Speech and voice characteristics in multiple sclerosis and cervical spinal cord injury: descriptive studies and effects of respiratory training

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ABSTRACT

Introduction and aims: Respiratory function may be impaired in multiple sclerosis (MS) and cervical spinal cord injury (CSCI), but few studies have reported voice and speech data before and after respiratory training in MS and CSCI. The aims of these studies were therefore to provide a detailed description of voice and speech following CSCI, and to evaluate effects of glossopharyngeal breathing (glossopharyngeal pistoning for lung insufflation, GI) and expiratory muscle strength training (EMST) on respiration, voice and speech, and communication in individuals with MS or CSCI.

Methods: Participants were 26 individuals with CSCI, a control group (CG) of 19 matched non-injured individuals, and six individuals with MS. The project included three group studies and two single subject studies (one being repeated across five participants). The following data were analyzed: respiratory, acoustic, aerodynamic, and anamnestic information, self-reported voice and speech function and limitations, and perceptual voice and speech assessment performed by experienced speech-language pathologists.

Results: A majority of the participants with CSCI experienced long-standing voice changes and used a range of strategies to compensate for the limited respiratory function. The Sw-VHI scores showed significantly more pronounced voice problems in the group with CSCI, and their results on maximum respiratory, voice, and speech performance tasks were significantly worse when compared with the CG. Participants with a vital capacity (VC) of less than 50 % of the expected performed significantly worse than participants with a VC above 50 % of the expected, and the level of injury had an impact on respiratory function in complete CSCI. The listeners rated the presence of the perceptual voice and speech characteristics to be low in the group with CSCI, but harshness and vocal fry were present to a higher degree, and in more participants with CSCI, and loudness was rated lower than normal compared with the CG. There were both short- and long-term effects on voice and speech, including increased loudness and improved phonatory stability in the seven individuals with CSCI who used GI. Long-term effects were particularly marked in the participant with MS, who showed continued improvements of respiration and speech up to the last follow up 20 months after intervention, both on habitual speech measures and when using GI. Following EMST, some of the five participants with MS showed increases in maximum expiratory pressure, maximum phonation time, loudness and phonatory stability, but the results suggested larger effect sizes in the two participants with mild MS, who were able to train with a higher load. Self-reports indicated effects on communicative participation in MS after GI and EMST.

Conclusions: CSCI can result in long-standing changes in voice function secondary to the respiratory impairment, especially in challenging speech tasks. Therefore, individuals with CSCI risk voice fatigue and restrictions in communicative participation. The voice and speech changes following CSCI are perceptually subtle, but can be identified by posing questions or using instruments about self-perceived limitations, and by including more challenging speech tasks in the assessment. GI can be considered in speech pathology intervention for patients with CSCI and MS. EMST may have additional positive effects, why more clinical investigations about the outcomes of this treatment are needed.

Key words: acoustic analysis, perceptual assessment, cervical spinal cord injury, communication, dysarthria, expiratory muscle training, glossopharyngeal breathing, maximum phonation time, multiple sclerosis, respiration, voice, self-reports, sound pressure level, speech, subglottal pressure, Voice Handicap Index, voice range profile

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SPEECH AND VOICE CHARACTERISTICS IN MULTIPLE SCLEROSIS AND CERVICAL SPINAL CORD INJURY: DESCRIPTIVE STUDIES AND EFFECTS OF RESPIRATORY TRAINING

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Stockholm 2013

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Key words: acoustic analysis, perceptual assessment, cervical spinal cord injury, communication, dysarthria, expiratory muscle training, glossopharyngeal breathing, maximum phonation time, multiple sclerosis, respiration, voice, self-reports, sound pressure level, speech , subglottal pressure, Voice Handicap Index, voice range profile

SAMMANFATTNING

Introduktion och syfte: Andningsfunktionen kan vara nedsatt vid multipel skleros (MS) och ryggmärgsskada (cervical spinal cord injury, CSCI), med påverkan på rösten och talet som följd. Träning med grodandning (glossopharyngeal pistoning for lung insufflation, GI) och av exspiratorisk muskelstyrka (expiratory muscle strength training, EMST) har resulterat i förbättrad andning, men effekter på röst och tal är beskrivna i få studier. Syftet med dessa studier var därför att ge en fördjupad beskrivning av röst och tal vid CSCI, och att undersöka effekterna av GI och EMST på andning, röst, tal och kommunikation vid MS och CSCI.

Metod: 26 personer med CSCI, 19 matchade icke-skadade personer i en kontrollgrupp (CG), och 6 personer med MS deltog i projektet, som omfattade tre gruppstudier och två fåpersonsstudier (varav en med fem deltagare). Analysen omfattade andningsdata, akustiska och aerodynamiska data, anamnestisk information, självrapporterad röst- och talfunktion och begränsningar i röst och tal, samt perceptuell bedömning utförd av erfarna logopeder.

Resultat: En majoritet av deltagarna med CSCI upplevde röstförändringar efter skadan och använde sig av olika strategier för att kompensera för den begränsade funktionen. Jämfört med CG, upplevde gruppen med CSCI signifikant mer röstproblem och presterade signifikant sämre på andnings-, röst- och taluppgifter som krävde maximal prestation. Deltagarna med en vitalkapacitet (VC) lägre än 50 % av det förväntade, presterade signifikant sämre än med de med VC över 50 % av det förväntade. Skadenivån hade betydelse för andningsfunktionen vid komplett CSCI. Logopederna skattade låg förekomst av de perceptuella röst- och talparametrarna i gruppen med CSCI, men skrap och knarr förekom i högre grad och hos fler deltagare med CSCI, än i CG. Röststyrkan skattades också som lägre i gruppen med CSCI. Deltagarna med CSCI som tränade GI uppvisade såväl korttids- som långtidseffekter på röst och tal, bland annat ökad intensitet och stabilitet i rösten. Bedömningar upp till 20 månader efter avslutad GI-träning visade att deltagaren med MS förbättrade sina andnings- och talprestationer kontinuerligt. Prestationerna var också markant bättre när han använde GI, än utan GI. Efter EMST noterades högre maximalt exspiratoriskt tryck, och för vissa deltagare med MS, längre maximal fonationstid, ökad röstintensitet och ökad röststabilitet. Resultaten antyder att effekten av träningen var större hos de två deltagarna med lättare MS, som orkade träna med ett högre motstånd. Deltagarna med MS uppgav att de upplevde GI och EMST positivt och att deras kommunikativa delaktighet ökade efter träningen.

Slutsatser: CSCI kan medföra förändringar av röstfunktionen sekundärt till den nedsatta andningsfunktionen, särskilt i uppgifter med ökad röstbelastning. Personer med CSCI riskerar därför rösttrötthet, dysfoni och begränsningar i sin kommunikativa delaktighet. Perceptuellt är förändringarna i rösten och talet oftast subtila. Röstförändringar och begränsningar i kommunikationen kan uppmärksammas med frågor om röstfunktionen eller självskattningsinstrument, eller med testuppgifter som medför en högre belastning. GI kan vara ett alternativ i logopedisk behandling av patienter med röst- och talsvårigheter efter CSCI och vid MS. Det är möjligt att EMST har flera generella och specifika positiva effekter, och metoden behöver därför utvärderas i fler studier.

LIST OF PUBLICATIONS

I. Johansson K, Nygren-Bonnier M, Klefbeck B, Schalling E. Effects of glossopharyngeal breathing on voice in individuals with cervical spinal cord injury. *Int J Ther Rehabil* 2011 18(9):501-510

II. Johansson K, Nygren-Bonnier M, Schalling E. Effects of glossopharyngeal breathing on speech and respiration in multiple sclerosis: a case report. *Mult Scler* 2012 18(6):905-8. Epub 2011 Dec 19

III. Johansson K, Kjellmer L, Schalling E, Hartelius L, Fredrikson S. "I can walk briskly and talk at the same time": Effects of Expiratory Muscle Strength Training on respiration and speech in multiple sclerosis. *J Med Speech Lang Pathol* 2012 20(4):70-76

IV. Johansson K, Seiger Å, Forsén M, Holmgren Nilsson J, Hartelius L, Schalling E. Speech Function after Cervical Spinal Cord Injury: Habitual and Maximum Speech Performance in Relation to Respiratory Function. 2013. *Manuscript submitted*

V. Johansson K, Hartelius L, Seiger Å, Schalling E. Speech Function after Cervical Spinal Cord Injury: Patient-Reported Outcomes and Perceptual Impressions. 2013. *Manuscript submitted*

CONTENTS

LIST OF ABBREVIATIONS AND DEFINITIONS

AMR	Alternate Motion Rate: syllables per second for the repetition of one syllable, for example /pa/ at maximum speed
cm H2O	Centimeter water
CG	Control Group
CSCI	Cervical Spinal Cord Injury
CVF0	Coefficient of Variation of the Fundamental frequency
dB	Decibel
EDSS	Expanded Disability Status Scale
EMG	ElectroMyoGraphy
EM(S)T	Expiratory Muscle (Strength) Training
EMST 150	Expiratory Muscle Strength Trainer ($0 - 150$ cm H_2O)
FIS-C	Fatigue Impact Scale – Communication
F0	Fundamental frequency
FVC	Forced Vital Capacity
GI	Glossopharyngeal pistoning for lung Insufflation, also called glossopharyngeal breathing or "frog breathing"
GIV	Glossopharyngeal Insufflation Volume
Hz	Hertz
ICC	Intra class Correlation Coefficient
IM(S)T	Inspiratory Muscle (Strength) Training
LSVT	Lee Silverman Voice Treatment
M	Mean
Md	Median
MEP	Maximum Expiratory Pressure
MFIS	Modified Fatigue Impact Scale
MFT	Maximum sustained Fricative Time
MIP	Maximum Inspiratory Pressure
MPT	Maximum Phonation Time
MS	Multiple Sclerosis
NMD	Neuromuscular Disorder
P	Participant
PCF	Peak Cough Flow
PEF	Peak Expiratory Flow
PEPTT	Positive Expiratory Pressure Threshold Trainer ($0 - 20$ cm H_2O)
P_S	Subglottal Pressure
QASD	Questionnaire on Acquired Speech Disorders (*sw.* SOFT)
QoL	Quality of Life
REL	Resting Expiratory Level
RM(S)T	Respiratory Muscle (Strength) Training
RPM	Respiratory Pressure Meter
RV	Residual Volume
SD	Standard Deviation
SMR	Sequential Motion Rate: syllables per second for the repetition of

	a syllable sequence, for example /pataka/ at maximum speed
SLP	Speech Language Pathologist
SOFT	Självsvarsformulär Om Förvärvade Talsvårigheter (*eng.* QASD)
SPL	Sound Pressure Level
SPSS	Statistical Package for Social Sciences
SRP	Speech Range Profile
Sw-VHI	Swedish validated version of the Voice Handicap Index
SWINT	Swedish Intelligibility Test
TLC	Total Lung Capacity
VAS	Visual Analogue Scale
VC	Vital Capacity
VC_{GI}	Vital Capacity supplemented by GIV
VRP	Voice Range Profile
WNL	Within Normal Limits
WPB	Words Per Breath
WPP	Words Per Phrase

1 INTRODUCTION

Patients with neuromuscular disorders (NMD) who are enrolled in speech interventions in the speech pathology clinic frequently comment that they cannot longer take deep breaths and that they run out of air when they speak.

Respiratory musculature may be affected in NMD (1-6), and respiratory dysfunction following NMD can also result in speech problems. Respiration is central as the driving force for speech production, and that explains why impaired respiratory function can have a negative impact on several aspects of speech production, such as phonation, resonance, articulation, and prosody. Symptoms related to respiratory dysfunction are not uncommon in dysarthria.

In dysarthria, the underlying neuropathology results in flaccid, spastic, or uncoordinated speech musculature. This leads to muscle weakness or rigidity, and movements which show abnormalities in "the strength, speed, range, steadiness, tone, or accuracy required for control of the speech production" (7). Depending on the underlying neuropathology, dysarthria can be categorized as flaccid, spastic, ataxic, hypokinetic, hyperkinetic, unilateral upper motor neuron, or mixed.

1.1 RESPIRATION AND SPEECH IN MS AND CSCI

Weak respiratory musculature leads to reduced inspiratory capacity and smaller lung volumes than expected. As a consequence, the recoil of the rib cage and the pressure on the air column at the following expiration will be reduced. Also, weakness of the respiratory musculature negatively affects the balance between inspiratory and expiratory forces needed to maintain a stable subglottal pressure during an utterance (8-10). Weakness in the expiratory musculature particularly affects the control of the subglottal pressure when speaking at lung volumes below resting expiratory level (REL), or when using a loud voice (8, 11, 12). Individuals may also experience speech dyspnea following respiratory muscle weakness (2, 13, 14).

Two neuromuscular disorders often affecting respiration are multiple sclerosis (MS) (15-19) and cervical spinal cord injury (CSCI) (2, 20-22). MS affects 4-5/100,000 individuals per year in Sweden depending on the geographic area (23, 24), and the prevalence is about 190/100,000 (25). Also in Sweden, the yearly incidence of CSCI is 1-2/100,000 individuals, with a prevalence of approximately 5000 individuals. Fifty-five percent of all traumatic spinal cord injuries are localized to the cervical spinal cord, and of these 75 % are localized to the lower cervical spinal cord (26). Contrary to the heterogeneous physiopathology in MS, respiratory dysfunction in CSCI is the result of transection of the cervical spinal cord which leads to paralysis of the muscles supplied

by the spinal nerves under the level of injury (2). At the level of injury of C4 and below, the innervation of the diaphragm is left intact and the patients can breathe without a ventilator. However, the muscles in the chest and abdomen are paralyzed, which explains why both inspiration and expiration are limited (2, 22). Respiratory dysfunction is therefore a frequent problem for patients with CSCI (2, 22), possibly resulting in a negative impact on speech function (1).

MS is an inflammatory disease resulting in multiple areas of demyelination in the nervous system. Because of this, nerve impulse conduction is slowed, causing muscle atrophy and weakness. The clinical picture is often complex and symptoms vary depending on the localization of the lesions (27, 28). Respiratory impairment is often described as a symptom in the late stages of MS, but studies have also shown that the respiratory musculature is already affected in mild MS resulting in lower inspiratory and expiratory pressures than expected (17). Respiratory involvement in MS seems to be more common in individuals where the lesions involve the cerebellum (18).
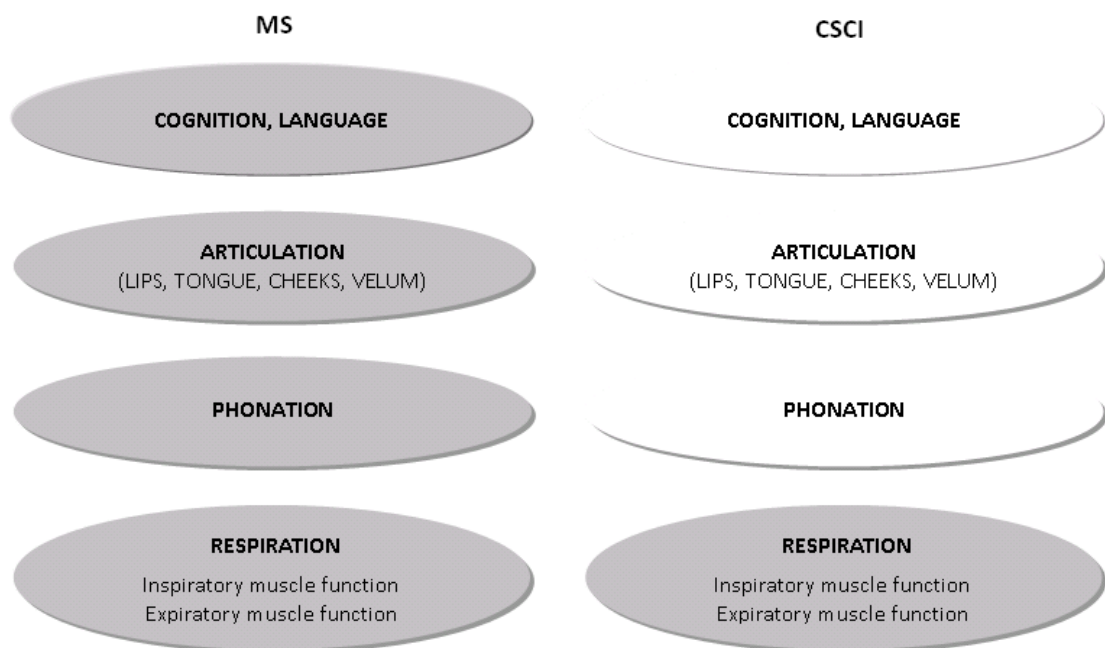
Figure 1. *Grey ellipses indicate functions related to voice, speech and communication that can be affected in multiple sclerosis (MS), to the left, and following cervical spinal cord injury (CSCI), to the right. White ellipses indicate functions primarily not affected in CSCI. (Schematic illustration by the author)*

Speech symptoms related to respiratory dysfunction have been reported in both MS and CSCI (1). In MS, one or more speech subsystems may be affected, sometimes making it difficult to rule out the isolated contribution of respiratory dysfunction (see figure 1). In a survey on speech and swallowing, 44% of the

informants with MS reported voice and speech dysfunction compared with before disease onset (29). Sixteen percent of these ranked the speech impairment as their largest problem. The most common voice and speech problems were weak voice (43%), imprecise articulation (33%) and difficulties initiating speech (34%).

In CSCI, the speech symptoms are more likely to be purely related to the respiratory dysfunction, except in injuries leading to an additional impact on the larynx (see figure 1). There are no exact reports of incidence of voice and speech problems in CSCI following injury. However, the participants in an interview study by Nygren-Bonnier et al. (14) reported that voice function was one of three main areas affected by respiratory impairment.

Both MS (27, 30) and CSCI (26, 31) affect young to middle-aged individuals with social and occupational obligations, who are therefore particularly dependent on their ability to communicate (32). As a result, there is a need for the development of speech pathology methods aimed at improving speech in these populations.

1.1.1 Respiratory function in MS

Respiratory impairment is most common during relapses or in advanced MS, when there are large lesions in several areas, or when areas in the medulla that are responsible for respiratory control are affected (3, 19). However, weakness in the respiratory muscles and decreased expiratory flows have also been reported in mild MS (17), and the prevalence of respiratory impairment increases with disease progression and severity. Grasso et al. (18) found respiratory dysfunction in 63% of their patients with MS; respiratory function was impaired in 36% of the ambulatory and in 83% of the non-ambulatory patients. Their explanation for the respiratory involvement in mild MS was the association found between respiratory function and cerebellar involvement.

Respiratory function and maximum lung pressures are reduced in 30 – 60% of individuals with MS (15, 16, 18, 33-38). Although some studies have shown reduced lung function (18, 39), it is generally relatively well preserved in MS with values for total lung capacity (TLC) and vital capacity (VC) within the normal ranges (15, 17, 40). Maximum lung pressures, however, are markedly reduced, indicating the early onset of respiratory muscle weakness (15-18, 33-38).

Maximum expiratory pressure (MEP) is more impaired, than the maximum inspiratory pressure (MIP). An explanation for this is that the disease progresses in a distal – proximal direction, affecting the thoracic spinal cord and the abdominal and intercostal muscles before affecting the diaphragm, which is innervated by the phrenic nerve that originates in the upper part of the cervical spinal cord (19). In subjects with mild MS, MEP is around 70% of the expected for a non-injured individual the same gender, age and height, and in moderate to severe MS, MEP is 40–50% of the expected

(15, 16). The corresponding percentages for MIP are 84% and 74% of the expected in subjects with mild MS and moderate to severe MS, respectively (15, 17).

1.1.2 Speech symptoms related to respiration in MS

Dysarthria is present in about 40 to 60% of the population of MS (29, 41-43) and there is a large variation in the symptomatology because of the variety of localization of lesions. Several types of dysarthria are possible, but a mixed ataxic – spastic dysarthria is believed to be the most common (44).

Descriptions of speech in MS include several respiratory-related aspects (see table 1). One of the earliest analyses of speech in MS found "irregular" breathing affecting voice intensity (45). Reduced respiratory support for speech and loudness control have been described by Farmakides and Boone (46), Darley et al. (38), Murdoch et al. (40), and Hartelius et al. (47). The prevalence of deviating perceptual voice and speech characteristics in two large studies is shown in table 1. Perceptually, impaired loudness control has been found to appear together with deviating pitch control in individuals with low VC (38).

Impaired respiratory support is expected to reduce the duration of maximum phonation and of breath phrases (1). Individuals with MS sustain a vowel with a significantly shorter duration, than healthy controls (38, 48, 49), and the participants with low VC in particular had shorter maximum phonation times (MPT) (38). Hartelius et al. (47) described both increased durations and decreased intra-utterance variability (more isochronous syllables) in combination with increased inter-utterance variability in participants with MS. For inter-stress-intervals there were increased durations and increased variability. These findings reflect inflexibility, as well as instability of temporal control due to cerebellar involvement.

The majority of studies of speech in MS have focused on different aspects of phonatory function. Various aspects of phonatory dysfunction, such as pitch breaks and harsh voice quality, as well as vocal fatigue, have been reported in 18–70% of individuals with mild – moderate MS (48-50), and in up to 90% of individuals with severe MS (51). Phonatory instability is also common in MS, with studies reporting a higher presence of jitter and shimmer in individuals with MS than in healthy control individuals (38, 46, 48-50, 52-54). Perceptually, a predominant voice characteristic in MS is harshness (38, 51).

Table 1. Two perceptual studies showing the ten most deviating perceptual speech characteristics in multiple sclerosis (MS). The protocol used by Darley et al. (38) included 25 speech dimensions. Hartelius et al. (41) used a 32-dimension protocol by Fitzgerald & Chenery (51), based on a protocol by Darley et al. (55).

Darley et al. (1972) N = 168	% of sample	Hartelius et al. (2000) N = 77	% of sample
Impaired loudness control	77	Precision of consonants	92
Harshness	72	Glottal fry	88
Defective articulation	46	Prolonged intervals	87
Impaired emphasis	39	Harshness	86
Impaired pitch control	37	General stress pattern	83
Decreased vital capacity	35	Loudness level	81
Hypernasality	24	Overall intelligibility	78
Inappropriate pitch level	24	Respiratory support	77
Breathiness	22	General rate	74
Increased breathing rate	11	Pitch variation	69

Darley et al. (38) found loudness control problems also occurring in participants with no neurological involvement and Hartelius et al. (53) found significantly higher variability in fundamental frequency (F0) and in sound pressure level (SPL) in their participants with MS and no or mild dysarthria, compared with healthy controls. Poor breath support may also have an impact on the individual's ability to signal syntactic, pragmatic and interactional information (56). There is no unequivocal relationship between the severity of speech symptoms and the score on the expanded disability status scale (EDSS) (57) or duration of the disease (49), but as with respiration, speech is generally more affected when the involvement of neurological functions is larger (38, 41). Also, impaired voice and speech function is more common when cerebellar function is involved, in fact, Grinker and Sahs, in Darley et al. (38), described MS speech to be a type "representing an ataxia of the vocal and respiratory mechanism" .

Murdoch et al. (40) found normal values for the total lung capacity in the participants with moderate to severe cerebellar MS, which is similar to that reported by Altintas et al. (17) in individuals with mild MS. However, the participants in the study by Murdoch and colleagues had a VC below normal limits (< 80% of the expected) in 7 of the 9 participants, and the respiratory kinematics showed restricted and abnormal movements of the rib cage and abdomen during speech tasks (reading, narration, alternate motion rate [AMR], sequential motion rate [SMR], and sustained vowels). The reduced VC was thought to be reflected by the finding that the participants initiated both reading and conversation utterances below the levels reported as normal, and had

slightly lower lung volume excursions compared with a group of healthy controls. The participants also showed irregular (bizarre) movements during inspiration and expiration, which were thought to be related to the breakdown in coordination between the rib cage compartment and the abdomen.

Apart from the neurophysiological impact on speech production, factors such as cognitive load and fatigue may affect an individual's ability to speak. In healthy individuals, speech tasks with a cognitive-linguistic loading affect speech breathing patterns, resulting in slower speaking rate, reduced number of syllables produced per breath group, greater lung volume expended per syllable than under a lower demanding condition, and a smaller volume of the abdomen at breath group termination (58). Demanding speech tasks could therefore pose a communication problem for individuals with MS.

1.1.3 Compensatory strategies

In NMD, there are also speech symptoms which are the result of the individual's conscious or unconscious way of coping with impaired speech function. For example, Scripture (45) found prolonged pauses secondary to irregular breathing in participants with MS, and named these efforts of coping with ataxia "anataxia". Maladaptive breathing strategies are, for example, producing few syllables or words when there is enough respiratory support for longer breath groups (7). An individual with limited respiratory function may compensate for insufficient respiratory support by "downstream" adaptations (1). Such coping strategies are, for example, on a laryngeal level, increasing vocal fold adduction, thereby increasing laryngeal resistance in order to extend the expiratory phase. This will however affect voice quality and intensity. Another strategy for economizing expiration may be increasing resistance in the oral and pharyngeal cavities by changing articulation. This will affect the distinctness of speech articulation and voice intensity, and result in decreased intelligibility (1). From the above, it can be hypothesized, that the maladaptive compensatory strategies themselves can cause exertion, which can lead to increased fatigue, and may further impair voice function and speech.

1.1.4 Respiratory function in CSCI

Vital capacity is decreased by 30-50% of the predicted based on gender and height following CSCI (2). Respiratory function is related to the level of injury and the completeness of injury (21), with higher levels of injury showing more restricted VC. However, as the proportion used for speech is estimated to be about 20% of the VC in healthy individuals (10), respiratory function will probably be sufficient for conversational speech (59). Poor respiratory support could however affect speech production in more demanding speech tasks. One way in which people with CSCI

compensate for the limited respiratory function is by initiating speech at high relative lung volumes (60). In that way they rely, at least in part, on elastic recoil for the generation of subglottal pressure (P_S). Individuals with CSCI also seem to rely more than non-injured individuals on the accessory muscles in the neck, the sternocleidomastoid and trapezius, during speech, both for inspiration and for expiration (61, 62) (see figure 2).
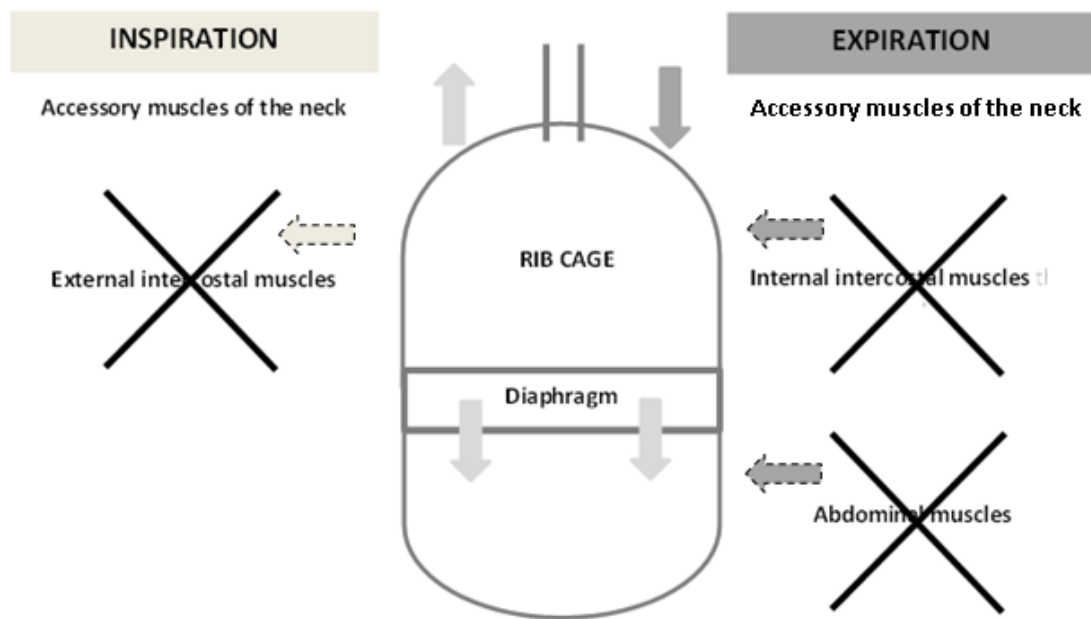
Figure 2. *Respiratory muscle function following cervical spinal cord injury (CSCI). Direction of displacement of abdomen and rib cage when different respiratory muscle groups contract. The direction of movement on inspiration is indicated with light grey arrows, and on expiration with dark grey arrows. Dotted arrows indicate movement direction if muscle function had been intact. (Schematic illustration by the author)*

1.1.5 Speech symptoms related to respiration in CSCI

Since respiratory dysfunction is a known consequence following CSCI, deviation in voice and speech function in individuals with CSCI are likely to be related to the impaired respiration. In Sweden, patients with CSCI are generally not referred to speech-language pathologists. This may be explained by the fact that most individuals with CSCI have intact articulation and are fully intelligible (63). Another contributing factor may be that voice and speech problems are still relatively small compared to the overall body dysfunction following quadriplegia.

Table 2. Studies including information about voice and speech function following cervical spinal cord injury (CSCI).

Study (N)	Respiration	Acoustic	Perceptual, clinical	Self-rating/ Interview
Hoit et al. (60) N = 10	Long inspirations Speech at high lung volume Individual speech breathing patterns		3/10 Short or long breath groups 2/10 ↓ Loudness 4/10 ↓ Voice quality: breathy, rough	Activation of neck muscles "to get extra breath"
Watson and Hixon (64) N = 3	↓ Inspiratory capacity ↓ VC	Short breath groups Deviant pause locations	Slow inspirations ↓ Loudness Imprecise articulation	
Klugman and Ross (43) N = 30				57%:↓ Speech QoL impact in 60% of these
Hixon and Hoit (1) Case, CSCI anecdotal	40% of predicted inspiratory capacity 4% predicted VC MIP -23 cm H2O MEP 2 cm H2O		↓ Loudness Short breath groups Slow inspirations Abnormal phrasing Loudness decay ↓ Adjusting loudness/stress	Speech dyspnea Breathing is fatiguing Voice "lacked power" Difficulty control service dog at distance Speaking-related fatigue
MacBean et al. (63) N = 24		↓ Speech rate ↓ MPT	Pitch, loudness Overall intelligibility	
Johansson, et al. (65) N = 7	↓ VC	↑ CVF0 ↓ MPT ↓ MFT	Harshness, phonatory instability, fry ↑ Pitch	Sw-VHI > 20 in four participants
Tamplin et al. (61) N = 6	s-EMG showed greater activation of accessory muscles in soft and loud voice and prephonatory EMG peaks	MPT within normal limits	Strained, breathy, rough	Md VHI 22.5
Nygren-Bonnier et al. (14) N = 33				Voice limitations due to noisy surrounding, fatigue or dyspnea
MacBean et al. (66) N = 17	M VC = 67% of the predicted (range: 35-125%)	In the group with atypical respiratory kinematic pattern: ↓ Syllables/breath ↓ Syllables/minute	Deviations in: - rate and phrase length - general stress patterns ↓ Loudness 35 % ↓ Respiration	

Note. VC – vital capacity, QoL – quality of life, MIP – maximum inspiratory pressure, MEP – maximum expiratory pressure, cm H₂O – centimeter water, MPT – maximum phonation time, CVF0 – coefficient of variation of F0, MFT – maximum duration of sustained fricative, Sw-VHI – Swedish validated version of the voice handicap index, s-EMG – surface electromyography, Md – median, M – mean

Although voice and speech issues in CSCI risk being neglected because they are relatively minor in comparison with the other sequelae of CSCI, speech function in individuals with CSCI has been investigated in studies using respiratory kinematics, electromyography (EMG), acoustic and perceptual analysis, and self-reports in order to to describe different aspects (see table 2). Speech following CSCI has been described as being characterized by mild dysprosody, reduced speech rate, and phonatory deviations (63). Individuals with CSCI have been found to speak at high relative lung volumes, thus relying on elastic recoil for the generation of subglottal pressure (60).

Clinical observations of speech following CSCI include reduced loudness (60, 64), imprecise consonants and slow inspirations (64), and also changes in voice quality parameters such as increased strain (61), breathiness, and roughness (60, 61).

Acoustic analysis of speech in individuals with CSCI has shown shorter duration of a maximally sustained vowel, lower sound pressure levels (66), shorter breath groups (64, 66), and lower speech rate (63, 66) than control subjects or compared to reference data. MacBean et al. (63) found less intelligible words per minute and thus lower communication efficiency ratio (CER) values for a group with CSCI, in comparison to a healthy control group.

1.1.6 Self-reports regarding voice and speech in MS and CSCI

Poor respiratory drive for speech can have a negative impact on an individual's ability to communicate, making it difficult to talk in a noisy environment, or fatiguing when talking for a long time. Also, in MS, other symptoms such as fatigue, depression, cognitive problems, and a lack of social support have been found to have an impact on the ability to participate in communicative situations (67, 68).

Participants with CSCI in a study by Nygren-Bonnier et al. (14) reported the consequences of impaired respiratory function in daily life. Although voice and speech symptoms following CSCI are often relatively mild, the study revealed that voice function was affected by the respiratory impairment, resulting in limitations such as difficulties being heard in a noisy surrounding, or when calling for help or calling children. When communicating in social situations, such as during phone calls and dinner parties, speech was affected by reduced endurance. They also perceived speech-related fatigue and dyspnea.

Small-sample studies have shown that individuals with CSCI present with relatively high ratings on the Voice Handicap Index (VHI) (61, 65), indicating that the respiratory dysfunction affects voice and speech to such a degree that it results in limitations in communicative participation.

1.2 INTERVENTION TARGETING SPEECH DYSFUNCTION RELATED TO RESPIRATION

Speech pathology interventions for respiratory – phonatory dysfunction in dysarthria include methods aimed at improving respiration, voice and speech function and reducing limitations in activity and in participation (1, 32, 69). A rule of thumb is that if the patient manages to produce an adequate loudness level and to adjust his or her speech breathing according to the speech demands, then respiratory function is probably sufficient for speech purposes, and intervention can focus on voice, articulation, speech naturalness, and compensatory strategies (1, 7, 32).

Farmakides and Boone (46) prescribed the use of expiratory exercises for patients with MS and sufficient respiratory drive for speech, so that "the patient can learn to utilize exhaled air effectively to produce louder phonation". They also reported that the different speech interventions were planned from the individual needs of each of their 68 patients with MS in the study. The authors recommended using "the most effective technique for improving speech: to speak louder." They also recommended working with specific respiratory exercises to develop an understanding of the normal process of respiration, and respiratory drills were used to extend the expiratory phase of respiration.

The Lee Silverman Voice Treatment (LSVT) is a method that also focuses on increased phonatory effort in to achieve increased sound pressure levels. Secondary positive effects, for example on articulation and speech rate, have also been shown as a result of LSVT (70). LSVT was originally developed for patients with hypokinetic dysarthria secondary to Parkinson's disease, but positive outcomes have also been shown in patients with dysarthria of other etiologies, for example in ataxic dysarthria in MS (71).

If the initial assessment indicates weak respiration, therapy should focus on improving respiratory function and respiratory support for speech, before any treatment for improved voice, speech and communicative participation is introduced. The intervention methods may focus on increasing the strength, timing, and endurance of the respiratory musculature, in order to increase and regulate the lung volume, and the subglottal pressure.

For example, training can focus on increased strength of respiratory muscles, so called Respiratory Muscle (Strength) Training (RM(S)T) (72), targeting the expiratory musculature (EM(S)T), or the inspiratory (IM(S)T). EMST is described in more detail later.

In a recent attempt to improve respiratory function, speech function, and mood in individuals with CSCI, a 12-week singing training program (3 times a week) was used

(73). The participants in the group that received singing therapy sustained a vowel with significantly longer duration and with a significantly higher sound pressure level when singing than participants in a control group, but no significant improvement was found in respiratory function. Both groups demonstrated improvements in mood after the 12-week intervention period. Thus, music and singing have positive effects on mood, but, although larger portions of the VC are used for singing, than in speech, singing probably does not challenge the respiratory mechanism enough to increase muscle strength.

When improved muscle function is not a possible goal, the treatment will instead often focus on optimizing respiratory function, and may include postural adjustments, respiratory prostheses, and respiratory techniques (1, 32). Examples of behavioral compensation are to initiate speech at relatively larger lung volumes, and terminate speech earlier in the expiratory phase (32), or train "inspiratory checking", which is a technique that was described by Netsell (74) for balancing the expiratory forces and thereby being able to produce longer breath phrases. An important part of therapy is to identify and train patients to substitute maladaptive compensatory breathing strategies with more well-functioning strategies (1, 44), such as glossopharyngeal breathing (described below) for individuals with paralyzed respiratory musculature.

1.3 INTERVENTION TARGETING RESPIRATION

1.3.1 Glossopharyngeal breathing

Glossopharyngeal breathing, also called glossopharyngeal pistoning for lung insufflation (GI), or "frog breathing" because of the way in which a person performing the breathing looks, is a method used to increase VC when inspiratory capacity is limited due to paralysis of the inspiratory muscles (75). The technique has been used for many years by breath-hold divers, to increase their lung volume above their total lung capacity, thus making it possible for them to stay under water for a longer time (76). In NMD, glossopharyngeal breathing has been used since the 1950s by patients with polio and respiratory dysfunction due to paresis of the inspiratory musculature (77, 78). By using glossopharyngeal breathing, these patients managed to increase alveolar ventilation, and to back-up artificial ventilation in case of failure, or simply make it possible to remain off a ventilator for short periods of time. After learning the technique, individuals have demonstrated a capacity to add liters of air to their VC, even beyond their inspiratory capacity (79). The presumable risks, such as fainting, following glossopharyngeal air stacking, are reported to be minimal (79).

Glossopharyngeal breathing is performed by first inhaling to total lung capacity. Thereafter, small volumes of air are gulped into the lungs with the movements of the

lips, tongue and cheeks to increase VC (77, 80, 81). One gulp of air takes approximately 0.6 sec and has to be repeated about 10 times to get a good ventilation (77). On average, individuals perform 14-20 gulps per breath (78, 79). The technique is relatively easy to learn, and there are instructional videos available on the internet (79, 82).

Glossopharyngeal breathing has resulted in larger lung volumes and capacities in healthy individuals (83, 84), and in patients, for example those with post-polio (77), CSCI (79, 85), Duchenne's muscular dystrophy (86) and stroke (87).

Patients with poliomyelitic paralysis who used glossopharyngeal breathing increased their ability to perform a functional cough and clear secretions, as well as increasing the volume of their voice and the length of their breath phrases (77). McKeever and Miller (87) found improved respiration and speech at assessment after a 5-month intervention when a patient with flaccid dysarthria and impaired respiratory support after a stroke was trained in glossopharyngeal breathing. The patient was able to sustain a vowel for significantly longer after the intervention, and could produce significantly longer utterances; it was suggested that increased lung volumes accounted for these improvements. VC was further increased when the participant maximized his VC capacity with glossopharyngeal breathing, but there were no speech measurements when using glossopharyngeal breathing.

The only report on implementing glossopharyngeal breathing into conversational speech was provided by Hixon and Hoit (1). The patient, who was highly skilled in using glossopharyngeal breathing, was able to extend utterances by using glossopharyngeal breathing intermittently in running speech.

1.3.2 Expiratory muscle strength training, EMST

The goal in EMST is increased strength in the expiratory muscles in order to increase the ability to generate intra-thoracic pressure, which can be used for coughing, the clearance of secretions, and for singing and speaking (10, 72). EMST is performed by exhaling forcefully from TLC against a resistance, using threshold resistance trainers. For the treatment to have effect, training is performed intensively at resistance levels of 70-80% of the individual's maximum muscle strength (72).

Since conversational speech is normally produced on lung volumes above REL in healthy individuals (8), increased expiratory muscle strength could have an impact on the production of loud voice and long utterances (1). Individuals could also potentially benefit from increased muscle strength for the production of prosodic aspects that require fast changes of the subglottal pressure, such as stress and inflections, particularly at phrase endings. As contraction of the diaphragm is dependent on abdominal muscle contraction, increased expiratory muscle strength could improve

inspiratory capacity, particularly in advanced stages of NMD, when pronounced weakness in the respiratory muscles is common and reduced lung function a frequent symptom (3, 5, 6, 10, 88). Also, increased expiratory muscle strength could have effects on the checking action of conversational speech (74), which is presumably more important in individuals with limited VC.

Significant improvement in respiratory muscle strength and lung function after EMST has been found in sedentary elders (89), and for patients with diagnoses, such as respiratory disorders (90). Improvements have also been seen in patients with weakness or loss of function in the respiratory musculature secondary to, for example, Parkinson's disease (91), Lance-Adams syndrome (92), MS (34-36, 93, 94), and in patients with spinal cord injury (95). The effects of EMST on speech and voice were studied in 17 individuals with MS and EDSS 1.5–6.5 (35). After the training period expiratory muscle strength improved significantly, and there were numerical differences in the voice parameters; however, no significant changes were shown.

Also, strength training of the inspiratory musculature has resulted in positive effects on muscle strength and respiration in patients with mild to moderate MS (33) and with severe MS (96), as well as in CSCI (97, 98).

To date, the proportion of individuals with MS or CSCI who are offered and receive speech pathology services is not known. Hartelius and Svensson (29) found that 44% of the participants with MS experienced voice and speech problems, but only 2% had received speech therapy. In the study by Johansson et al. (99) the participants with CSCI reported that voice and speech issues had not been identified by health-care professionals. Possibly because individuals with MS and CSCI have many physical symptoms that are more prominent and have greater impacts on daily life, voice and speech functions risk being neglected, even though impairment in this area may limit the ability to communicate.

1.4   RATIONALE FOR THE INCLUDED STUDIES

This doctoral project was motivated by previously established knowledge about respiratory impairment in MS and CSCI and the relationship between respiratory function and speech and voice production. The limited number of studies of effects of respiratory training on speech and voice function in MS and CSCI further motivated the following studies.

2 AIMS

The overall aim of this project was to investigate voice, speech and communication in individuals with MS and CSCI before and after respiratory training using objective respiratory, acoustic, aerodynamic and perceptual methods of analysis. Also, self-reported data including interviews and self-rating questionnaires were included. The study was motivated by the fact that only a minority of individuals with voice and speech problems following MS and CSCI are identified, and there have been few descriptions of voice, speech and communication in MS and CSCI before and after respiratory training.

More specifically the aims were:

1) To provide an in-depth description of voice and speech function in individuals with CSCI, as measured with respiratory, acoustic, and aerodynamic methods (Study I and IV)

2) To evaluate effects on respiration, voice, speech and communication following GI in individuals with CSCI or MS (Study I and II)

3) To evaluate effects on respiration, voice, speech and communication of EMST in individuals with MS (Study III)

4) To describe how individuals with MS or CSCI perceive their voice, speech and communicative function (Study I, II, III, and V)

5) To investigate how experienced listeners rate voice and speech perceptual parameters in speech in individuals with CSCI (Study I and V).

3 MATERIAL AND METHODS

3.1 PARTICIPANTS

3.1.1 Participants with MS

In studies II and III, the participants with MS were recruited via the Department of Neurology at Karolinska University Hospital. Background data included a measure of disease severity (EDSS, 57), self-rated fatigue (100), and self-rated impact of fatigue on communication (101). All participants gave their written informed consent. In study II, the participant was a 47-year old male severely restricted by his MS (EDSS 9.0), with quadriplegia and severely affected respiration and dysphonia. The participants in study III had mild–moderate MS: EDSS was 6.5–8 for participants 1–3, and 2–3 for participants 4 and 5 (see table 3).

3.1.2 Participants with CSCI

The participants with CSCI in study I were recruited via a study investigating effects of glossopharyngeal breathing in CSCI (79). Inclusion criteria were CSCI >12 months post-injury, level of injury C7 and above with at least partly spared diaphragm function and no other conditions that could affect respiration, voice and speech, and age 18–65 years. Background data in the group with CSCI included gender, age, height, weight, time post-injury, level and completeness of injury.

The participants with CSCI in studies IV and V were recruited via Spinalis, Rehab Station, Stockholm, a clinic with a near complete prevalence population available. Individuals with CSCI were first asked by their primary physician at Spinalis if they were interested in being contacted about the study. Those who agreed (N=25) were contacted by the project leader, informed about the study and asked to participate. Twenty-two individuals agreed to participate, and of these one dropped out before assessment because of work, one withdrew because of disease-related medical problems, and one was excluded because her age was over 65 years. In all, 19 individuals with CSCI participated. The reasons for declining to participate in the group with CSCI included lack of time, participation in other studies, no economic compensation, or did not find the study of importance.

3.1.3 Control group

For studies IV and V, non-injured individuals were recruited to form a control group (CG) via professional and social networks. For minimal inter-group variability in the independent variables, the control participants were carefully matched on an individual level with the participants with CSCI. The participants were matched for gender, age,

height, and weight, in that order (see table 3). The groups did not differ significantly on these variables. As in the group with CSCI, some individuals who were interested in participating withdrew because of the distance to the clinic where the assessments were performed, and some because there was no economic compensation.

Table 3. Characteristics of the participants with cervical spinal cord injury (CSCI), the matched non-injured control group (CG), and the participants with multiple sclerosis (MS) in the five studies.

Study	Etiology	N	Characteristics		Aspects related to condition		Description of speech
I	CSCI	7	Md Age (range) 41 years (30–65)		Level of injury C4–C7		
	Females	3			Md time post-injury		
	Males	4			30 years (3–51)		
II	MS	1	47 year-old male		EDSS 9.0	Fatigue No	Dysphonia related to limited respiration
III	MS	5	Age (years)		EDSS	Fatigue	Dysarthria
	P1: Female		58		6.5	No	Mild
	P2: Female		62		7	Yes	Moderate
	P3: Male		74		8	No	Mild
	P4: Male		31		2	No	Speech dyspnea
	P5: Male		48		3	Yes	Mild dysphonia
IV, V	CSCI	19		M (SD)	Level of injury C3–C7		
	Females	9	Age (years)	41 (10.9)	12 complete		
	Males	10	Height (cm)	174 (10.0)	M time post-injury		
			Weight (kg)	64 (13.9)	15 years (10.9)		
	CG	19	Age (years)	41 (12.2)			
	Females	9	Height (cm)	171 (9.2)			
	Males	10	Weight (kg)	68 (12.5)			

Note. Md – median, C – cervical vertebra, EDSS – expanded disability status scale, P – participant, M – mean, SD – standard deviation

3.2 DATA COLLECTION

3.2.1 Procedure

For an overview of the data collection, see tables 4-6.

Questionnaires (see table 4) were completed by the participants at home, and collected at the beginning of the assessment session. Participants with CSCI

completed a Swedish validated version of the Voice Handicap Index (Sw-VHI) (102), and the participants with MS used the Questionnaire on Acquired Speech Disorders (QASD, *sw*. SOFT) for self-ratings (103). The Participants with MS also performed self-ratings on the Modified Fatigue Impact Scale (MFIS) (100) and the Fatigue Communication Scale (FIS-C) (101). Information on relevant background aspects were collected via questions during an interview.

Speech samples (see table 5) were recorded in a sound-proof booth. The recordings followed standardized routines, making it possible to perform computer-based acoustic analyses of the speech signal. The speech and voice samples included three trials of a maximum sustained /ɑ/ and three of a maximum sustained /s/, two repetitions of syllables at high rate (SMR/AMR), reading of phrases for the assessment of articulation, and nonsense-sentences for the assessment of intelligibility (104), two trials of counting as far as possible in one breath, text reading with and without background noise, narrative speech, and vocalizations for the voice range profiles (VRP). The participants performed the VRP according to Hallin et al. (105). For the elicitation of the VRP, repeated vocalizations were performed in a soft and loud voice across the whole pitch range to reach the extreme F0 and SPL-levels of the participant's voice range (106).

For the estimation of P_S, the intra-oral pressure during the production of a string of 7 syllables consisting of voiceless stops and vowels, /pae/, in three loudness conditions – habitual, loud and soft – was registered. The audio signal was recorded simultaneously (see table 6).

Respiratory testing included spirometry and measurement of respiratory pressures (see table 6). Respiratory measures were collected with a MicroLoop Spirometer (Care Fusion, San Diego, USA), and the spirometry was performed according to the American Thoracic Society Standards (107). The participants performed maximum exhalations, forceful maximum exhalations, and coughing, until there were three trials resulting in values within a 10% range for each respiratory parameter. In study I, VC after a maximal GI- cycle was measured to obtain the VC_{GI}, which is the VC with the glossopharyngeal insufflation volume (GIV) added (79). For each of the respiratory measures VC, forced vital capacity (FVC), peak expiratory flow (PEF), peak cough flow (PCF), MEP and MIP, the best performance of three within a 10% range was used. In study III, respiratory pressures were tested using a respiratory pressure meter Micro RPM (MicroMedical, Basingstoke, UK).

For the evaluation of speech function, a Swedish standardized dysarthria test was used (108).

3.2.2 Collection of probe data and data at follow-ups

In studies II and III, probe data were collected continuously during the intervention period. Also, data were collected at follow-ups after the intervention period. The sessions included assessments and training, starting with audio registrations in a sound-proof booth followed by measurements of respiratory pressures and exertion during the last week's training, self-rated on the Borg CR-10 scale (109). After the measurements were taken, the participant's resistance trainer was adjusted to correspond to 70–80% of the actual MEP (72), the training logg was checked, and instructions were given for the subsequent week. At the last follow-up during the intervention period, the participants were given three open questions covering their experiences of the training and the effects on speech and on life in general. The questions were answered in writing and participants returned the answers to administrative staff in the clinic.

Table 4. Test materials and outcome measures for self-reports.

Task	Measure	Study
Perceived exertion	Ratings on Borg CR-10	III
Rated voice function on Sw-VHI	Test and subtest scores	I, V
Rated speech function on QASD (*sw.* SOFT)	Test and subtest scores	II III
Rated perceived fatigue on MFIS	Test score	
Rated impact of fatigue on communication on FIS-C	Test score	
Questions about the treatment	Written answers	II, III
Interview	Answers transcribed verbatim	V

Note: Sw-VHI – Swedish validated version of the voice handicap index, QASD – questionnaire on acquired speech disorders, SOFT – självsvarsformulär om förvärvade talsvårigheter, MFIS - modified fatigue impact scale, FIS-C – fatigue impact scale - communication

Table 5. Speech tasks and outcome measures related to voice and speech function.

Task	Measure	Study
Max sustained fricative /s/ Counting maximally in one breath Blowing bubbles through a straw in 5 cm H_2O	Max duration (seconds)	I, II, III, IV
Max sustained vowel /ɑ/	Max duration (seconds) F0 (Hz) SPL (dB) CVF0 (%)	
Repetition of syllables at maximum speed	Articulatory rate (syllables/second)	

Text reading, with or without
  background noise
Narration, description of picture
  sequence
	SRP (text reading +narration):
	Area (STdB)
	M F0 (Hz)
	M SPL (dB)
	Min and max F0 (Hz)
	Min and max SPL (dB)
	Rate (syllables/second, WPM)
	Breath phrase (seconds, syllables or
	  word/phrase)	

VRP: vocalizations in extreme pitch and intensity	Area (STdB) M F0 (Hz) M SPL (dB) Min and max F0 (Hz) Min and max SPL (dB)	IV
Perceptual ratings by 2 judges Perceptual ratings by 4 judges	Presence of voice, speech and prosody parameters (mm VAS) - " -	I V

SWINT: reading 10 nonsense
  sentences
	Intelligibility (% correctly
	  transcribed words)
		V

Clinical dysarthria test	Test and subtest scores	III, IV

Note: Max – maximum, cm H_2O – centimeter water, F0 – fundamental frequency, Hz – hertz, SPL – sound pressure level, dB – decibel, CVF0 – coefficient of variation of F0, SRP – speech range profile, STdB – semitones x decibel, M – mean, Min – minimum, WPM – words per minute, VRP – voice range profile, VAS – visual analogue scale, SWINT – Swedish Intelligibility Test

Table 6. Tasks and outcome measures related to respiratory and aerodynamic data.

Task	Measure	Study
Max exhalation after a deep inhalation	VC (liters)	I
Forced max exhalation after a deep inhalation	FVC (liters) PEF (liters per minute)	II IV
Forceful cough after deep inhalation	PCF (liters per minute)	
Maximum exhalation after a deep inspiration supplemented with GIV	VC_{GI} (liters)	I
Forced exhalation after a deep inhalation Forced inhalation after a deep exhalation	MEP (cm H_2O) MIP (cm H_2O)	III
Production of seven /pæ/ in habitual, loud and soft voice	P_S (cm H_2O) SPL (dB)	I IV

Note: Max – maximum, VC - vital capacity, FVC – forced vital capacity, PEF – peak expiratory flow, PCF – peak cough flow, GIV – glossopharyngeal insufflation volume, VC_{GI} – VC supplemented by GIV, GI – glossopharyngeal pistoning for lung insufflation (glossopharyngeal breathing), MEP – maximum expiratory pressure, MIP – maximum inspiratory pressure, P_S – subglottal pressure, cm H_2O – centimeter water, SPL – sound pressure level, dB – decibel

3.3   AUDIO RECORDINGS AND ANALYSES OF SPEECH SAMPLES

3.3.1   Instrumentation

For the speech recordings, a head-mounted electret microphone (MKE2, Sennheiser, Wedemark Wennebostel, Germany) at a 15 cm distance from the participant's lips was used. Speech was recorded in the Phog software (Electronix NG Hitech AB, Täby, Sweden). The acoustic analyses were carried out in the Soundswell software (Electronix NG Hitech AB, Täby, Sweden), and the speech range profiles and voice range profiles were analyzed in Phog.

3.3.2   Acoustic analyses

The longest durations of the maximally sustained fricative and vowel were measured. Duration was also measured for the best trial of counting in one breath. The maximum syllable rate was calculated from the middle 5 seconds of the best trial of the repetitions for the syllables /pa/, /ta/, /ka/, and /pataka/. The middle three seconds of the vowel with the longest duration was used for the measurements of mean FO and mean SPL, and for the calculation of the coefficient of variation of F0 (CVF0). The speech range profiles were based on the recordings of text reading and narration.

The values for the speech area and for the mean, minimum, and maximum F0 and SPL were measured in Phog (Electronix NG Hitech, Täby, Sweden). Speech rate was calculated by excluding all pauses >250 milliseconds (110) and then dividing the total duration in the total number of words for the passage. To calculate the number of words per breath (WPB, or words per phrase, WPP), the total number of words of the text or the narration was divided by the number of phrases. The inspiratory junctures were identified perceptually from the recordings with the visual support of the oscillogram and SPL-registrations in the Soundswell files (111).

In study I, acoustic analyses were repeated for 10% of the speech material to calculate the intra-reliability. The Pearson product moment correlation coefficient calculated for the duplicated analyses was $r = 0.997$.

Voice range profiles provided the maximum area of the voice with the F0 on the x-axis and the SPL on the y-axis (112). Values for the maximum voice area, mean F0, and mean SPL were generated by the Phog software. Minimum and maximum F0 and SPL were measured manually directly in Phog.

3.3.3 Intelligibility

Intelligibility was determined using the Swedish Intelligibility Test (SWINT) (104). For each participant 12 nonsense sentences were recorded. One naïve judge first listened to the two random SWINT sentences recorded, and then listened to the 10 nonsense test sentences for each participant, one at a time. Each sentence was only listened to once, and was directly transcribed orthographically. Two weeks after the listening and transcription of all participants' recorded sentences, the judge listened to and transcribed the 4 randomly duplicated recordings (two from the group with CSCI and two from the CG). Intra-rater agreement was 100%.

3.3.4 Perceptual assessment

In studies I and V, speech and voice parameters were perceptually assessed by experienced listeners according to methods described by Schalling, Hammarberg (113). Two speech language pathologists (SLPs) with several years of experience in assessing speech patients with motor speech disorders performed the perceptual assessments in study I. In study V, four experienced SLPs performed the assessments; in study V, one of these judges was the same as in study I. In both studies, the judges performed the assessments independently, rating a number of parameters on VAS. The protocol in study I was adapted from Schalling et al. (113), and included one overall and seven voice and speech parameters rated on 100 mm VAS, and the parameters pitch, speech rate, and loudness rated on 200 mm VAS (see table 7). In study V, five parameters that were related to restricted respiratory dysfunction and

therefore expected to be present in the speech of the group with CSCI were added, constituting a 16-item protocol; one overall parameter and 12 voice and speech parameters rated on 100 mm VAS, and the parameters pitch loudness and rate rated on 200 mm VAS (see table 8).

For the intra-rater reliability, the recordings of all the seven participants in study I were duplicated and all recordings were randomized. The Pearson product moment correlation coefficient for intra- and inter-reliability was found to be high, $r = 0.98$.

Table 7. Protocol for perceptual assessment in study I. Parameters rated on Visual Analog Scale (VAS)

Parameters rated on 100 mm VAS End points: 0 = not at all, 100 = very much.	Parameters rated on 200 mm VAS End points: -100 mm lower than normal, 0 = normal, 100 higher than normal
Degree of overall deviation Phonatory instability Harshness Strain Glottal fry Short phrases Monotony Imprecise articulation	Loudness Pitch Speech rate

In study V, six recordings from the group with CSCI and six from the CG, i.e. 30% of the recordings, were duplicated, and all of the recordings were randomized. The intra-class correlation coefficient (ICC) was used for the calculations of intra- and inter-reliability. For the intra-rater reliability, the mean ICC for the four judges was >0.6 for 12 parameters, ranging from 0.92 for the parameter "loudness decay" to 0.64 for "monotony". "Imprecise articulation" and "loudness" had ICC coefficients <0.6. For "short phrases" and "long pauses" the ICC coefficient was not possible to calculate, due to the limited variability in the ratings. ICC coefficient values for inter-rater reliability were between 0.60 and 0.72 for nine of the 16 parameters.

Table 8. Protocol for perceptual assessment in study V. Parameters rated on Visual Analog Scale (VAS)

Parameters rated on 100 mm VAS End points: 0 = not at all, 100 = very much.	Parameters rated on 200 mm VAS End points: -100 mm lower than normal, 0 = normal, 100 higher than normal
Impaired respiratory support Breathiness Phonatory instability Harshness Strained-strangled Glottal fry Imprecise articulation Loudness decay Short phrases Unexpected pauses Long pauses Monotony Speech deviates	Pitch Loudness Speech rate

3.4 ESTIMATION OF SUBGLOTTAL PRESSURE, P_S

The P_S is generated by the respiratory apparatus, and is directly related to voice loudness: the higher the P_S, the higher the SPL (114). Therefore, it is possible that individuals with NMD that affect their respiratory function generate P_S that differs from that of individuals with normal respiratory function. They may also have difficulties adjusting their respiratory apparatus for the generation of different P_S depending on the intended sound pressure level.

The P_S is estimated from the intra-oral pressure during the production of the syllable /pæ/. When the bilabial plosive /p/ is produced, the oral tract is closed at the lips, the glottis is open, and the pressure in the oral cavity equals the pressure in the lungs (115, 116). For the vowel /æ/, vocal folds adduct and are set into vibration by the pressure beneath the glottis, the P_S.

For the registration of intra-oral pressure during production of the syllable /pæ/, the participants held a plastic catheter between the lips and about one cm in to the mouth. The plastic catheter was attached to a differential transducer (model 600 D-014, range 0-25 cm H2O, Autotran Inc.). The sound was captured via a cardiod microphone (AKG G420), mounted on a neck set, fixed at a distance of 8 cm (study I) and at 10 cm from the lips (study IV), and calibrated for a distance of 30 cm. Intra-oral pressure

during the production of /pæ/, was registered in Swell in study I, and in Pico Scope (Pico Technology, Cambridgeshire, UK) in study IV, and the sound was recorded in Phog (Electronix NG Hitech AB, Täby, Sweden) in both studies.

The registration and analysis of the P_S and the SPL followed a protocol used earlier (114-117). The participants produced a string of 7 /pæ/ in habitual loudness and at a steady pace of 1.5 syllables per second. This was repeated in a loud voice and in a soft voice, until three sets of seven adequate pressure curves, in each loudness condition, were registered. Simultaneous feedback was provided on the monitor, allowing the quality of the registered pressure curves to be determined.

P_S was estimated from the maximum points of the middle five pressure peaks in the syllable string, in Swell (study I) and in Pico Scope (study IV). The same settings in Swell and in Pico Scope were used for all measurements. Maximum SPL was measured for each of the vowels from the SPL-channel in Swell. Mean P_S and mean SPL were calculated for all three syllable strings in each loudness condition (habitual, loud, and soft voice).

3.5 SELF-REPORTED INFORMATION AND QUESTIONNAIRES

All five studies included the collection of background data, to control for inclusion and exclusion criteria and to obtain information related to the neurological condition, such as duration of the condition, magnitude of symptoms etc. In study V, 19 predefined questions were formulated for background information. Questions included personal data (age, height, and weight), data on the CSCI (time post injury, level and completeness of injury), social factors (family situation, profession, and employment), and history of voice and speech problems and on professional help regarding voice and speech issues. The answers were transcribed verbatim at the time of the interview.

Studies I and V used Sw-VHI to obtain information about the participants' self-perceived voice problems. Sw-VHI is a Swedish validated version of the VHI (118), which consists of thirty statements divided into three subscales: a physical, a functional and an emotional subscale. Each statement is rated on a five-point interval scale; 0 represents never, 1 = almost never, 2 = sometimes, 3 = almost always, and 4 = always. The maximum total score is 120, which indicates severe problems related to voice dysfunction. A total score >20 indicates a voice disorder (102).

The participants with MS in studies II and III rated their perceived speech problems using QASD (*sw*. SOFT; Självsvarsformulär Om Förvärvade Talsvårigheter) (103). QASD consists of thirty statements, with each statement rated on a four-point scale with 0 = definitely false, 1 = sometimes true, 2 = mostly true,

and 3 = definitely true. The questionnaire includes three sections. Part A includes 12 statements about how speech and language is perceived by the respondent, part B consists of 12 statements about communicative participation, and part C has 6 statements about personal and environmental factors.

The MFIS (100) and the FIS-C (101) were used to gather information regarding how the participants with MS perceived fatigue and its impact on communication. The MFIS includes 21 statements rated on a five-point scale, where 0 = never, 1 = seldom, 2 = sometimes, 3 = often, 4 = almost always. The FIS-C is a questionnaire with 20 statements, where the degree of problems with communication the last month because of fatigue is rated on a five-point scale: 0 = no, 1 = small, 2 = moderate, 3 = large, 4 = extreme (problems).

Exertion was rated on Borg CR-10 scale, a logarithmic scale running from 0 = not at all, to 10 = extremely strong (109). It is also possible to mark an 11, which indicates the absolute maximum of perceived exertion.

3.6   RESPIRATORY INTERVENTION

3.6.1   Glossopharyngeal breathing

In study I, the participants received instructions about glossopharyngeal breathing from a physiotherapist, and thereafter performed glossopharyngeal breathing (glossopharyngeal pistoning for lung insufflation, GI) on their own. The intervention consisted of 10 GI-cycles, 4 days a week, for 8 weeks, and the physiotherapist performed follow-up visits once a week. In study II, instructions were also given by a physiotherapist. The participant trained at home on his own, performing at least 3 GI-cycles 3 times a day 3 days a week for 7 weeks. After this intervention period, the participant performed GI when necessary.

3.6.2   Expiratory Muscle Strength Training, EMST

The expiratory muscle strength training (EMST) was performed by exhaling forcefully against a resistance provided by a resistance trainer (see figure 3). The five participants (P1-P5) in study III received instructions in the technique and thereafter trained with the PEP-threshold trainer ("PEPTT") which allows for resistances up to 20 cm H_2O (Respironics, Cedar Grove, NJ) at home following a protocol which consisted of 5 sets of forceful exhalations, five days a week, for six weeks (91). Each week, the participants came in to adjust the level of resistance, if needed, as the load should be set to about 75–80% of the individual's maximum capacity, in order to increase muscle strength (72).
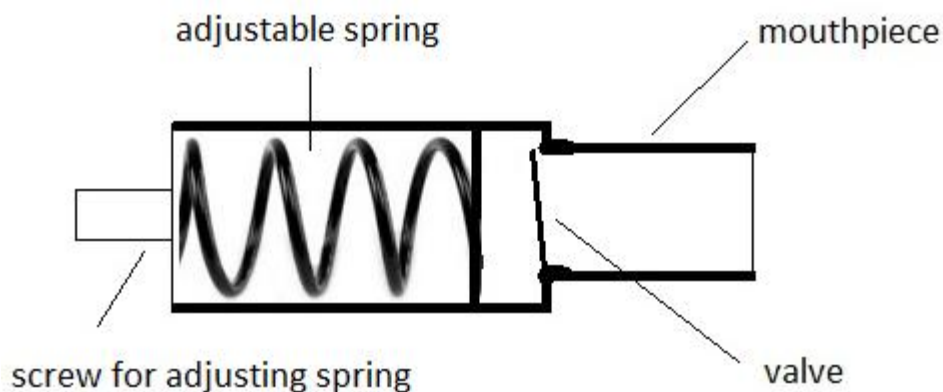
Figure 3. *Components of a resistance trainer. (Schematic illustration by the author)*

P1–P3 completed phase B. After a period with no training, P4 and P5, who had lower EDSS scores (2 and 3), resumed training using the "EMST 150", allowing for resistances up to 150 cm H_2O (Aspire Products, Gainesville, FL). P4 and P5 trained until their performance reached a plateau.

To obtain information regarding perceived training load, the participants rated the degree of perceived exertion on the Borg CR-10 scale (79, 109). At each follow-up, they were asked to rate the perceived exertion during the previous weeks' training.

3.7 STATISTICAL ANALYSES

Statistical analyses included descriptive statistics and comparisons pre and post treatment. Calculations of median and range were performed in studies I and III. Calculations of mean and standard deviations were performed in studies II, IV and V. In study III Cohen's d $(SD_2 - SD_1) / (M_2 - M_1)$ was used to indicate the effect size of change in performance after intervention. In study I, non-parametric statistical methods were used for the comparison between performance pre- and post-treatment. In study IV, parametric methods were used for the comparison between groups. For the analyses of inter- and intra-rater reliability, the Pearson product moment correlation coefficient was used in study I, and the ICC in study V. Regression analysis was used in study IV to investigate the relation between background factors, respiratory variables and speech variables.

All statistical analyses were performed in Excel and in Statistical Package for Social Sciences (SPSS) (119).

3.8   ETHICAL APPROVALS

The studies in the doctoral project were approved by the Regional Ethics Committee of Stockholm: Dnr 2006/127/32 (study I), Dnr 2009/565-31 (studies II-III) and Dnr 2010/2088-31/2 (study IV and V), and Dnr 2013/328-32 (additional application covering inclusion of the CG).

4 RESULTS

4.1 STUDY I

The aim of this study was to describe voice, speech and self-rated communication in 7 individuals with CSCI before and after an 8-week GI-intervention, and when using GI.

The main findings before the 8-week GI-intervention were that phonatory stability, as measured with CVF0 was higher than reference data (113), and the maximum duration of sustained phonation and of sustained fricative were shorter than reference data (120, 121). The perceptual parameters phonatory instability, harshness, and glottal fry were present to a higher degree than other voice and speech parameters. Also, the perceptual ratings showed vocal loudness lower than normal and pitch that was higher than normal. After the 8-week GI-intervention the participants used a significantly louder voice in the production of a 7-syllable string and when reading with background noise. Also, phonation was stabilized, as shown by the significant decrease in CVF0, to levels of vocally healthy reference individuals, and the participants' breath phrases increased from 9.5 to 12.8 words per breath during reading. Perceptually, pitch was normalized. Self-ratings on the Sw-VHI indicated voice disorder in four participants before, and in three participants after the 8-week intervention.

With maximized VC after a GI-cycle, VC increased by 0.6 liters and P_S in loud voice by 4.5 cm H_2O, and SPL increased by 4.6 dB in habitual loudness and 7.2 dB in loud voice.

4.2 STUDY II

The aim of this study was to evaluate effects of GI on respiration and speech in a 47-year old male with quadriplegia due to advanced MS. The participant was referred for voice therapy, but was not able to change his breathing or his voice behavior in any way, such as taking a deeper breath to improve voice quality, increasing loudness or prolonging phonation. Results from GI in individuals with MS and quadriplegia have not been previously reported.

The study used a single-subject study design, with baseline measurements before and after a 7-week GI-intervention. A physiotherapist instructed the participant in the GI-technique. The participant quickly learned how to "frog breathe", and trained 3 x 3 GI-cycles 3 days per week for 7 weeks. After that, he used GI when necessary, which he reported to be typically in the morning and before making phone calls. Respiration and speech were always better after having maximized his VC with a GI-cycle. When maximized with GI, VC increased by up to 60%, he could sustain a vowel 2.5 times

longer, and he could speak in breath phrases twice as long. Some examples are provided in figures 4 and 5. At follow-up assessments 6, 12 and 20 months after the intervention period was completed, further improvement was noted in respiration and in speech when the participant used frog breathing. Despite a severe upper respiratory infection during the intervention period, positive changes remained, or continued improvement was noted, up to 30 months post treatment. The patient experienced a positive impact on his life as a result of the treatment; for example, was able to speak on the phone again.

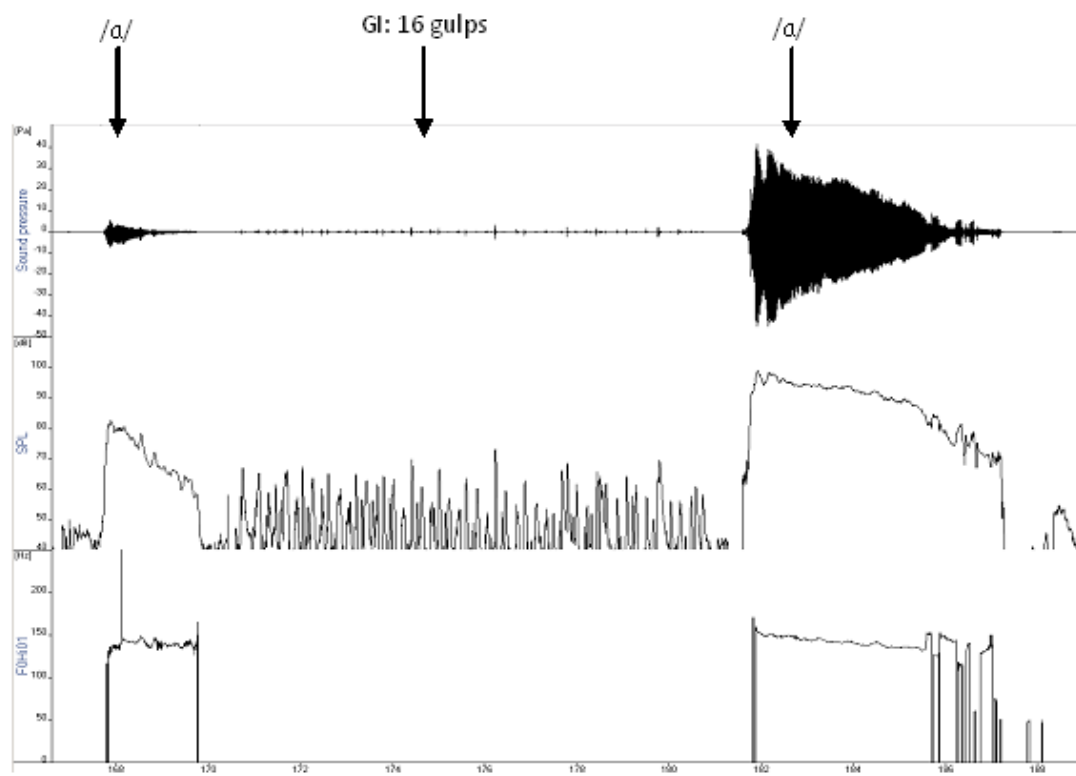Figure 4. *Probe data in study II: the sequence shows phonation without glossopharyngeal breathing (glossopharyngeal pistoning for lung insufflation, GI), a maximal GI-cycle, and phonation at VC$_{GI}$ when the participant with MS sustains a vowel /a/ maximally. The upper panel shows the waveform, the central panel the sound pressure level (SPL) and the lower panel the fundamental frequency (F0) in Swell.*
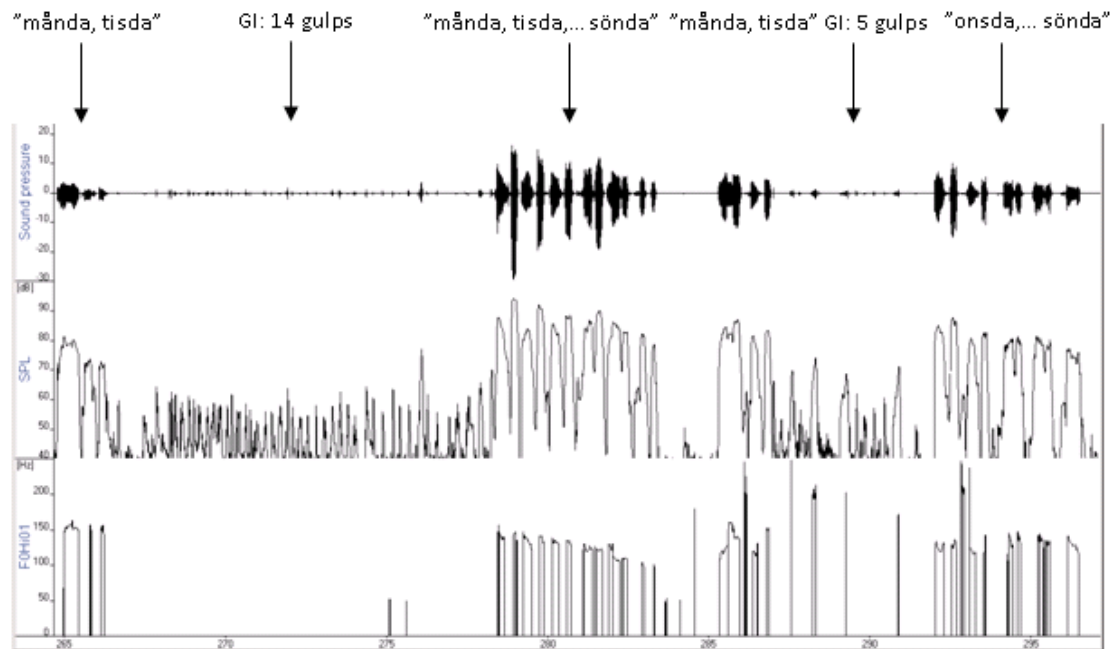
Figure 5. *Probe data in study II: speech without glossopharyngeal breathing (glossopharyngeal pistoning for lung insufflation, GI), a maximal GI-cycle, and speech at VC_{GI}, when the participant with MS counts as many days as he can in one breath. The upper panel shows the waveform, the central panel the sound pressure level (SPL) and the lower panel the fundamental frequency (F0) in Swell.*

4.3 STUDY III

The aim of study III was to evaluate effects of EMST on respiratory muscle strength and speech in individuals with MS. This study also used a single-subject study design (A1–B–A2) across 5 participants (P1–P5), with an EDSS score of 2 to 8. The participants trained their expiratory muscle strength by performing forceful exhalations in a positive expiratory pressure threshold trainer (PEPTT), with a maximum resistance of 20 cm H_2O (phase B). After a period of no training (phase A2), participants P4 and P5, who had the lowest EDSS scores (2 and 3), continued training using the "EMST 150", which offers a maximum resistance of 150 cm H_2O (phase C). Thus, the design was A1–B–A2–A3–C–A4 for P4 and P5. When they reached a plateau in their production of MEP, a period of no training followed (phase A4).

All participants increased their expiratory muscle strength following EMST. Voice intensity increased for P1–P4 during prolongation of a vowel, and for P1and P3–P5 in reading. P1, P3, and P4 increased their duration of a maximally sustained vowel. P2, P4, and P5 increased their phonatory stability. During phase C the improvement was more marked for P4, who demonstrated more stable and improved performances in all speech parameters. All five participants reported that they experienced positive effects of the intervention, such as improved breathing, improved voice and speech (sustained phonation, voice loudness and voice stability), and increased communicative

participation. Also, P5 spontaneously mentioned a decrease in nocturnal coughing and in misdirected swallows of saliva.

4.4 STUDY IV

Study I raised questions about how voice and speech function was affected by CSCI. Therefore, the aims of studies IV and V were to obtain in-depth analyses of voice and speech function in a larger sample with CSCI, and to compare the results in this group with those of a matched non-injured CG. In study IV, respiratory, aerodynamic, acoustic and clinical data were analyzed using quantitative methods.

Performances varied in both groups, but the individuals with CSCI performed significantly worse than the CG on maximum performance respiratory, voice, and speech tasks. The group with CSCI had smaller lung volume and softer voice; they produced a significantly smaller maximum voice area, and they reached significantly lower maximum levels of F0 and SPL during the vocalizations. The participants with a VC of less than 50% of the expected performed significantly worse on several speech tasks, compared with participants who had a VC that was greater than 50% of the expected. The level of injury had an impact on respiratory function in individuals with a complete injury, but other physical aspects did not correlate with respiration or speech. Data for P_S showed that the individuals with CSCI initiated speech (a 7-syllable train) with a numerically higher P_S than the CG, and that P_S decayed over the phrase more than for the CG. Despite this, the group with CSCI produced a significantly softer voice than the CG.

4.5 STUDY V

The aim of this study was to further investigate some of the findings from study I, indicating higher self-perceived voice problems and an increased presence of some perceptual parameters following CSCI, by investigating patient-reported and perceptual assessments of voice and speech in a larger sample with CSCI, as compared with a non-injured CG.

A major part of the group with CSCI reported longstanding changes in voice function and limitations in participation related to these changes. The reports included changes in voice and speech (89%), limitations related to restrictions in voice and speech (39%), 74% mentioned compensatory strategies to adapt speech to the restricted respiratory function, and 58% experienced occupational limitations related to their voice function. One out of 19 had been referred to an SLP. As a comparison, only a few participants in the CG reported limitations related to their voice and speech.

The Sw-VHI total score and the scores for the two subscales "Functional" and "Physical" were significantly higher in the group with CSCI than in the CG, which indicates that individuals with CSCI have more voice problems than non-injured individuals. The perceptual mean ratings of the four judges (in mm VAS) indicated the presence of more pronounced irregular phonation, such as harshness (6.1 mm) and glottal fry (8.9 mm). Loudness was perceived as lower than normal (-7.9 mm) in the group with CSCI. Some individuals with CSCI were also perceived to have impaired respiratory support. The results of the perceptual ratings have to be interpreted cautiously, since the ICC coefficient for inter-rater reliability was low for several of the perceptual parameters. However, the results indicate that perceptual parameters representing aspects of irregular voice quality are the most prominent in both the group with CSCI and in the CG and that these parameters are also present in a larger number of the group with CSCI. The results showed large variations in self-ratings on the Sw-VHI and of perceptual assessments in both the group with CSCI and in the CG.

5 DISCUSSION

This doctoral project has investigated voice, speech and communicative participation following MS and CSCI, both before and after respiratory training. The main findings were that: 1) the components of speech production affected in CSCI were, above all, related to challenging speech tasks and situations, 2) habitual and maximum performance, as well as performance optimized with various compensatory strategies, risk straining the voice mechanism, thereby putting the individual at risk for increased voice fatigue and dysphonia, 3) there were both instant and long-term effects on speech when applying GI, and 4) expiratory muscle strength training (EMST) seems to positively affect some components of speech in individuals with MS.

The findings indicate that individuals with MS and CSCI who have speech problems as a result of impaired respiration should be identified, since respiratory training can improve their voice and speech function, and thus increase communicative participation. The findings of this study add to the body of evidence for respiratory intervention, and also raise questions for further research in the area of speech restrictions related to respiratory dysfunction following neurological conditions.

5.1 SELECTION OF PARTICIPANTS

Not all of the individuals with CSCI who were contacted by their primary physician about participation in the studies IV and V wanted to be contacted again to receive more information about the study. Subsequently, about 80% of those who had agreed to get more information about the study participated, and 20% declined. Studies in various populations on participants' reasons for deciding to participate or not have included the following barriers for participating: additional demands on the patient (appointments, travelling and additional costs), concerns about information and consent (122), current health status, or personal factors (123). A major reason for participating in research studies seems to be altruism, that participating is seen as a civic duty, but is also a way of benefiting the health of family members and friends (124).

Some reported their decision to decline to participate due to a lack of time, or because there was no economic compensation. Some indicated that they had decided not to participate in studies, because of their health condition, while others mentioned that they did not find the purpose of the study motivating. Regardless of the individual reason for participating or not, there is a risk that the individuals agreeing to participate were those who were the least affected by their injury, or were experiencing voice problems. Vice versa, some individuals may have engaged in the study because they did not experience voice problems, but wanted to get a "voice check", particularly since

voice and speech function following CSCI, to date, is not routinely identified by the health-care practice.

Thus, a larger sample would have been desirable to minimize this risk of biases. However, because of the size of the population of individuals with CSCI in the Stockholm area, the possible number of individuals meeting the inclusion criteria for this study was limited. Similar aspects regarding the recruitment of participants were described in Nygren-Bonnier et al. (79), a study with similar inclusion criteria and a similar sample size.

The non-injured individuals invited to participate in the control group (CG) reported the same reasons for not participating as the individuals with CSCI, with the exception of health condition. As for participants with CSCI, those in the CG may also have had different reasons for accepting to participate. For example, individuals with some voice issues may have been more motivated to participate. This could have been the case in study I, where all participants were asked if they wanted an SLP assessment of their voice and speech in addition to the respiratory measurements, but only about one third accepted the offer. It is possible that these individuals were motivated because of limitations in their voice and speech function. The same might be true for some of the participants in the CG, who also had ratings above the cut-off for voice disorder on the Sw-VHI. This further emphasizes the importance of considering factors that may influence inclination to participate, as this may have an impact on the generalizability of the study results.

5.2 SUBGLOTTAL PRESSURE, P_S

One important task of the respiratory system is to generate and maintain a stable P_S during speech production (8, 59). The threshold subglottal pressure (P_S) for phonation is 2–3 cm H_2O (114) and a basic demand for speech production is the ability to sustain a P_S of 5 cm H_2O for at least 5 seconds (59). The mean P_S generated for the five middle syllables in the seven-syllable train by the participants with CSCI in studies I and IV was within normal limits (114), and the mean P_S generated by the group with CSCI for the production of the syllable-train did not differ significantly from that of the CG. However, the resulting SPL was significantly lower for all three loudness conditions in the group with CSCI compared with the CG.

Figure 6 shows mean P_S and mean SPL for each of the analyzed five middle syllables in the registered 7-syllable string. The group with CSCI tended to initiate the syllable train at a higher P_S than the CG, particularly in the production of a loud voice. It can also be observed that the decay was steeper for the individuals with CSCI than for the CG. This may reflect findings similar to those reported by Hoit et al. (60), who

34

noted that their ten participants with CSCI initiated speech on higher lung volume levels. It was assumed that this was a way to benefit of the larger recoil effect at higher lung volumes for the generation of P_S.
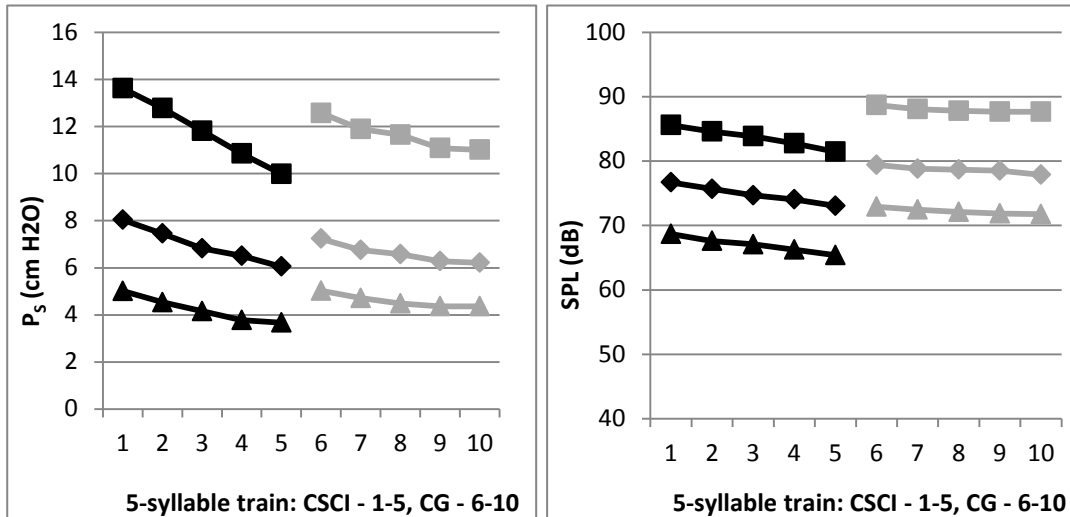

Figure 6. *Subglottal pressure (P_S) and sound pressure level (SPL) for the middle five syllables in the 7-syllable train in the three loudness conditions: loud (squares), habitual (diamonds) and soft (triangles) voice. Mean values for each syllable are shown in black for the participants with cervical spinal cord injury (CSCI), and in gray for the control group (CG).*

In healthy individuals, the recoil forces generate a higher P_S than needed during the first part of the expiratory phase in conversational speech (1). These recoil forces of the rib cage are balanced by activating the inspiratory muscles (10). In non-injured individuals, the external inter-costal muscles serve as "brakes" (74). After CSCI, these muscles are paralyzed and the only functioning inspiratory muscles are the diaphragm and the accessory muscles of the neck (61). However, as the magnitude of the contraction of the diaphragm relies on the activity of the abdominal and external inter-costal musculature, its role as a "brake" during expiration is reduced (10).

When the lung volume decreases to levels below REL, P_S is maintained by increasing the activity of the abdominal and internal inter-costal musculature (8). This musculature is paralyzed in individuals with CSCI (2, 20), but some active expiration is possible by activating of the accessory muscles in the neck (62). Therefore, following CSCI elastic recoil is the major force for the generation of P_S, and the ability to maintain a stable P_S over even a short phrase is impaired, as seen in figure 6.

5.3 HABITUAL VS MAXIMUM PERFORMANCE

The difficulties with controlling P_S can explain why the individuals with CSCI in study IV performed worse on several speech tasks than the participants in the CG. The group with CSCI had a significantly lower speech rate in reading when compared with the CG. Also, the speech range profiles showed that men with CSCI produced significantly lower maximum F0-levels compared with men in the CG. The rest of the speech measures for habitual performance did not differ significantly between the group with CSCI and the CG, but the speech range profiles, which were based on the text reading and the narration in habitual loudness, showed numerically smaller speech areas in men with CSCI than in men in the CG. The larger number of complete injuries in men than in women with CSCI could explain this numerical difference only existing for men.

Above all, the individuals with CSCI performed significantly worse than the participants in the CG on the maximum performance tasks, such as using loud voice, sustaining speech sounds maximally, and vocalizing at extreme F0 and SPL levels. Larger lung volume is used for loud speaking conditions (8, 12, 120). While the lung volume required for conversational speech corresponds to about 20% of the VC, public speaking uses about 65% of the VC (8). As the participants with CSCI had VC ranging from 30% to 65% of the expected volume, several of them can be expected to have great difficulties producing a loud enough voice and maintaining a loud voice for longer phrases. These problems were also reported by the participants with CSCI in study V. Also, the fact that larger lung volume is needed to generate the P_S up to 20 cm H_2O required for challenging speech, explains why the group with a VC below 50% of the expected performed significantly worse on several speech measures than those individuals with a VC >50% of the expected.

Thus, the reduced performance on both habitual and maximum performance speech tasks in individuals with CSCI, as compared with non-injured individuals, could be explained by the difficulty described by individuals with CSCI with regard to generating and maintaining a sufficient P_S.

5.4 COMPENSATORY STRATEGIES

The participants in study V reported a variety of ways to deal with their limited respiration during the production of speech, and the pattern of initiating speech at a higher P_S may reflect at least two of these strategies.

Some participants with CSCI reported working harder or forcing themselves to produce speech in demanding speech tasks. When respiratory muscles are paralyzed, it may be difficult to adjust the respiratory apparatus for the coordination of respiration and phonation during a specific speech task. Therefore, individuals with CSCI have to

rely on a limited respiratory apparatus for the production of speech. Also, impaired sensation could result in distorted feedback from the musculature. One possible way to compensate for these difficulties in controlling the musculature could be to increase the physical effort. This strategy could be used for the production of a loud enough voice over a longer phrase. As reported by the individuals with CSCI, it was necessary to try to inhale deeper when they planned to utter a long phrase.

Even during the relatively short test phrase, a 7-syllable string for a total duration of 3–4 seconds, participants with CSCI had difficulties maintaining a steady P_S. Considering that the mean utterance length in a reading task is about 13 syllables per breath in healthy subjects (125), it is possible that the P_S at phrase endings in running speech declines to pressure levels that are not sufficient to generate loud enough phonation.

In summary, the above include demands for excessive effort in individuals who have to rely on the accessory muscles of the neck for speech production (61, 62). Tamplin et al. (61) found pre-phonatory activity in the accessory muscles of the neck. This indicates that people with CSCI use a lot of effort even before speech is initiated. Strain in these muscles may lead to further fatigue, but the tension may also spread into the muscles of the throat and of the larynx. Therefore, individuals with CSCI risk straining their laryngeal muscles and damaging their vocal folds (126, 127).

5.5 PERCEPTUAL ASSESSMENTS

5.5.1 Reliability

The results of the perceptual assessment have to be interpreted with caution. While inter-reliability was excellent in study I, as calculated with Pearson's correlation coefficient, the inter-rater reliability in study V, as calculated with the ICC, was low for a majority of the perceptual parameters. In study I, two SLPs with several years of experience in assessing speech in patients with dysarthria performed the perceptual assessments, while the assessments in study V were performed by four highly experienced SLPs. One of the two judges in study I also participated in the perceptual assessment in study V. In both studies, the judges independently performed the assessments, rating a number of parameters on VAS. Intra-judge reliability was good in both studies, which may reflect the fact that the listeners were experienced in perceptual assessment. Thus, the procedure and the listener experience were very similar in both studies.

There are several possible explanations for the low inter-rater reliability in study V (128). Firstly, the changes in voice and speech following CSCI were relatively small, and therefore difficult to identify perceptually. The parameters that received high

ratings were essentially the same in the group with CSCI as in the CG. This indicates that these aspects may also be present in the speech of non-injured individuals, and therefore regarded to be within normal limits.

Also, 2 of the 19 in the CG scored >20, a cut-off point for voice disorders (102), which may indicate that the CG also included some individuals with subtle voice problems. There was a large variability in both the group with CSCI and the CG, but the individuals with CSCI scored significantly higher on the Sw-VHI than the CG on a group level. Altogether, this shows that the groups overlapped regarding voice handicap, which might explain the unexpected high prevalence of perceptual dysfunction in the CG and thus, possibly regarding the presence of perceptual aspects. It is possible that this overlap in perceptual characteristics between the group with CSCI and the CG could have been reduced if there been an inclusion criterion of a Sw-VHI score <20 for the CG.

Secondly, the variability in the ratings could also reflect the judges' limited experience in assessing voice and speech in this particular population. Listener training before the perceptual assessment could have resulted in different ratings and may have strengthened the inter-rater reliability (129, 130). It is also possible that an initial procedure where the listeners identify prominent parameters in the speech of individuals with CSCI could have resulted both in a partially different set of perceptual parameters, and higher agreement between the judges. A similar procedure was used in Schalling et al. (113).

Yet another explanation for the difference in inter-rater reliability is that voice and speech were possibly more affected in the participants in study I than in those in study V. In study I, the participants were recruited from a larger study (79). All participants were asked if they were also interested in seeing an SLP for measurements of voice and speech, in addition to the respiratory measurements. About one third of the participants in the study enrolled in study I, and it is possible that part of their motivation was caused by perceived voice problems.

Therefore, these results should be taken as an indicator for further work in perceptual assessments of speech following CSCI. Perceptual assessment of voice and speech in CSCI has not been reported in many studies. Similar results were found in a study of 24 participants with CSCI and 31 healthy controls (63). The authors did not find significant differences at a group level for the majority of perceptual speech dimensions, but the results indicated that speech was characterized by deviations in variation and maintenance of pitch, general stress pattern, breath support for speech, intermittent breathiness, and hyponasality. Rating speech and voice parameters perceptually can be considered a natural, effective, and clinically useful and accessible method (131). However, in the assessment of voice and speech following CSCI,

parameters, rating methods and rating scale properties may need to be revised, in order to define the parameters that distinguish disordered voice and speech in individuals with CSCI.

5.5.2 Perceptual voice and speech characteristics

The perceptual assessments in studies I and V indicated that the perceptual parameters harshness and glottal fry were present to a higher degree than other perceptual parameters in the speech of the individuals with CSCI (see figure 7). The mean ratings for these parameters were numerically lower in study V than in study I, but they were present in a larger number of individuals with CSCI, than in non-injured participants. In both studies, loudness was also rated as lower than normal in those individuals with CSCI.
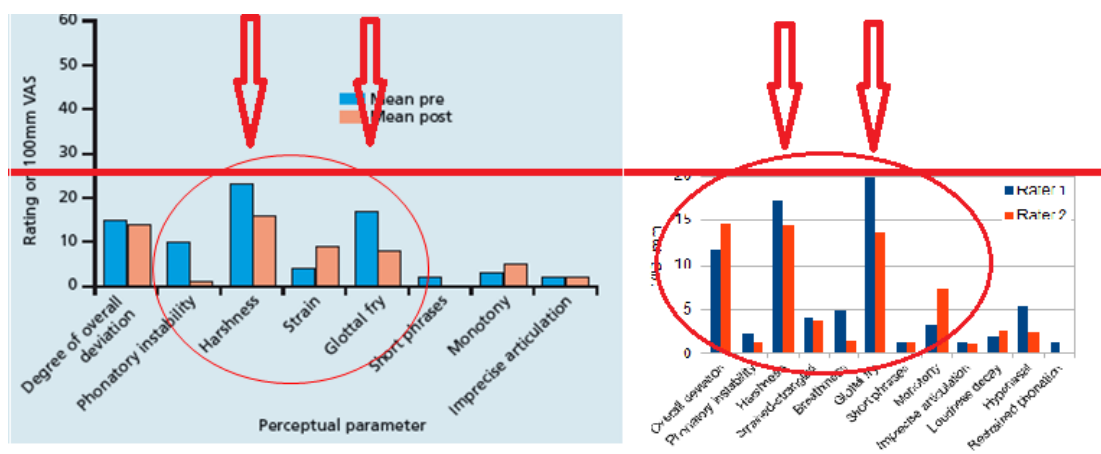
Figure 7. *Most prominent voice and speech characteristics (as indicated by the arrows). The left panel shows the result of the perceptual assessment in study I, and the right panel shows the results of a pilot study including the 12 first participants with CSCI in study V.*

In healthy individuals, the larynx' vertical position descends with increased lung volume, which leads to a less hyperfunctional voice (132). This "tracheal pull" on the larynx exerts an abductory force on the glottis and thereby prevents hyperfunctional phonation. This has a positive effect on the partials of the voice, intensifying them; due to more prominent partials, the voice quality becomes clearer (133). Iwarsson and Sundberg (132) showed that when healthy individuals speak at high lung volumes, and use abdominal muscle support, the tracheal pull on the larynx increases.

This is probably the opposite to the voice production in individuals with CSCI, where VC is smaller than expected and abdominal support absent. In addition, since individuals with CSCI rely on the accessory muscles of the neck for both active

inspiration (61) and active expiration (62), a large amount of muscular effort is used in the neck area for the control of the P_S, thereby risking muscular tension spreading into the muscles of the throat and larynx (134).

When air leaks out, caused by the recoil forces, one method of compensating for an insufficient respiratory support is to save air by increasing the resistance in the larynx or in the pharyngeal and oral cavities (1). Strategies such as this may explain the higher presence of harshness and glottal fry in those individuals with CSCI in studies I and V.

In contrast, an individual could use glottal abduction as a strategy, when the P_S is too high because of the recoil forces. The voice might, in that case, be characterized by a higher degree of breathiness, as air leaks out. In study V, breathiness was perceived in some individuals with CSCI.

5.6   EFFECTS OF GLOSSOPHARYNGEAL BREATHING ON SPEECH

Speech results after GI in individuals with CSCI, and with quadriplegia following MS, have not been previously reported. Existing descriptions of speech following GI are limited to two studies: Hixon and Hoit (1) reported on a participant with quadriplegia who used GI in running speech, while the other study reported one patient with flaccid dysarthria following bilateral medullary stroke (87). In this individual, respiratory function was identified as the main contributing cause for the dysarthria, and oral motor function was sufficient for GI. Speech outcomes were assessed over a 5-month GI-intervention. The participant increased his VC, MPT and the length of his breath phrases during reading and conversation, and VC was larger when maximized with GI. Thus, GI may lead to an increased duration of phonation and of phrases, and louder speech, at least following stroke.

It takes seconds to perform a maximal GI-cycle of about 10–14 gulps of air, which means that considerable training may be needed to incorporate GI into running speech. These two factors probably contribute to the limited use of this technique in speech pathology. Another important factor is that the clinician needs to be familiar with the technique and how to teach it. For the individual reported by (1), the use of GI was automatized and the patient was not always aware of using the technique. Some individuals may, however, need extensive speech training to implement the GI technique fully into their speech.

The participant with MS in study II was completely quadriplegic, and had very poor expiratory drive for speech. The results of the study showed dramatic improvement. The participant was assessed at follow-ups up to 20 months after completion of the 7-week intervention. His voice and speech improved over time, with

and without GI. As a result of the marked effects that GI had on his voice and speech, the participant took up speaking on the telephone again. He typically used GI in the mornings "to pump myself up". The study design with several follow-up assessments up to 20 months after the intervention period made it possible to show that the effect of GI on respiration and speech increased over time.

Also, the results indicate that GI may be a means to maintain respiratory function in individuals with quadriplegia. The participant had a bad cold during most of the intervention period. This may be reflected in his values for FVC and PCF, which became worse during habitual performance than before the intervention period. However, with VC maximized with GI, his values increased. The findings suggest that this individual was to be able to maintain his respiratory function and speech function with the help of GI.

Not surprisingly the increase in lung volume after a GI-cycle has immediate effects on P_S and SPL. The more the rib cage is stretched by the GIV, the larger the elastic forces of the maximally extended rib cage will be. Therefore, since the participants with CSCI in study I already produced median P_S within normal limits, the P_S generated after a maximal GI-cycle exceeded 20 cm H_2O in some individuals. This is higher than necessary for conversational speech, but may be of use for loud speech. The participant with MS in study II also increased his SPL after a maximal GI-cycle, but with his markedly restricted VC; the gain in SPL resulted in normal levels for conversational speech.

Nygren-Bonnier et al. (79) reported positive effects of GI on pulmonary function and chest expansion up to three months after intervention period in their participants with CSCI. Although the chest expansion was not measured in the participant in study II, it is possible that such changes accounted for the long-term effects on respiration and speech. Given the positive effects on the stretching of the rib cage, it is possible that GI could have effects in other populations with NMD, such as Parkinson's disease, where rigidity of the respiratory musculature is common (135). Since relatively well-preserved orofacial, pharyngeal and laryngeal function is required for GI, individuals with Parkinson's disease would probably need to learn the technique early in the course of their disease, when muscle function in the face, mouth, pharynx and larynx are not yet too severely affected.

To date, there are no reports on the effects of GI in patients with different NMDs who start training early in the disease progression. It has been shown that respiratory muscle strength and ventilation are affected early in MS (17). As weak respiratory muscles lead to decreased inspiratory capacity, there is a risk that this in turn leads to decreased respiratory function. It would therefore be interesting to evaluate the effects on ventilation, fatigue, and voice and speech in individuals with mild MS.

5.7 EFFECTS OF EMST ON SPEECH

Healthy adults performing EMST for 4 weeks increased their MEP by 29 to 41% (35, 136). Similar relative increases in MEP (31% to 41%), following EMST, have been reported for individuals with mild to moderate MS (34, 94). In individuals with moderate to severe MS somewhat smaller relative increases in MEP (18% to 36%) have been observed (36, 93). Thus, there is evidence suggesting that EMST is a method with the possibility to reduce the effects of muscle weakness in MS.

Few studies have evaluated the effects of EMST on voice and speech (35, 92, 134). In Roy et al. (134), there was no effect on self-rated VHI following a 6-week EMST in 20 teachers with voice disorders. Jones (92) showed increases in MPT, WPM and communicative efficiency score (CES) after a 6-month intervention with EMST in their participant with Lance-Adams Syndrome and flaccid dysarthria. Chiara, Martin (35) found a numerical, but not significant, increase in MPT and WPM in their group with 17 individuals with mild to moderate MS after an 8-week intervention with EMST. They hypothesized that incoordination of the laryngeal and respiratory muscle could explain the lack of significant voice and speech changes.

In study III, P1–P3 with moderate MS trained with a lower resistance, about 40–50% of their MEP. Despite this rather low resistance, they had to work hard to exhale forcefully. The changes in MEP were the largest in P4 and P5 when they trained with the "EMST 150", which offers resistance up to 150 cm H_2O. However, even these participants with mild MS had difficulty reaching the levels of resistance recommended for an improvement to occur (136). Despite the sub-optimal training load, all five participants with MS improved their expiratory muscle strength, as indicated by their increased MEP, and the training resulted in positive effects on some speech parameters (MPT, CVF0, and SPL). With the single-subject study design it was possible to visualize a decrease in variability in the participants' performances. Also, the participants' self-reports indicated clinical changes in both improved voice and speech function, and improved participation.

There are two possible explanations for the observed and perceived changes in voice, speech and communication in study III: improved coordination of expiration and an overall effect on ventilation.

Cerebellar involvement, which is frequent in MS, may lead to instability in the voice and speech production (47, 53). During the assessments in study III, it was often noted that the participants with MS had difficulties when performing coordinated exhalations through the mouth-piece of the resistance trainer. The intensive training with many repetitions of forceful expiration could therefore have contributed to a better control of the act of exhaling, and resulted in a decreased variability in performance.

This was particularly noted in P4, who markedly reduced his variability of F0 when sustaining a vowel maximally.

Ventilation is reduced in individuals with MS (17). EMST consists of multiple repetitions of inhaling to TLC and then exhaling forcefully, performed several days a week. It is possible that the participants' ventilation improved as an effect of these deep breaths, and this could have resulted in positive effects on the participants' general well-being. Such effects could have accounted for the changes noted in P1 and P3 with moderate MS, and in P5 who perceived restrictions in participation from his fatigue.

An interesting finding was the spontaneous report of P5 that his nocturnal coughing and misdirected saliva swallows had disappeared. Similar findings have been reported in individuals with Parkinson's disease following EMST (135). It is believed that transference accounts for these effects on other muscle groups and functions than those which are the primary goal for EMST (72).

In summary, the results of study III suggest the need for further evaluation of speech effects of EMST in individuals with NMD, in isolation or in conjunction with specific voice and speech exercises. It is possible that EMST can have several different effects on voice, speech and communication in individuals with MS, depending on the neurological involvement of the disease. The goals of the treatment may be different for a severely restricted population than for individuals in early stages of MS. When considering EMST for speech pathology intervention in MS, it is therefore important to specify in what ways the training is expected to contribute to the improvement of voice, speech, or communication.

In addition, EMST is an interesting training method because compliance with this method is high. The participants in study III all expressed their satisfaction with the intervention, which they found easy to perform. EMST is easy to learn and to perform independently or with assistance, changes in performance are easily quantified (92), and the cognitive load is minimal. Feedback on performance is immediate (valve opens or not). An additional factor is intensity: with several training sessions per week, it is possible that the training can develop into a routine.

6 CONCLUSIONS

The following conclusions were drawn:

- Following CSCI, there are longstanding voice changes, resulting in limitations in voice and speech function, and restrictions in communicative participation.

- Above all, the restricted respiratory function after CSCI leads to limitations in challenging speech tasks, such as loud speech or long phrases, and individuals with a VC <50% perform significantly worse on demanding speech tasks, than individuals with a VC >50%.

- Individuals with complete injuries seem to have most voice, speech and communication difficulties, and may be at greater risk for voice fatigue and damage.

- Individuals with CSCI adapt to their voice dysfunction by using a variety of strategies, including speech breathing strategies, compensatory strategies, and avoidance.

- The compensatory strategies used by individuals with CSCI put them at further risk of increased voice fatigue, and possibly of functional organic voice damage.

- GI-training may have positive short- and long-term effects on respiration, voice and speech in individuals with CSCI and MS.

- The changes in some voice and speech measures indicate the positive effects of EMST in individuals with MS: these changes may be more marked in individuals in the early stages of MS.

- The answers to the questions on social validity raise questions about a more general effect from EMST on ventilation, and thereby positive effects on well-being and overall function in the individuals with MS.

- Valid assessment tasks must be defined, in order to identify individuals with CSCI who would benefit from speech therapy interventions (assessment, counseling, voice and speech training).

7 FURTHER STUDIES

- Further research should investigate the effects of speech pathology intervention on voice function, voice fatigue, and communicative participation in individuals with CSCI.

- The results of studies I, IV and V indicate the importance of identifying individuals with CSCI experiencing voice changes and voice fatigue. In particular, studies should focus on individuals with a VC below 50% of the expected, to assess the long-term effects on voice and speech, and possible cut-off for a voice disorder.

- Test materials to detect voice and speech changes in CSCI need to be carefully selected and further developed.

- The results of the extended follow up period in study II suggest that glossopharyngeal breathing may be important for the maintenance of respiratory function and voice function in severe MS, despite the progression of disease. The study should be duplicated with additional subjects with the same condition.

- It is possible that the perceived positive effects on communication were partly due to the more general effects of EMST, for example improved ventilation, in the individuals with MS. This motivates further investigation into the additional effects of EMST in individuals with MS.

- As there were larger effects of EMST in participants with MS and lower EDSS-scores, it would be interesting to study effects of EMST in a group of mild MS. Also, the effects of EMST using low resistance load should be assessed in individuals with moderate to advanced MS.

- Since stiffness in respiratory muscles is common in disorders such as Parkinson's disease (PD), the long-term effects of GI on respiration and voice and speech function in patients who start to train in early stages of PD should be investigated.

- Also, investigations combining respiratory training methods, such as GI and EMST, with speech pathology intervention, such as voice training, should be studied.

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