tissue

Connective tissue is one of the fundamental tissue structures within the body. It was described by Lodesh et al. (2000) as the most abundant protein in the animal kingdom, and it makes up between 20-25% of the protein in the human body. Connective tissue is a collective term for the tissues that connect, bind and support the anatomical structures, organs and tissues of the body (Kamrani et al., 2023). The most prominent feature of

connective tissue is the extracellular matrix (ECM), and it is within the matrix where the collagen fibres are found.

In a person with normal collagen production, the collagen fibres are closely packed, long, thin fibres (Fig.2) that run parallel to each other or create a mesh-like structure. Reticular fibres are also found in the ECM and are recognised within the literature as collagen fibre. These fibres are present within the basement membrane of epithelial tissues, which helps to influence the tissue into the correct shape and size (Khalilgharibi and Mao, 2021). Together, The reticular and collagen fibres generate functional and structural support for connective tissue.

On a molecular level, a hypermobile person experiences a genetic mutation that disrupts the organisation of their collagen fibres (Pliego-Arreaga et al., 2024; Pereria and Bridges, 2021). Given that these proteins are a rudimental component within the extracellular matrix which creates the structural scaffold of connective tissue, it is unsurprising that this mutation would cause laxity in connective tissue as it compromises the structural integrity of the supportive and connective tissues such as muscles, ligaments and tendons (Pliego-Arreaga et al., 2024). Collagen is found continuously through the muscles of the body in the collagenous basement membrane called the endomysium, which exists between each and every muscle cell. Muscles are constructed by the grouping of muscle cells to create a muscle fibre. Several of these fibres are grouped together to create a fascicle, which is grouped to create a muscle (Kamrani et al. 2023).

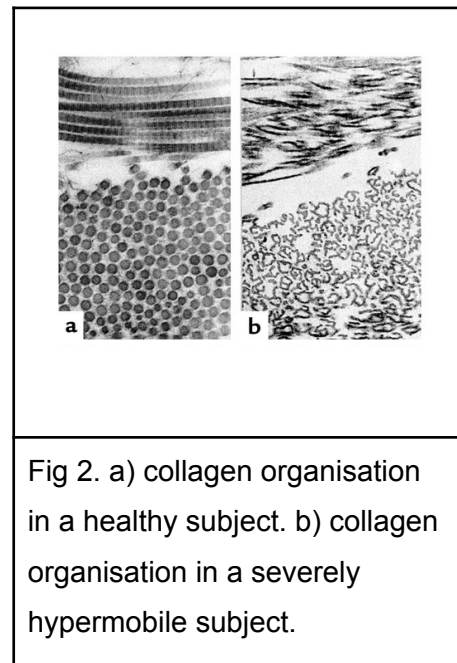


Fig 2. a) collagen organisation in a healthy subject. b) collagen organisation in a severely hypermobile subject.

The hypermobility spectrum disorder diagnosis relies heavily on observing hypermobility in the joints recognised in the Beighton Score (BS). The diagnostic criteria have shown a need for a categorisation outside of the BS with peripheral-HSD diagnosis, including hypermobility in the ankles.

An argument can be made that because hypermobility exists due to the disruption in the organisations of collagen and with collagen existing throughout the entire musculoskeletal

system due to its role in the ECM of connective tissue, hypermobility can be present within the larynx (Rimmer, Giddings, Cavalli and Hartley, 2008).

Connective Tissue Structures of the Larynx and Vocal Tract

We now know that hypermobility can be found in any connective tissue in the body, and so to understand the possible effects of hypermobility on the voice, we must understand the connective tissue structures of the larynx.

The larynx is a cartilaginous structure held in place by ligaments and muscles in the anterior aspect of the neck and acts to connect the pharynx and the trachea. Though the primary function of the larynx is to protect the respiratory tract from foreign objects, humans have evolved to use the vocal folds for protection and complex communication. The larynx can finitely alter the respiratory tract's shape and length whilst regulating subglottic pressure and influencing airflow for phonation (Benner, Sharma, Sharm, 2023). The larynx consists of three paired and three unpaired cartilages and intrinsic and extrinsic muscles and ligaments to create a stable but flexible structure.

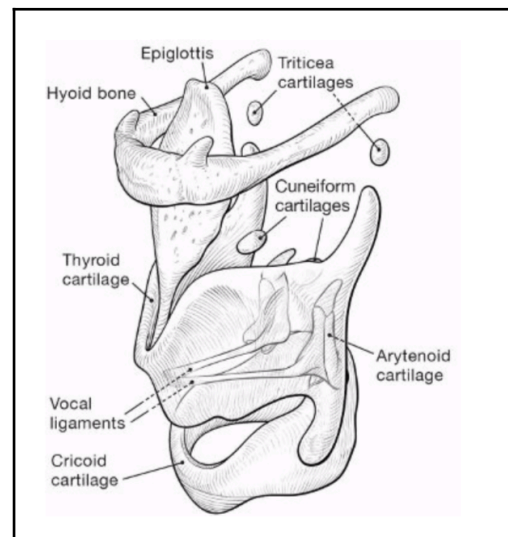


Fig.3 Cartilage structures of the larynx

The cricoid, thyroid and arytenoid cartilages create the framework of the larynx (Fig.3). The most inferior structure of the larynx is the cricoid cartilage, which is the body's only full ring of cartilage in the respiratory system and connects the trachea and larynx. On the cricoid superior posterior sits the arytenoid cartilage, and sitting above the cricoid connected via the cricothyroid joint is the thyroid cartilage. The cricoid and thyroid cartilage are also connected by each end of the vocal cords by proxy of the arytenoids on the posterior side. Within this framework is the epiglottis, a leaf-like shaped cartilage that stands upright and is attached at its base to the inner surface of the thyroid cartilage. We must also consider the hyoid bone when looking at the structure of the larynx. Although the hyoid is not a structure of the larynx, the whole larynx is suspended from this bone, and many of the extrinsic muscles of the larynx anchor to this bone (Benner, Sharma and Sharma, 2023).

Cricoarytenoid joint

On the posterior plane of the Cricoid sits the Arytenoids, two pyramid-shaped cartilages that can be found on the articular facet, creating the cricoarytenoid joint. The arytenoids have three surfaces: the medial, posterior, and anterolateral and two processes: the vocal and muscular. The arytenoids have several muscular attachments that all contribute to their movements (Andaloro, Sharma, La Mantia, 2023); these muscles are intrinsic muscles of the larynx and serve to either abduct, adduct, lengthen or shorten the vocal folds (Dimon, 2018).

The medial surface is the only surface that does not have a muscular attachment, instead, it is covered by the mucosa, creating the intercartilaginous border of the glottis (Andaloro, Sharma, La Mantia, 2023). The vocal process is on the anterior plane of the arytenoid, its' thin lateral tip serves as the posterior attachment point for the vocal ligament, which is connected anteriorly to the inner surface of the thyroid cartilage and supports the vocal fold (Fig.4). The muscular process is much wider than the vocal process and has two muscular attachments, the first is the thyroarytenoid (TA), a bilateral and symmetrically paired

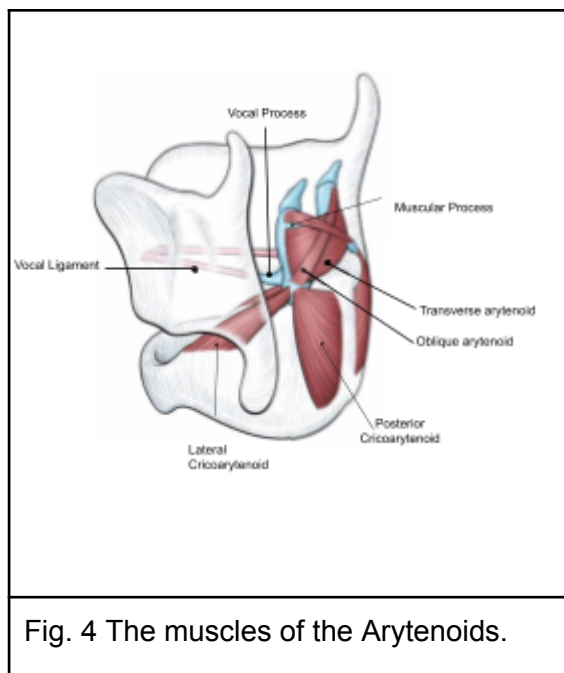


Fig. 4 The muscles of the Arytenoids.

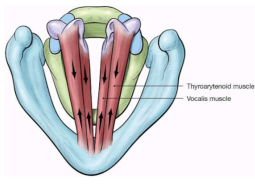
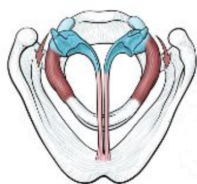
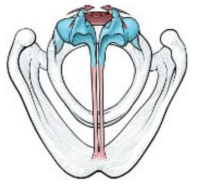
muscle. The TA and the vocal ligament are the tensor muscles of the larynx (Dimond, 2018) when they contract, they draw the arytenoids towards the thyroid cartilage, shortening and relaxing the vocal folds (Fig.5). When the TA stiffens and slightly adducts the vocal folds, this alters the tone and volume of the voice by altering the amplitude of vibration in the vocal folds cover (Andaloro, Sharma, La Mantia, 2023).

The second attachment is the posterior cricoarytenoid muscle, the primary abductor of the vocal folds. When the posterior

cricoarytenoid muscle contracts, it causes the arytenoids to rotate laterally (Fig.6), abducting the vocal folds and opening the rima glottidis, (Benner, Sharma, Sharma, 2023) this is the space between the vocal folds. Safi et al (2017) looked at three EDS patients experiencing compromised airways and abnormal voices. A laryngoscopy found that all three patients had

prolapsed arytenoid which were rescripting the respiratory cycle. The weakness shown in the connective tissues in the respiratory tract from EDS may imply hypermobility in the muscles of the posterior cricoarytenoids and their antagonistic pairs, as the correct balance of these muscles should keep the arytenoids in a midline position on the cricoid.

The antagonistic muscle to the posterior cricoarytenoid is the lateral cricoarytenoid (Fig.4). When this muscle contracts it causes the arytenoids to rotate medially, adducting the vocal folds and thereby closing the rima glottidis but leaving a triangular opening between the arytenoids, (Fig.7) this position is used for vocalisations such as whispering (Dimond, 2018). To close this gap for full phonation, the interarytenoid must contract (Fig.8). They are found on the posterior surface and consist of the only unpaired intrinsic muscles of the larynx: the transverse and oblique arytenoids (Fig.4). When these muscles contract, they pull the arytenoids together across the medial surface, causing full closure along the entire length of the vocal folds (Dimond, 2018). Lam, Wood and Birchall (2021) had a hypermobile patient within their case series who presented with vocal problems but, at first look on a laryngoscopy, showed seemingly healthy looking vocal folds. Further investigation showed when sniffing, the patient's arytenoids would show a wide excursion, implying weakness or laxity in the interarytenoids.

			
Fig.5 Tensors of the larynx	Fig.6 Effect of contraction of the posterior cricoarytenoid muscles on the glottis.	Fig.7 Effect of contraction of the lateral cricoarytenoid muscles on the glottis.	Fig.8 Effect of contraction of the interarytenoid muscles on the glottis.

On the apex of each arytenoid is the second set of paired cartilages called the corniculate cartilages. These small cone-shaped structures articulate in tandem with the arytenoids and serve as an attachment point for the aryepiglottic fold (Dimon, 2018). The last set of paired cartilages of the larynx are the Cuneiform cartilages. These cartilages do not attach directly to any part of the larynx's cartilaginous structures; they are found superiorly in either side of

the aryepiglottic folds and give support to the the vocal folds and epiglottis (Suárez-Quintanilla, Fernández Cabrera, Sharma, 2023).

The epiglottis plays an important role in the protection of the airways. The epiglottis can influence the shape of the vocal tract at the supraglottic level through involvement from the aryepiglottic muscle. During swallowing, the superior part of the aryepiglottic muscles contract, pulling the epiglottis downwards to close the laryngeal inlet. Whilst the inferior portion lubricates the vocal folds by squeezing the mucus glands. The thyroepiglottic muscle works antagonistically to the aryepiglottic muscles and opens the aryepiglottic sphincter (AES) created by the aryepiglottic folds. Singers can control these muscles and influence the diameter of the AES. Yanagisawa and Estill (1989) state that the narrowing of the AES creates a bright and brassy sound in the voice that they termed 'twang'. Sundberg and Tahél (2010) found that this narrowing boosted the higher frequencies of the voice.

Cricothyroid joint

The cricoarytenoid joint and muscles do not control vocal fold tension alone. The cricothyroid joint allows for movement of the thyroid cartilage in relation to the cricoid cartilage, which in turn can alter the vocal fold body-cover relationship (Dimon, 2018). The cricothyroid muscle attaches to the anterior external surface of the thyroid and cricoid cartilage, covering the cricothyroid space. When this muscle contracts, it closes the cricothyroid space by depressing the thyroid, which pivots from the cricothyroid joint (Andaloro, Sharma, La Mantia, 2023). The forward tilt of the thyroid cartilage increases the distance between the thyroid prominence and the arytenoids, elongating the vocal folds and stretching and tightening the vocal ligament. This movement raises pitch during phonation (Flynn and Vickerton, 2024).

Extrinsic muscles of the larynx

The extrinsic muscles of the larynx also manipulate sound production by elevating, depressing, and stabilising the laryngeal structure (Benner, Sharma, Sharma, 2023). The extrinsic muscles that connect directly to the larynx create a complex system of bilateral antagonistic pairs that suspend the larynx in the neck. An imbalance in these pairings can be detrimental to the singer's ability to control their timbre.

The sternothyroid muscle connects the thyroid to the sternum and causes the larynx to depress during contraction. This elongates the vocal tract, directly affecting the timbre of the voice by amplifying and resonating the lower frequencies (Echternach et al., 2016). The thyrohyoid muscle is the antagonistic pair to the sternothyroid and elevates the larynx on contraction. It inferiorly attaches bilaterally to the thyroid and superiorly to the hyoid bone. The primary function of this movement is to protect the airways during swallowing as the larynx is lifted away from the path of food bolus (Dimon, 2018). Singers can intentionally shorten their vocal tract to amplify the higher frequencies of their voice (Wolfe, Garnier and Smith, 2008). The Stylopharyneus muscle is the only direct connection of the larynx to the skull. It connects the styloid process to the posterior border of the thyroid cartilage and to the walls of the pharynx. This muscle elevates the larynx (Allen, Minuello, Murek, 2023) but also aids in tilting the thyroid cartilage (Dimon2018). The activation of this muscle helps to shorten the vocal tract, boosting the higher frequencies (Wolfe et al., 2008) and lengthening the vocal folds, which heightens the pitch of the voice (Flynn and Vickerton, 2024).

There are many muscles that influence the movement of the larynx without direct attachment to the larynx. The first set of muscles to consider are the suprahyoid muscles. This set of muscles connects the hyoid bone to the jaw, creating the floor of the mouth. This set of muscles, comprising the stylohyoid, hyoglossus, geniohyoid, mylohyoid and digastric, creates a pulling force on the hyoid bone towards the jaw, elevating the larynx (Allen, Minuello, Murek, 2023). They work antagonistically with the infrahyoid muscles, which comprise the omohyoid and sternohyoid. These muscles cause a depression of the larynx by contracting and pulling downwards on the hyoid bone (Dimon, 2018).

An alteration in the homeostasis of the antagonistic pairs in the extrinsic muscles can directly affect the larynx. The first case of hypermobility in the soft tissue of the suprahyoid muscles was reported in 2015 in a clinical case report of a 44-year-old female (Chatzoudis, Kelly, Jones). This caused a subluxation of the larynx with a rotation to the left, which would leave her unable to breathe and sadly needed surgical intervention. Other cases of hyoid bone dislocation (Arulanandam et al., 2016) and hyoid subluxation (Lam, Good and Birchall, 2021) have been reported. This report shows the detrimental effects of hypermobility in extrinsic muscles on the larynx. Though all of these reports may show extreme cases, it showcases the direct impact of extrinsic laxity and instability on the larynx.

The extracellular matrix of the Vocal Folds

The vocal folds can create an astounding variety of modes during phonation which is partly credited to the unique structure of the vocal folds. Our physiological understanding of this structure comes from Hirano's muscle viscoelastic-aerodynamic model, more commonly known as the cover-body theory of phonation (Benboujji and Hartnick, 2021). The vocal folds are made of layers of connective tissue called lamina propria sandwiched by the epithelium and the thyroarytenoid muscle. The cover consists of the epithelium, the basement membrane and the superficial lamina propria, which Hirano described as a critical structure to maintain vibration whilst phonating (Hirano, 1975, 1974). The body consists of the thyroarytenoid muscle, the intermediate lamina propria, and the deep lamina propria. The intermediate and deep lamina propria, also known as the vocal ligament, act as a transitional layer between the cover and body.

Collagen and elastin are widely recognised as fundamental to the elasticity and flexibility of the vocal folds. We have previously discussed that the role of reticular fibres is to help shape and size the basement membranes of epithelial tissues (Khalilgharibi and Mao, 2021) and that collagen acts as a foundational protein of all connective tissue. Khalilgharibi and Mao (2021) highlighted the importance of structural accuracy for a muscle to function correctly. Therefore, we must consider that an individual who suffers from HSD could experience a disruption in the homeostasis of the vocal folds ECM, which may result in interference of the vibratory function, which would affect the individual's ability to regulate airflow and ultimately affect phonation.

Further effects of hypermobility on muscles

Hypermobility is caused by a disruption of collagen structures in soft tissues, which can cause joint instability. The most prominent of these are tested by the Beighton score, but we can see in many cases presented above that instability can occur in any soft tissue in the body, increasing the risk of laxatives or subluxations (Castori, Tinkle, Levy, Grahame, Malfait, and Hakim, 2017). Due to its instability, a hypermobile joint is more susceptible to macro and microtrauma. Hypermobile people are able to overextend their joints due to the lack of passive tension and kinesthetic feedback due to their altered sense of proprioception, meaning they are unable to feel where their limbs are in space (Atwel et al., 2021). This can be considered microtrauma as it places unnecessary repetitive stress on the joint and

causes inflammation (Pliego-Arreaga et al., 2024). Repetitive microtrauma in one area may cause more specific joint disorders, one regularly since in hypermobile people is temporomandibular joint dysfunction (Castori et al., 2017), which I have seen diagnosed many times within my teaching practice. Macro trauma, such as dislocation and soft tissue injury, is more likely to occur due to the joint's ability to move beyond the normal physiological axes.

In a survey of 276 professionally trained adult singers diagnosed with HSD or Hypermobile Ehlers-Danlos Syndrome, 74.6% of participants reported having experienced voice difficulties (Jeffery, Postavaru, Matei, Meizel, 2021). Dysphonia in hypermobile presenting patients has been hypothesised to be multifactorial. Researchers have found many underlying causes to dysphonia including hypermobility of the vocal fold themselves (Castori et al, 2010; Hunter, Morgan and Bird, 1998), hypermobility of the extrinsic muscles (Chatzoudis, Kelly, Jones, 2015; Arulanandam et al., 2016; Lam, Good and Birchall, 2021) and hypermobility of the intrinsic muscles of the larynx (Safi et al., 2017; Arulanandam, 2016).

Though only a small range of joints are tested in the most recognised test for joint hypermobility, hypermobility can occur anywhere in the body, including the larynx. Vocal coaches would benefit from understanding the symptoms of Hypermobile spectrum disorder to allow for insights into how hypermobility can cause voice problems. I wish to further my research into the mechanisms of the active and passive forces of antagonistic muscles to understand how to safely strengthen lax muscles and then use this information to develop interventions for hypermobile singers.

Further research questions

Does the central nervous system play a role in the actions of hypermobile joints?

How does applying external forces aid in strengthening active and passive muscles?

Is it possible to create a vocal exercise regime to increase the strength of intrinsic and extrinsic muscles of the larynx and counter the effects of hypermobility?

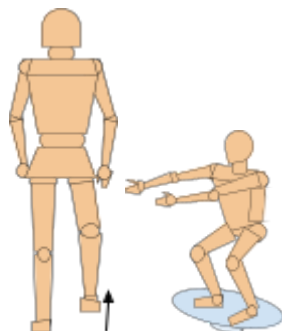
Appendix

Appendix 1- a compendium of exercises for increased body strength and proprioception in hypermobile singers

Knees

If your student presents with hypermobility for the knee and has a tendency to lock their knees when they sing, firstly bring awareness to this behaviour using mirrors and use these exercises where appropriate

For increased Proprioception in the knee



Exercise 1 (pictured left)

Balance on one foot simply. When this balance is established, continue this movement with your eyes shut.

Exercise 2 (pictured right)

Whilst standing on a balance board, gently squat and maintain your balance. The difficulty of this exercise can be increased by shutting the eyes, but it is suggested that this is done under supervision.

Postural exercise for use in singing practice



Exercise 1

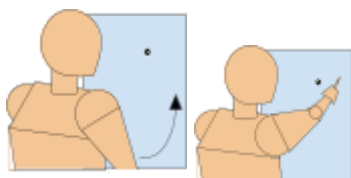
If a student has particularly hypermobile knees. This is a position that they can safely phonate in. Get them to stand in a semi, seated position against a wall. This encourages proprioception, strengthening and muscle memory whilst using the wall as a stabiliser for the spine.

Shoulders

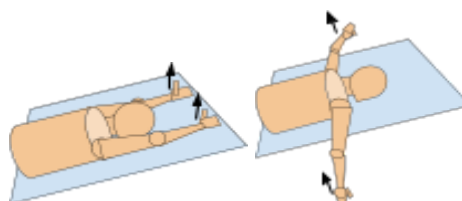
If your student presents with shoulder hypermobility, this could significantly affect their posture presenting as rounded shoulders.

Firstly, bring awareness to this behaviour using mirrors and use these exercises where appropriate

For increased proprioception



For strength.

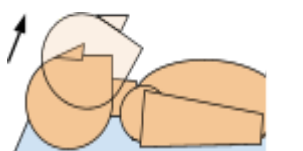


Exercise 1 Place a target on a wall and stand an arm's width away from it. One at a time, raise your arms and touch the target. Once you have done a few repetitions, do the same with your eyes closed.	This strengthening exercise is intended to rebalance the scapular from a kyphosis position back to neutral. Exercise 1 (pictured top) Overhead arm raise. Lie face down on the floor and raise the arms above the head with the thumbs lifted to the ceiling. Contract the scapular and raise the arms off the floor. Hold this pose for 3 to 5 seconds and then release. Exercise 2 (pictured bottom) Repeat exercise 1, but with the arms out to the side
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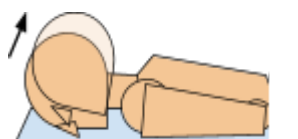
Neck

If your student presents with hypermobility of the Cervical spine that may present in the neck in postures such as kyphosis. These exercises will help to rebalance the muscles of the neck first, bring awareness to this behaviour using mirrors and use these exercises where appropriate.

Neck Flexor Strength



Neck Retractor Strength



Purpose

This exercise helps to rebalance the strength in the neck for those singers who tend to collapse into their necks or throw their heads back when they sing.

Exercise

Lay on your back with a pillow under your head and your arms relaxed at your side. Lift your head off the pillow and bring the chin to the chest. Hold for 3 to 5 seconds and release.

Purpose

This exercise helps rebalance the strength of the neck muscles for those with kyphosis.

Exercise

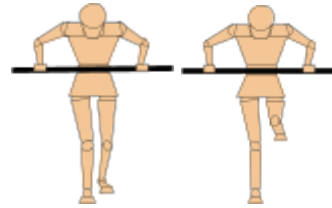
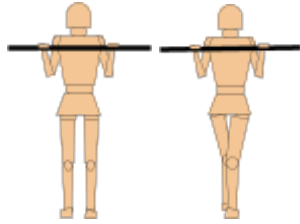
Lie face down with your arms at your side. You can place a towel under the forehead for comfort. Tuck in the chin and lift the head off the floor whilst the shoulders and chest remain on the floor. Hold for 3 to 5 seconds and then release

Trunk

The trunk can be affected by hypermobility through an imbalance in the hips, knees and spine. Begin by bringing awareness to the observed behaviour via the use of mirrors and then undergo these proprioception exercises.

Exercise 1

Exercise 2



Take a bar and hold it out in front of your chest. From here, walk forward, placing your feet from toe to heel, as if you were walking on a balancing beam. The aim is to keep the bar horizontal at all times. Once this movement improves, undergo this exercise with your eyes closed.

Take the bar and hold it against your stomach and rotate forward slightly at the hips, as if to lean over the bar. Keep the bar connected to the stomach and lift a foot behind you without rotating the trunk. Do this on both sides.

Appendix 2 - Search Criteria from A study into interventions for post-pubescent singers who present with hypermobility.

Database >	Medline		Cochrane Library		Pubmed		SAGE		Journal of Voice	
Search term	Total (n)	Relevant (n)	Total (n)	Relevant (n)	Total (n)	Relevant (n)	Total (n)	Relevant (n)	Total (n)	Relevant (n)
[Hypermobility OR "Joint Hypermobility" OR "generalised joint hypermobility" OR GJH OR "Hypermobility spectrum disorder" OR HSD] AND [Posture OR balance OR voice OR anxiety OR proprioception OR singers OR singing	23	1	0	0	102,653	-	0	0	0	0
Hypermobility and singers	10	0	1	0	4	2	0	0	11	3
Hypermobility and posture	19	0	4	1	36	4	0	0	7	3
Hypermobility and voices	5	0	0	0	9	2	0	0	11	3
Hypermobility and balance	28	0	13	2	12	0	0	0	24	3
Hypermobility and proprioception	5	0	10	1	22	6	0	0	0	0
Joint laxity and singers	2	0	0	0	3	0	0	0	23	2

Joint laxity and posture	5	0	0	1	4	0	0	0	10	2
Joint laxity and voices	5	0	0	0	0	0	0	0	33	2
Joint laxity and balance	7	0	0	0	10	0	0	0	1	0
Joint laxity and proprioception	1	0	0	1	10	3	0	0	3	2
Hypermobility voices	5	0	0	0	2	1	0	0	3	2
Trends in hypermobility	9	0	2	0	6	0	0	0	3	0
Anxiety AND Hypermobility	11	0	10	1	30	6	0	0	4	2
Posture and singing	7	1	2	1	6	1	0	0	566	-
Duplications				3		7				15
Used Documents		1		5		18	0			9

Appendix 3 - Initial consultation of students regarding hypermobility.

- > Did you have any 'double jointedness' as a child?
- > Have you experienced any ankle trauma? This includes breaks, sprains, dislocations or subluxations.
- > Have you experienced any knee trauma, such as a dislocation or any pain from prolonged standing?
- > Have you experienced any shoulder trauma, such as subluxation or dislocation?
- > Do you experience any performance anxiety, and if so, what are your symptoms? (this may open for questions on anxiety disorders which should only be explored if the student offers the information)
- > Are you particularly clumsy?
- > Do any of your immediate family suffer from hypermobility?

9 Appendix 4 - Search term criteria

Database>	Pubmed		Cochrane Library		Science direct		Journal of Voice	
Search Term	Total (n)	Relevant (n)	Total (n)	Relevant (n)	Total (n)	Relevant (n)	Total (n)	Relevant (n)
hypermobility and hypermobile spectrum disorder classification	2	1	4	1	27	3	0	0
Hypermobility spectrum disorder OR HSD AND Parameters OR diagnosis OR traits	21	4	6	1	56	3	2	2
hypermobility and hypermobility spectrum disorder and larynx	0	0	0	0	40	2	5	2
Laryngological presentations AND "HSD" OR "Hypermobility spectrum disorder" OR "EDS" OR Ehlers-danlos syndrome	0	0	0	0	1	0	0	0
Genetic Traits OR "Genetic basis" Hypermobility spectrum disorder"	9	2	3	3	252	0	1	0
collagen and "connective tissue" OR "cell membranes"	523	8	2	0	35	3	1	0
connective tissue AND "larynx" OR "voice box" OR "voice" OR "singing"	64	4	0	0	67	2	54	1
Anatomy of connective tissue	84	1	2	0	89	2	374	0
Extracellular Matrix AND "Vocal fold" OR "Vocal Cords" OR "Vocal ligament" OR "Larynx"	32	2	4	0	41	1	274	1
Hypermobility spectrum disorder and connective tissue	73	1	0	0	53	1	1	0
Intrinsic muscles of the larynx	387	6	4	1	310	6	70	1
Extrinsic muscles of the larynx	304	6	5	1	421	6	70	2
Duplications		6		0		8		2
paper used		10		0		14		1

Images

Figure 1 - Mao. J, Bristow. J (2001) The collagen fibril and EDS. The Journal of Clinical Investigation: American Society for Clinical Investigation.

Figure 2 - Gohn. J (2011) *Collagen fibrils and EDS*. Available at;
https://en.wikipedia.org/wiki/Ehlers%E2%80%93Danlos_syndrome#/media/File:JCIO112881.f2.jpg

Figure 3 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.26, illus.

Figure 4 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.36, illus.

Figure 5 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.41, illus.

Figure 6 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.37, illus.

Figure 7 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.38, illus.

Figure 8 - Dimon. T, (2018) *Anatomy of the Voice*. California: North Atlantic Books. pp.39, illus.

Bibliography

Andaloro. C, Sharma. P, La Mantia. I (2023) Head and Neck: Larynx Arytenoid Cartilage. *StatPearls [Internet]*. Treasure Island (FL): StatPearls Available at; <https://www.ncbi.nlm.nih.gov/books/NBK513252/> Accessed 05th March

Arboleda BM, Frederick AL. (2008) Considerations for maintenance of postural alignment for voice production. *Journal of Voice* .22 (1):pp90-9. doi: 10.1016/j.jvoice.2006.08.001

Arulanandam, S., Hakim, A.J., Aziz, Q., Sandhu, G. and Birchall, M.A. (2017), Laryngological presentations of Ehlers–Danlos syndrome: case series of nine patients from two London tertiary referral centres. *Clin Otolaryngol*, 42: pp860-863. <https://doi.org/10.1111/coa.12708>

Atwell. K, Michael. W, Dubey. J, James.S, Martonffy. A, Anderson. S, Rudin.N, Schrager. S, (2021) Diagnosis and Management of Hypermobility Spectrum Disorders in Primary Care. *The Journal of the American Board of Family Medicine*, 34(4) pp:838-848; DOI: <https://doi.org/10.3122/jabfm.2021.04.200374>

Barroso, J., Sandelowski, M. and Voils, C.I. (2006), Research results have expiration dates, ensuring timely systematic reviews. *Journal of Evaluation in Clinical Practice*, 12: pp454-462. <https://doi.org/10.1111/j.1365-2753.2006.00729.x>

Benboujja. F, Hartnick. C (2021) Quantitative evaluation of the human vocal fold extracellular matrix using multiphoton microscopy and optical coherence tomography. *Sci Rep* 2440(11). <https://doi.org/10.1038/s41598-021-82157-9>

Benner A, Sharma P, Sharma S. (2023) Anatomy, Head and Neck: Cervical, Respiratory, Larynx, and Cricothytenoid StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; Available from: <https://www.ncbi.nlm.nih.gov/books/NBK538307/>

Bloom, B. S. (1956). Taxonomy of educational objectives: Cognitive and affective domains. New York: David McKay.

- Borry, P., Schotsmans, P. and Dierickx, K. (2006), Evidence-based medicine and its role in ethical decision-making. *Journal of Evaluation in Clinical Practice*, 12 (3): pp.306-311. <https://doi.org/10.1111/j.1365-2753.2006.00548.x>
- Boyer, E. (1990). *Scholarship reconsidered: Priorities of the professoriate*. Princeton, NJ: Carnegie
- Breehl L, Caban O. (2023) Physiology, Puberty. StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK534827>, Accessed; Feb 13th 2023.
- Briney, K. (2015). *Data Management for Researchers: Organize, Maintain and Share Your Data for Research Success*. United Kingdom: Pelagic Publishing.
- Brown, S. P., Miller, W. C., Eason, J. M. (2006). *Exercise Physiology: Basis of Human Movement in Health and Disease*. United Kingdom: Lippincott Williams & Wilkins. pp405-28
- Carroll. M.B (2023) Hypermobility spectrum disorders: A review. *Rheumatology and immunology research*. 4(2)pp60-68
- Castori. M, Tinkle.B, Levy. H, Grahame. R, Malfait. F, Hakim.A. (2017). A Frameworkfor the classification of Joint Hypermobility and related conditions.Am JMed Genet Part C Semin Med Genet. 175:pp 148-157.
- Castori. M, Camerota. F, Celletti. C, Danese. C, Santilli. V, Saraceni. V.M, Grammatico P. (2010) Natural history and manifestations of the hypermobility type Ehlers–Danlos syndrome: A pilot study on 21 patients. *Am J Med Genet* 152 pp556–564.
- Chandler J, Hopewell S. (2013) Cochrane methods – twenty years experience in developing systematic review methods. *Systematic Reviews*; 2: pp 76
- Chatzoudis. D, Kelly. T.J, Lancaster. J, Jones. T.M (2015) Upper airway obstruction in a patient with ehlers-danlors syndrome. *The annals of The Royal College of Surgeons England*. 3(97) pp 50-51. <https://doi.org/10.1308/003588414X14055925061793>
- Clarke. T, Holmes. P, Feeley. G, et al. (2013) Pointing to performance ability: examining hypermobility and proprioception in musicians. *Proc Int Symp Perform Sci*. pp.605-610

- Clayton, J. A. (2016). Studying both sexes: A guiding principle for biomedicine. *The FASEB Journal*, 30(2), pp519–524. doi:10.1096/fj.15-279554
- Clayton, J. A. (2018). Applying the new SABV (sex as a biological variable) policy to research and clinical care. *Physiology & Behavior*, 187, pp2–5. doi:10.1016/j.physbeh.2017.08.012
- Clifford, H. (2021) *Almost seventy per cent of university drama applicants in 2020 identified as female, Drama And Theatre*. Available at: <https://www.dramaandtheatre.co.uk/news/article/almost-seventy-per-cent-of-university-drama-applicants-in-2020-identified-as-female> (Accessed: 02 February 2024).
- Clinch. J, Deere. K, Sayer.s A, Palmer. S, Riddoch. C, Tobias. JH, Clark. EM. (2011) Epidemiology of generalized joint laxity (hypermobility) in fourteen-year-old children from the UK: a population-based evaluation. *Arthritis Rheum*. 63(9): pp. 2819-27. DOI: 10.1002/art.30435.
- Coussens. M, Banica. T, Lapauw. B, De Wandele. I, Rombaut. L, Malfait. F, Calders. P. (2022) Bone parameters in hypermobile Ehlers-Danlos syndrome and hypermobility spectrum disorder: A comparative cross-sectional study. *Bone*. 166: doi: 10.1016/j.bone.2022.116583.
- Dimon, T. (2018). *Anatomy of the Voice: An Illustrated Guide for Singers, Vocal Coaches, and Speech Therapists*. United States: North Atlantic Books.
- Dinet, J., Favart, M. and Passerault, J.M. (2004), Searching for information in an online public access catalogue (OPAC): the impacts of information search expertise on the use of Boolean operators. *Journal of Computer Assisted Learning*, 20: pp338-346. doi.org/10.1111/j.1365-2729.2004.00093.x
- Echternach. M, Burk. F, Burdumy. M, Traser. L, Richter. B (2016) Morphometric Differences of Vocal Tract Articulators in Different Loudness Conditions in Singing. *PLOS ONE* 11(4): <https://doi.org/10.1371/journal.pone.0153792>
- Feldenkrais, M. (1949). *Body and mature behaviour; a study of anxiety, sex, gravitation and learning*. International Universities Press.

- Fikree.A, Aziz. Q, Grahame. R (2013) Joint hypermobility syndrome. *Rheum Dis Clin Norh Am* 39: pp 419-430
- Flynn W, Vickerton P. (2024) Anatomy, Head and Neck: Larynx Cartilage. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing Available from: <https://www.ncbi.nlm.nih.gov/books/NBK553185/>. Accessed: 23rd March 24
- Folci. M, Capsoni. F, (2016). Arthralgias, fatigue, paresthesias and visceral pain: Can joint hypermobility solve the puzzle? A case report. *BMC Musculoskeletal Disorders*. 17. (58.)pp10-15 doi: 1186/s12891-016-0905-2.
- Grahame. R, (1999) Joint hypermobility and genetic collagen disorders: are they related? *Arch Dis Child*. 80 pp188-191. <https://doi.org/10.1136/adc.80.2.188>
- Grahame, R. (2013) Joint hypermobility: emerging disease or illness behaviour? *Clinical Medicine* 13 (6)pp50-52
- Hakim. A.J, Grahame. R, (2003) A simple questionnaire to detect hypermobility: an adjunct to the assessment of patients with diffuse musculoskeletal pain. *Int J Clin Pract*. 57: pp163-166
- Hirano, M. (1974) Morphological structure of the vocal cord as a vibrator and its variations. *Folia Phoniatr. Logop*. 26, pp89–94.
- Hirano, M. (1975) Phonosurgery; basic & clinical investigations. *Otologia* 1, pp239–442.
- Huber, M. T., & Morreale, S. P. (2002). Situating the scholarship of teaching and learning. In M. T. Huber & S. P. Morreale (Eds.), *Disciplinary styles in the scholarship of teaching: Exploring common ground* (pp. 1-24). Washington, DC: American Association for Higher Education and the Carnegie Foundation for the Advancement of Teaching.
- Hunter. A, Morgan. A.W, Bird. H.A, (1998) A survey of Ehlers-Danlos syndrome: hearing, voice, speech and swallowing difficulties. Is there an underlying relationship?, *Rheumatology*, 7(37), pp 803–804, <https://doi.org/10.1093/rheumatology/37.7.803>
- Jansson A, Saartok T, Werner S, Renstrom P. (2004) General joint laxity in 1845 Swedish school children of different ages: age- and gender-specific distributions. *Acta Paediatr*.93: pp. 1202–6.
- Jeffery T, Postavaru GI, Matei R, Meizel K. (2021) 'I Have Had to Stop Singing Because I Can't Take the Pain': Experiences of Voice, Ability, and Loss in Singers With

Hypermobility Spectrum Disorders. *J Voice*(21)00407-0. doi:
10.1016/j.jvoice.2021.11.017.

Jindal P, Narayan A, Ganesan S, MacDermid JC. (2016) Muscle strength differences in healthy young adults with and without generalized joint hypermobility: a cross-sectional study. *BMC Sports Sci Med Rehabil.* 25(8)pp1-9 doi: 10.1186/s13102-016-0037-x.

Khalilgharibi. N, Mao. Y (2021) To form and function: on the role of basement membrane mechanics in tissue development, homeostasis and disease. *Open Biology*, 2(11)
<https://doi.org/10.1098/rsob.200360>

Kamrani. P, Marston. G, Arbor. T.C, Jan. A.(2024) Anatomy, Connective Tissue. *StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan–*. PMID: 30860769.
Accessed 23rd February

Lam, C.M., Wood, G. and Birchall, M.A. (2022) ‘Laryngological presentations and patient-reported outcome measures in Ehlers–Danlos syndrome’, *The Journal of Laryngology & Otology*, 136(10), pp. 947–951. doi:10.1017/S0022215121004072.

Lame, G. (2019) ‘Systematic Literature Reviews: An Introduction’, in *proceedings of the 22nd International Conference on Engineering Design (ICED19)*, Delft, The Netherlands, 5-8 August 2019. doi:10.1017/dsi.2019.169

Lowth, M and Tidy, M (2019) *Hypermobility Syndrome* Available at:
<https://patient.info/bones-joints-muscles/hypermobility-syndrome-leaflet#nav-0>
(Accessed: 07th of February 2024)

Morlino. S, Castori. M, (2023) Placing joint hypermobility in context: traits, disorders and syndromes, *British Medical Bulletin*, 147(1) pp90–107,
<https://doi.org/10.1093/bmb/ldad013>

Malfait, A. J. Hakim, A. De Paepe, R. Grahame, (2006) The genetic basis of the joint hypermobility syndromes, *Rheumatology*, 45(5), pp502–507,
<https://doi.org/10.1093/rheumatology/kei268>

Meizel. K (2020) Lost voices *Multivocality*. Open University Press. pp177-196

Nightingale, A. (2009) A guide to systematic literature reviews. *Surgery (Oxford)* 27(9):
pp381-384; doi.org/10.1016/j.mpsur.2009.07.005.

- Patino. C.M, Ferreira. J.C. (2018) Inclusion and exclusion criteria in research studies: definitions and why they matter. *J Bras Pneumol.* 44(2): pp84. doi: 10.1590/s1806-37562018000000088.
- Pereira, C., Bridges, A. (2021). Too Flexible To Feel Good: A Practical Roadmap to Managing Hypermobility. United States: Victory Belt Publishing. pp23-24
- Pliego-Arreaga. R, Cervantes-Montelongo. J.A, Silva-Martínez. G.A, Tristán-Flores. F.E, Pantoja-Hernández. M.A, Maldonado-Coronado. J.R. (2024) Joint Hypermobility Syndrome and Membrane Proteins: A Comprehensive Review. *Biomolecules.* 14(4):pp47. doi: 10.3390/biom14040472. PMID: 38672488; PMCID: PMC11048254.
- Prager. E.M, Chambers. K.E, Plotkin. J.L, McArthur. D.L, Bandrowski. A.E, Bansal. N, Martone. M.E, Bergstrom. H.C, Bernalov. A, Graf. C. (2018) Improving transparency and scientific rigor in academic publishing. *Brain Behav.* 9(1): doi: 10.1002/brb3.1141.
- Remvig, L., Jensen, D.V. and Ward, R.C. (2007). Are diagnostic criteria for general joint hypermobility and benign joint hypermobility syndrome based on reproducible and valid tests? A review of the literature. *The Journal of rheumatology*, 34(4), pp.798-803.
- Rimmer. J, Giddings. C.E.B, Cavalli. L, Hartley. B.E.J, (2008) Dysphonia—A rare early symptom of Ehlers–Danlos syndrome?, *International Journal of Pediatric Otorhinolaryngology*, 12(72) pp1889-1892 <https://doi.org/10.1016/j.ijporl.2008.09.018>
- Rubin, J.S., Blake, E. & Mathieson, L. (2006). The effects of posture on voice. In Rubin et al, Eds. *Diagnosis and Treatment of Voice Disorders*. San Diego, CA: Plural Publishing.
- Safi. F.A, Alyosif. M.D, Imam. S, et al (2017) Arytenoid prolapse in 3 patients with ehler-danlos syndrome leading to respiratory compromise. *Mayo Clin Proc.* (92) pp851-853, doi.org/10.1016/j.mayocp.2017.01.016
- Schön, D., (1991). The Reflective Practitioner: How Professionals Think in Action. London: Ashgate Publishing Limited
- Schneider, C., Dennehy, C.A., Saxon, K.G. (1997) Exercise physiology principles applied to vocal performance. *J Voice.* 11(3):pp332-7 doi: 10.1016/s0892-1997(97)80012-4
- Seow. C.C, Chow. P.K, Khong. K.S. (1999) A study of joint mobility in a normal population. *Ann Acad Med Singap.* 28(2): pp231-6. PMID: 10497673.

- Shulman, L. S. (1999). Taking Learning Seriously. *Change: The Magazine of Higher Learning*, 31(4); pp10–17. doi.org/10.1080/00091389909602695
- Shulman, L. S. (2000). Inventing the future. In P. Hutchings (Ed.), *Opening lines: Approaches to the scholarship of teaching and learning*. Retrieved from <http://www.carnegiefoundation.org/elibrary/inventing-future-opening-lines-approaches-scholarship-teaching-and-learning>
- Suárez-Quintanilla. J, Fernández Cabrera. A, Sharma. S. (2023) Anatomy, Head and Neck: Larynx. *StatPearls [Internet]*. Treasure Island (FL): StatPearls Publishing; Available from: <https://www.ncbi.nlm.nih.gov/books/NBK538202/>
- Sundberg. J, Thalén. M, (2010) What is “Twang”?, *Journal of Voice* 24(6) pp 654-660 doi.org/10.1016/j.jvoice.2009.03.003.
- Titze, I.R (1982) Sensory feedback in voice production. *The NATS Bulletin*, 38, (4), pp32
- Tinkle, B. T, (2020) ‘Symptomatic Joint Hypermobility’, *Best Practice & Research Clinical Rheumatology*, 34(3), <https://doi.org/10.1016/j.berh.2020.101508>
- Tuval-Mashiach, Rivka. (2016). Raising the Curtain: The Importance of Transparency in Qualitative Research. *Qualitative Psychology*. 4. 10.1037/qup0000062.
- Twohig, I. (2024) *Iterative research: What it is, how to do it and tools you can use*, *Indeemo*. Available at: [https://indeemo.com/blog/iterative-research#:~:text=Essentially%2C%20the%20iteration%20component%20of,steps%20\(Medium%2C%202019\).](https://indeemo.com/blog/iterative-research#:~:text=Essentially%2C%20the%20iteration%20component%20of,steps%20(Medium%2C%202019).) (Accessed: 18 April 2024).
- Wolfe. J, Garnier. M, Smith. J. (2008) Vocal tract resonances in speech, singing, and playing musical instruments. *HFSP J.* 3(1) pp6-23. doi: 10.2976/1.2998482.
- Yamada. K, Watanabe. A, Takeshita. H, Fujita. A, Miyake. N, Matsumoto. N, Matsumoto. KI. (2019) Measurement of Serum Tenascin-X in Joint Hypermobility Syndrome Patients. *Biol Pharm Bull.* 42(9) pp1596-1599. doi: 10.1248/bpb.b19-00168.
- Yanagisawa. E, Estil. J, (1989) The contribution of aryepiglottic constriction to “ringing” voice quality—A videolaryngoscopic study with acoustic analysis. *Journal of Voice.* 4(3) pp.342-350 doi.org/10.1016/S0892-1997(89)80057-8.

