

Thesis for doctoral degree (Ph.D.)  
2008

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*Life begins when we inhale our first breath  
and ends when we exhale our last*

*To my family*



## ABSTRACT

**Introduction and aims:** The technique of glossopharyngeal breathing was introduced already in the 1950's, but today, few health professionals are aware of its uses. The technique is performed by using the glossopharyngeal muscles to piston boluses of air into the lungs. It has been used by patients to improve ventilation and cough function and by breath-hold divers to prolong their period of time under water. The aims of the present thesis were to determine whether healthy women, elite swimmers, people with cervical spinal cord injury (CSCI) and children with spinal muscular atrophy (SMA) type II were able to learn glossopharyngeal pistoning for lung insufflation (GI) and if so, to describe and evaluate the immediate and long-term effects after a training period of GI on pulmonary function and chest expansion.

**Methods:** In Study I 26 healthy women were recruited; 16 were randomly assigned to the training group (TG) and nine to the control group (CG). In Study II 26 elite swimmers were recruited, 16 men and 10 women. In Study III 25 participants with CSCI were recruited, 20 men and five women. In Study IV 11 children with SMA type II were recruited, eight boys and three girls. All participants performed 10-15 cycles of GI, three to four times a week for five to eight weeks. Pulmonary function tests and chest expansion were measured before and after the training period and also three months after training completed.

**Results:** All of the healthy participants in Study I and II, with the exception of one woman, were able to learn GI. Five of the participants with CSCI and six of the children with SMA type II were not able to perform the technique. The participants in all studies who did learn GI were able to exceed their vital capacity (VC) by 23% and to increase their chest expansion. The men with CSCI had a higher glossopharyngeal insufflation volume (GIV) than the male swimmers in relation to their VC and their chest expansion was also improved in relation to their normal chest expansion. VC increased in the TG compared to the CG in Study I,  $p < 0.01$ .

VC increased for the female swimmers and chest expansion increased for all the swimmers after the training period. Most of the pulmonary function variables and chest expansion increased in participants with CSCI. Some pulmonary function variables tended to improve in the children with SMA type II and chest expansion tended to increase at the level of the processus xiphoideus. Some of the participants reported temporary symptoms when performing the technique, such as dizziness, tension in the chest and some participants even fainted. The improvements in VC persisted three months after training completed.

**Conclusions:** Nearly all of the healthy participants, most of the participants with CSCI and half of the children with SMA type II were able to learn GI. They all performed the technique without any major discomfort. Performance of a training period of five to eight weeks of GI produced positive effects on pulmonary function and chest expansion both in the healthy participants, the participants with CSCI and the children with SMA type II. The improvements were still noticeable three months after training, regardless of whether the participants had continued to train or not.

**Key words:** airway clearance, breathing exercise, buoyancy, chest expansion, cough, glossopharyngeal breathing, maximum insufflation capacity, pulmonary function, physiotherapy

## SAMMANFATTNING

**Introduktion och syfte:** Glossopharyngeal breathing tekniken introducerades redan under 50-talet, men idag är det få inom sjukvården som har vetskap om dess användning. Tekniken utförs genom att använda de glossopharyngeala musklerna för att trycka ner små luftvolymmer till lungorna. Den har använts av patienter med syfte att förbättra ventilation och hostfunktion och av fridykare för att kunna vara under vatten längre. Syftet med avhandlingen var att fastställa om friska kvinnor, elitsimmare, personer med cervikal ryggmärgsskada (CSCI) och barn med spinal muskelatrofi typ II kunde lära sig glossopharyngeal pistoning för lunginsufflation (GI), och om de lärde sig, att utvärdera direkta och långtidseffekter efter en träningsperiod med GI på lungfunktion och bröstkorgsrörlighet.

**Metod:** I Studie I rekryterades 26 friska kvinnor, 16 randomiserades till en träningsgrupp (TG) och nio till en kontroll grupp (CG). I Studie II, rekryterades 26 elitsimmare, 16 män och 10 kvinnor. I Studie III rekryterades 25 personer med CSCI, 20 män och fem kvinnor. I Studie IV rekryterades 11 barn med SMA, typ II, åtta pojkar och tre flickor. Alla deltagare utförde 10-15 omgångar av GI, tre till fyra gånger per vecka under fem till åtta veckor. Lungfunktion och bröstkorgsrörlighet mättes före och efter träningsperioden och även tre månader efter avslutad träning.

**Resultat:** Alla deltagare i Studie I och II med undantag för en kvinna lärde sig GI. Fem av deltagarna med CSCI och sex av barnen med SMA typ II klarade inte av att utföra. Deltagarna i alla studier som lärde sig GI kunde överstiga sin VC med 23 % och också att öka bröstkorgsrörligheten med GI. Männerna med CSCI hade högre glossopharyngeal insufflationsvolym (GIV) än de manliga simmarna i relation till deras VC och även deras bröstkorgsrörlighet var bättre i relation till deras normala bröstkorgsrörlighet. VC ökade i TG jämfört med CG i Studie I,  $p < 0.01$ . VC ökade hos de kvinnliga simmarna och bröstkorgsrörligheten ökade för alla simmarna efter träningsperioden. De flesta lungfunktionsvariabler och bröstkorgsrörligheten ökade hos deltagarna med CSCI. Några av lungfunktionsvariablerna tenderade att öka hos barnen med SMA typ II och bröstkorgsrörligheten tenderade att öka på processus xiphoides nivå. Några av deltagarna rapporterade tillfälliga symtom när de utförde tekniken som yrsel, spänning i bröstkorgen och några svimmade till och med. Förbättringarna av VC kvarstod tre månader efter avslutad träning.

**Konklusion:** Nästan alla friska deltagare, de flesta deltagare med CSCI och hälften av barnen med SMA typ II kunde lära sig GI. De utförde alla tekniken utan något större obehag. Genomförandet av en träningsperiod på mellan fem till åtta veckor gav positiva effekter på lungfunktion och bröstkorgsrörlighet hos de friska deltagarna, deltagarna med CSCI och barnen med SMA typ II. Förbättringarna kvarstod tre månader efter avslutad träningsperiod, oavsett om deltagarna hade fortsatt träna eller inte.

**Nyckelord:** andningsträning, bröstkorgsrörlighet, flytförmåga, sekretmobilisering, glossopharyngeal breathing, hosta, lungfunktion, maximal insufflationskapacitet, sjukgymnastik

## LIST OF PUBLICATIONS

- I. Nygren-Bonnier M, Lindholm P, Markström A, Skedinger M, Mattsson E, Klefbeck B. Effects of glossopharyngeal pistoning for lung insufflation on vital capacity in healthy women. *Am J Phys Med Rehabil* 2007; 86(4): 290-4.
- II. Nygren-Bonnier M, Gullstrand L, Klefbeck B, Lindholm P. Effects of glossopharyngeal pistoning for lung insufflation in elite swimmers. *Med Sci Sports Exerc* 2007; 39(5): 836-41.
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## LIST OF ABBREVIATIONS

CC	Chest expansion at the level of the fourth costae
CG	Control group
CI	Confidence interval
CSCI	Cervical spinal cord injury
CV	Coefficient of variance
CX	Chest expansion at the level of the processus xiphoideus
ERV	Expiratory reserve volume
FEV <sub>1</sub>	Forced expiratory volume in one second
FRC	Functional residual capacity
FVC	Forced vital capacity
DLCO	Diffusion capacity
GI	Glossopharyngeal pistoning for lung insufflation
GIV	Glossopharyngeal insufflation volume
HW	Hydrostatic weight
ICF	International classification of functioning, disability and health
IVC	Inspiratory vital capacity
MEP	Maximum expiratory pressure
MIC	Maximum insufflation capacity
MIP	Maximum inspiratory pressure
NMD	Neuromuscular disorders
PCF	Peak cough flow
PEF	Peak expiratory flow
PEP	Positive expiratory pressure
RV	Residual volume
RMT	Respiratory muscle training
ROM	Range of motion
SD	Standard deviation
SMA	Spinal muscular atrophy
SNIP	Sniff nasal inspiratory pressure
TG	Training group
TLC	Total lung capacity
VA	Alveolar ventilation
VC	Vital capacity
VC <sub>GI</sub>	Vital capacity supplemented by GIV
Vt	Tidal volume

# 1 INTRODUCTION

Respiration is the motor of life. There are many different reasons, caused by a range of different diseases, for normal respiration to become negatively affected in one way or another. One example of such a negative effect is that people with neuromuscular disorders (NMD) often have respiratory muscles weakness that can lead to an inability of the weak respiratory muscles to generate normal levels of pressure and flow during inspiration and expiration. In this way the ability to cough and airway clearance can be reduced, which in turn might result in respiratory complications. To prevent these complications it is important to clear secretions and therefore chest physiotherapy with its many airway clearance techniques is of great importance. Many manual techniques have been developed for this purpose over the years and today there are even technical devices on the market. However, the old techniques should not be forgotten. One of these is the self-administered technique known as glossopharyngeal breathing; this technique was first described in the 1950s and in addition to clinical use, divers have also used this technique as a fundamental tool, to prolong the length of stay under water; it is now practised by breath-hold divers all over the world. Today, there are few health professionals (e.g. physicians, physiotherapists, respiratory therapists, speech therapists) who are familiar with this technique and importantly, few who are also capable of teaching the technique to patients. Given the rarity of purveyors of glossopharyngeal breathing and the benefits it can bring to its users, it is of great importance to highlight this technique.

## 1.1 RESPIRATION- GENERAL ASPECTS

Respiration can be divided into three phases: cellular respiration, transport of respiratory gases and ventilation of the gas exchange organs (breathing). The respiratory muscles are the mechanical components of the breathing system. They can be divided into three major groups, the inspiratory muscles, the expiratory muscles and the accessory muscles of respiration.<sup>1-3</sup>

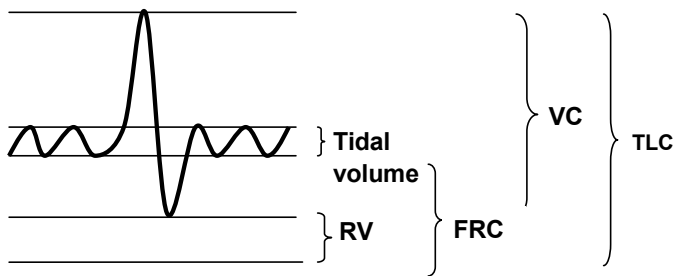
Normal respiration is dependent upon the coordinated efforts of the diaphragm, abdominal muscles, intercostals muscles and accessory muscles, as well as muscles of the upper airway, to maintain patency of the airway for gas exchange. The following movements occur in breathing: during inspiration the diaphragm contracts, the abdominal contents are forced in a caudal direction and the vertical dimension of the chest cavity is increased. In addition the rib margins are lifted and moved out, causing an increase in transverse diameter of thorax.<sup>4</sup> During normal tidal breathing the diaphragm moves about 1 cm, but on forced inspiration and expiration it can move up to 10 cm. The diaphragm contributes approximately 70% to inspiratory tidal volume in healthy people. The phrenic nerve originates from cervical nerve roots 3 (C3), 4 (C4) and 5 (C5) and innervates the diaphragm.<sup>1</sup> Expiration during rest and light exercise represents a predominately passive process of air movements out of the lungs because of the elastic recoil of the stretched lung tissue and relaxation of the inspiratory muscles.<sup>2,3</sup> Normal breathing consists of varying tidal volumes with intermittent deep breaths or sighs. Sighs could be described as periodic hyperinflation, which is a reflex to prevent closure of lung units.<sup>5</sup>

The thoracic cage is as elastic as the lungs. The lung tends to move inward and the chest wall springs outward. Under equilibrium conditions, the chest wall is pulled inward while the lungs are pulled outward, the two opposing actions balancing each other. The chest wall function comes from the passive stiffness of the structural components of the wall (the abdomen, diaphragm and rib cage). For a normal adult, the chest should be stiff enough to resist deformation and inward collapse during inspiration, but not so stiff to affect ventilation negatively. Functional residual capacity (FRC) is the equilibrium volume when the elastic recoil of the lungs is balanced by the normal tendency for the chest wall to spring out.<sup>2,3</sup>

Transpulmonary pressure is the difference in pressure between the inside and outside of the lung. Compliance is the slope of the pressure-volume curve, or the volume change per unit pressure change. The compliance falls if the lung remains unventilated for a long period of time and if the volume is low. This is partly caused by atelectasis of some lung units, but increases in surface tension also occur.<sup>3</sup>

The mucous membranes in the upper airways normally produce a small amount of secretion every day, which can increase in volume if the airways become infected. Normally, any excess secretions can be cleared by coughing. The function of a cough is to clear inhaled foreign materials captured in the mucociliary system, retained secretions or excessive secretions associated with respiratory infection or impaired mucociliary clearance and aspirated materials.<sup>6,7</sup> An effective cough depends on four mechanisms: 1) normal sensory pathways from the lung receptors to the brain to realise mucus retention, 2) deep inspiration to 80-90% of normal vital capacity, 3) glottis closure to hold in the inspired air followed by 4) rapid contraction of expiratory muscles in coordination with sudden glottis opening.<sup>6,8,9</sup> In healthy people, peak expiratory cough flow of at least 360 l / min can be generated.<sup>10,11</sup> To achieve an effective cough, there is a need for insufflation capacity of over one litre and an abdominal pressure, which is produced by a contraction of chest- and abdominal muscles. The maximum insufflation capacity (MIC) is the maximum amount of air inhaled and it is a function of oropharyngeal, laryngeal muscle and inspiratory muscle function to some degree of pulmonary compliance.<sup>5</sup>

Normal breathing is called tidal volume ( $V_t$ ) and it is the volume inspired or expired per breath. The maximal volume expired after maximum inspiration is the vital capacity (VC). The remaining gas in the lungs after a maximal expiration is the residual volume (RV). Total lung capacity (TLC) is the volume in the lungs after maximum inspiration ( $VC + RV = TLC$ ). The volume of gas in the lung after a tidal expiration is the FRC (figure 1).<sup>2,3</sup>



**Figure 1.** Diagram of the lung volumes. RV, residual volume; FRC, functional residual capacity; VC, vital capacity; TLC, total lung capacity

## 1.2 PULMONARY FUNCTION AND TRAINING IN HEALTHY PEOPLE INCLUDING ATHLETES

The four main systems that enable oxygen transport are the heart, the blood, muscles and the lungs. The first three respond positively to exercise, however the lungs do not adapt to exercise. The only stimuli that increase lung growth seem to be hypoxia, particularly in the post natal period, and substantial experimental lung resection.<sup>12, 13</sup> Opinions differ as to whether it is possible to improve pulmonary function by any form of training.<sup>2, 14-20</sup> It is not clear if larger lung volumes have developed due to genetic selection or training of the respiratory muscles during singing or swimming.<sup>17, 18, 21</sup> One accepted fact regarding the response of the lungs to exercise is that large lung volumes generally reflect genetic influences and body size characteristics because exercise training does not appreciably change static lung volumes.<sup>2</sup>

The respiratory system works harder during swimming than in other sports,<sup>17</sup> but it is not clear whether elite swimmers have naturally larger lung volumes or whether they develop these larger volumes as a result of training respiratory muscles during swimming.<sup>17, 22</sup> Armour et al.<sup>17</sup> compared groups of elite swimmers, elite runners and controls and found significantly larger lung volumes in the swimmers than the others. However Mahler et al.<sup>15</sup> reported that static lung volumes and maximal inspiratory/expiratory flow rates were the same for elite athletes as they were for untrained people.

Studies have shown that some singers, wind-instrument players and swimmers have large lung volumes compared to a normal population.<sup>17, 21, 23</sup> Fanta et al.<sup>20</sup> have reported that VC can be increased to a small extent in healthy adults (non-athletes) by making inhalations to TLC with the glottis open.<sup>20</sup> Respiratory muscle training (RMT) has also shown various results, with some studies indicating that VC may be increased in adults by RMT.<sup>24, 25</sup> Leith and Bradley<sup>24</sup> showed that normal subjects could improve their ventilatory muscle strength and endurance with a specific ventilatory muscle training program. This is in contrast to other studies of RMT, which do not show any changes in lung volumes.<sup>26, 27</sup>

In competitive swimming it is of a great importance to reduce the total drag (dynamic and static) to reach optimal swimming velocity. For the total drag of the swimmers the volume of air in the lungs and to a lesser extent, that in the intestine and other cavities is important. By streamlining and making other corrections of the body shape, the frontal and eddy resistance will be reduced during swimming.<sup>28</sup> Passive drag measurements show great differences in drag at different ranges of velocities when the swimmers has inhaled and exhaled lungs.<sup>29</sup> Manipulation with different volumes in the lungs would therefore influence the total drag to be overcome during competitive swimming and a larger total lung capacity would theoretically be beneficial. Furthermore, swimmers competing over short distances (50 m and 100 m) try to minimize the number of breaths, in order to reduce the disturbances in stroking rhythm and body balance caused by breathing.

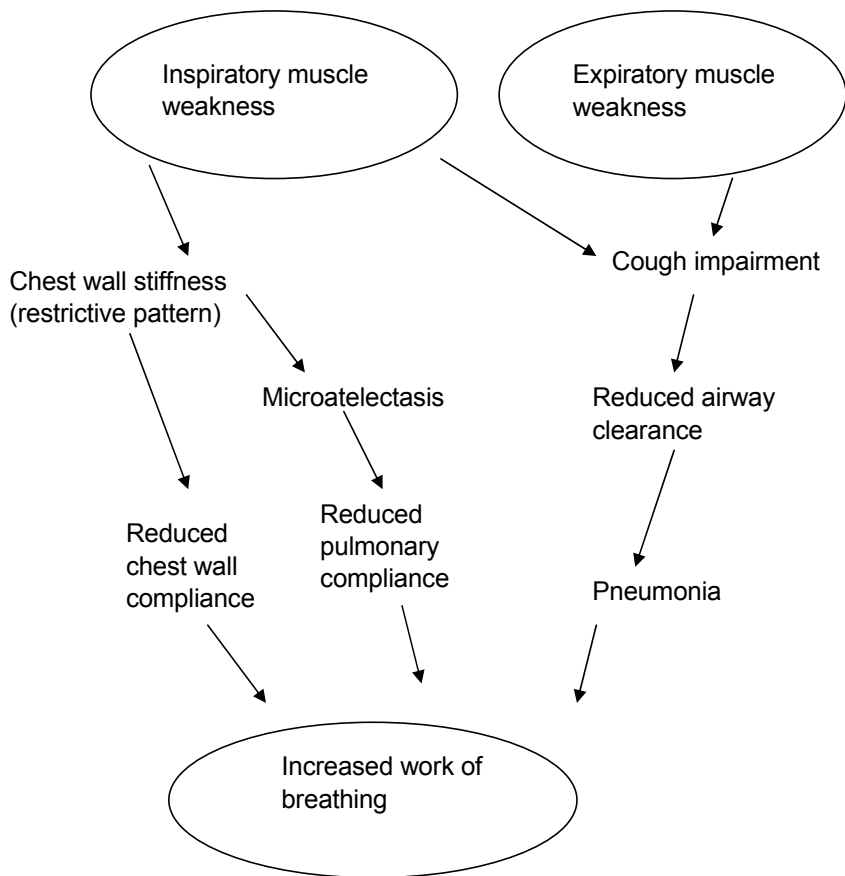
### **1.3 PULMONARY FUNCTION IN PEOPLE WITH NEUROMUSCULAR DISORDERS (NMD)**

In contrast to healthy people and athletes there are many disorders that affect respiration in a negative way. The diseases can be divided in to obstructive diseases such as chronic obstructive pulmonary disease (COPD) and asthma and to restrictive disorders, such as lung fibrosis (affecting the lung) and those suffers from NMD (healthy lung). Diseases within NMD can be divided into diseases of the central nervous system (cerebral cortex, brainstem, basal ganglia and spinal cord) and in disease of the peripheral nervous system (motor nerves, neuromuscular junction and myopathies).<sup>30,31</sup> For patients with NMD, one of the main causes of morbidity and mortality is due to complications of respiratory muscle dysfunction.<sup>32</sup>

People with NMD can have severe inspiratory and expiratory muscle weakness, which diminishes tidal volumes, sighs, and cough flows, resulting in little expansion of the lungs and chest wall. Inspiratory muscle weakness results in a decreased VC that prevents lung expansion to the predicted VC.<sup>33</sup> It is normal for healthy people take deep breaths or sigh regularly and thereby they stretch the respiratory structures this is in contrast to people with NMD. In the absence of deep intermittent deep breaths and sighs, the VC decreases. When the VC decreases, some areas of the lungs remain under ventilated and microatelectasis occurs.<sup>34</sup> This decreases lung and chest wall compliance and range of motion (ROM) which also may be related to alteration in the elastic properties of lung tissues caused by the diminished lung volumes (figure 2).<sup>32,35-37</sup> All told, may develop restrictive respiratory failure.<sup>1,30</sup>

The inspiratory muscle weakness associated with NMD also causes a reduction in inspiratory volume which leads to a reduced cough efficiency. The reduction of expiratory muscle length and the elastic recoil of the respiratory system will be reduced successively. This result in a reduced peak cough flow (PCF) and reduced MIC.<sup>33</sup> This often causes retention of secretions with mucus plugging which may lead to severe pulmonary complications such as pneumonia and then further to respiratory failure. All of this causes an increased work of breathing (figure 2).<sup>30,32</sup> The measurement of VC and PCF is fundamental in the evaluation of pulmonary function.

Cough insufficiency aside from hypoventilation, is the major cause of respiratory complications for people with NMD. Due to inspiratory muscle weakness the volume of air that can be inspired is restricted,<sup>38</sup> which leads to expiratory muscle length tension and that limits chest wall recoil forces.<sup>33, 38, 39</sup> Therefore, intrathoracic pressure, expiratory flow and volume are also limited and this might reduce airway clearance in the expiratory phase. A peak expiratory cough flow of at least 160 l / min has been used as a threshold for people with NMD. However even with a PCF below 270 l / min are considered at risk for respiratory complications, because their PCF can fall below 160 l / min during infections.<sup>40</sup>



**Figure 2.** The effects of inspiratory and expiratory muscle weakness.



### **1.3.1 Pulmonary function in people with cervical spinal cord injury (CSCI)**

People with cervical spinal cord injury (CSCI) with lesions lower than C4-C6, whose phrenic nerves are completely or at least partially intact, can contract their diaphragm. Although their intercostals muscles will probably be weak, they should still be able to function and stabilize the rib cage.<sup>36, 41</sup> This leads to reduced inspiratory function by the lack of chest wall motion or paradoxical motion of the chest wall.<sup>42, 43</sup> VC will decrease while the lungs and thorax will become restricted and passive recoil of the lung and chest wall may occur.<sup>44-46</sup> It is likely that the respiratory muscle might be over used and fatigue in such a scenario.<sup>47, 48</sup>

People with CSCI generate lower intrathoracic pressure than normals and lower expiratory pressure reduces the cough efficiency. The reduced ability to cough and clear pulmonary secretions effectively, leads to reduced airway clearance. As a result of this, atelectasis, retention of secretions, infection and impaired gas exchange may develop.<sup>49</sup>

Pulmonary function tests in people with (CSCI) with acute and complete injuries have shown significant reductions in spirometric parameters. VC can be reduced to 20- 50 % of predicted values.<sup>49</sup> The reduction of VC during the acute phase, appear to improve to some extent and then stabilise over the first six months after injury.<sup>50, 51</sup> The higher and more complete the level of on injury, the greater the respiratory muscle impairment.<sup>51, 52</sup> VC also decreases with age and with duration of spinal cord injury.<sup>53, 54</sup> As a consequence of these respiratory abnormalities, pulmonary complications are one of the most common causes of mortality and morbidity.<sup>55-61</sup>

### **1.3.2 Pulmonary function in children with spinal muscular atrophy (SMA) type II**

There are number of neuromuscular disease that may affect the respiratory muscles of children: one of these is spinal muscular atrophies (SMA). SMA is a group of autosomal recessive neuromuscular disorders characterised by degeneration of spinal cord motor neurons, resulting in progressive muscular atrophy and weakness. The International Spinal Muscular Atrophy Consortium classified the first three types according to establish criterias; type I (severe), type II (intermediate), type III (mild) and type IV(adult), the last a mild form. The age of onset for type II is 7-18 months.<sup>62</sup>

The respiratory problems for children with SMA type II include an impaired cough resulting in reduced airway clearance, microatelectasis, recurrent chest infections, hypoventilation during sleep, and chest wall and lung underdevelopment.<sup>62, 63</sup> In patients with SMA type II, the respiratory disability involves mainly the intercostals muscles with relative sparing of the diaphragm. This give rise to the characteristic pattern of paradoxical breathing associated with the disease. The thoraco-abdominal asynchrony of the respiratory muscles increases the work of breathing.<sup>64</sup> If the lung volumes decrease during the inspiration phase because of the intercostal muscle weakness this could result in underdevelopment of the lungs and chest wall and in bone deformity. The paradoxical breathing may also cause pectus excavatum, a depression of

the sternum in relation to the costal cartilages, to develop if untreated. In turn, the small tidal volumes and the impaired ability to take deep breaths can cause thoracic muscle contracture.<sup>65-69</sup>

Children with SMA also tend to develop spinal deformities due to the respiratory muscle weakness, with scoliosis being the most severe problem. Scoliosis impairs lung function by reducing the mechanical efficiency of the chest wall.<sup>62</sup> Aside from this the children with SMA type II have various grades of reduced range of mandibular motion.<sup>70</sup> A study by Granger et al.<sup>70</sup> showed that children with SMA had one-half the range of opening and protrusion compared to a control group of healthy children. Together with weak masticatory and facial muscles these mandibular problems might lead to nutritional problems.

## **1.4 PHYSIOTHERAPY**

Many definitions of physiotherapy have been proposed across the years and the World Confederation for Physical Therapy (WCPT) defines it thus: “Physical therapy provides services to individuals and populations to develop maintain and restore maximum movement and functional ability throughout the lifespan. Physical therapy is concerned with identifying and maximising quality of life and movement potential within the spheres of promotion, prevention, treatment/intervention, habilitation and rehabilitation. This encompasses physical, psychological, emotional, and social well being”.<sup>71</sup> Different kinds of theoretical frameworks have been developed during the years. Hislop<sup>72</sup> introduced an approach to physiotherapy with influences from system theory and developed the Model of Pathokinesiology. She conceptualised movement as occurring at six different levels: cell, tissues, organs, systems, people and family. The human body and its environment are presented as a communicative system ordered in hierarchies. Tyni-Lenné<sup>73</sup> developed Hislops’ model further when she added a movement hierarchy in her own model. Based on the biopsychosocial model a standardised language for classification of disability was developed, the international classification of functioning, disability and health (ICF) and today it is used by many physiotherapists. ICF is a multipurpose classification of health and health-related states developed by the World Health Organization (WHO) and designed to provide a unified and standard language and framework. ICF can be used as a conceptual model to understand and explain functioning and disability and covers the following dimensions: body structures or functions, activities and participation, and environmental factors (personal factors).<sup>74</sup>

## **1.5 CHEST PHYSIOTHERAPY**

Chest physiotherapy can be defined as the external application of a combination of forces to increase airway clearance and improve ventilation.<sup>75</sup> Clearance of airway secretions is determined by mucociliary debris transport and a sufficient cough mechanism. In healthy people these mechanisms are normally effective and efficient, but will become overloaded if these systems malfunction or are in the presence of excessive bronchial secretions. A document was found on an Assyrian tablet and it is

says to be one of the first reported treatment for airway clearance: “ *If the patient suffers from hissing cough, if his wind-pipe is full of murmurs, if he coughs, if he has coughing fits, if he has phlegm: bray together roses and mustard in purified oil, drop it on his tongue, fill moreover, a tube with it and blow it into his nostrils. Thereafter he shall drink several times beer of the finest quality; thus he will recover* ”.<sup>76</sup>

Different airway clearance techniques have been developed independently during the years. A variety of interventions are used to enhance airway clearance with the goal of improving lung mechanics and gas exchange, and preventing atelectasis and infections. The techniques, intensity, duration and frequency are different between physiotherapists in different parts of the world, and have changed across the years.

Chest physiotherapy has been evaluated by using measurements of airflow, changes in gas exchange, measurements of pulmonary mucus clearance and measuring the volume of expectorated mucus.<sup>75</sup> Techniques described during the years are; postural drainage, breathing exercises (deep breathing, pursed-lip breathing), manual techniques such as percussion, vibration, clapping, shaking, active cycle of breathing techniques (ACBT), autogenic drainage, positive expiratory pressure (PEP) oscillating PEP (flutter), high pressure PEP, inspiratory resistance positive expiratory pressure IR-PEP or RMT, incentive spirometry, physical exercise, continuous positive airway pressure (CPAP), BilevelPAP and mechanical insufflation exsufflation.<sup>7, 75, 77</sup> Glossopharyngeal breathing is a technique that has not have been highlighted over the most recent decades.<sup>7, 75, 77-79</sup>

The basic concepts of chest physiotherapy in paediatric patients are identical to those in adults. However, the child’s growth and development results in continuing changes in respiratory structure and function, and the requirement for different applications of chest physiotherapy in each age group. Difference in airway mucus composition and respiratory mechanics exist and are more important in infancy than in children and adolescents. Other differences are that children have higher compliance of airway walls, smaller airway diameter, fewer alveolar collateral channels, lower elastic recoil pressure and higher chest wall compliance the latter being due to the fact that they have less musculature and less stiffness of the rib cage, while children with neuromuscular disorders have even less musculature.<sup>80, 81</sup> Voluntary cooperation is another thing to have in mind when treating children.<sup>80</sup> The age, sex, the child’s maturity and psychological adjustment all affect the ability to use different techniques. Airway clearance techniques appear to be of benefit in the care of children with neuromuscular disease.<sup>80</sup>

### **1.5.1 Chest physiotherapy in people with NMD**

Respiratory treatment for people with NMD consists of chest physiotherapy (as described above), including assisted coughs. However, given that the patients often have healthy lungs but weak respiratory muscles, other techniques could also be necessary for these patients, such as periodic hyperinsufflation using intermittent positive pressure ventilation and assisted ventilation.<sup>10, 82</sup> Active and passive exercise is also important to help increase or restore the chest wall range of motion.<sup>37</sup> The goals with chest physiotherapy for these patients are to improve or maintain ventilation, chest wall range of motion, cough efficiency and as a result of that decrease the work of breathing.<sup>30, 75, 77, 78</sup>

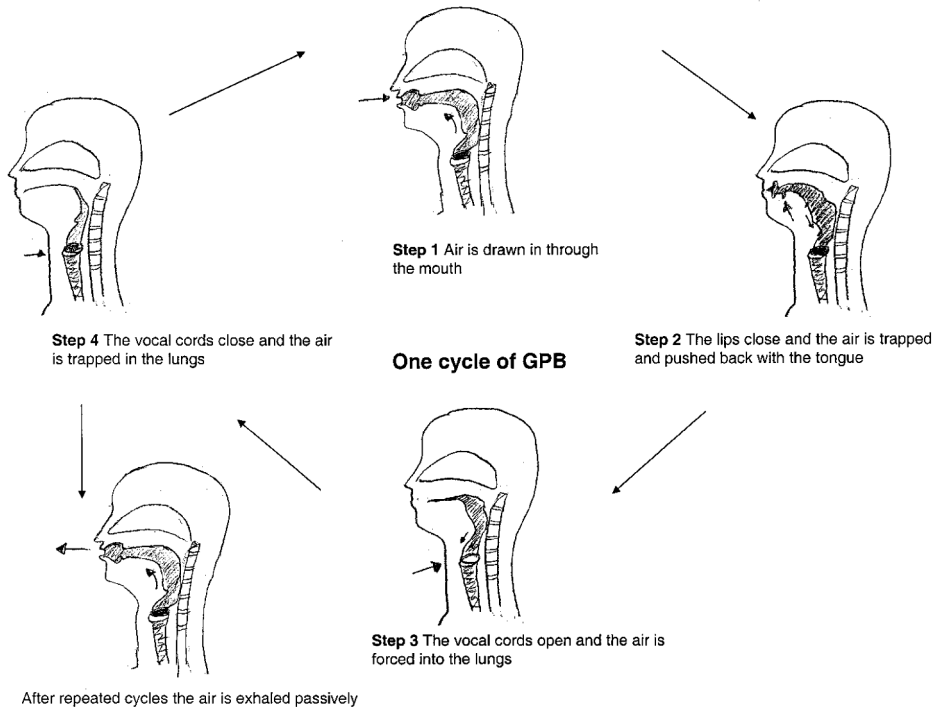
To increase the MIC in patients with NMD and weak inspiratory muscles, air stacking is often a useful approach. Air stacking is the act of deep lung insufflation allowing the patient to approach MIC, and is facilitated by the stacking of sequentially delivered volumes of air that is held with a closed glottis. Air stacking can be performed in different ways, either with a manual resuscitator bag, or with a volume ventilator.<sup>32, 34</sup> A mechanical tool for airway clearance is the mechanical insufflator-exsufflator that produces a pressure limited phase that insufflates the lungs, followed by a rapid reversal to negative pressure that removes the insufflated volume at an expiratory flow sufficient to clear secretions.<sup>34, 83</sup> This has been helpful for people with NMD with a weak cough and intact upper airways.<sup>84, 85</sup>

Expiratory muscles can be manually assisted by providing thoraco-abdominal thrusts to a maximally inflated lung and an assisted cough.<sup>86</sup> By combining MIC with spontaneous cough manoeuvres the patient can use the stored elastic recoil energy developed with lung and chest wall expansion to produce a PCF that is sufficient to clear secretions.<sup>33</sup> Pulmonary compliance and the inspiration phase are important to generate adequate insufflation volumes and producing maximal PCF. To increase MIC for the patients with a reduced VC without any mechanical aids, the self-administrated technique, glossopharyngeal breathing can be used.<sup>34, 87</sup>

## 1.6 GLOSSOPHARYNGEAL BREATHING

The first report of glossopharyngeal breathing was published by Dail in 1951<sup>88</sup> in patients with poliomyelitic paralysis. It is a technique that is performed by using the muscles of the mouth, cheeks, lips, tongue, soft palate, larynx, and pharynx to piston boluses of air into the lungs. The tongue is the main organ of this breathing technique. The tongue is pushed upwards and backwards forcing the air into the pharynx. The larynx opens and the air pass into the trachea where it is trapped by closure of the larynx.<sup>89</sup> This pistoning action is the mechanism of each gulp. A gulp is defined as the boluses of air projected into the trachea by the pistoning action of the tongue.<sup>79, 90-94</sup> In glossopharyngeal breathing the patients first perform a TLC manoeuvre and then beyond that they perform cycles of e.g. 6-10 gulps of air for example, followed by the relaxing of the larynx and passive expulsion of the air (figure 3). Some people perform glossopharyngeal breathing with the mouth (pinch their nose) while others perform glossopharyngeal breathing with their mouth closed, drawing the air through the nose.<sup>79, 92, 95</sup> In virtue of its resemblance to the mode of breathing of frogs it is also known as “frog breathing”.<sup>92</sup> It is an alternative technique of breathing which maintains an adequate ventilation when respiratory muscles are weak.<sup>79, 90-94, 96, 97</sup>

Glossopharyngeal breathing is used by breath-hold divers to help them increase their lung volume above their normal TLC and thereby increase diving performance.<sup>98-101</sup> Large lung volumes have been reported in competitive breath-hold divers,<sup>98</sup> but whether this is a result of selection of genetically gifted individuals or whether it is a training effect from glossopharyngeal breathing is not known.



**Figure 3.** One cycle of glossopharyngeal breathing (GPB) (own drawing)

Glossopharyngeal breathing has been called many different names such as frog breathing, buccal pumping, lung packing and carpa. For the patients it is most commonly known as glossopharyngeal breathing. The terminology has also been different in different studies. In most studies it is called glossopharyngeal breathing,<sup>90, 93, 94, 96, 97, 102-107</sup> but in studies involving breath-hold divers it is known as glossopharyngeal insufflation or inhalation<sup>98, 101, 108, 109</sup> The word breathing means both the inhalation and exhalation phase of one breath but when performing glossopharyngeal insufflation/inhalation the focus is on the insufflation phase.<sup>98</sup>

### 1.6.1 Physiological effects of glossopharyngeal breathing

The mechanics and risks of glossopharyngeal breathing are described in previous studies. In a study by Collier et al.<sup>91</sup> it was shown that the resistive work is rather small. The effort of expanding the thorax is greater than in the lungs. High intrathoracic pressure can develop and a concurrent depressant effect on the arterial pressures. When a person performs glossopharyngeal breathing the intrathoracic pressure increases. This will have a similar effect to a valsalva manoeuvre, of when a person voluntarily strain to increase the intrathoracic pressure, this increase may cause orthostatic syncope from the reduction in venous return.<sup>109</sup> Collier et al.<sup>91</sup> showed that in patients with reduced respiratory muscle function, the increase in

pulmonary gas caused a drop in arterial blood pressure. Many reports have shown that significant hemodynamic abnormalities occur during glossopharyngeal breathing. Arterial blood pressures falls and heart rate increases in healthy people.<sup>109, 110</sup> In a study by Loring et al.<sup>108</sup> the tranpulmonary pressures increased up to 80 cm of H<sub>2</sub>O with glossopharyngeal breathing and the intrapulmonary pressures increased up to 109 of cm H<sub>2</sub>O. Those results indicated that some healthy individuals are able to withstand repeated insufflations to transpulmonary pressures greater than normal exposition. Contra indications have in earlier studies have included; bulbar dysfunction, failing cardiac function or diseases that affect the lungs.<sup>111, 112</sup>

## **1.7 RATIONALE FOR THE THESIS**

The cause of morbidity and mortality for people with NMD is mainly due to respiratory complications brought on by a reduced coughing ability, and therefore a reduced ability to clear secretions that can lead to severe respiratory conditions. The overall treatment goals for patients with NMD with reduced pulmonary function are to improve or maintain ventilation, chest wall range of motion, improve cough force and to improve and maintain the lung volumes.

It is well known that the MIC and PCF can be increased when patients with reduced lung volumes perform glossopharyngeal breathing,<sup>90, 91</sup> however it is unclear if there are any long-term or sustained effects on pulmonary function and chest expansion after a training period of glossopharyngeal breathing. If it is possible to improve pulmonary function and chest expansion both immediate (while performing glossopharyngeal breathing) and in long-term (after a training period) it would be of great value for the patients to enhance the capability to clear secretions and reduce pneumonia.

Most studies up to now have focused on patients with a reduced VC to achieve a volume closer to their predicted VC while performing glossopharyngeal breathing.<sup>95, 105</sup> To our knowledge no study has investigated the effects of on pulmonary function when performed above TLC after a specific training concept.

Glossopharyngeal breathing is a technique that on one hand can help to improve performance in the sporting arena and on the other can be considered as an authentic life-saving manoeuvre in patients with restrictive respiratory failure. Glossopharyngeal breathing is a part of the potential breathing capacity of humans. To benefit and optimize the use of glossopharyngeal breathing in patients it is of great importance to achieve the ultimate knowledge of its physiology, mechanisms and effects of specific training concepts.

## 2 AIMS

The overall aims of this thesis were to evaluate if healthy women, elite swimmers, people with cervical spinal cord injury (CSCI) and children with spinal muscular atrophy (SMA) type II were able to learn glossopharyngeal pistoning for lung insufflation (GI) and if learned, to describe and evaluate the immediate and the long term effects after a training period of GI on pulmonary function and chest expansion.

The specific aims were

- To evaluate if the participants were able to learn GI and if it the technique caused any discomfort (Study I-IV)
- To evaluate the immediate effects and the long term effects after a training period of GI on pulmonary function and chest expansion (Study I-IV)
- To evaluate the effects of GI three month after completed training period (Study I-IV)
- To compare the effects of GI on pulmonary function and chest expansion between male swimmers and men with CSCI (Study II and III)
- To compare the effects of GI on pulmonary function and chest expansion between female and male swimmers (Study II)

## **3 MATERIAL AND METHODS**

### **3.1 STUDY POPULATION AND DROP OUTS**

In Study I the criteria for inclusion into the study were: women aged 18-60 years, who were healthy and non-smokers. A sample of participants was recruited from a physiotherapy clinic at a university hospital. Approximately 70 female physiotherapists answered a study recruitment advertisement, and of these 26 were accepted into the study. The training group (TG) consisted of two thirds of the participants with the control group (CG) making up the remaining third, the participants were randomly assigned either to the TG (n=17) or to the CG (n=9).

In Study II the inclusion criteria's were: male and female elite swimmers, who were aged between 18- 35 years. A sample of swimmers from different swim clubs in the Stockholm area was asked to participate. Thirty-five participants from Swedish elite swimmers who had competed in the Swedish, European or World Championships or in the Olympic Games in 2004 accepted to participate. The swimmers competed with different strokes and at different distances, and all trained in the pool between eight to ten times per week. Initially the male group consisted of 20 participants, and the female group consisted of 11 participants. However four male swimmers dropped out, one having contracted an airway infection while the other three left the study due to lack of time. One female swimmer also dropped out because of lack of time. Finally the study population consisted of 16 males and 10 females.

In Study III the inclusion criteria were: a spinal cord injury with a lesion between the level of C4 and C8, with time between injury and inclusion of at least one year, an American Spinal Injury Association (ASIA)<sup>113</sup> impairment classification of A, B or C, ventilatory independence, an age of between 16-70 years. Exclusion criteria were: chronic obstructive pulmonary disease (COPD), symptomatic infection characterized by fever, the inability to perform GI because of the presence of severe conditions, trauma or cognitive dysfunction. Invitation letters were sent to 111 people at the spinal cord injuries clinic at the Karolinska University Hospital in Stockholm and 37 people volunteered. The reasons for the people who declined participation were due to lack of time and low motivation. In the introductory stage of the study 12 participants dropped out due to upper airway infection or lack of time. Finally 25 participants completed the study protocol.

In Study IV the inclusion criteria for the study were: children diagnosed with SMA type II and who were aged between 6-16 years. Exclusion criteria included the presence of a tracheotomy, a symptomatic upper airway infection characterized by fever, cognitive dysfunction, or if the child was too disabled to perform GI due to the presence of severe symptoms. All children with spinal muscular atrophy type II (n=11) at the National Respiratory Centre (NRC), Danderyd, Sweden were asked to participate and all 11 children entered the study.



**Table 1.** Baseline characteristics of the participants in Study I-IV.

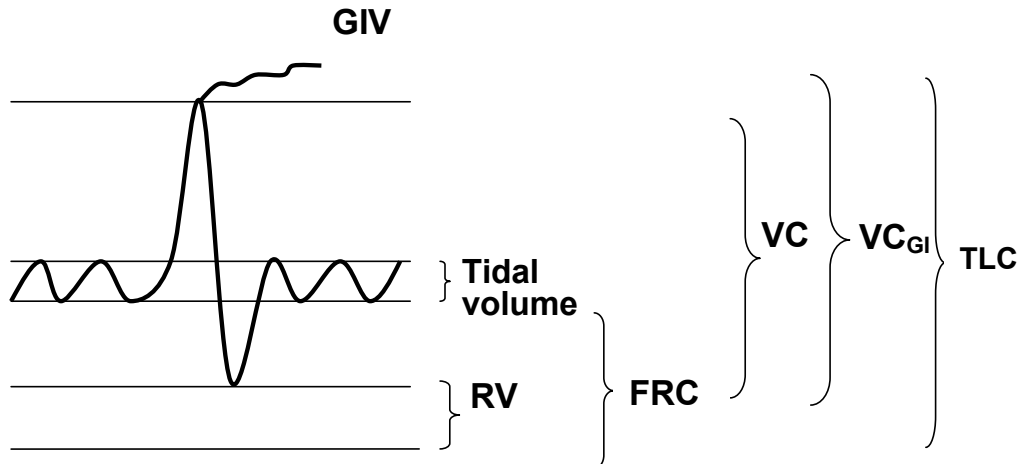
	Study I (n=26)	Study II (n=26)	Study III (n=25)	Study IV (n=11)
Age (yrs)	37 (25-58)	21 (17-30)	47 (21-70)	11 (6-15)
Gender (male/female)	0/26	16/10	20/5	8/3
VC % Predicted	121 (95-156)	128 (107-151)	67 (23-95)	33(11-68)
TLC % Predicted	115 (100-135)	125 (102-154)	81 (59-109)	55 (30-79)
RV % Predicted	108 (88-131)	118 (70-186)	120 (30-197)	156 (60-259)

VC, vital capacity; TLC, total lung capacity; RV, residual volume.

### 3.2 TERMINOLOGY AND DEFINITIONS

The focus in Studies I-IV was on the insufflation phase of breathing and therefore the term *glossopharyngeal pistonning for lung insufflation (GI)* was used. *Pistonning* is the world used to explain mechanism for gulping. Glossopharyngeal insufflation volume (GIV) is the lung volume over the VC added by GI.  $VC_{GI}$  is the vital capacity supplemented by GIV, thus  $VC_{GI} = VC + GIV$  (figure 3).<sup>114, 115</sup>

Within this thesis the term *immediate effects* refers to any effect noted during the performance of a GI manoeuvre, thus the measurements are taken at  $VC_{GI}$ . Similarly the term *long-term effect* is defined as the effects noted after a training period of GI.



**Figure 4.** Diagram of lung volumes including glossopharyngeal insufflation volume (GIV) and vital capacity supplemented with GIV =  $VC_{GI}$  (modified from Lindholm and Nyren<sup>98</sup>).

### **3.3 PROCEDURE**

Each participant in Study I-IV received individual instruction on the GI technique from the same physiotherapist. They watched an instructional video, received written information and practiced the technique with the instructor. The children in Study IV also practiced the technique together with their parents and/or their personal assistant.

The participants first carried out a maximal inhalation and then performed GI using as many gulps of air as possible without discomfort. Finally, they relaxed the larynx and air was passively expelled (figure 3). All participants gulped through the mouth; a nasal clip was used if needed to avoid air leakage past the soft palate. All carried out a short warm-up with stretching exercises for the chest. Participants performed GI in a sitting or supine position. In each study the participants had to exceed VC by the limit of at least five percent in order to be able to manage GI.

In Study I, the TG performed 15-30 cycles of GI three times a week for six weeks. The CG did not perform GI. TG and CG were followed up once a week.

In Study II, the swimmers performed 10 cycles of GI four times a week for five weeks. TG and CG were followed up once a week.

In Study III, the participants performed 10 cycles of GI. They performed GI, at least four times a week for eight weeks.

In Study IV the children performed 10 cycles of GI. They performed GI, at least four times a week for eight weeks. The children performed GI on their own or together with their parents or their personal assistant.

In Study III and IV, the participants were followed up at least three times during the eight weeks.

After the training period the healthy women in Study I were instructed not to continue to train. The elite swimmers in Study II were divided into two groups after the training period: half were instructed to continue to train and the other half not to. The participants in Study III and IV were instructed to continue to train if they wanted to.

### **3.4 MEASUREMENTS**

All measurements were performed by the same test leader before and after training. Participants were blinded to their own results throughout the whole study. Some intermediate measurements were taken during 'follow ups'. In addition measurements of pulmonary function and chest expansion were taken three month after participants completed the training period. All measurements were measured at the body function level according to ICF.

### 3.4.1 Pulmonary function tests

#### 3.4.1.1 Spirometry

Static and dynamic spirometry was carried out in Study I and II including measurements of VC, expiratory reserve volume (ERV), FRC, RV, TLC, forced vital capacity (FVC) and forced expiratory volume in one second ( $FEV_1$ ) using a body plethysmograph (Master Screen Body, Jaeger). The coefficient of variance (CV) of test-retest measurements of VC (within-day) (n=50) was 1.1%.

A portable infrared interruption flow sensor (SpirobankG, [www.spirometry.com](http://www.spirometry.com), MIR, Italy) was used to measure VC before a training session. It was subsequently used to measure  $VC_{GI}$  each week and to evaluate the participants' technique. In Study I the CG performed measurements of the VC each week.

Static and dynamic spirometry in Study III and IV, including measurements of VC, ERV, FRC, RV, TLC, FVC,  $FEV_1$ , diffusion capacity (DLCO) and alveolar ventilation (VA) were carried out using a Vmax 229 spirometer (SensorMedics, Yorba Linda, CA, USA). Lung volumes were measured using the nitrogen washout method.

A portable infrared interruption flow sensor (Micro Loop; Micro Medical, UK) was used for 'follow up' measurements of  $VC_{GI}$  during the eight week training period, along with measurements of VC before and after a session, enabling calculation of GIV according to  $VC_{GI} = VC + GIV$ . PCF were measured by coughing into a tight-fitting full face mask (Laerdal Medical AB, Sweden) connected to the Micro Loop.

All measurements were performed in accordance with the American Thoracic Society standard (ATS).<sup>116</sup> Reference values from Quanjer et al.<sup>117</sup> were used for comparison. For the participating children reference values were taken from Polgar et al.<sup>118</sup>



**Figure 5a and 5b.** Static and dynamic spirometry measured in a body plethysmograph (5a) or with the Vmax 229 (5b).

#### 3.4.1.2 Maximal expiratory and inspiratory pressure and sniff nasal pressure

Mouth pressure in Study III was measured using a mouth pressure meter (Precision Medical Ltd, UK) and in Study IV, a Micro RPM (respiratory pressure meter) (Micro Medical, UK) was used. To measure the maximum expiratory pressure (MEP) participants performed a maximum expiratory effort after a maximum inspiration. The maximum inspiratory pressure (MIP) was measured by exerting the maximum inspiratory effort after a maximum expiration. The value for MEP and MIP was taken as the highest value after three or more attempts.<sup>119</sup> In addition to the measurement of MIP in Study IV, maximal sniff nasal inspiratory pressure (SNIP) manoeuvres were performed from functional residual capacity with the participating children seated without a nose clip. Pressures were measured at the nose via a pressure line inserted into the Micro RPM. Repeated sniffs were performed until no further increase in pressure was seen, with the highest value being recorded as the SNIP.<sup>120-122</sup>

#### 3.4.2 Chest expansion

Chest expansion was measured in Study I-IV at the level of the processus xiphoideus (CX) and at the fourth costae (CC) using a tape measure. The participants were instructed to perform a maximal exhalation (to RV) followed by an inhalation to TLC. Chest expansion was calculated as the difference between circumferences at RV and TLC.<sup>123, 124</sup> Chest expansion was also measured after gulping to TLC<sub>GI</sub>.



**Figure 6.** Chest expansion measured at the level of the processus xiphoideus

#### 3.4.3 Hydrostatic weight and buoyancy

Hydrostatic weight (HW) and buoyancy were measured in Study II. Buoyancy is the lifting power of an object in a fluid. According to Archimedes' principle, the buoyancy of an object reflects its ability to sink or float in water and is measured by the weight difference between an object and an equal volume of water.<sup>2</sup>

The weight of each participant in water was measured after full exhalation at RV,  $HW_{RV}$ , (figure 7a) and after a full inhalation at TLC,  $HW_{TLC}$  (figure 7b).  $HW_{RV}$  was measured in a procedure in which the lungs were nearly fully emptied with head above the surface, followed by emptying of the expiratory reserve volume when the participant was fully submerged. The upper body was held in a position perpendicular to the water surface, and hip and knee joints were held at 90 degrees. The top of the head was held at a distance of 30 cm below the water surface.  $HW_{TLC}$  measurements were carried out using the same procedure, with the attachment of a 10 kilogram weight to a belt at the swimmer's waist. Participants inhaled to full TLC with the head above the surface and then submerged. The participants were weighed with a calibrated strain gauge (Model KRG-4, Bofors Electronic, Sweden), and the weighing was repeated three to five times each to obtain reliable results. The CVs of test-retest measurements (within-day) for the two hydrostatic values were 1.5 % at both exhaled and inhaled conditions for the males, and 3.6 % and 1.2 % for the females at exhaled and inhaled conditions, respectively. The CVs were calculated between the second and third trials. The water temperature was  $28 \pm 0.5$  °C.



**Figure 7a and 7b.** Hydrostatic weight (HW) after full exhalation at residual volume (RV),  $HW_{RV}$ , (7a), while figure 7b shows how an extra weight is added when measuring HW at full inhalation at total lung capacity (TLC),  $HW_{TLC}$ .

### 3.4.4 Body composition

Body composition evaluation quantifies the major structural components of the body including; muscle, bone and fat and it was measured in Study II. The percentage of body fat was calculated after measurements at four skin fold sites<sup>125</sup> using a Harpenden caliper (Harpenden Skinfold Calipers, H.E. Morse CO, British Indicators, Ltd). The radioulnar and femoral condylar widths were also measured, making it possible to calculate the relative amounts of skeleton and fat-free tissue.<sup>126</sup>

### 3.4.5 Other assessments

The number of pistoning gulps in each cycle and number of cycles were recorded in a training diary, along with any remarks on the training. The participants graded their perceived tension in the chest on the Borg CR -10 scale<sup>127</sup> while performing GI during the training sessions.

The participants in Study III were also asked to grade their ability to cough and their ability to clear secretion before and after the training period. They were asked: “How is your cough function affected?” and “How is your ability to eliminate secretion affected?” The answers were also rated on the Borg CR-10 scale.<sup>127</sup>

### 3.5 STATISTICAL METHODS

Results in Study I-IV were either presented as mean and/or median values, with either one standard deviation (SD) and/or range and 95% confidence interval (CI). Table 2 presents the type of statistical analysis used in Study I-III.

**Table 2.** Analytical statistics performed in Study I-III.

	Study I	Study II	Study III
Students t-test	x	x	x
Wilcoxon signed rank test			x
Repeated measures ANOVA	x		
Pearson correlation coefficients		x	
Multiple regression		x	

In all four studies the level of statistical significance was set at  $p < 0.05$ . STATISTICA software (7.0, Stat Soft Inc, Tulsa, OK, USA) was used for all analysis.

The strength of the correlation coefficient was valued according to Domholdt,<sup>128</sup> 0.00-0.25 little, if any, 0.26-0.46 low, 0.50-0.69 moderate, 0.70-0.89 high, 0.90-1.00 very high.

### 3.6 ETHICAL APPROVAL

All participants gave their informed written consent to participate. The studies were approved by the regional research ethics committee in Stockholm and by the local ethic committee at Karolinska University Hospital.

## **4 RESULTS**

### **4.1.1 The ability to learn**

Most of the participants were able to learn GI, however some could not. The number of participants in each study that not could exceed their VC when trying to perform GI were as follows; in Study I - one woman, in Study III - five participants (four men and one woman), and in Study IV - six children (three boys and three girls). These participants were therefore excluded from further analysis.

Sixteen women completed Study I, 26 swimmers completed Study II, and 20 participants completed Study III. In Study IV, one of the children who was able to learn the technique, did not complete the study protocol as he lacked motivation; some measurements of GIV and PCF with GI were still taken from this child, but he was excluded from further analysis. Therefore completed measurements in this group were taken from only four children.

Some of the participants in studies 1-IV learned the GI technique immediately, while for others it took up to three weeks.

### **4.1.2 Discomfort**

In Study I, seven participants experienced some temporary adverse symptoms of the technique including dizziness, a hacking cough, headache, and yawning. None of the participants fainted during the manoeuvres.

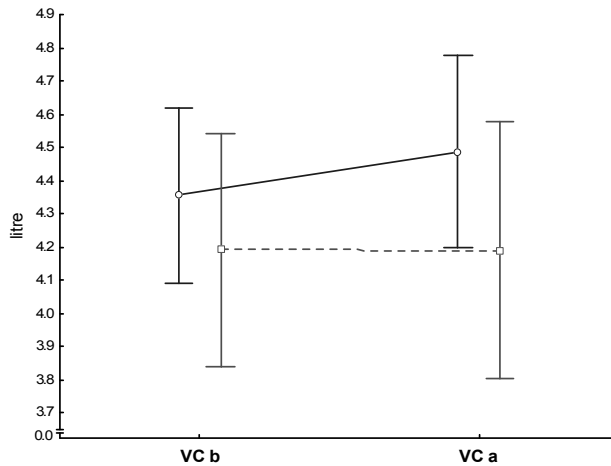
In Study II, participants occasionally reported that performance of GI caused temporary symptoms such as dizziness, a hacking cough, headache and yawning, tension in the chest, sweating. Four participants reported isolated episodes of fainting during GI.

In Study III, participants occasionally reported that during, or shortly after performing GI, temporary symptoms such as dizziness, local paresthesia, and tension in the chest occurred. Three participants reported episodes of syncope during GI and two reported that they were close to syncope.

In Study IV, two of the children occasionally reported that during or shortly after performing GI, temporarily symptoms such as dizziness and tension in the chest occurred.

### 4.1.3 Pulmonary function

In Study I, VC increased after the six-week period by 0.13 l ( $p<0.001$ ) in the TG, while it did not change in the CG (figure 9). Statistical comparison between the groups indicated that there was a significant increase in the TG compared to the CG ( $p<0.01$ ). The increase in VC in the TG persisted for at least 12 weeks after the study,  $4.47 \pm 0.61$  l.



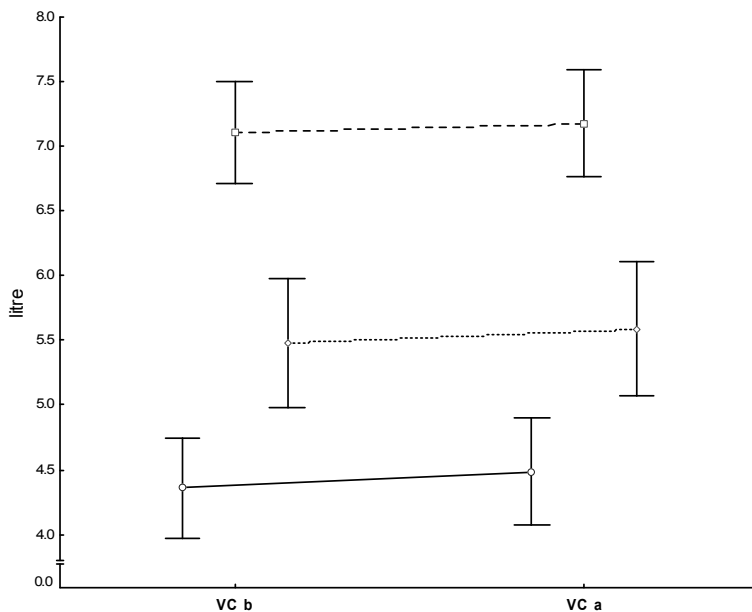
**Figure 9.** Measurements of vital capacity (VC) before (b) and after (a) a six-week period of glossopharyngeal pistoning for lung insufflation (GI) performance. Values are presented as means (SD). Training group  $\circ$  (TG,  $n=16$ ). VC increased in the TG,  $p<0.001$ . Control group  $\square$  (CG,  $n=9$ ). There was significant difference in effect between groups,  $p<0.01$  after six weeks.

In Study II, VC increased significantly in the female group during the training period. The increase was 0.11 l (from  $5.48 \pm 0.68$  l to  $5.59 \pm 0.63$  l),  $p<0.05$ . There was no significant change in VC for the male group.

Average maximal  $VC_{GI}$ , as measured weekly with the portable spirometer, was  $9.05 \pm 1.74$  l in the male group and  $6.07 \pm 0.89$  l in the female group. These results are 22.6 % ( $1.64 \pm 0.55$  l)  $p<0.001$  and 14.8 % ( $0.74 \pm 0.26$  l),  $p<0.001$  higher than the average VCs in the two groups respectively.

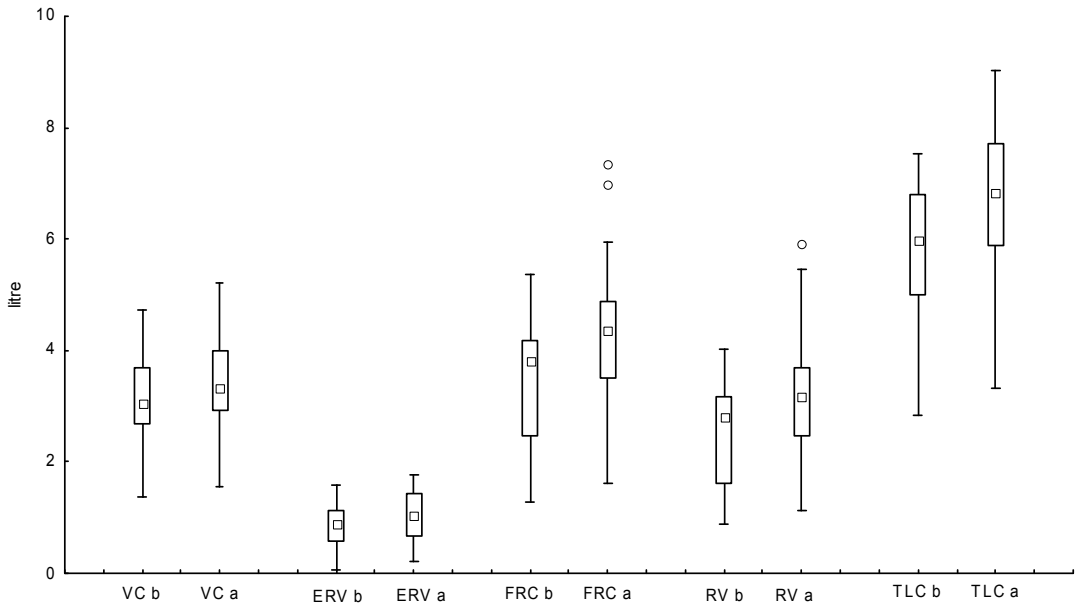


VC increased on an average of 0.16 (CI 0.05-0.16) l,  $p < 0.01$  in females in the TG in Study I, female swimmers in Study II and male swimmers in Study II after the training period (figure 10), but there were no differences between the three groups in effects of VC after the training period.



**Figure 10.** Vital capacity before (b) and after (a) the training period for female in Study I ○ (n=16), female swimmers in Study II ◇ (n=10) and male swimmers in Study II □ (n=16),  $p < 0.01$ .

In Study III, it was found that VC ( $p<0.001$ ), ERV ( $p<0.01$ ), FRC ( $p<0.0001$ ), RV ( $p<0.001$ ) and TLC ( $p<0.0001$ ) all increased significantly after the training period (figure 10). Mean GIV above VC was  $0.88 \pm 0.5$  l (an increase of 28%). PCF changed from before and after the performance of GI from  $395 \pm 83$  l/min to  $424 \pm 101$  l/min, ( $p = 0.057$ ). There was a significant correlation,  $r = 0.54$ , ( $p<0.01$ ) between GIV and PCF. DLCO did not change ( $7.26 \pm 1.51$  mmol /kPa/min), but VA increased significantly from  $5.29 \pm 1.2$  l to  $5.47 \pm 1.2$  l ( $p<0.05$ ). Neither MIP nor MEP changed.



**Figure 10.** Pulmonary function variables before (b) and after (a) eight weeks of glossopharyngeal pistoning for lung insufflation (GI), ( $n=20$ ). VC, vital capacity; ERV, expiratory reserve volume; FRC, functional residual capacity; TLC, total lung capacity; RV, residual volume. There were significant increases in all parameters; VC  $p<0.001$ , ERV  $p<0.01$ , FRC  $p<0.001$ , RV  $p<0.001$ , TLC  $p<0.001$ .

In Study IV, the median GIV for the five children who learned the technique was 0.28 (range 0.15-0.98) l. The median change in PCF with GI for four of the five children who learned the technique was 40 (13-87) l / min. The per-protocol analysis showed that there were no changes in VC, FRC, TLC, RV, MEP, MIP or SNIP. There were positive effects on the mean differences in IVC at 0.13 (CI 0.03-0.23) l and PEF at 116 (CI 60-173) l / min.

#### 4.1.4 Chest expansion

The mean differences of changes in chest expansion in Study I-III are presented in table 3.

In Study IV, there was a tendency to increase in chest expansion at CX before and after eight weeks, mean difference of 0.7 (CI -0.18-1.66) cm, but here were no changes at CC, mean difference of -0.09 (CI -1.26-1.09) cm. With GI there were positive changes at CX, mean difference of 1.50 (CI 0.16-2.84) cm and at CC, mean difference of 1.79 (CI 0.85-2.73) cm.

**Table 3.** Mean difference of changes in chest expansion in cm before and after a training period and with glossopharyngeal pistoning for lung insufflation (GI).

	Study I (n=15)	p-value	Study II male (n=16)	p-value	Study II female (n=10)	p-value	Study III (n=20)	p-value
CX	-0.24 ±1.2	ns	1.0 ± 0.6	<0.001	0.6±0.8	< 0.05	1.1±1.0	<0.001
CC	-0.19 ±1.2	ns	0.8± 0.7	<0.001	0.8±1.0	<0.05	0.8±0.8	<0.001
CX GI	1.0±0.8	<0.001	2.3±0.9	<0.001	1.6±0.4	<0.001	2.1±0.9	<0.001
CX GI	1.0±0.7	<0.001	1.6±0.9	<0.001	0.4±0.3	<0.01	1.6±0.7	<0.001

Values are presented as mean difference and one standard deviation. CX, chest expansion at the level of processus xiphoideus; CC, chest expansion at the level of the fourth costae.

#### 4.1.5 Training statistics

Training statistics of training compliance, cycles of GI, gulps and Borg CR-10 are presented in table 4.

**Table 4.** Training statistics

	Study I (n=16)	Study II (n=26)	Study III (n=20)	Study IV (n=4)
Training compliance (%)	84 (50-100)	80 (57-97)	87 (50-100)	100 (100-100)
Cycles of GI (number)	19 (15-30)	10 (8-12)	10 (5-15)	10 (6-10)
Gulps (number)	10 (2-42)	14 (4-35)	14 (3-42)	11 (5-20)
Borg CR-10 (score)	4 /1.5-8)	4 (0.5-9)	4 (2-10)	7 (3-10)
Nasal clip (number)	5	8	6	0

Values are presented as mean and range in studies I-III and as median and range in Study IV. GI, glossopharyngeal pistoning for lung insufflation (GI).

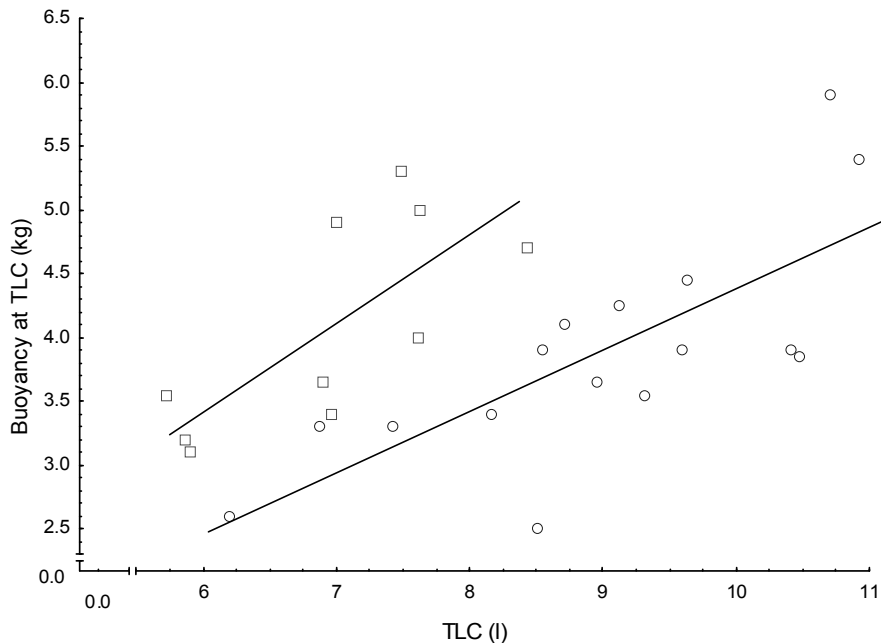
#### 4.1.6 Experience of GI

In Study III, the participants significantly improved their rating of the two questions concerning cough function and ability to clear secretions. In answer to the former, the average reply moved from 7 (1.5-10) (strongly affected) to 3.5 (2-10) ( $p<0.01$ ) (moderately affected), and the latter question's reply changed from 7 (0-10) (strongly affected) to 4 (1-9) ( $p<0.01$ ) (moderately affected).

#### 4.1.7 Lung volumes and Buoyancy

In Study II, anthropometrical results such as body mass, relative amounts of fat, muscle, bone and fat-free tissue did not change during the training period.

In Study II, the buoyancy (lifting force) increased significantly from  $3.87 \pm 0.88$  kg to  $4.04 \pm 0.89$  kg ( $p<0.05$ ) for the male group, and from  $4.08 \pm 0.82$  kg to  $4.45 \pm 0.92$  kg ( $p<0.01$ ) for the female group. TLC correlated with buoyancy at  $HW_{TLC}$  (lifting force) for both the male group ( $r=0.75$ ) and the female group ( $r=0.73$ ) (figure 11). Multiple regression analysis showed that the variables TLC and fat% explained about 70% of the variation in the buoyancy in both the male group and the female group.

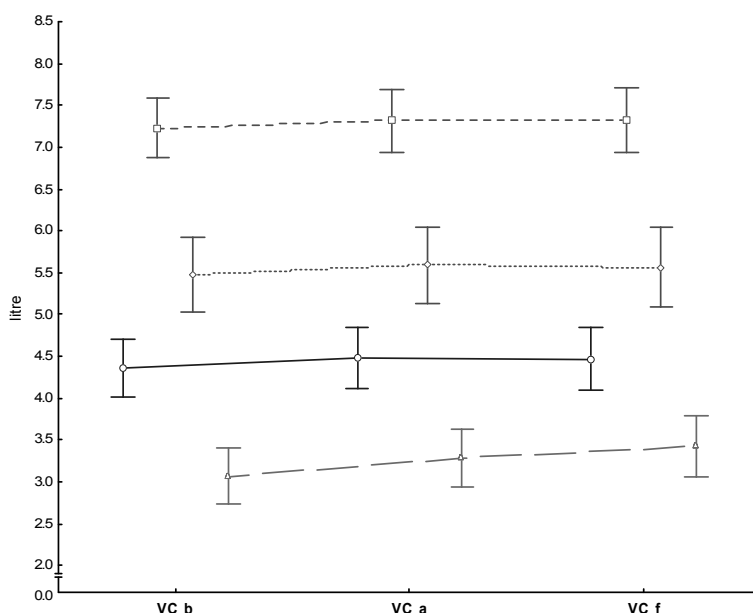


**Figure 11.** Correlation between buoyancy at hydrostatic weight (HW) at total lung capacity (TLC) $HW_{TLC}$  (lifting force) and TLC for both the male group ○ ( $n=16$ ),  $r=0.75$  and the female group □ ( $n=10$ ),  $r=0.73$ .

## 4.2 FURTHER RESULTS

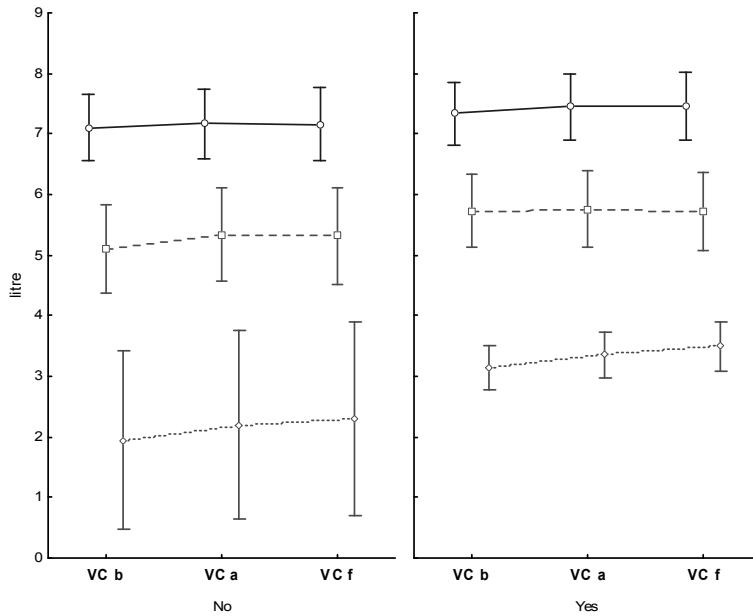
### 4.2.1 Three months follow up

After three months the numbers of participants who had continued to train were as follows; no women in Study I, four female swimmers, seven male swimmers, 17 participants with CSCI and all the four children with SMA type II. The effects of IVC remained for the four children three months after completed training. In figure 12 the three months follow ups are presented irrespective of whether the participants had continued training or not in females in the TG in Study I, female swimmers in Study II, male swimmers in Study II and participants with CSCI in Study III. VC increased with an average of 0.16 (0.07-0.25) l,  $p < 0.001$  and there were no interaction between the groups.



**Figure 12.** Vital capacity (VC) before (b), after (a) and at follow up (f) three month after completed training period separated for healthy women ○ (n=16), female swimmers ◇ (n=10), male swimmers □ (n=16) and participants with cervical spinal cord injury △ (n=20),  $p < 0.001$ .

When separating the groups in figure 12, except the healthy women in Study I, if they had continued to train or not the VC increased in average 0.18 (CI 0.06-0.30) l,  $p < 0.01$  and there were no difference in effect on VC regardless if they had continued to train or not (figure 13).



**Figure 13.** Vital capacity (VC) before (b), after (a) and at follow up (f) three month after completed training period separated for those who did not continue to train (No) (n= 15) after the training period and for those who did (Yes) (n=31). Divided into female elite swimmers □ (n=10), male swimmers ○ (n=16) and participants with cervical spinal cord injury ◇ (n=20),  $p < 0.01$ .

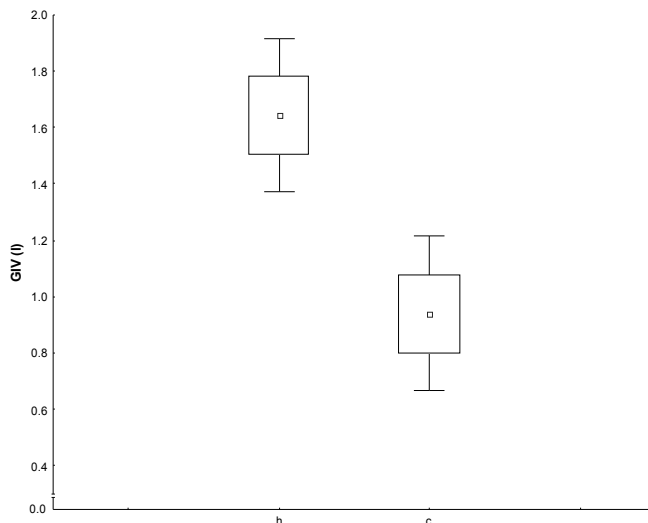
#### 4.2.2 Comparison between male swimmers and men with cervical spinal cord injury (CSCI)

The comparisons between male swimmers and men with CSCI are presented in table 5.

**Table 5.** Pulmonary function and chest expansion variables in male swimmers in Study II and men with cervical spinal cord injury (CSCI) in Study III.

	Male swimmers (n= 16)	Men with CSCI (n=16)	p-value
GIV (l)	1.64 ± 0.55	0.93± 0.56	<0.01
% GIV of VC	23 ± 7	27±15	Ns
Difference of VC (l)	0.07±0.22	0.27±0.25	<0.05
Difference of VC (%)	1±3	9 ± 8	<0.001
CX GI (cm)	11.1 ± 1.7	6.6 ± 1.8	<0.001
CC GI (cm)	8.9 ± 1.5	5.9 ± 1.3	<0.001
CX GI% of CX	26 ± 10	50 ± 27	<0.01
CC GI % of CC	21 ± 11	40 ± 29	<0.001

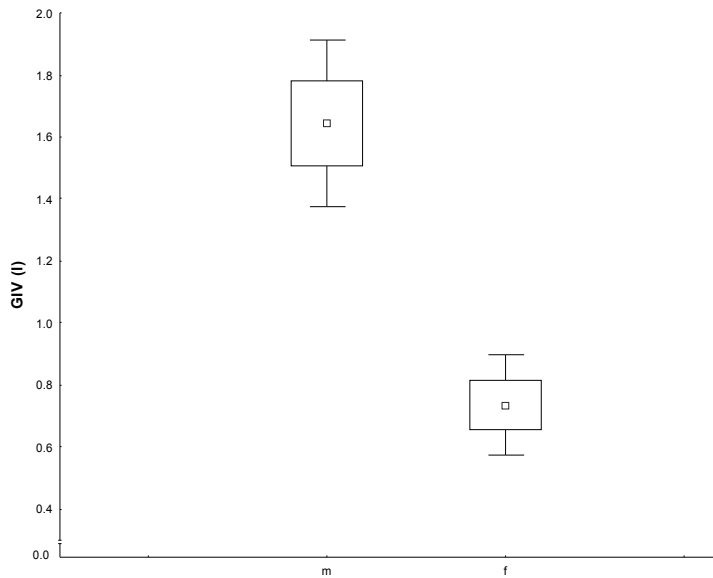
GIV, glossopharyngeal insufflation volume; VC, vital capacity; CX, chest expansion at the level of processus xiphoideus; CC, chest expansion at the level of the fourth costae; GI, glossopharyngeal pistoning for lung insufflation.



**Figure 14.** Glossopharyngeal insufflation volume (GIV) in healthy men (h) (n= 16) and men with cervical spinal cord injury (c), (n= 16), p<0.01.

### 4.2.3 Comparison between male and female swimmers

There was no difference between male or female swimmers in percentage of predicted VC, TLC or RV. There was a significant difference in GIV between men and women of  $1.64 \pm 0.55$  l compared to  $0.74 \pm 0.26$  l respectively,  $p < 0.05$  (figure 15), and on comparison of the % GIV of VC, the men also had a higher volume when considering their normal VC of 23 % compared to 15 % ( $p < 0.01$ ). The men had a significantly higher chest expansion with GI than the women at both CX and CC level, at  $9.1 \pm 1.3$  cm compared to  $11.1 \pm 1.7$  cm and  $7.6 \pm 1.0$  cm compared to  $8.9 \pm 1.5$  cm,  $p < 0.001$ .



**Figure 15.** Glossopharyngeal insufflation volume (GIV) in male swimmers (m) (n=16) and in female swimmers (f) (n= 10),  $p < 0.05$ .



## 5 DISCUSSION

GI is a technique that could be of benefit for people with totally different prerequisites. Study I-IV have shown that performance of GI can be beneficial in the clinical setting for people with reduced pulmonary function, who are at risk of severe pulmonary complications. However it can also be beneficial for athletes, who although often having an above average pulmonary function in the first place, can also use the technique effectively to further stretch their physiological limits and achieve better results.

The aim of this thesis was to first evaluate if healthy people, and athletes who are well trained and extremely motivated to enhance their vital capacity, were able to learn GI. From there the studies moved on to also evaluate the specific training concept and the risks or discomforts associated with it. The next step was to transfer this knowledge and evaluate the benefit of the technique in people with CSCI and also in children with SMA type II.

### 5.1 FINDINGS

All participants in Study I and II were able to learn GI, with the exception of one woman. Five of the participants with CSCI and six of the children with SMA type II were not able to manage the technique because they were not able to exceed their VC. The participants who learned GI were able to exceed their VC by a mean value of 23%. Pulmonary function parameters increased in healthy women, female swimmers and participants with CSCI, while positive changes were also seen among the children with SMA type II. Chest expansion with GI was significantly greater for all participants in all four studies. In addition, buoyancy increased for the elite swimmers. GI was performed without any major discomfort among the participants. Three months after training completed, the positive changes persisted for all participants, regardless of whether they had continued to train or not.

Some participants that not were able to learn GI, thus, they could not exceed their VC, which is in line with the findings of similar studies.<sup>103</sup> Proper and repeated instruction on the correct pistoning action and motivation are important for learning.<sup>93, 103</sup> More than half of the children (55%) in Study IV were not able to learn GI, however for the five children who did learn, it appeared that motivation, in this case in the form of competition, played an important role. Some of the participants learned the technique immediately, while for others it could take up to several weeks. Other studies have also shown that some people learn at the first lesson and for others it can take several months, while some even learn it on their own.<sup>87, 94</sup>

All of the participants received the same instruction. It appeared that the video film was the best tool for learning, while the feedback they received when they practiced together with their instructor was also very effective. The performance of particular sounds was also a good learning tool, as was the filling of the cheeks with air, with participants then trying to push/move it in different directions. Occasionally the

participants pushed air down through the oesophagus instead of the trachea and this also gave them a direct feedback signal that they had performed the technique incorrectly.

The portable spirometer was useful as a teaching tool, giving direct quantitation of efficacy and many of the participants become motivated to beat their own records. Other studies have shown that common problems while learning GI include an open nasal passage or glottis allowing the air to escape, swallowing air into the oesophagus rather than trachea, difficulty in shaping their mouth, an inability to coordinate movements of the tongue, or making an incorrect sound.<sup>93-95</sup>

When the participants were asked how they would describe their performance of GI they expressed it thus: “it feels like ping pong balls of air are forced down to the lungs” or “like squeezing the air against the palate backwards” or “swallowing air, but to the lungs instead of to the stomach”.

The ability to insufflate a large GIV does not seem dependent on training intensity (numbers of gulps, cycles of gulps and training sessions/week). More likely it seems dependent on the function of the vocal cords and their ability to maintain the intrathoracic pressure, while the strength and function of the glossopharyngeal muscles might also influence the performance of the GI technique.<sup>91</sup> The different anatomical prerequisites for men and women could also influence how well the technique is learned. Healthy men had a higher GIV compared to healthy women in Studies I and II. The five children who learned GI were all boys and they seemed to be competitive in wanting to increase their MIC. It would seem that motivation did have a great impact on learning.

For the children in Study IV, the reduced mandibular range of motion and masticatory and facial muscle weaknesses seemed to have a negative effect on the ability to learn GI and perform the correct pistonning actions.<sup>70</sup> The children in Study IV had various grades of reduced range of mandibular motion and it appeared that the six children that could not learn GI were more affected in this respect than those who learned the technique.

The participants had rather high training compliance, and the four children in Study IV had 100 % compliance. The numbers of cycles of GI varied between Study I and the other studies and this was dependant on the training concept, which was modified during the period of the studies. The numbers of gulps varied between all the participants, and some filled each gulp with the maximum amount of air possible, while some did not, a behaviour which was also noted in another study.<sup>89</sup> Ratings on the Borg CR-10 scale were almost the same in Study I-III, however the children rated tension in the chest as being higher during performance of GI. Some of the participants in Study I-III used a nasal clip during training to avoid leakage of air and this is consistent with some people having difficulties in closing the larynx.<sup>89</sup>

The training concept investigated in this thesis has not, to our knowledge, been practiced in any other study. However a case report by Warren<sup>95</sup> did outline a similar training concept, with the patient training for three to four days per week, over five weeks.

Some of the participants reported temporary symptoms during or after the training sessions such as dizziness, tension in the chest, a hacking cough, headache, and yawning. All together seven participants fainted while performing GI, which emphasises how important it is for susceptible people to take a supine training position to avoid falling and subsequent injury in the event of becoming unconscious. The orthostatic syncope is probably due to the increase in intrathoracic pressure due to the reduction in venous return, especially if the participant is standing upright while performing GI.<sup>91, 98, 109, 129</sup> The participants who fainted or felt other discomfort while performing GI, did not have higher Borg CR-10<sup>127</sup> ratings than the other participants, suggesting that the risk for syncope cannot be predicted by rating chest tension.

Forceful straining (e.g. in labour) has been shown to cause pneumomediastinum with subcutaneous emphysema in some individuals,<sup>130, 131</sup> and it is possible that GI may cause pneumothorax in susceptible individuals due to the increased stress on the pulmonary tissue. However, this method has been used by numerous adult patients and athletes for decades and very few complications have been reported, despite transpulmonary as high as 80 cm H<sub>2</sub>O being developed.<sup>108</sup> From our data, and from an extensive review of the literature, it does not appear that GI or air stacking to lung volumes greater than inspiratory capacity increases the risk of pulmonary trauma.<sup>11, 79, 82, 92, 93, 132</sup> This method, if learned and practised properly, should not entail any major risk compared to other physical activities. Therefore, the technique can be considered as a safe method for all people to perform and any benefits should outweigh the relatively small risks.<sup>99, 108</sup>

The effect of GI was investigated in healthy people and in adults and children with NMD. The individuals with NMD, in contrast to healthy people, have reduced chest-wall compliance, while the rib-cage may be stiff due to weak inspiratory muscles which prevent stretching of the chest and lungs to the predicted TLC.<sup>34, 39, 45</sup>

The VC increased in Study I and also for the female swimmers in Study II. The mean VC of the subjects in the TG in Study I increased by 3 % after the six-week period and by 2 % in Study II after five weeks of training. When including the groups of healthy women in Study I, female swimmers in Study II and male swimmers in Study II in the same analysis, there are no differences between the groups concerning the increase of VC. Other authors have also showed a small increase in the mean VC of healthy subjects after inspiratory muscle training.<sup>20, 24</sup> However, the training in the present Study differs from others in that our training consisted of maximal inhalation to TLC, followed by GI. This may result in a stretching effect rather than an increase in muscle strength. In cases of lobectomy the remaining pulmonary tissue expands.<sup>13</sup> This suggests that lung volume in healthy subjects is limited either by the magnitude of chest wall expansion or by resistance to diaphragm movement. The healthy participants probably need more intensive training and longer training periods to achieve greater increases in VC. The male swimmers in Study II had larger lung volumes than those predicted from their height, weight and body mass index (BMI). It could therefore be argued that the elite swimmers in the present study comprise a pre-selected population with a genetic heritage.<sup>17, 22, 133</sup>

Chest expansion increased with GI in Study I and II in healthy people. After the training period the chest expansion increased only for the swimmers. For the women in Study I these results it may not be of clinical importance, but for the swimmers it could be of value to their swimming performance.

The measure of buoyancy in the swimmers is another way to measure lung volume, as it is the air in the lung and in other cavities that decides the buoyancy. Figure 11 shows clearly that there is a high positive correlation between TLC and buoyancy, especially in the male group, whose body fat percentage is lower than the female group. Increases in buoyancy decrease passive drag and generates body roll, as has been shown by manipulation of the volume of air in the lungs.<sup>29, 134</sup> Increased buoyancy might increase swimming velocity for elite swimmers. The residual volume or buoyancy after exhalation did not change, nor did the percentage of body fat, and we believe that the findings are due to an increased ability to fill the lungs with air.

Furthermore, we hypothesise that if a swimmer can increase their TLC after a period of GI performance, they would achieve a higher water position. It is also possible that the increased chest volume would allow higher tidal volumes at reduced breathing frequency. This would also raise the average position of the swimmer relative to the water surface during a race. Today a 50 m freestyle race at international level is usually swum with one breath or without breathing. Swimmers with a larger lung volume have higher buoyancy and a greater store of oxygen available during such a sprint. If the swimmers use GI when they are warming up before a race, it might allow them to maximally fill their lungs. Studies have shown that after performing GI at one training session the VC increased, which represents a warm up effect.<sup>99, 115</sup>

In Study III that involved people with CSCI, chest expansion increased both during GI and after the training period and also showed positive changes for the children in Study IV, which supports the increase in the pulmonary function parameters. GI will add a substantial volume of gas to the lungs, and the major effect of the added gas is an expansion of the chest.<sup>108, 114, 115</sup> This chest expansion, beyond 'normal' TLC, results in a stretching effect of the respiratory system. Studies on people with CSCI by Fugel Myer et al.<sup>135</sup> and Huldtgren et al.<sup>136</sup> were the first to show that is possible to maintain or restore optimal mobility of the chest, and improve cough function by adding an extra volume of air using a manual resuscitator to reach the MIC.

For people with NMD there is a changed length-tension relationship for the respiratory muscles that leads to a loss of ROM due to muscle weakness.<sup>37, 39</sup> ROM for the extremities is a basic treatment to maintain joint mobility for patients with muscular weakness,<sup>37</sup> however ROM for the chest wall is unfortunately not always performed. The present thesis showed that lung and chest wall ROM can be maintained or even improved using deep insufflations as performed during GI. These results suggest that it is important for people with NMD to receive regular ROM mobilisation of the lungs and chest wall to maintain or prevent a further decrease of volumes for lungs and chest wall.<sup>37</sup> Periodic hyperinsufflation, used together with an abdominal belt to prevent paradoxical breathing with thoraco-abdominal asynchrony, was investigated in a study by Ioos et al.<sup>66</sup> for respiratory management in children with SMA. This allowed regular stretching and better ventilation of the upper pulmonary areas,<sup>66</sup> however chest expansion was not measured. A study of the effects of abdominal binding during

breathing exercise showed that static lung volumes decreased, VC increased and FRC remained unchanged.<sup>137</sup>

In Study III, VC, ERV, FRC and TLC increased significantly. This is obviously of benefit, as cervical spinal cord injury patients are characterised by a decrease in these parameters and an increase in RV.<sup>48, 138</sup> The participants performed maximal GI manoeuvres several times a week and therefore the increase in TLC might be explained as a result of stretching the chest wall and the structures surrounding the rib-cage as previously discussed.

In Study III and IV there were no changes in MEP, MIP, or in SNIP for the children in Study IV. However, one of the purposes of using GI was to stretch the chest wall and thereby create conditions for larger lung volume; therefore, it was not expected that any changes in respiratory muscle strength would be achieved.

In a study by Rutchik et al.<sup>139</sup> subjects with CSCI participated in an eight week resistive inspiratory muscle training (IMT) program. The participants had similar predicted values of their pulmonary function as in Study III. They had similar increases after training in FVC, VC, TLC and FRC and also in MIP. However, in the present study, MIP and MEP did not change. The increased RV in Study III also supports the notion that we did not train the expiratory muscles. Some studies in people with CSCI show that RMT can improve ventilatory function.<sup>139, 140</sup> In a review article of Van Houtte et al.<sup>141</sup> they reported that RMT tended to improve expiratory muscle strength, VC and RV.

The increases in pulmonary function seen in Study I-IV were probably due to increased chest wall compliance or pulmonary compliance, thus increased FRC and the ability to inhale a larger VC with unchanged respiratory muscle strength. To further elucidate the relative contribution of chest wall and lung tissue with compliance would require measurements of transpulmonary pressure.<sup>108</sup>

No one can cough without an adequate pre cough volume to generate an increased intrathoracic pressure and an increased expiratory flow. In Study III and IV the participants tended to improve their PCF values and in Study III the GIV correlated moderately with PCF. Kang and Bach<sup>33</sup> also showed that VC and MIC correlated significantly with PCF, strengthening our results.

Abdominal thrust may also increase PCF,<sup>33</sup> however in Study I-IV it was not investigated, since the intention was to increase PCF through GI alone. The participants had PEF and PCF values of over 270 l / min which is enough for an effective cough according to Bach and Saporito.<sup>40</sup> Studies have shown that the PCF increases more with GI than with maximum inspiratory techniques.<sup>33</sup> Some studies also show it is important to combine techniques to achieve the most effective cough.<sup>8, 32</sup> It is of great importance to regularly perform GI, to maintain ROM in the chest wall and thereby also maintain pulmonary compliance, as this will increase the pre-cough volume and thus be of benefit for the PCF.<sup>37, 38</sup>

A study by Dohna-Schwake et al.<sup>67</sup> showed that an IVC < 1.1 l and a PCFs < 160l/min could predict severe chest infections in paediatric neuromuscular disorders. The results from Study IV indicate that both IVC and PCF increased with GI, however some of the values were still under the values mention above.

The participants in Study III notably improved the rating of their ability to cough and clear secretions after the GI intervention. These results might be due to the fact that once having learned the technique, they were then able to use GI as they felt necessary, increasing their lung volume and thus achieving a more effective cough and clearing secretions more effectively.<sup>105</sup> Seventeen of the 20 participants with CSCI and all four children with SMA type II continued to train after three month, which also indicates that they found the technique useful. Few studies have investigated the patients' subjective experience of their breathing or what they think of different treatments. A study by Spungen et al.<sup>142</sup> used a standard respiratory questionnaire to determine prevalence of respiratory symptoms and among participants with a complete CSCI: problems with phlegm, cough and wheeze were reported. In a study by Dail et al.<sup>93</sup> the patients were interviewed about their attitudes toward glossopharyngeal breathing. Positive comments on the use of GI, to improve cough function and stretch the chest wall included that it felt safe to have a non mechanical technique that required no equipment in case of power cut and that they could perform GI independently while negative comments concerned cosmetic effects and the difficulties encountered in learning the technique.<sup>93</sup>

The positive effects on pulmonary function and chest expansion persisted three months after completed training, regardless of whether the participants had continued to train or not. For those who did continue to train, they often did so at different frequencies, with some training each day, while others did so once a week or less. Therefore, the training intensity up to three months did not seem to matter. It would be of interest to follow up one year after training in order to evaluate if the training was still having an effect or not. An explanation for the persistent benefit seen in those subjects who did not continue to train might be that they continued to expand their chest and lungs while taking deep breaths and sighs, so in effect were training passively.

When comparing male swimmers to men with CSCI, a difference was noted between the predicted values in VC and TLC. The men with CSCI had about half of the predicted value of VC as the male swimmers. However the predicted RV did not differ and that is probably due to the fact that men with CSCI had a weakness of the expiratory muscles. In absolute terms of litres volume, there was a difference in GIV between these groups, with the male swimmers having a higher GIV. However, in relative terms when GIV was related to the percentage VC there was no difference between the groups. Similar result was evident with chest expansion, however there was a significantly increase also in percent to CX GI and CC GI. As the men with CSCI have reduced pulmonary function, they have the potential to make a greater increase. Consequently, the men with CSCI had greater increases in VC than the male swimmers after the training period. These results are desirable, as participants with CSCI stand to benefit the most from all of the improvements that GI training can bring about.

When comparing female and male swimmers there are no difference in their predicted values in pulmonary function but in absolute terms of litres volume, which is in line with other studies.<sup>143-145</sup> However it was found that the men could generate a larger GIV both in absolute and in relative terms when GIV was related to the percentage VC and could also increase their chest expansion more with GI. The male swimmers seemed to push themselves more than the female swimmers when they performed GI. Men have larger lungs than women<sup>143-145</sup> and probably more muscle strength: the fact that they have better prerequisites to achieve larger volumes might be one explanation for these observations.

## 5.2 METHODOLOGICAL CONSIDERATIONS

In study I, female physiotherapists were chosen because it was convenient to have all of the participants working at the same place as there was a need for many follow-ups. The CG was used to eliminate the learning effect of blowing into a spirometer. The elite swimmers were recruited from the Stockholm area, again so that all of the follow ups could be carried out locally and within the training period. In Study III and IV, the attempt was to recruit groups that were as homogenous as possible, therefore people with CSCI and children with SMA type II were recruited. People from the Stockholm area were also chosen in these studies, again because of the follow ups. Two thirds of the approached people with CSCI declined to participate, mainly due to lack of time and because of low motivation. This might seem like a large number who declined participation, however these people with CSCI might also be tired of all examinations and other studies they are invited to. For the studies in this thesis it was very important that the participants were high motivated.

Study I, which was carried out on healthy women, was the first study to be designed and the interventions were chosen to be in common with ordinary training techniques in healthy people. The training concept was modified and developed during the studies. Before Study II three male swimmers were interviewed on their training effects and its intensity; they felt, like the women in study I, that performing 15 cycles of GI was too much and therefore the recommended number of cycles was changed to 10. The same thoughts were expressed by the two people with CSCI who were tested before Study III.

The method of measuring pulmonary function was changed from a body plethysmograph to a Vmax 229, due to difficulties for the wheelchair bound participants in entering the body plethysmograph.

Chest expansion as measured with a tape measure is inexpensive and easy to use in the clinical settings, however the reliability of this method has been discussed. A study by Bockenbauer et al.<sup>123</sup> showed that a tape measure was highly reliable to assess thoracic excursion (chest expansion). In the present studies the coefficient of variance (CV) of test-retest measurements at the level of CX (n=50) was 0.8% and it was 0.4 % for measurements at CC.

Another important variable to consider was the ability to perform the measurements especially on the children with SMA type II. For those children with a decreased range of mandibular motion, it was difficult to fit the mouthpiece, to perform GI and then blow in the spirometer. They also seemed to find it complicated to hold the insufflated air and then put their lips around the mouth piece, which might also have been difficult for other participants.

### **5.2.1 Statistical considerations**

The power analyses indicate that there were too few participants in Study I and II, however when including the female from the TG in Study I, female swimmers in Study II and male swimmers in Study III in the same analyses it strengthens the results and in that way the power detects the effect of training. In Study III there was enough power, and in Study IV there was a low power. However there were significant changes in Study I, II and III individually. Most of the result variables were normal distributed, thus parametric statistics was used. For the children it was only possible to use descriptive statistics because of the small sample number. If a much larger sample of children were recruited, then hopefully more would be able to perform GI; the present results (and those from studies in adults) indicate that the technique is beneficial and so it is likely that statistical confidence would also increase if a larger study population were recruited.

To be able to generalise the results of this thesis after the training period and after three months there is a need for a larger sample in all studies. Which factors that predict the ability to learn GI are also still unknown. However, if once learned the technique, it seems like there are positive effects on pulmonary function and chest expansion to achieve.



### 5.3 CLINICAL IMPLICATIONS

The results from the present studies show that the participants were able to increase their volume of air with GI and to increase their chest expansion. It also shows that following a period of training with GI, there are positive effects on pulmonary function and chest expansion and that the effects persist for at least three months after an intensive training period. Other studies have also shown the similar effects to both the physical and psychological advantage of the participants, along with other positive benefits of the technique such as the fact that it allows patients to inhale deeper breaths, improve cough efficacy, improve speech, maintain lung compliance and increase the time free from a ventilator.<sup>79, 90-94, 96, 97, 104, 111, 146-148</sup> Some positive psychological effects are that it is an independent and non-mechanical technique,<sup>90</sup> which gives the patients on a ventilator a greater freedom, giving them the potential to be more socially active, and also reducing the worry of ventilator loss in the case of a power cut. Limitations of the technique reported in previous studies include the fact that it is not possible to perform GI during sleep or when eating, while some patients think there are cosmetic disadvantages.<sup>112</sup>

GI is a technique that can be used in different ways. It can be used to an immediate effect when the patient needs to cough, to increase volume in order to open closed airways, or to maintain adequate breathing. Additionally, it is a technique that can increase or maintain the chest wall range of motion, which in the long term might prevent the decrease of lung volumes.

GI require none of the ordinary respiratory muscles and no mechanical equipment to achieve a more effective cough and to increase or maintain chest wall range of motion.<sup>92</sup> If the chest wall range of motion is maintained or even increased, then the patient will also be better adapted to maintain and increase lung volumes. All of these factors might be valuable in helping to reach the goal of the prevention of severe pulmonary complications. The GI technique is easy to perform once learned, it is not time-consuming and it can be practiced without any assistance. GI is useful as a complement to respiratory aids and it would be beneficial if it were a part of all respiratory therapists' airway clearance techniques.

Motivation and encouragement of those patients who already practice the technique is also essential in order to maintain an ongoing benefit. Respiratory complications have an economic impact for hospitals treating in patients with CSCI. GI seems to be a valuable tool in the prevention of such complications and is therefore of great importance in the rehabilitation<sup>149, 150</sup> and also in reducing the costs of treating these patients.

GI seems also to be a useful technique in sports, as already shown in breath-hold diving. It also might be of benefit for the swimmers in purpose to gain a better water position and to perform short race without taking a breath. In that area there are more to explore.

## **5.4 FUTURE STUDIES**

In the present studies we carried out a follow up of the participants three months after a training period of GI. However it would be of great value to evaluate the effects after longer periods of time, for example up to one or two years later. Such results could provide direction towards the most beneficial way of training and to give the most persistent effects. It would also be of interest to be able to predict which patients will be able to learn the technique and to whom the technique would have the most benefit. GI is a technique that could be used by other types of patients, for example those with a stiff chest wall or reduced chest wall range of motion, such as patients with ankylosing spondylitis or those patients who have undergone some kind of surgery in the thorax. Further studies on children with different neuromuscular disorders are also needed. It is also important to investigate whether GI can prevent pulmonary complications and if it is cost effective in rehabilitation. Furthermore the patients own subjective experience of respiratory problems and the effects of GI would be valuable to explore.

## 6 CONCLUSIONS

- Nearly all of the healthy participants, most of the participants with cervical spinal cord injury (CSCI) and half of the children with spinal muscular atrophy (SMA) type II were able to learn glossopharyngeal pistoning for lung insufflation (GI). They all performed the technique without any major discomfort.

### Immediate effects of GI

- The VC was exceeded by 23 % for all participants using GI.
- Chest expansion increased for all participants using GI.

### Effects after five to eight weeks of training GI

- VC increased for the healthy women and female swimmers. Nearly all pulmonary function variables improved in people with CSCI. Some pulmonary function variables tended to improve in the children with SMA type II.
- Chest expansion increased in the elite swimmers, in people with CSCI and tended to increase in children with SMA type II at the level of the processus xiphoideus.

### Follow up three months after completed training period

- The effects of GI on pulmonary VC remained three months after the training period, regardless of whether the participants had continued to train or not.

### Comparison between the participants

- The men with CSCI had a higher glossopharyngeal insufflation volume (GIV) than the male swimmers in relation to their VC and chest expansion with GI in relation to their normal chest expansion.
- Men with CSCI had a larger increase in VC than the male swimmers after training period.
- The male swimmers had a higher GIV than the female swimmers, while the male swimmers also increased their chest expansion more with GI.

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