Inter and Intra-rater Reliability of the Manual Assessment of Respiratory Motion (‘MARM’ technique) in Adults

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DECLARATION

Name of candidate: Martin Ludwig

This Research Project entitled "Inter and Intra-rater Reliability of the Manual Assessment of Respiratory Motion ('MARM' technique) in Adults" is submitted in partial fulfilment for the requirements for the Unitec degree of Master of Osteopathy

CANDIDATE'S DECLARATION

I confirm that:

- This Thesis/Dissertation/Research Project represents my own work;
- Research for this work has been conducted in accordance with the Unitec Research Ethics Committee Policy and Procedures, and has fulfilled any requirements set for this project by the Unitec Research Ethics Committee.

  Research Ethics Committee Approval Number: 2011-1230

Candidate Signature: …………………………………………….Date: ………………………….

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PREFACE

Dysfunctional breathing is a topic that is receiving increasing attention in health care research. And within good reason: the respiratory system is a basic life support system that is involved in numerous processes that allow the human body to maintain the state of homeostasis, and in this respect, forms a central part in health, or the lack thereof. The concept of dysfunctional breathing is currently not completely understood and is undergoing research that finds its relevance in many health care disciplines ranging from medical care over psychology to manual therapy.

The Manual Assessment of Respiratory Motion is a palpatory technique that allows the tester to gain a perception of the volume of a breath and the relative contribution of the upper thorax and lower thorax/abdomen. (Courtney and Cohen 2008). The manual assessment of respiratory motion was first developed for, and applied in a study involving the 5 year outcome of breathing and relaxation therapy in cardiac patients in the 1980s (Courtney, Cohen et al. 2009). It is of particular interest in a research setting as it includes a form of graphic notation from which quantitative data can be calculated (Courtney, Cohen et al. 2009). Previous reliability studies have demonstrated good inter and intra-rater reliability of the technique (van Dixhoorn 1997; Courtney, Van Dixhoorn et al. 2008; Courtney, Cohen et al. 2009). However, no study has yet examined the reliability of the technique on actual patients.

This thesis is arranged in three main sections: Section 1 is a literature review that outlines the respiratory system and breathing dysfunction; examines current assessments of breathing function; and critically discusses existing research on the manual assessment of respiratory motion. Section 2 of the thesis contains a manuscript formatted for submission to the Journal of Manual and Manipulative Therapies. Section 3 (appendices) contains other material supplementary to the thesis. The aim of this thesis is primarily concerned with further examining the reliability of an existing manual assessment of dysfunctional breathing

Secondarily, it explores the relationships between various assessments of breathing function.

Section 2: Manuscript

SECTION 1: LITERATURE REVIEW

1. INTRODUCTION

Dysfunctional breathing (DB) is a proposed umbrella term that describes the production of symptoms that result from abnormal breathing patterns (Thomas 2000). The term is an attempt to broaden the classical term “hyperventilation syndrome” which has gained wide acceptance, both in clinical practice and research studies. Hyperventilation is defined as breathing in excess of metabolic requirements and it can lead to hypocapnia (Warburton and Jack 2006). However, studies have shown that many symptoms related to abnormal breathing patterns are independent of hypocapnia (Thomas 2000), and so DB has been introduced as an all-encompassing diagnosis (Warburton and Jack 2006). DB poses a challenge in terms of definition as it incorporates multiple symptoms related to a variety of causes, which are not well understood.

Dysfunctional breathing can be described as shallow, irregular and/or rapid breathing, inability to take a deep breath, frequent sighing and/or yawning, and thoracic dominant breathing (Thomas 2000; Courtney and Greenwood 2009; Courtney, van Dixhoorn *et al.* 2011). One of the principles of manual medicine is that the musculoskeletal system is the major expender of body energy; the greater the activity of this system, the greater the overall demand for energy. A dysfunction that reduces the efficiency of this system increases its demand for energy during normal activity. For example, restrictions in two major joints in one limb can increase energy expenditure during walking by as much as 300% (DeStefano 2010). The process of breathing involves complex muscular contraction and relaxation, motion of fascial planes and the movement of nearly 150 joints (D'Alonzo and Krachman 2003). A person takes approximately 17 thousand breaths per day (Fritz, Paholsky *et al.* 1999). It can be deduced that small deficits in breathing efficiency could amount to differences in energy expenditure over the course of the day. Additionally the altered pressure exerted on spinal and visceral elements as a result of abnormal breathing patterns may cause spinal instability and altered organ function (Hodges et al. 2003; McGill et al. 1995)

Dysfunctional breathing is commonly observed in patients with respiratory or cardiac diseases such as asthma, chronic pulmonary obstructive disease and ischaemic heart disease (Warburton and Jack 2006). It has also been observed in patients with conditions that have no clear pathophysiological relationship to the respiratory system for example temporomandibular dysfunction, fibromyalgia, chronic fatigue syndrome and irritable bowel syndrome (Chaitow 2007). Finally, DB may also cause symptoms in patients with no underlying condition (Courtney, van Dixhoorn *et al.* 2011), suggesting that abnormal breathing may generate morbidity in the absence of pathology. Symptoms that have been related to DB include neck and head pain, anxiety and panic attacks, chronic fatigue, gastrointestinal dysfunction (Chaitow 2007), spinal instability (Hagman, Janson *et al.* 2008), and hypertension (Goldsmith et al. 2011).

It is thought that causative factors of DB may include biomechanical, biochemical and psychological influences. The condition thus manifests itself in various dimensions including, but not limited to, breathing pattern disturbances, biochemical abnormalities and breathing symptoms, all of which may or may not occur simultaneously (Courtney, Greenwood *et al.* 2011). Consequently, the diagnosis of DB would require a multi-factorial approach that entails several measures in order to address the multiple dimensions of the condition. Questionnaires cover the signs and symptoms of DB, and as such are useful in the detection of the condition. Instruments like the capnograph, a device that measures the carbon dioxide concentration at the end of exhalation, can provide biochemical information associated with DB. Palpation techniques like the Manual Assessment of Respiratory Motion are potentially useful in detecting abnormal breathing patterns.

This review explores the respiratory system, and the aetiology and consequences of breathing dysfunction. The review also describes and critically evaluates relevant diagnostic tools for the assessment of different forms of breathing dysfunction.

2. THE RESPIRATORY SYSTEM: STRUCTURE AND FUNCTION

2.1 Background

The respiratory system is a basic life support system of the body. It helps to maintain homeostasis, which is the ability to maintain stable internal conditions (West 2008). On a physiological level it plays a vital role in the elimination of various substances such as carbon dioxide (CO_2), toxins and drugs. It is the body's means of supplying oxygen to cells and has a strong role in maintaining an acid-base metabolic balance. It also plays a role in venous and lymphatic return to the chest. Furthermore it affects motor control, postural stability and psychological regulation (D'Alonzo and Krachman 2003).

The muscles of breathing are uniquely supplied by both voluntary and autonomic nerves. Breathing rhythm is generated autonomically by clusters of neurons called respiratory centres in the pons and in the medulla. These centres are influenced by central and peripheral chemoreceptors and mechanoreceptors (West 2008). Input from higher brain centres, such as the cortex and limbic system, has the ability to alter the output of the respiratory centres in terms of timing and rhythm (Perna, Caldirola *et al.* 2004). Voluntary control is required for the production of speech, creating abdominal pressurization during defecation and physical work, and the expression of emotions for example laughing or crying (Fritz, Paholsky *et al.* 1999; D'Alonzo and Krachman 2003). Autonomic control primarily regulates breathing rate and blood supply of the lungs as well as vital reflexes such as coughing and sneezing (West 2008). The varying abdominal pressures during inhalation and exhalation also assist in the digestive process (West 2008). Functionality of the system relies on two processes: respiration and ventilation. Respiration is the gaseous exchange between lung tissue and blood, and cells and blood whilst ventilation refers to the mechanical action of inhalation and exhalation (West 2008). A healthy adult takes approximately 12 breaths per minute, with each breath containing approximately 500 millilitres of air (Fritz, Paholsky *et al.* 1999). This accounts for 17 280 breaths and around 8 640 litres of air per day.

2.2 Anatomy and Physiology of Breathing

The respiratory system is composed of the nose, pharynx, larynx, trachea, bronchial tree, alveoli and the pleurae (Marieb 2004). The process of breathing involves complex muscular contraction and relaxation, motion of fascial planes and the movement of nearly 150 joints (D'Alonzo and Krachman 2003). Respiratory muscles are active primarily in inhalation but also have a role in forced exhalation for example strenuous activity and coughing (Rochester 1993). The diaphragm is the primary respiratory muscle, whilst other breathing muscles are referred to as accessory respiratory muscles. During inhalation the diaphragm contracts and flattens, increasing the volume of the thoracic cavity and thus causing a decrease in intrapleural pressure and a flow of air into the lungs. During exhalation the diaphragm relaxes and domes, reducing thoracic cavity volume and increasing intrapleural pressure, which, in addition to the gravitational pull on the thoracic cage and the elastic recoil of the ribs, causes air to be forced out of the lungs. Accessory muscles involved in deep or forced inhalation include the external intercostals, scalene, upper trapezius, sternocleidomastoid and pectoralis minor muscles (De Troyer and Estenne 1988). Muscles involved in forced expiration include the abdominal wall musculature. The respiratory musculature transmits its force through the 12 pairs of ribs and indirectly though attachments to the sternum and all vertebrae from the first thoracic to third lumbar (De Troyer and Estenne 1988), hence breathing also has a direct impact on spinal stability and vice versa (Hodges 2003). Normal muscle function and joint mobility provides the structural foundation for movement and force transmission involved with breathing (Fritz, Paholsky *et al.* 1999).

Healthy respiratory function relies on several anatomical features, other than the respiratory muscles. The continuous beating of hair-like processes called cilia project foreign bodies upwards so that they may be expelled through the mouth or nose. Also, a thin membrane, called the respiratory membrane, separates air within the alveoli from the blood within the pulmonary capillaries. The respiratory membrane consists of alveolar and capillary walls and permits gaseous diffusion. Lung compliance, the distensibility of the lungs, is necessary to allow them to be stretched and filled with air during inhalation. (Fritz, Paholsky *et al.* 1999).

2.3 The Psychology of Breathing

Breathing is influenced by emotion and stress. Ronald Ley (1999), monitoring CO_2 in exhaled air, found that subjecting college students to brief mental stressors such as mental calculations increased their breathing rate in the direction of hyperventilation. Emotional states such as grief, suppressed anger, frustration and anxiety may maintain dysfunctional breathing patterns indefinitely (Gilbert 1998). Conversely, it is apparent that breathing can influence emotional state. A study by Philippot, Chapelle *et al.* (2002) demonstrated that specific emotional states such as anger, fear, joy and sadness could be induced by manipulating participants' breathing patterns (Philippot, Chapelle *et al.* 2002). Breathing has been shown to calm both mind and body, increase resilience in stressful situations and dampen levels of psychological and physiological arousal (McCaul, Solomons *et al.* 1979). Numerous studies have also shown that conscious control of breathing improves anxiety, depression and panic disorder (Ley 1999; Ley and Timmons 1999; Brown and Gerbarg 2005). Hence, specific breathing patterns are not only physical manifestations that are subject to emotional states, but may also play a role in perpetuating that emotional state (Philippot, Chapelle *et al.* 2002).

3. BREATHING DYSFUNCTION

3.1 Background

Breathing is suggested to be dysfunctional when it is inefficient or when it is insufficient for adapting to environmental conditions and changing requirements of the individual (Courtney 2009).

Possible causes of diminished breathing function are musculoskeletal dysfunction, disease, chronic psychological stress and other factors that affect respiratory drive and control (Courtney 2009). The breathing pattern in dysfunctional breathing often includes over-breathing, irregularity and unsteadiness of respiratory effort, predominant use of chest and accessory muscles of respiration during quiet breathing, rather than of the diaphragm, and frequent sighing (Han, Stegen *et al.* 1997). Altered breathing mechanics can change respiratory chemistry and therefore pH, causing smooth muscle constriction, altered electrolyte balance and decreased tissue oxygenation (Levitsky 2003). These changes can potentially affect any body system and cause a wide range of symptoms (Thomson, Adams *et al.* 1997).

In a review of breathing associated with chronic pain Chaitow (2007) attributes a diverse array of symptoms to breathing pattern disorders including, but not limited to, neck and head pain, chronic fatigue, anxiety and panic attacks, cardiovascular distress, gastro-intestinal dysfunction, lowered pain threshold, spinal instability and hypertension. Furthermore, there is a clear connection between respiratory (diaphragmatic) dysfunction and pelvic floor problems (high- or low-tone), potentially involving associated effects including stress incontinence, prostatic symptoms, interstitial cystitis and chronic pelvic pain (Lee and Lee 2004).

3.2 Dysfunctional Breathing Patterns

Dysfunctional breathing patterns include dysfunctions of breathing co-ordination (Lewis 1953; Lum 1981; Folgering 1999), abnormalities of volume and timing (Wientjes 1992) and excessive or reduced variability of breathing (Loveridge, West *et al.* 1984; Sody, Kiderman *et al.* 2008).

3.2.1 Dysfunctions of Breathing Co-ordination

Dysfunctions of breathing co-ordination comprise thoracic or upper rib breathing and paradoxical breathing. Thoracic breathing occurs when accessory muscles of breathing produce an upper rib cage motion during inhalation that exceeds lower rib cage and abdominal motion. This form of breathing pattern may be a normal functional adaptation to increased ventilatory demand as during physical activity (Sharp, Goldberg *et al.* 1975; De Troyer 1983). However, its persistence during quiet breathing can be considered dysfunctional (Lewitt 1980). Asynchronous or paradoxical breathing takes place when either the thoracic or abdominal compartment size decreases during inspiration (Jubran and Tobin 1992). Thoraco-abdominal asynchrony is generally considered to be dysfunctional because of its association with respiratory distress and with weakness and hypertonicity of respiratory muscles (Wolfson, Strohl *et al.* 1983; Jubran and Tobin 1992).

3.2.2 Abnormalities in Timing and Volume

Abnormalities in timing and volume are also indicative of DB and may contribute toward symptom production. Tobin, Chadha, *et al.* (1983) found that respiration rate and mean inspiratory flow was increased above normal in patients with COPD, restrictive lung disease and pulmonary hypertension. Patients with COPD often had major fluctuations of expiratory frequency and periodic fluctuations of end expiratory volume. Furthermore, the authors found that the breathing of chronic anxiety sufferers was characterised by frequent sighs, and occasionally episodic rapid rates alternating with apneas.

Hyperinflation occurs when the lungs retain a larger amount of air after exhalation. This may occur as a result of pathology for example emphysema for example(Wolfson, Strohl *et al.* 1983).

3.2.3 Abnormalities in Variability of Breathing

Variablity or irregularity of breathing is a normal homeostatic function of the respiratory system to some extent (Baldwin, Suki *et al.* 2004). Excessive irregularity of breathing, where the breathing pattern is unstructured and random, is a reliable indicator for disordered breathing as it is a sign that the respiratory control mechanisms are struggling to maintain homeostasis (Baldwin, Suki *et al.* 2004; Perna, Caldirola et al. 2004). In contrast, decreased variability of breathing demonstrates a lack of flexibility and responsiveness of the respiratory system which may lead to inappropriate matching of ventilation to conditions of rest and activity (Loveridge, West *et al.* 1984; Jubran and Tobin 1992).

3.3 Aetiology

There are several factors that can contribute to an inefficient and compromised respiratory system. These include muscular factors, such as decreased efficiency and strength of respiratory muscles; behavioural and psychological factors, such as anxiety; and the influence of chronic disease, including asthma and chronic obstructive pulmonary disease (Derenne, Macklem *et al.* 1978; De Troyer and Estenne 1988; Teixeira, Cherin *et al.* 2005).

3.3.1 Muscular Factors

The major biomechanical factors that contribute towards breathing dysfunction include abnormal function of the diaphragm and abdominal musculature as well as hyperinflation (Hruska 1997).

As with all muscles, power and effectiveness of the diaphragm are related to its resting length (Caruana, Petrie et al. 2001). If it is in a hypertonic and shortened state, the effectiveness of its contraction is limited (De Troyer and Estenne 1988; Finucane, Panizza et al. 2005). This inefficiency in turn leads to increased recruitment of accessory breathing muscles and contributes towards thoracic and paradoxical breathing (Hruska 1997). Furthermore, the diaphragm provides proprioceptive input that influences the perception of breathing. A shortened diaphragm may alter the sensation of breathing and thereby contribute to an increased respiratory drive (Frazier and Revelette 1991; Zhang and Davenport 2003). An increased respiratory drive, if persistent, further increases hypertonicity of breathing muscles, due to the increased frequency of contraction, which again leads to inefficiency and fatigability of the respiratory system (Gorini and Ginanni 1990; Rochester 1993; Hruska 1997). For example in one study, Finucane, Panizza et al. (2005) assessed the efficiency of the normal human diaphragm with hyperinflation in 5 healthy non-smoking men using digital recordings of oesophageal, gastric and mouth pressures, diaphragm electromyogram (EMG), as well as fluoroscopic images of the diaphragm and adjacent chest wall. The results suggest that a 25% decrease in diaphragm length is associated with an 80% decrease in diaphragm efficiency and requires an approximately fourfold increase in activation. Multiple regression analysis showed that diaphragm efficiency was highly correlated with diaphragm length ($p < 0.001$) (Finucane, Panizza et al. 2005).

Hyperinflation also decreases the efficiency of the respiratory system. When the system is in a state of hyperinflation the inspiratory muscles are shortened (Wolfson, Strohl *et al.* 1983; De Troyer and Estenne 1988; Hruska 1997). When the lungs are at 30% of inspiratory capacity, the costal part of the diaphragm has flattened and no longer expands the rib cage. Above 30% inspiratory capacity, contraction of the costal diaphragm pulls the ribcage inwards and has a deflating action (Wolfson, Strohl *et al.* 1983). At enlarged volumes there is also a tendency for the motion of the abdomen to reverse its timing so that inspiration is accompanied by inward motion of the abdomen and expiration is accompanied by outward motion of the abdomen i.e. paradoxical breathing (Wolfson, Strohl *et al.* 1983).

Dysfunction of the abdominal musculature may also have a detrimental effect on diaphragmatic function and thus breathing function (Hruska 1997). Hypertonic abdominal muscles limit diaphragmatic shortening during the inspiration phase and weak abdominal muscles fail to assist full diaphragmatic relaxation during exhalation. Abdominal weakness can lead to breathlessness and abnormal breathing patterns (Sharp, Goldberg *et al.* 1975; De Troyer 1983; Cahalin, Braga *et al.* 2002).

3.3.2 Behavioural and Psychological Factors

Psychological and emotional states, such depression, can disrupt normal breathing patterns independently of chemoreceptor and mechanoreceptor input (Boiten 1998; Jack, Rossiter *et al.* 2003). Persistent activation of the 'fear' network involving the cortex, hippocampus and amygdala can lead to disruptions in neurotransmitter, and receptor balance potentially resulting in panic disorder, an anxiety disorder linked with dyspnoea in terms of sensation of increased breathing effort and suffocation (Perna, Caldirola *et al.* 2004). In a study by Wolf (1994) the diaphragmatic movements of 17 subjects with a history of respiratory distress following emotional stress were examined fluoroscopically during engagement in a discussion of known emotional sensitivity. The ensuing emotional stress led to hyperventilation with continuously increasing hyperinflation of the lungs as well as hypertonicity and flattening of the diaphragm in all subjects (Wolf 1994). No statistical analysis of recorded data was provided in the article.

3.3.3 Chronic Disease

Suboptimal breathing biomechanics and breathing disorders are a feature of many chronic multi-symptom syndromes and diseases such as fibromyalgia, irritable bowel syndrome, anxiety disorder, multiple sclerosis, asthma, and chronic obstructive pulmonary disease (COPD) (Chaitow 2007).

Cardiorespiratory disease places higher demands on the respiratory system which adapts to maintain homeostasis. With respect to DB, changes occur in respiratory control, drive and pattern, for example chronic decreases in arterial oxygen tension associated with right-sided heart failure lead to an increased respiratory drive causing increased recruitment of secondary respiratory muscles and a thoracic dominant breathing pattern (Caruana, Petrie *et al.* 2001; Weitzenblum 2003; Hagman, Janson *et al.* 2008).

A study by Mutluay, Gürses *et al.* (2005) examined the effects of multiple sclerosis on respiratory functions in 38 patients with a definite diagnosis of multiple sclerosis. The patients were assessed using spirometry, and maximal inspiratory and expiratory mouth pressures (MIP and MEP respectively). Expected values from the healthy general population were used to evaluate findings of spirometry and maximal mouth pressures. Significant reductions in respiratory functions were found in both measurements ($p < 0.01$). However, reductions in MIP and MEP (-23% and -40% respectively) were larger than reductions in spirometry measures (6-9%). These results demonstrate that multiple sclerosis impairs respiratory functions as shown by a reduction in maximal mouth pressures. This impairment increased with multiple sclerosis-induced disability level but was found to be independent from duration of disease. The three most common respiratory problems in multiple sclerosis were found to be respiratory muscle weakness, bulbar function impairment and abnormalities of breathing control. Furthermore the study found that, compared to the healthy population, patients with multiple sclerosis tend to have a more superficial and faster breathing rate (Mutluay, Gürses *et al.* 2005).

The link between DB and asthma has been established in several studies where, in one case report, 42% of patients attending a hospital asthma clinic showed evidence of hyperventilation disorder as assessed by capnographic responses and Nijmegen questionnaire scores (McClean, Howells *et al.* 1999). Thomas, McKinley *et al.* (2001) evaluated 7033 patients with a diagnosis of asthma for symptoms related to breathing dysfunction using the Nijmegen Questionnaire. The authors found that about one third of women and one fifth of men with asthma had symptoms suggestive of dysfunctional breathing. Similar links are noted between DB and COPD. Tobin, Chadha, *et al.* (1983) found that in 28 patients with COPD, 14 with restrictive lung disease and 7 with pulmonary hypertension, respiration rate and mean inspiratory flow was increased above normal. Patients with COPD often had major fluctuations of expiratory timing, periodic fluctuations of end expiratory level of carbon dioxide, and asynchrony between ribcage and abdominal movements.

The adaptations associated with chronic conditions are the body's attempt to maintain homeostasis, yet they may also contribute to an exacerbation of physical symptoms (Gallego, Benammou *et al.* 1997; Mandak and McConnell 1998; Cahalin, Braga *et al.* 2002). Hence, diagnosis of the appropriate breathing dysfunction may prove to be beneficial in the management of these conditions.

3.4 Consequences

Several consequences of breathing dysfunction have been suggested including dyspnoea, adverse fluid dynamics, impaired posture and motor control and musculoskeletal pain.

3.4.1 Dyspnoea

Dyspnoea is a complex phenomenon that can be described as the sensation of respiratory distress or discomfort. It varies in intensity and quality of dysfunction depending on the underlying pathophysiological mechanism (Simon and Schwartzstein 1990; Elliot, Adams *et al.* 1991; Mahler, Harver *et al.* 1996). Dyspnoea often leads to increased recruitment of accessory muscles of breathing due to the increase in breathing effort, which causes a thoracic dominant breathing pattern (Simon and Schwartzstein 1990). The disparity between the intended motion of breathing and the actual achieved breathing motion contributes to the sensation of an “unsatisfying breath” which forms one dimension of dyspnoea (Manning and Schwartzstein 1995).

3.4.2 Adverse Fluid Dynamics

The alternating pressure gradients during inhalation and exhalation create rhythmic pressure fluctuations between the thorax and the abdomen that aid fluidic movement of blood and lymph between the two compartments (Dornhorst, Howard *et al.* 1952; DeBoer, Karemaker *et al.* 1987). Alterations in breathing pattern, in particular asynchronous motion, affect the amplitude of rhythmic fluctuations of blood volume, which activate homeostatic reflexes involved in maximizing optimal cardiorespiratory interaction and regulating blood pressure (Bernardi 2001; Yasuma and Hyano 2004). Byeon, Choi *et al.* (2012) observed superior and inferior vena cava collapsibility in relation to diaphragmatic breathing over a period of 2 years and found that the extent of abdominal breathing affects venous return via the inferior vena cava. Hence, the altered pressure differentials during a lack of abdominal breathing limit fluctuations in central venous pressure. This directly affects lymph drainage due to the decreased effect of venous return on the transmural pressure in lymphatic vessels (Lattuada and Hedenstierna 2006; Hedenstierna and Lattuada 2008).

3.4.3 Impaired Posture and Motor Control

The function of the respiratory muscles is not limited to breathing. These muscles play important parts in other everyday functions such as swallowing, speech production, valsalva manoeuvres, spinal stabilisation and movement of the trunk and the limbs. As breathing does not cease during most of these functions, a considerable amount of motor control is required to co-ordinate them with breathing (Rassler and Kohl 2000; Gandevia, Butler *et al.* 2002). Compromised respiratory musculature and abnormal breathing patterns adversely affect postural balance and proprioceptive function of the lower limbs and neuro-muscular co-ordination needed for prevention of spinal and other types of muscular-skeletal injury. Sakellari, Bronstein *et al.* (1997) investigated the effects of hyperventilation on sway in normal subjects and found that hyperventilation disrupts mechanisms mediating vestibular compensation, which are commonly associated with controlling balance and decreasing sway. Subsequently, hyperventialtion lead to an increase in sway. A recent study by Janssens, Brumagne *et al.* (2013) found that in 20 individuals with COPD, especially those with inspiratory weakness, there was a decreased reliance on back muscle proprioceptive signals resulting in decreased postural stability and hence an increased risk of falls.

Forward head posture, which is commonly associated with breathing dysfunction (Cuccia, Lotti *et al.* 2008), may have several adverse effects on the biomechanics of the head, neck and jaw and is associated with temporo-mandibular joint syndrome, neck pain and headache (Hruska 1997).

3.4.4 Musculo-Skeletal Pain

Dysfunctional breathing patterns have been associated with musculoskeletal pain in various regions, such as the chest wall (Wheatley 1975), neck and shoulder region (Perri and Halford 2004; Kapreli and Vourazanis 2009), and lower back (Chaitow 2004). In their study "Disorders of breathing and continence have a stronger association with back pain than obesity and physical activity" Smith, Russell, *et al.* (2006) conducted a cross-sectional analysis using multinomial logistic regression to model four levels of back pain (self-reported) in relation to body mass index, activity level, incontinence, allergies and breathing difficulties in 38 050 females. The authors found that unlike obesity and physical activity, disorders of incontinence and breathing were strongly related to frequent back pain (Odds ratios: 2.5 and 2.0 respectively compared to females with no incontinence or breathing difficulties).

4. ASSESSMENT OF BREATHING DYSFUNCTION

The clinical assessment of dysfunctional breathing or hyperventilation syndrome has previously been limited to hypocapnia and the symptoms thereof. To gain a more complete picture of DB, biomechanical and biochemical aspects as well as breathing symptoms need to be evaluated (Courtney and Cohen 2006; Courtney, van Dixhoorn *et al.* 2011). As such there are multiple assessment tools available to evaluate these specific aspects. Measures that assess psychological aspects of breathing and breathing symptoms are questionnaire-based for example the Nijmegen questionnaire (van Dixhoorn and Duivenvoorden 1985) and the Self-Evaluation of Breathing Questionnaire (Courtney and Greenwood 20009). Biochemical and physiological measures are generally instrument-based and include capnography (Johnson, Schweitzer *et al.* 2011), breath holding time (Delapille, Verin *et al.* 2001), breathing frequency (Folgering 1999), and spirometry (Lee 2009). Finally, biomechanical measures can also be instrument-based for example respiratory inductive plethysmography (RIP) (Brüllmann, Fritsch *et al.* 2010), but this aspect may also be evaluated using palpation, as for example in the manual assessment of respiratory motion (MARM) (Courtney, Van Dixhoorn *et al.* 2008). Any measurement of breathing of which the participant is aware may alter their normal breathing pattern, as the awareness of breathing potentially shifts breathing regulation from autonomic to voluntary control. The extent of this change remains unknown to date.

4.1 Questionnaire-based Assessments

4.1.1 The Nijmegen Questionnaire

The Nijmegen questionnaire (NQ) was originally developed as a symptom checklist to identify persons with hyperventilation syndrome. The NQ is a 16-item questionnaire asking about the frequency of incidence of complaints and indicated on a 5-point ordinal scale:

never = 0, rarely = 1, seldom = 2, often = 3 and very often = 4 (van Dixhoorn and Duivenvoorden 1985). A higher score indicates a greater degree of breathing dysfunction. A cut-off score of 23 is commonly used as a criterion of abnormal values. This is based on the original validation study that differentiated patients with a positive versus negative hyperventilation provocation test. However, the provocation test is no longer considered to be a valid test of hyperventilation syndrome due to its low specificity (Hornsveld and Garssen 1996).

In one efficacy study by van Dixhoorn and Duivenvoorden (1985), 75 patients were recruited on the basis of the diagnosis of hyperventilation, which was based on the complaint patterns described by the GP, specialist or therapist who referred the patient. The complaint patterns included high thoracic, rapid and irregularity in breathing, frequent sighing, etc. The reference group consisted of 76 professional individuals in the fields of yoga and physical therapy. All participants completed the questionnaire and were blinded as to the purpose of the study. The results revealed the sensitivity of the NQ to be 91% and the specificity to be 95% when comparing hyperventilation patients to non-hyperventilation individuals (van Dixhoorn and Duivenvoorden 1985). The research also established a three-dimensional structure of the questionnaire, the dimensions being dyspnoea, peripheral tetany and central tetany (van Dixhoorn and Duivenvoorden 1985). So, of the 16 NQ items, only four relate to breathing sensations directly, the others relate to neurological, psychological and cardiovascular symptoms (van Dixhoorn and Duivenvoorden 1985; Courtney and Greenwood 2009). According to Courtney, van Dixhoorn, *et al.* (2011) the NQ has been designed mainly to recognise symptoms of hyperventilation, and does not sufficiently identify all possible symptoms of DB. A copy of the NQ is provided in Appendix A.

4.1.2 The Self Evaluation of Breathing Questionnaire

Another questionnaire-based assessment of DB is the Self Evaluation of Breathing Questionnaire (SEBQ), which has been developed to cover a wider range of potential symptoms of breathing dysfunction than the NQ (Courtney and Greenwood 2009). The questionnaire’s development was based on multiple studies, the clinical experience of the development team as well as the online questionnaire: “How Good is your Breathing, Take our free Breathing Test and See” (White 2005). Studies considered in the SEBQ development included early papers by Burton (1993) and Howell (1990) who both describe a suggested link between hyperventilation disorder or dysfunctional breathing and specific symptoms. Other considered research addressed physiological and psychological aspects of breathing symptoms (Fried and Grimaldi 1993; Wilhelm, Gevirtz *et al.* 2001).

In their investigation of the SEBQ Courtney and Greenwood (2009) suggest that breathing symptoms associated with dysfunctional breathing arising from predominantly biomechanical aspects might be distinguishable from symptoms arising from factors reflecting chemoreceptor input. Two dimensions of the SEBQ were identified: “lack of air” and “perception of inappropriate or restricted breathing”. These dimensions appear to represent different aspects of dysfunctional breathing symptoms than those assessed by the NQ. As such the SEBQ has the potential to evaluate the response of separate dimensions of breathing symptoms more discriminately. A study by Mitchell (2011) showed high test-retest reliability of the SEBQ in a heterogenous sample of generally healthy individuals who took part in an online survey (ICC=0.88; 95% CI=0.84 to 0.91). To date there have been no studies that examined the validity of the questionnaire. A copy of the SEBQ is provided in Appendix B.

4.2 Instrument-based biochemical and physiological assessments

4.2.1 Capnography

Capnography is the measurement of carbon dioxide (CO_2) being exhaled with each breath. CO_2 is a by-product of cellular metabolism, brought to the lungs by the venous circulation and exhaled. The amount of exhaled CO_2 measured is a function of both CO_2 production and elimination. A capnograph measures the CO_2 content in exhaled air (known as End Tidal CO_2 or $ETCO_2$) via chemical reaction or actual CO_2 molecule measurement (Johnson, Schweitzer *et al.* 2011). $ETCO_2$ closely reflects arterial CO_2 in people with normal cardiopulmonary function. Capnography is used in critical care settings and is considered an accurate, time sensitive, arterial CO_2 measure (McLaughlin, Goldsmith *et al.* 2011). People with an accelerated rate of breathing that results in an increased minute volume, as is often the case in dysfunctional breathing, eliminate more CO_2 and have as a consequence a lower $ETCO_2$. Because of this, capnography could potentially be used as another diagnostic tool for DB. It should not be used as the sole diagnostic tool for DB, since instances of DB where no hypocapnia is exhibited would be missed (Thomas 2000).

McLaughlin *et al*. (2011) investigated the correlation between breathing dysfunction and musculoskeletal pain, as well as improvements in breathing function following breathing awareness training and manual therapy. The $ETCO_2$ of 29 patients with back and/or neck pain was measured pre- and post-intervention using capnography. At baseline, they reported that all patients had a below normal $ETCO_2$. Furthermore, they noted significant improvements in post-intervention $ETCO_2$ (Mean improvements = 6 mmHg, 95%CI = 5.0,7.2; $p < 0.001$) Numeric Pain Rating Scale (66% of patients improved according to the minimum clinically important difference (MCIP)) and Patient Specific Functional Scale values (79% of patients improved according to the MCIP). Two factors need to be acknowledged in the context of a lack of confirmatory evidence: the absence of a control group, and a conflict of interest of one of the authors in this study, the first author being a distributor for and a minor shareholder in the capnograph manufacturer: Better Physiology™.

4.2.2 Spirometry

Another option for evaluating aspects of breathing function is using spirometry, which can distinguish between normal and abnormal airway function. Spirometry measures the amount of flow and the volume of inhaled and exhaled air. Specific values of abnormal airway function can help in the diagnosis of pulmonary disease and the stages thereof (Lee 2009).

4.3 Instrument and palpation-based biomechanical assessments

4.3.1 Respiratory Inductive Plethysmography (RIP)

Since its introduction in 1977, the respiratory inductive plethysmograph (RIP) has become a valuable tool for monitoring ventilation and breathing patterns with multiple applications, including evaluation of sleep-related breathing disorder, respiratory monitoring in critical care and anaesthesia as well as other areas of physiologic and clinical research (Brüllmann, Fritsch *et al.* 2010). RIP is a method for non-invasive respiratory monitoring using inductance coils within elasticated bands to measure the respiratory excursion of the ribcage and abdomen (Duffty, Spriet *et al.* 1981; Brown, Aun *et al.* 1998). The device is based on the principle that the sum of volume changes of the rib cage and the abdomen equals the total inspiratory and expiratory flow at the airway opening making it a less-invasive alternative to its oral counterparts for example spirometry, and therefore possibly less likely to change breathing. The RIP signals from the rib cage and abdomen can be analysed separately to determine their relative contribution to ventilation (Leino, Nunes *et al.* 2001). RIP is relatively simple to perform and requires minimal patient cooperation (Mayer, Clayton *et al.* 2003).

Tobin *et al.* (1983) examined breathing patterns in non-smoking subjects without breathing pathology (‘normal’) (n=65), asymptomatic smokers (n=22), asymptomatic (n=17) and symptomatic (n=15) asthmatic patient, patients with chronic obstructive pulmonary disease (COPD) (n=28), restrictive lung disease (RLD) (n=14), primary pulmonary hypertension (PPH) (n=7) and anxiety state (n=13) using RIP. The authors found significant (see Table 1) increases in frequency of breathing and heightened respiratory drive in smokers, patients affected by COPD, RLD, anxiety state and PPH compared to normal subjects. Inspection of recorded breathing patterns revealed that patients with COPD and symptomatic asthma displayed greater asynchronous motion between the rib cage and the abdomen than the ‘normal’ group. Differences in age did not affect the breathing pattern in normal subjects (Tobin, Chadha et al. 1983). In conclusion, components of breathing such as the frequency and respiratory drive as well as asynchronous motion between the rib cage and abdomen are abnormal in selected respiratory conditions. Furthermore, these components can be assessed by physical examination.

Table 1

	Normal	Smokers	COPD	RLD	Anxiety	PPH
Frequency	16.6 (2.8)	18.3 (3.0)*	23.3 (3.3)**	27.9 (7.9)**	18.3 (2.8)*	25.1 (6.4)**
Respiratory Drive	250 (58)	345 (95)**	479 (132)**	434 (74)**	310 (135)**	461 (126)**

*Significant breathing pattern components (values indicate Mean +- SD) *p < 0.05, **p < 0.001*

4.3.2 Breath Holding Time

Breath holding time (BHT) is an indicator of a person's ventilatory response to biochemical, biomechanical, and psychological factors (Delapille, Verin *et al.* 2001). A shortened BHT may indicate abnormalities in respiratory control (Lin, Lally *et al.* 1974), and is commonly observed in individuals with tendencies towards hyperventilation and dysfunctional breathing (Jack, Darke *et al.* 1998; Warburton and Jack 2006). BHT varies markedly depending on how it is performed. Holding the breath after inhalation or exhalation, hyperventilation prior to assessment, and the size of the breath taken at the beginning of the breath hold all contribute toward BHT variation (Mithoefer 1965). The Buteyko method employs a standardized procedure that is used for evaluating and monitoring dysfunctional breathing: the 'BHT control pause'. The BHT control pause is a post-expiratory breath hold (tidal volume) and is performed with two slight variations. In one variation the breath is held until the first urge to breathe and in another variation until the first involuntary motion of the respiratory muscles (Courtney and Cohen 2008). BHT Control Pause levels below 20 s BHT are proposed to indicate the presence of dysfunctional breathing (Stalmatski 1999; Stark and Stark 2002).

Courtney and Cohen (2008) examined the relationship between BHT (control pause protocol) and other measures of dysfunctional breathing. They found that the BHT was shorter in people with abnormal spirometry (FEV1 or FVC < 15% below predicted), and coincided with upper thoracic breathing (as assessed by the MARM). Furthermore the authors found a negative correlation between the BHT and the end tidal carbon dioxide ($ETCO_2$) levels (Courtney and Cohen 2008). Because of the correlation of BHT with spirometry and abnormal breathing patterns, as well as its observed presence in dysfunctional breathing, BHT assessment may potentially form a part of the diagnostic procedure for DB.

4.3.3 Breathing Depth & Frequency

Depth and frequency of breathing determines how much oxygen and carbon dioxide are exchanged during breathing. These variables may be assessed using spirometry (Lee 2009). The two variables are matched according to the body's metabolic need. A mismatch of the variables, because of abnormal control or execution of breathing, leads to homeostatic imbalance if the minute volume is altered (Marieb 2004). The increased breathing frequency during hyperventilation causes respiratory alkalosis due to carbon dioxide deficiency (Folgering 1999). Increased breathing depth as exhibited during frequent sighing has a similar effect on respiratory alkalosis (Courtney and Cohen 2008). Significant respiratory alkalosis can reduce cerebral circulation and interfere with nerve transmission causing confusion, dizziness, light-headedness and sensory alterations (Gilbert 1998).

An upper thoracic breathing pattern has been found to be associated with hyperventilation symptoms in asthmatics and individuals with panic disorder (Carr, Lehrer *et al.* 1992). Hyperventilation is an important aspect of dysfunctional breathing that has established physiological effects (Courtney 2011). Measuring the depth and frequency of breathing thus also forms an important part in the diagnosis of DB.

4.3.4 An Osteopathic approach to assessment

Chaitow, *et al.* (2002), provide an osteopathic approach to the evaluation of respiratory function based on three elements: category (structures and pattern involved in breathing), locus of abdominal motion (extent of abdominal excursion during inspiration), and rate of breathing. Additionally, the authors describe a series of palpations and observation methods followed by an evaluation of posture, head position and relevant aspects of the shoulder girdle. Although this approach appears to be a complete and thorough way of assessing the respiratory system, it also seems to be time and effort consuming. Furthermore, there is no research confirming the reproducibility and validity of this approach. A faster and more practical way of assessment could prove beneficial in terms osteopathic practice.

4.3.5 The Manual Assessment of Respiratory Motion

The Manual Assessment of Respiratory Motion (MARM) is a technique to assess biomechanical aspects of a breathing pattern on the basis of palpation. It can be used to measure the relative distribution of breathing motion between the upper rib cage and the lower rib cage and abdomen as well as other aspects of breathing such as rate and regularity. The procedure takes three minutes to perform (Courtney, Van Dixhoorn *et al.* 2008). According to Courtney, Cohen *et al.* (2009), the MARM appears to be a reliable and valid as well as cost-efficient and easily applicable tool to establish breathing pattern disturbances. Furthermore, it is useful as a clinical and research tool as quantitative data (scores) can be derived from the assessment, with a higher score indicating a greater degree of breathing dysfunction. The procedure was first developed and applied in a follow-up study of breathing and relaxation therapy with cardiac patients in the 1980s. Later tests of inter-examiner reliability have indicated its potential as a clinical and research tool for evaluating breathing pattern (Courtney, Van Dixhoorn *et al.* 2008).

The relationship between dysfunctional breathing patterns and breathing symptoms was investigated in a recent study by Courtney, van Dixhoorn, *et al.* (2011). The MARM (breathing patterns) and the NQ (breathing symptoms) were used to measure the effect of a whole body breathing intervention (WBB) on 62 patients with medically unexplained complaints that appeared to be associated with breathing dysfunction. The WBB is a breathing and relaxation protocol.

The NQ was subdivided into 4 categories on the basis of principal component analysis and actual content. The subscores were: tension (‘anxiety’, ‘tension’, ‘palpitations’, ‘chest pain’), central neurovascular (‘blurred vision’, ‘dizziness’) peripheral neurovascular (‘tingling’, ‘stiffness’, ‘cold extremities’) and dyspnoea (‘tight feeling in chest’, ‘shortness of breath’, ‘faster or deeper breathing’, ‘unable to breathe deeply’).

The results revealed two observations that indicate that there may be a relationship between an abnormal breathing pattern and symptoms of dyspnoea. Firstly, a calculation of the changes in measurements revealed that changes in MARM balance were only correlated with changes in NQ dyspnoea but not with changes in other NQ subscales. Secondly, analysis of variance (ANOVA) revealed that although there were decreased NQ total scores in patients with and without initial abnormal MARM scores after treatment, the scores decreased more in patients with initial abnormal MARM scores. However, even though all subjects with high initial NQ dyspnoea scores demonstrated abnormal initial MARM values, abnormal initial MARM values were also present without elevated initial NQ dyspnoea scores (Courtney, van Dixhoorn *et al.* 2011). Therefore, breathing pattern disturbance and breathing symptoms may be related, but there appears to be no direct or immediate causal relationship. Perhaps prolonged abnormal breathing patterns lead to the production of breathing symptoms, as indicated by the finding that all subjects with breathing symptoms presented with an abnormal breathing pattern. Finally, classification of abnormal or normal breathing on both measurements (NQ and MARM) agreed in 74% ($p < 0.001$) of patients (Courtney, van Dixhoorn *et al.* 2011). This gives some indication that both tools may be useful in the diagnosis of breathing dysfunction.

In one of their studies Courtney, *et al.* (2008) investigated the MARM and compared it to respiratory induction plethysmography (RIP). The study involved two practitioners performing both assessments on 12 subjects who consciously altered their breathing and posture to achieve nine conditions arising from combinations of three breathing and three postural instructions (Table 2).

Table 2

1. Breathe normally-sit in your normalposture (BN-NP)	4. Breathe normally-sit in slumpedposture (BN-SP)	7. Breathe abdominally-sit in slumpedposture (BA-SP)
2. Breathe thoracically-sit in your normalposture (BT-NP)	5. Breathe normally-sit in erect posture (BN-EP)	8. Breathe thoracically-sit erect posture (BT-EP)
3. Breathe abdominally-sit in your normalposture (BA-NP)	6. Breathe thoracically-sit in slumpedposture (BT-SP)	9. Breathe abdominally-sit erect posture (BA-EP)

The nine breathing and posture instructions used in the study (Courtney, Van Dixhoorn et al. 2008).

The MARM measurements utilized were ‘volume’ (ribcage and abdominal excursion), ‘balance’ (difference between rig cage excursion and abdominal excursion) and ‘percent rib cage (%RC)’ (the percentage of relative rib cage contribution). [Appendix C].

The RIP measurements used were ‘percentage of ribcage motion (RIP %RC)’ (the percentage of relative rib cage contribution), ‘mean phase relation of total breath (MPRTB)’ (the percentage of direction in which ribcage and abdomen move in opposite directions during the respiratory cycle), and ‘peak inspiratory flow ribcage (PIFRC)’ (reflects respiratory drive in the thoracic compartment)

The inter-rater reliability for the MARM %RC and balance variables was very large (r = 0.844, p <0.01 and r = 0.85, $p < 0.01$ respectively) which indicated significant agreement between the two examiners.

The strongest correlation between the two measurements was found between measures of upper ribcage contribution i.e. RIP %RC motion and MARM %RC (r = 0.60, $p < 0.01$) and MARM balance (r = 0.59, $p < 0.01$) measures. Other correlations were positive but weak. The correlation with the RIP measure of upper ribcage contribution indicates that the MARM appears to be a reliable tool to measure thoracic dominance.

Instructions to breathe abdominally resulted in lesser rib cage involvement and vice versa for instructions to breathe thoracically. An ANOVA of voluntary breathing (dependent variable) and posture (independent variable) showed that each measurement was able to detect changes in voluntary breathing. The MARM %RC and balance measures displayed a higher ability to differentiate between the three breathing patterns than the RIP %RC measure (MARM %RC ($F(2,22) = 191.2$, $p = .0001$, partial $n^2 = .946$) and MARM balance ($F(2,22) = 189.4$, $p = .0001$, partial $n^2 = .945$) compared to RIP %RC ($F(2,16) = 12.89$, $p = .0001$, partial $n^2 = .617$). A possible explanation is that RIP can only be used to measure horizontal expansion, yet the ribcage also expands vertically during inhalation. Hence, the MARM may be better suited to specifically distinguish vertical ribcage motion than RIP.

With respect to the ability of the MARM to distinguish vertical ribcage motion, one may argue, as the authors acknowledge, that there is the possibility of an observer bias in terms of the visual information gained by seeing the patient's chest movement, where the RIP measures were collected electronically. The extent to which the participants were able to accurately reproduce a given breathing pattern could be questioned. However, the variation between participants for all parameters was markedly small, indicating that they were, in fact, able to accurately reproduce a given breathing pattern. Despite the correlations between MARM and RIP measures, the conclusions of the study could be challenged due to the limited number of examiners and test subjects.

In addition to comparing the MARM to a questionnaire based (NQ), and instrument based (RIP) assessment, Courtney, Cohen *et al.* (2009) also compared the MARM to the Hi-Lo Breathing assessment, a method of distinguishing between abdominal or thoracic motion dominance during breathing based on palpatory findings. An additional aim was to establish the relationship between experience and ability, by comparing the ability of students and practitioners to perform these assessments.

The study involved 56 osteopaths and osteopathic students who performed both assessments on each other. All participants took part in a two-hour training session during which they were taught to simulate abnormal breathing patterns, and to apply the MARM and Hi-Lo techniques of breathing pattern assessment. They each assessed random combinations of thoracic, abdominal or paradoxical breathing (Hi-Lo only). Furthermore all participants completed a survey that reflected on the experience of using the assessments.

The authors found that both students and practitioners were able to accurately assess the simulated breathing patterns for both assessments in terms of percentage of correct rating (Table 2). The survey showed that both groups had slightly more positive impressions of the MARM than the Hi-Lo assessment (Courtney, Cohen *et al.* 2009). The results reflected some agreement between the two types of assessment for evaluation of thoracic breathing ($k = 0.29$, $p = 0.001$). Assessors, using the MARM, could distinguish between thoracic and abdominal dominant breathing, yet, in the absence of a reference measure, there is a lack of data on the validity of the MARM, i.e. how accurately the assessment can establish the actual contribution of the rib cage or the abdomen. Similarly, the validity of this assessment in a clinical context is unknown. The breathing patterns in this study were simulated and may provide the assessor with information that varies from a real breathing pattern, for instance the trunk excursions during a breath may be clearer during a simulated breath.

Measure	Students (% correct)	Practitioners (% correct)	Total (% correct)
MARM thoracic	82	88	85
MARM abdominal	87	78	83
Hi-Lo thoracic	94	97	96
Hi-Lo abdominal	93	96	95
Hi-Lo paradoxical	68	78	73

Table 3*: Performance on specific MARM and Hi-Lo measures (Courtney, Cohen et al. 2009).*

The quality appraisal tool for studies of diagnostic reliability (QAREL) was used to test the quality of the two studies that assessed the reliability of the MARM (Courtney, Van Dixhoorn et al. 2008; Courtney, Cohen et al. 2009) . The QAREL is a checklist that includes 11 items that encompass key principles for the quality of studies of diagnostic reliability (Lucas, Macaskill et al. 2010). Items cover the spectrum of subjects, spectrum of examiners, examiner blinding, order effects of examination, suitability of the time interval among repeated measurements, appropriate test application and interpretation, and appropriate statistical analysis. Both, raters and subjects were representative of the sample which the authors intended the results to be applied to. The order of examination was varied and the tests were applied correctly in appropriate time intervals and interpreted properly in both cases. Appropriate statistical measures of agreement were also used in both studies. The drawback of both studies is that the raters were not blinded to visual clues and previous findings during the re-test phase. Overall the QAREL evaluation shows that combined results of both studies is sufficient to indicate the MARM as a reliable diagnostic tool in assessing breathing pattern disturbances. The QAREL checklists for both studies are provided in Appendices D and E.

4.3.6 Relationships between Breathing Assessments

The relationships between the various non-invasive assessments of breathing have not been explored to a large extent. One study by Courtney, Greenwood *et al* (2011) compared various screening tools proposed to identify DB including the NQ, SEBQ, breath-holding time (BHT), capnography, Hi-Lo breathing assessment, MARM, and spirometry in their ability to measure distinct or associated aspects of breathing functionality. Eighty-four self-referred or practitioner-referred individuals with concerns about their breathing were assessed using the screening tools, and correlations between these measures were determined. There were no statistically significant correlation between spirometry and other screening tools except for a moderate correlation with BHT ($r = 0.31$, $p < 0.004$). There were no statistically significant correlations between breathing symptoms (questionnaires) and physiological and biomechanical aspects (other measures) of breathing (Courtney and Greenwood 2009).

However, other studies found several correlations between assessments of breathing. Courtney and Cohen (2008) assessed 83 adults, healthy or with suspected breathing dysfunction, using spirometry, BHT, the NQ, capnography ($ETCO_2$), and the MARM and correlated BHT with each of the measurements. The authors found a small negative correlation between BHT and $ETCO_2$ (r = -0.24, $p < 0.05$), and a moderate negative correlation between BHT and thoracic dominant breathing pattern (r = -0.41, $p < 0.03$). This indicates that BHT may represent a functional aspect in abnormal breathing. The study by Courtney, van Dixhoorn *et al.* (2011) discussed previously, on the effects of breathing retraining on dysfunctional breathing as assessed by the MARM and the NQ demonstrated a small correlation between the MARM and the NQ (r = 0.26, $p < 0.05$). Another finding was that symptoms of dyspnoea were only elevated in individuals with abnormal MARM scores.

So the lack of correlation of various breathing assessments may be, as the authors suggest, due to the fact that the samples used in the studies tended to represent individuals with only mild signs of dysfunctional breathing. Correlations not evident in this sample with mild DB may be stronger in individuals with more severe DB, or in a sample that demonstrates a greater variety of DB. Additionally, relationships that do exist between breathing pattern and symptoms may be non-linear (Courtney and Greenwood 2009). On the other hand, there is a possibility that some measures do not indicate breathing dysfunction with dependent clinical consequences. In conclusion, each tool may provide diagnostic information about a specific aspect of DB but, no single tool is all encompassing. The observation that several measures of dysfunctional breathing do not necessarily correlate more highly, further emphasises the need for research in this area.

5. CONCLUSION

A review of literature reveals a lack of complete characterisation of DB and the need for clinically useful means to diagnose it due to its ability to produce symptoms. The current use of various diagnostic tools in isolation may not be sufficient for a diagnosis (Han, Stegen *et* *al.* 1997; Thomas, McKinley *et al.* 2001; Hagman, Janson *et al.* 2008; Courtney, Greenwood *et al.* 2011). Hence, a specific characterisation and associated standardized method of diagnosis would prove beneficial in the detection, management and treatment of the condition (Courtney, Cohen *et al.* 2009). Perhaps a reason why a standardized diagnostic tool has not been established to diagnose DB is, as Courtney (2011) suggests, that DB occurs in a multi-dimensional clinical presentation, notably biomechanical, biochemical, psychological and physiological, each of which may or may not co-exist. In light of this, the prospect of a single diagnostic tool appears unlikely. However, different diagnostic tools may be used in combination to provide a more comprehensive way to diagnose DB. This study will provide further information on the utility of the MARM as one diagnostic tool in the evaluation of the biomechanical aspects of DB by examining the reliability of the tool when applied to the assessment of non-simulated breathing patterns. In this way the results will provide a more clinically realistic view on the reliability of the assessment compared to previous studies. Furthermore, this study will further examine the relationship between several non-invasive measures of breathing function, including breath holding time, breathing rate, symptoms (assessed by the NQ and SEBQ), and the MARM.

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SECTION 2: MANUSCRIPT

Inter and Intra-rater Reliability of the Manual Assessment of Respiratory Motion ('MARM' technique) in Adults

MANUSCRIPT

1. ABSTRACT

Aim: The primary aim of this study was to investigate the inter- and intra-rater reliability of a 'manual assessment of breathing' (MARM). A secondary aim was to explore the relationship between Breath Holding Time (BHT), Breathing Rate (BR), breathing compartment dominance (thoracic and abdominal), and two questionnaires concerned with symptoms of breathing (Nijmegen (NQ) and Self Evaluation of Breathing Questionnaires (SEBQ)).

Methods: Practitioners (n=6) received introductory level instruction in MARM assessment. They then rated the mechanical breathing pattern of 16 participants with nil to moderate breathing dysfunction. Participants completed two breathing questionnaires (NQ and SEBQ) and were also assessed for Breath Holding Time, Breathing Rate and Abdominal Excursion using Respiratory Inductive Plethysmography.

Results: The inter-rater and intra-rater reliability of the MARM was poor (inter-rater: $ICC_{[2,1]}$ range = -0.18 to 0.36; intra-rater: $ICC_{[2,1]}$ range = -0.51 to 0.92). Inter and intra-rater reliability of the MARM was insufficient to warrant clinical use. Moderate correlations were identified between Breathing Rate (BR) and Breath Holding Time (BHT) ($r = 0.5$; $p = 0.04$), between BHT and the Self Evaluation of Breathing Questionnaire (SEBQ) ($r = 0.5$; $p = 0.05$), and between the SEBQ and the Nijmegen Questionnaire (NQ) ($r = 0.6$; $p = 0.01$).

Conclusions: Raters in this study demonstrated poor inter-rater reliability of the MARM. The intra-rater reliability is higher for some raters indicating that the MARM may be more useful for individual practice, when the same rater is undertaking assessment of breathing.

KEYWORDS

Breathing Dysfunction, Breathing Assessment, Respiration, Measurement, Physical Examination

2. INTRODUCTION

It has been suggested that 'optimal' breathing occurs when there is an even distribution of breathing effort between the two main functional compartments of the body involved in breathing, the upper rib cage and the lower rib cage/abdomen [1]. An uneven breathing distribution may be considered to be unnecessary, laborious and dysfunctional [1].

Although there is no consensus definition of what constitutes DB, the prevalence of dysfunctional breathing (DB) has been estimated to be as high as 5-11% in the general population, around 30% in people with asthma and up to 83% in people with anxiety [2, 3]. Impairment of breathing function may negatively affect people's lives by hindering the process of homeostasis, creating undesirable symptoms and compromising health [2]. Abnormal breathing patterns have been associated with a range of symptoms including: breathlessness, chest tightness, chest pain, light-headedness, paraesthesiae, and anxiety [4].

Several measurements of breathing dysfunction have been reported in the literature. However, relationships between measurements to identify DB remain unclear. DB may include hyperventilation [5], breathing pattern abnormalities [6], poor breathing control, and the presence of breathing symptoms [7]. The common perception of hyperventilation-associated hypocapnia as the sole contributor towards symptoms of dysfunctional breathing has been disputed [3, 8, 9]. It has been reported by several authors that DB symptoms can exist in the absence of hypocapnia [10-12]. In a recent study, Courtney, Greenwood et al. [10] found that individuals may exhibit only one aspect of breathing dysfunction and that the different aspects do not necessarily correlate.

To gain a more complete picture of DB, biochemical, biomechanical and psychological factors all need to be evaluated [9, 13]. The assessment tools available to evaluate the various aspects of breathing dysfunction range from instrument-based measurement tools including capnography [14], respiratory inductive plethysmography [15] and spirometry [16], to simple assessment methods including breath holding time, breathing rate, questionnaires of breathing symptoms and motion palpation of chest and abdominal wall movement [10]. A standardized and validated method for assessing the biomechanical aspects of breathing patterns would be clinically useful in assessing biomechanical factors associated with DB and consequently enhance its diagnosis and management.

The Manual Assessment of Respiratory Motion (MARM) is a palpation-based assessment of chest wall movement during breathing and was first developed and applied in a follow-up study of breathing and relaxation therapy with cardiac patients in the 1980s [1]. MARM allows the examiner to record a graphic representation of their impression of the direction of relative dominance of upper ribcage motion to lower ribcage/abdomen motion [13]. According to a reliability study by Courtney, Cohen et al. [17] the MARM is reliable and valid, as well as a user-friendly tool to assess breathing pattern disturbances. Additionally, a good level of inter-rater agreement in the assessment of breathing pattern disturbance using the MARM has been shown ($r = 0.85$, $p = 0.01$) [1]. However, the study on inter-rater agreement was conducted using healthy participants who simulated dysfunctional breathing patterns. Simulated breathing patterns may provide the assessor with information that varies from a natural breathing pattern for example the pattern may be more obvious in presentation. To date, there has been no study aimed at investigating the reliability of the MARM utilizing participants with known breathing pattern disturbances and using raters outside of the MARM developers.

Therefore, the aim of this study was to: 1) investigate the inter and intra-rater reliability in determining mechanical breathing pattern abnormalities using the MARM; and 2) quantify the correlation between 6 different measurements of breathing function (Nijmegen Questionnaire (NQ); Self Evaluation of Breathing Questionnaire (SEBQ); Breath Holding Time; Breathing Rate; and Abdominal Excursion as assessed by Respiratory Inductive Plethysmography) and MARM scores.

3. Methods

3.1 Sample

Participantswere recruited over a period of three months from three sources: 1) patients attending an osteopathy teaching clinic (Unitec Institute of Technology, Auckland) 2) two Auckland-based physiotherapy clinics specialising in breathing dysfunction; and 3) from tertiary students attending the local campus. All participants were required to be over 18 years and able to read and understand English. Exclusion criteria were: surgery involving the trunk within the last 6 months; known disease or illness involving incapacitation or considerable amounts of bed rest. People who responded to advertising and met the inclusion-exclusion criteria were provided with an information sheet about the study and gave written informed consent. The study was approved by the Unitec Research Ethics Committee (Approval number: 2012-1230) [see Appendix F].

Raters (n=6) were recruited from practitioners attending a 2-day instructional course on the assessment and treatment of breathing dysfunction by a co-developer of the MARM. Of the 6 raters recruited, 3 were physiotherapists specialising in breathing therapy, and 3 were osteopaths. All practitioners were experienced clinicians and held current annual practicing certificate with their respective regulatory authorities. The workshop took place 3 weeks prior to the first testing session.

3.2 Data Collection

All raters attended a 2-hour instructional session in using the MARM and the corresponding method of recording as part of the workshop on the assessment and treatment of dysfunctional breathing. A written protocol including the MARM notation template was provided to all raters during each testing session [see Appendix G]. The raters read the breathing directions to the participants for each item as per protocol. To improve reliability the protocol included ample variation in size and depth of breathing in order to observe a range between maximal and minimal inhalation. The protocol was suggested, following enquiry, by the original developer of the MARM (van Dixhoorn, J 2012, personal conversation, 19th May). The variables collected were: ‘Sit Easy 1’, ‘Sit Straight’, ‘Breathe Deep’, Breathe Wide’, and ‘Sit Easy 2’. The MARM variables: ‘balance’, ‘area’, and ‘%RC’ were then calculated for each of the variables [See Appendix C for a calculation and description of the variables].

Breathing rate, lower ribcage/abdominal excursion, breath holding time (BHT) and MARM measurements were assessed during three separate testing sessions, between 2 and 3 weeks apart. Participants being assessed were randonly allocated to one of three testing sessions.

The raters were blinded as to which participant they were testing using participant draping and uniform clothing in an attempt to minimise the presence of non-clinically relevant cues that may bias MARM interpretation. After completing the MARM testing procedure on each participant, the raters left the room. The participant sitting arrangement was randomised in an attempt to minimise recall bias. Only two of the six raters re-entered the room during each session and completed a second round of the MARM on each participant. Different raters completed the re-test for each of the three sessions. Each rater therefore performed re-tests in only one testing session.The smaller number of re-tests was used to avoid prolonged testing procedure for reasons of potential participant fatigue.

Breathing rate, lower ribcage/abdominal excursion and BHT of each participant was assessed using a respiratory inductive plethysmography belt (Model: ML856; ADInstruments Pty Ltd, NSW). Data were recorded using a digital data acquisition system (Powerlab 8/SP; ADInstruments Pty Ltd, NSW).

The “control pause” protocol described by Courtney and Cohen [18] was used to assess BHT. Participants were asked to perform a post-expiratory breath hold (tidal volume) and indicate when they felt the first involuntary contraction of the respiratory muscles. Two variables were extracted for BHT: the elapsed time when participants reported the first involuntary contraction of inspiratory muscles; and the elapsed time observed at point of inflection for the trace recorded from by respiratory inductive plethysmography belt.

Breathing symptoms were assessed using the SEBQ and NQ questionnaires. Participants completed the two questionnaires following the test sessions using web-based questionnaire forms.

3.3. Data Analysis

To address the first aim, inter-rater and intra-rater reliability was assessed for breathing specialists, manual therapists and between all raters, by calculating the Intraclass Correlation Coefficient using the two-way random model ($ICC_{[2,1]}$) and interpreted according to the descriptors of effect size suggested by Hopkins [19]. A correlation threshold of $r > 0.6$ was used to estimate clinical significance [19]. As recommended by Hopkins [20], within-subject variability as well as systematic change (test-restest bias) were calculated using standard errors of measurement (SEM) and change in mean respectively. According to Hopkins [20] SEM is directly proportional to Bland-Altman Limits of Agreement [21].The coefficient of variation (CV) was calculated in order to express variation of measurement as a percentage rather than a raw number [19].

The second aim, to observe the relationship between the collected variables of breathing function, was explored by calculating Pearson correlation coefficients between all variables. An exploratory factor analysis, using principal axis factoring extraction and varimax with Kaiser normalization rotation, was used to describe variability among observed, correlated variables. Factor loading was interpreted according to descriptors of magnitude by Comrey and Lee [22]. Normality among the single variables was assessed using skewness and kurtosis. Statistical analysis was undertaken using SPSS (SPSS Inc. Released 2008. SPSS Statistics for Windows, Version 17.0. Chicago: SPSS Inc.)

4. Results

A total of 16 participants and 6 raters were recruited for this study. All raters completed the data collection process. All participants completed the questionnaires and attended their scheduled data collection session.

4.1 Reliability

4.1.1 Inter-Rater Reliability

As can be seen in figures 1 and 2, no MARM variables reached the clinical significance threshold of correlation ($ICC_{[2,1]}$ range = -0.18 to 0.36). The breathing specialist rater group achieved overall higher reliability. The highest intraclass correlation coefficient calculated was for the 'Width Breath % RC' variable of the breathing specialist rater group: $ICC_{[2,1]}$= 0.36; 95% CI = -0.01 to 0.73; SEM = 11.6 ; and CV = 28%. [A specific depiction of all variables can be seen in Appendix H]

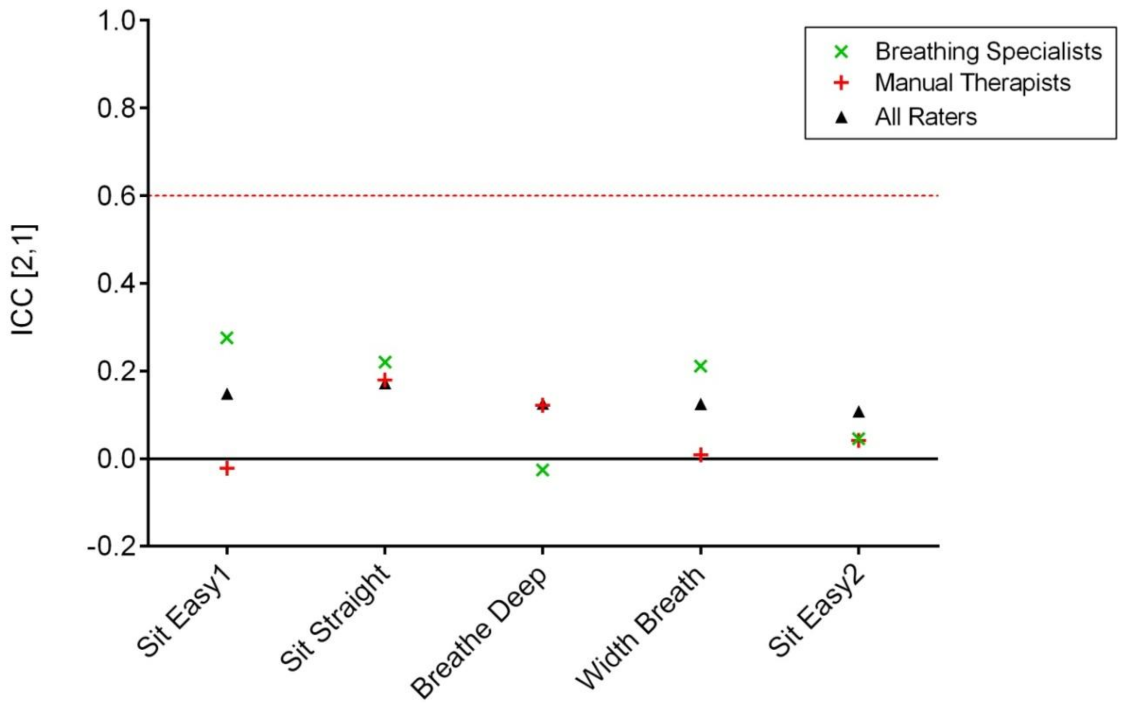


Fig.1: *Summary of reliability for the five breathing variables. The red dotted line indicates the clinical significance threshold of correlation.*

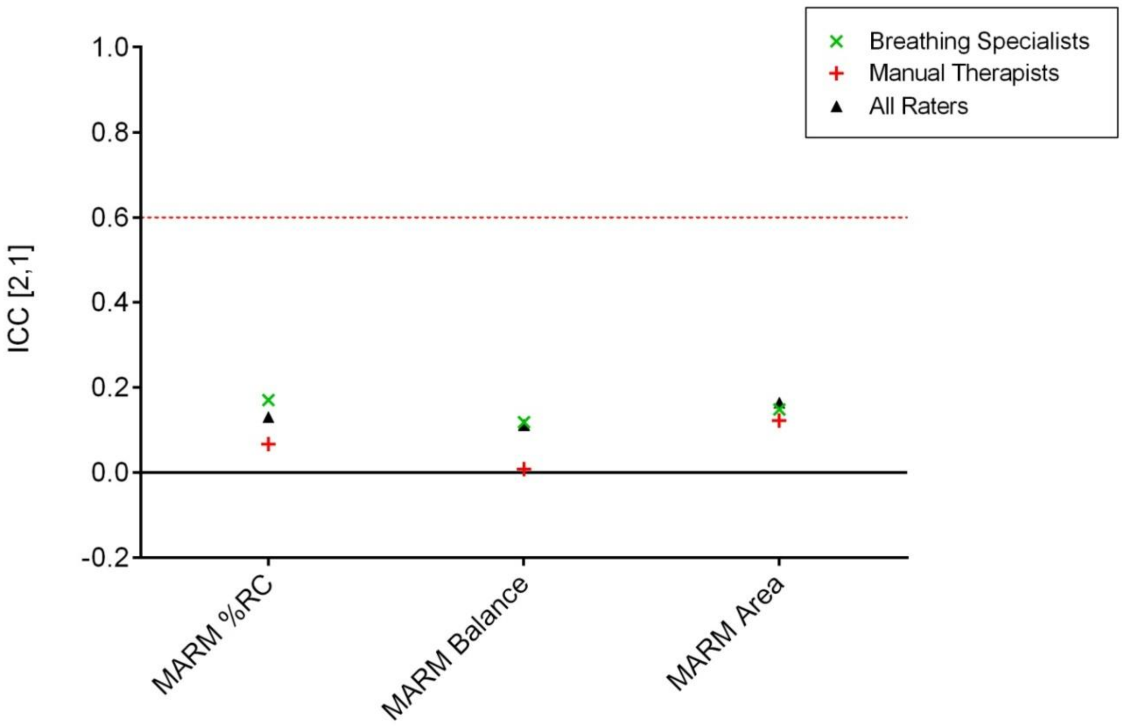


***Fig. 2**: Summary of reliability for the three variables calculated from each breathing variable. The red dotted line indicates the clinical significance threshold of correlation.*

The calculation of ICCs assumes interval or ratio data i.e. that each rating category is equally wide [23]. This trait may not be inherent to the MARM notation. Consequently the variables were standardised by calculating z-scores in order to achieve ordered-category ratings. However, calculation of the ICC from the standardised variables did not increase the inter-rater reliability significantly.

4.1.2 Intra-Rater Reliability

As can be seen in figures 3 and 4 the intra-rater reliability was higher than the inter-rater reliability but still fell below the threshold considered acceptable for clinical significance ($ICC_{[2,1]}$ range = -0.51 to 0.92). The breathing specialist rater group achieved higher reliability on average. The highest intraclass correlation coefficient calculated was for the 'Width Breath AREA' variable of the manual therapist rater group: $ICC_{[2,1]}$= 0.92; 95%CI = 0.78 to 0.97; SEM = 7.8; CV = 11%. The 'Width Breath AREA' variable for the breathing specialist rater group was the only other variable to reach significance (95%CI > 0.6). [A specific depiction of all variables can be seen in Appendix I]

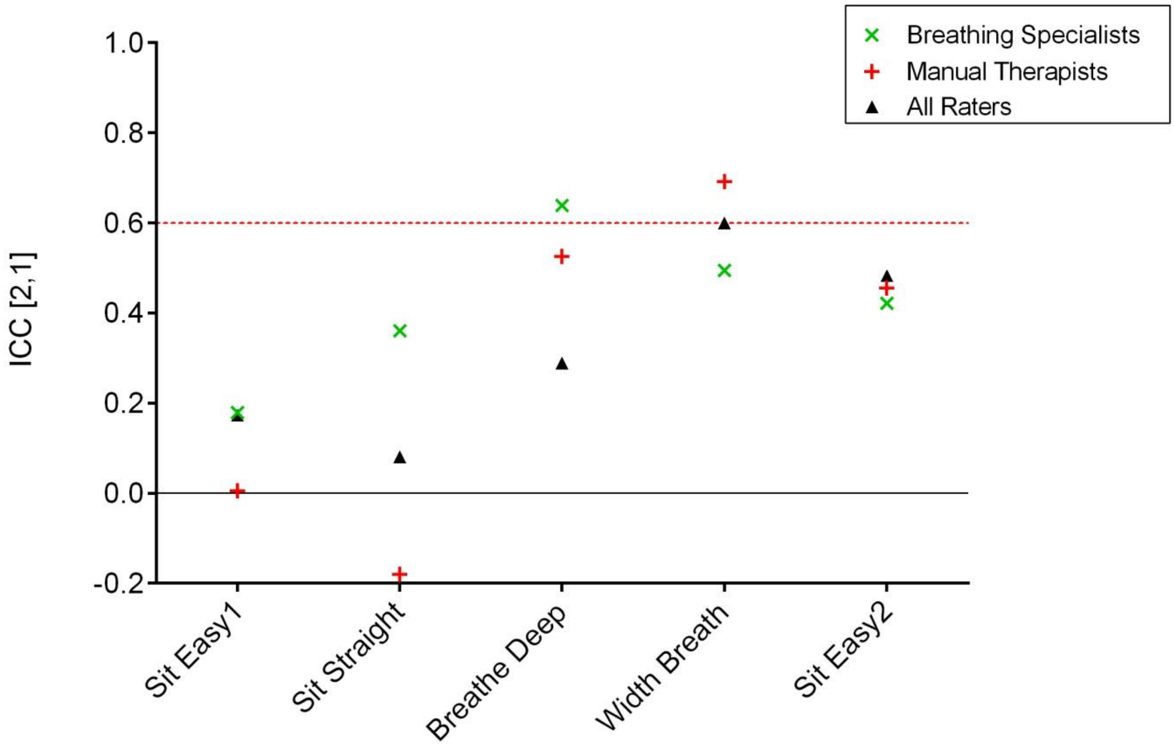


***Fig.3**: Summary of reliability for the five breathing variables. The red dotted line indicates the clinical significance threshold of correlation.*

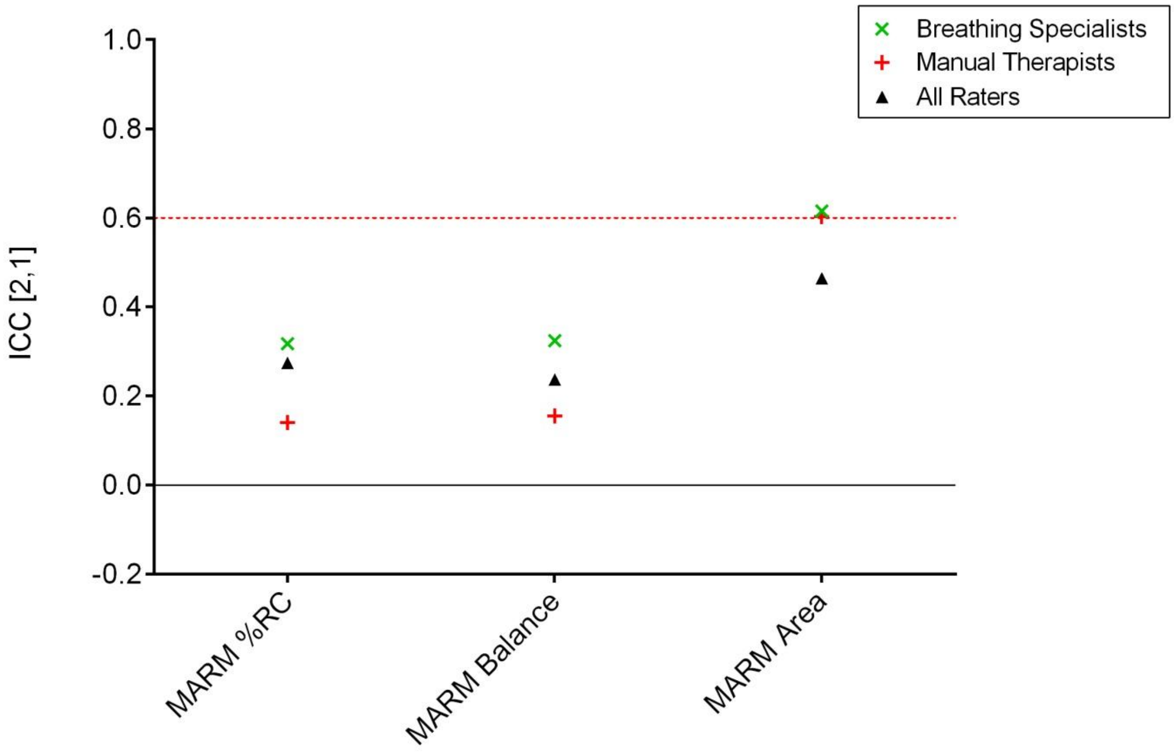


Fig. 4: *Summary of reliability for the three variables calculated from each breathing variable. The red dotted line indicates the clinical significance threshold of correlation.*

Calculation of within-subject variance demonstrated moderate variation. The standard error of measurement for all testers was Mean = 11.7; Range = 7.9,17.5 (CV: '%RC' = 18-30%; 'Area' = 11-24%). The CV for the 'balance' variable could not be calculated due to negative numbers. Calculation of systematic change demonstrated a statistically significant ($p < 0.001$) retest score decrease in the MARM 'area' variables. The MARM '%RC' and 'balance' variables did not show consistent significant retest score differences.

4.2 Correlation between Measures of Breathing Function

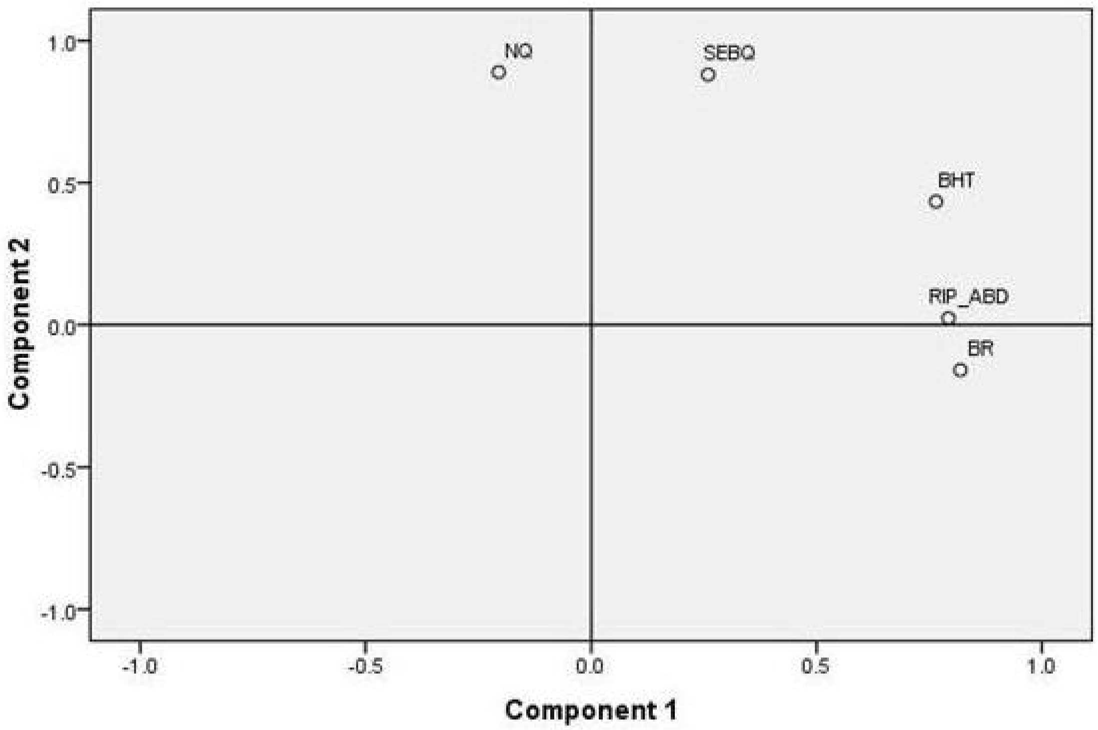
Participants in this study predominantly presented with none to minor breathing dysfunction according to the SEBQ and NQ scores. The mean score for the SEBQ was 19.7 out of 75 with a minimum score of 6 and a maximum score of 36 (DB cut-off score not defined as of yet). The mean score for NQ was 19.5 out of 64 with a maximum score of 41 and a minimum score of 6 (NQ cut-off score for DB is set at 23).

Moderate positive correlations were found between Breathing Rate (BR) and Breath Holding Time (BHT) ($r = 0.52$, $p = 0.03$), between BHT and the Self Evaluation of Breathing Questionnaire (SEBQ) ($r = 0.50$, $p = 0.05$), and between the SEBQ and the Nijmegen Questionnaire (NQ) ($r = 0.60$, $p = 0.01$). Other correlations between the measures of breathing function were trivial. MARM scores were excluded due to the low test reliability.

In all cases except 2 the perceived duration until the first diaphragmatic contraction during the breath hold was longer than the objective recorded first contraction (RIP). The time difference between the two measures was Median = 8.4 s; Mean = 7.8 s, SD = +- 1.8 s; Range = -3.9, 23.2).

Visual inspection of data revealed that the participant with the highest symptom (SEBQ) score (36) also had the highest NQ, lowest BHT, highest BR and lowest RIP abdominal excursion (RIP_ABD) scores; whereas the participant with the lowest SEBQ score demonstrated the opposite findings.

Factor analysis revealed two distinct components. ‘Component 1’ accounts for 40% of variance with an Eigenvalue of 1.99. The variables BHT, BR and RIP_ABD all load highly in ‘Component 1’. BHT also loads moderately in ‘Component 2’, along with SEBQ and NQ.



‘Component 2’ accounts for 36% of variance with an Eigenvalue of 1.78. Figure 5 shows a scatterplot of the individual variable loadings in relation to each other.

Fig. 5*: Component plot displaying loadings of five measurements of breathing function NQ (Nijmegen Questionnaire), SEBQ (Self Evaluation of Breathing Questionnaire), BHT (Breath Holding Time), BR (Breathing Rate) & RIP_ABD (Abdominal Excursion as measured by RIP)*

5. Discussion

The primary aim of this study was to investigate the inter and intra-rater reliability in determining mechanical breathing pattern abnormalities using the MARM. Contrary to previous studies [1, 24], we showed that the inter-rater reliability was poor for all variables of the MARM. Courtney, Van Dixhoorn et al. [1] utilised 2 raters to assess 12 participants using the MARM. The results demonstrated very high inter-rater agreement for measures of the upper rib cage relative to lower rib cage/abdomen motion during breathing ($r = 0.85$, $p = 0.01$) but not for measures of area ($r = 0.13$). A subsequent study by van Dixhoorn [24] using 20 assessors reported very high inter-rater reliability for measures of area (ICC = 0.84, 95%CI = 0.78-0.89). There are several possible explanations as to why this disparity between previous investigations and the current study occurred.

For one, skill acquisition may play a pivotal role in the application of the MARM. Successful performance of clinical skills is interdependent with prior clinical skill acquisition which involves the repetitive performance of intended cognitive or psychomotor skills as well as specific informative feedback [25]. Raters using the MARM may need to build a repertoire of clinical experiences across the possible range of the measurement tool in order to make an assertive decision on their rating. Furthermore, raters require feedback during the development of experience in order to adjust their process of assessment to a common standard (“calibration”). The process of skill acquisition emphasizes difficulties in the application of the MARM as a practitioner may not simply be able to pick the tool up and use it. Additionally, practitioners who develop new evaluative skills in isolation will not be able to calibrate their findings with other practitioners. Whilst the raters involved in the previous study were experienced in using the MARM [1], raters in this study were trained in the assessment but did not have any experience in utilising it. Raters with clinical experience in working with people with breathing disorders (breathing specialists) demonstrated higher overall reliability (inter and intra-rater) than raters who had no prior experience, which suggests that experience may influence the reliability of the MARM.

Secondly, variability of breathing patterns may result in instability of the assessed variables. In a study of reliability, the tested trait may change independently of, or in response to, the application of the test. Reliability can only be attributed to agreement among raters if the stability of the test variable is known and controlled [26]. The variability of a breathing pattern in terms of rate and depth is innate to a healthy respiratory system as the pattern is continuously adapted to maintain homeostasis [27]. Hence, a characteristic of dysfunctional breathing may be an inability to change. The influences of patient breathing awareness on breathing pattern consistency have been demonstrated to alter spontaneous breathing pattern, mainly the breathing frequency [35]. A lack of stability in breathing pattern would cause discrepancy within and between raters. Raters could be reporting the breathing events accurately but the events are changing between separate ratings. The participants in the previous reliability studies [1, 24] were simulating a given sequence of breathing patterns and were able to reproduce each breathing pattern consistently as shown by analysis of within-subject variance calculated from data recorded by a second objective measure (RIP). It is possible that simulated breathing patterns are clearer in presentation and more consistent, than regular breathing, and thus produce more reliable assessment results. Compared to previous studies [1, 24], this study was intended to be more representative of the clinical situation as symptomatic patients were utilised. However, participants in this study predominantly exhibited none to minor symptoms of breathing dysfunction according to the

SEBQ and NQ scores. Knowledge of the stability of a test variable can only be obtained if there is a reference standard [26]. It is unclear whether the participants in the current study demonstrated stable and consistent patterns of breathing in the absence of reference standard. Currently there is no reference standard that has been shown to coincide with the MARM. Hence, it is not possible to attribute poor reliability to raters or changes in the breathing patterns of the participants

Another potential reason for the disparity of results compared to previous studies is the utilization of blinding. A strength of this study was that contrary to previous reliability studies, raters were blinded to the presence of visual cues that may bias MARM interpretation (for example contraction of accessory breathing muscles, and mouth breathing). A design limitation of previous studies is that the raters may have interpreted their findings according to cues that were not part of the MARM assessment. Visual blinding also potentially minimized the effect of recall bias which is necessary to establish that a retest's findings rely solely on the test procedure and are not influenced by other factors.

The intra-rater reliability was overall higher than the inter-rater reliability especially for the last three variables on the protocol ('deep breathing', 'width breathing' and 'sit easy2'). The first two variables ('sit easy' and 'sit straight') demonstrated poor reliability. Participants may have been able to reproduce the last three breathing events more consistently. A reason for this may be that the 'deep breathing' and 'width breathing' variables both lie at the maximal end of breathing depth and may be easier to reproduce as they are dependent on the maximal inhalation capability rather than the current breathing pattern. Another possible explanation for the higher reliability of the last three variables is that both participants and raters may have calibrated their breathing and method of notation, respectively, from the previous breathing events. Raters were more consistent within themselves but not with other raters. This emphasises the need for raters to calibrate their notation method with other raters.

The second aim was to quantify the correlation between 6 different measurements of breathing function utilised in this study. In line with a previous study by Courtney and Greenwood [28] there was a high positive correlation between the SEBQ and the NQ. Although the two questionnaires measure different aspects of breathing related symptoms [28], these aspects appear to be related to each other as demonstrated by their correlation. This suggests that symptoms associated with the sensation of dyspnoea may commonly be accompanied by symptoms arising from hypocapnia.

The high negative correlation of breath holding time (BHT) with breathing rate (BR) indicates that a short breath holding time may be accompanied by an elevated breathing rate and *vice versa*. On a physiological basis the time between breathing intervals is dependent on the firing threshold of chemoreceptors. When the partial pressure of carbon dioxide in the blood reaches a certain level the need to breathe rises[29]. This threshold is different between individuals [29] and may be indicative of breathing-related symptoms as shown by the high positive correlation between BHT and the SEBQ.

The BHT variable varied considerably between the subjective sensation of first diaphragmatic contraction and the objective recorded first contraction (RIP). Participants almost consistently perceived a longer duration until the first diaphragmatic contraction than the actual recorded first contraction. Should BHT be utilised as a measure in future studies this fact needs to be taken into consideration.

The extent of abdominal excursion (RIP_ABD) during breathing did not correlate significantly with any other variable. Dominance of breathing compartments (upper ribcage and abdominal excursion) have been associated with poor diaphragmatic function[30] and breathing dysfunction[31] and may thus represent important variables in the assessment of breathing dysfunction. Unfortunately, the ratio between the upper ribcage and abdominal excursion during breathing could not be included as a correlate since the appropriate measure (MARM) did not demonstrate reliability.

The inclusion of the factor analysis in this study was intended as a preliminary exploration of the correlations of potential variables indicative of breathing dysfunction for purpose of hypothesis generation. This method of analysis provides a way to identify relationships between multiple variables [34]. Such analysis could be used to identify the relationships between different assessment tools and therefore could contribute to addressing the question regarding which assessment tools may be useful in the diagnosis of breathing dysfunction.

The apparent association between symptoms and signs of breathing dysfunction observed in participants at the maximal ends of breathing symptom scores were not reflected in the factor analysis. An observation from the factor analysis revealed that variables relating to possible signs of breathing dysfunction (BR, BHT, RIP_ABD) were located in a different dimension to variables relating to symptoms of breathing dysfunctions (SEBQ, NQ). This would indicate that breathing rate, breath holding time and breathing pattern may not be directly related to symptoms of breathing.

However, the results of the factor analysis do not hold any significant weight due to the low sample size (n=16). According to Bryant and Yarnold [32] the sample size for factor analysis should be at least five times the number of variables. The subjects-to-variables ratio should be 5 or greater. Furthermore, every analysis should be based on a minimum of 100 observations regardless of the subjects-to-variables ratio. Larger samples tend to minimize the probability of errors, maximize the accuracy of population estimates, and increase the generalizability of the results [33].

6. Conclusion

Raters in this study demonstrated poor inter-rater reliability of the MARM. The intra-rater reliability is higher which means it may be a useful tool for individual practice, where the same rater is involved in the analysis of the results. Due to the subjectivity of the tool it is not likely to be useful in a research setting.

This study has made some initial advances at interpreting the relationship between various potential contributing factors of dysfunctional breathing and breathing symptoms. A drawback of this study is the lack of a reference standard for the MARM in order to test the reliability of the tool when comparing findings to an established measurement tool. Future studies should be directed at exploring the relationships between the various measures of breathing function further in order to establish or dissolve potential links between the possible contributing factors of breathing dysfunction.

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SECTION 3: APPENDICES

Appendix A: The Nijmegen Questionnaire

Nijmegen Questionnaire

A score of over 23 out of 64 suggest a positive diagnosis of hyperventilation syndrome.

	Never 0	Rarely 1	Sometimes 2	Often 3	Very Often 4
Chest pain					
Feeling tense					
Blurred vision					
Dizzy spells					
Feeling confused					
Faster or deeper breathing					
Short of breath					
Tight feelings in chest					
Bloated feeling in stomach					
Tingling fingers					
Unable to breathe deeply					
Stiff fingers or arms					
Tight feelings round mouth					
Cold hands or feet					
Palpitations					
Feeling of anxiety					

Appendix B: The Self Evaluation of Breathing Questionnaire

The Self Evaluation of Breathing Questionnaire (SEBQ): Version 2

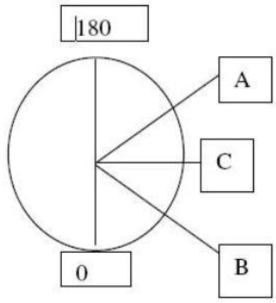
Scoring this questionnaire: (0) never/not true at all; (1) occasionally/a bit true; (2) frequently/mostly true; and, (3) very frequently/very true

1. I get easily breathless out of proportion to my fitness
2. I notice myself breathing shallowly
3. I get short of breath reading and talking
4. I notice myself sighing
5. I notice myself yawning
6. I feel I cannot get a deep or satisfying breath
7. I notice that I am breathing irregularly
8. My breathing feels stuck or restricted
9. My rib cage feels tight and can't expand
10. I notice that I am breathing quickly
11. I get breathless when I am anxious
12. I find myself holding my breath
13. I feel breathless in association with other physical symptoms
14. I have trouble coordinating my breathing when I am speaking
15. I can't catch my breath
16. I feel that the air is stuffy, as if not enough air in the room
17. I get breathless even when I am resting
18. My breath feels like it does not go in all the way
19. My breath feels like it does not go out all the way
20. My breathing is heavy
21. I feel that I am breathing more
22. My breathing requires work
23. My breathing requires effort
24. I find myself breathing through my mouth during the day
25. I breathe through my mouth at night while I sleep

Resource: (Courtney & Greenwood, 2009)

Appendix C: The Manual Assessment of Respiratory Motion

Fig.1. The MARM Graphic Notation



Variables Calculated From MARM Graphic Notation

Variable	Description	Calculation
Area of Breathing	Angle formed between upper line and lower line	Angle A B
Balance	Difference between angle made by horizontal axis (C) and upper line (A) and horizontal line (C) and lower line (B)	AC-CB
Percent rib cage motion	area above horizontal / total area between upper line and lower line x 100	AC/AB X 100

Fig.1.The MARM Graphic Notation(Courtney, Van Dixhoorn et al. 2008).

Instructions on how to use the MARM (Courtney, Van Dixhoorn et al. 2008):

"Sit behind the subject and place both your hands on the lower lateral rib cage so that your whole hand rests firmly and comfortably and does not restrict breathing motion. Your thumbs should be approximately parallel to the spine, pointing vertically and your hand comfortably open with fingers spread so that the little finger approaches a horizontal orientation. Note that the 4th and 5th fingers reach below the lower ribs and can feel abdominal expansion. You will make an assessment of the extent of overall vertical motion your hands feel relative to the overall lateral motion. Also decide if the motion is predominantly upper rib cage, lower rib cage/abdomen orrelatively balanced. Use this information to determine the relative distance from the horizontal line of the upper and lower lines of the MARM diagram. The upper line will be further from the horizontal and closer to the top if there is more vertical and upper rib cage motion. The lower line will be further from the horizontal and closer to the bottom if there is more lateral and lower rib cage/abdomen motion. Finally get a sense of the overall magnitude and freedom of rib cage motion. Place lines further apart to represent greater overall motion and closer for less motion."

Appendix D: QAREL Checklist (Courtney, van Dixhoorn, & Cohen, 2008)

Table 1: Quality Appraisal of Diagnostic Reliability (QAREL) Checklist for the *Evaluation of Breathing Pattern: Comparison of a Manual Assessment of Respiratory Motion (MARM) and Respiratory Induction Plethysmography* study (Courtney, van Dixhoorn, & Cohen, 2008)

Item	Yes	No	Unclear	N/A
1. Was the test evaluated in a sample of subjects who were representative of those to whom the authors intended the results to be applied? (DEF: 3, 4, 5, 7, 8, 9)	☒	☐	☐	
2. Was the test performed by raters who were representative of those to whom the authors intended the results to be applied? (DEF 3, 4, 6, 7, 8, 9)	☒	☐	☐	
3. Were raters blinded to the findings of other raters during the study? (DEF 10)	☒	☐	☐	☐
4. Were raters blinded to their own prior findings of the test under evaluation? (DEF 11)	☐	☐	☐	☒
5. Were raters blinded to the results of the reference standard for the target disorder (or variable) being evaluated? (DEF 12)	☐	☐	☒	☐
6. Were raters blinded to clinical information that was not intended to be provided as part of the testing procedure or study design? (DEF 13)	☐	☐	☐	☒
7. Were raters blinded to additional cues that were not part of the test? (DEF 14)	☒	☐	☐	
8. Was the order of examination varied? (DEF 15, 16)	☐	☐	☒	☐
9. Was the time interval between repeated measurements compatible with the stability (or theoretical stability) of the variable being measured? (DEF 17)	☒	☐	☐	
10. Was the test applied correctly and interpreted appropriately? (DEF 18)	☒	☐	☐	
11. Were appropriate statistical measures of agreement used? (DEF 19, 20, 21)	☒	☐	☐	
TOTAL	7	-	2	2

DEF numbers relate to items on the QAREL Data Extraction Form

Appendix E: QAREL Checklist (Courtney, Cohen, & Reece, 2009)

Table 2: Quality Appraisal of Diagnostic Reliability (QAREL) Checklist for the *Comparison of the Manual Assessment of Respiratory Motion (MARM) and the Hi Lo Breathing Assessment in determining a simulated breathing pattern study* (Courtney, Cohen, & Reece, 2009)

Item	**Yes**	**No**	**Unclear**	**N/A**
12. Was the test evaluated in a sample of subjects who were representative of those to whom the authors intended the results to be applied? (DEF: 3, 4, 5, 7, 8, 9)	☒	☐	☐	
13. Was the test performed by raters who were representative of those to whom the authors intended the results to be applied? (DEF 3, 4, 6, 7, 8, 9)	☒	☐	☐	
14. Were raters blinded to the findings of other raters during the study? (DEF 10)	☐	☐	☐	☒
15. Were raters blinded to their own prior findings of the test under evaluation? (DEF 11)	☐	☐	☒	☐
16. Were raters blinded to the results of the reference standard for the target disorder (or variable) being evaluated? (DEF 12)	☐	☐	☒	☐
17. Were raters blinded to clinical information that was not intended to be provided as part of the testing procedure or study design? (DEF 13)	☐	☐	☐	☒
18. Were raters blinded to additional cues that were not part of the test? (DEF 14)	☒	☐	☐	
19. Was the order of examination varied? (DEF 15, 16)	☒	☐	☐	☐
20. Was the time interval between repeated measurements compatible with the stability (or theoretical stability) of the variable being measured? (DEF 17)	☒	☐	☐	
21. Was the test applied correctly and interpreted appropriately? (DEF 18)	☒	☐	☐	
22. Were appropriate statistical measures of agreement used? (DEF 19, 20, 21)	☒	☐	☐	
TOTAL	7		2	2

DEF numbers relate to items on the QAREL Data Extraction Form

Appendix F: Ethics Approval

Martin Ludwig
35 Puriri Ave
Greenlane
Auckland 1051

20.10.2011

Dear Martin,

Your file number for this application: 2011-1230
Title: The Clinical Utility of the Manual Assessment of Respiratory Motion in People with Breathing Dysfunction.

Your application for ethics approval has been reviewed by the Unitec Research Ethics Committee (UREC) and has been approved for the following period:

Start date: 19.10.2011
Finish date: 19.10.2012

Please note that:

1. **The above dates must be referred to on the information AND consent forms given to all participants.**

2. **You must inform UREC, in advance, of any ethically-relevant deviation in the project. This may require additional approval.**

You may now commence your research according to the protocols approved by UREC. We wish you every success with your project.

Yours sincerely,

pp-

Scott Wilson
Deputy Chair, UREC

cc: Catherine Bacon
Cynthia Almeida

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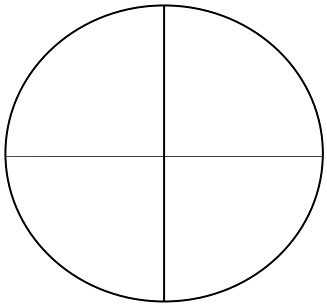
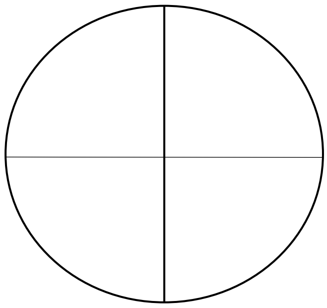
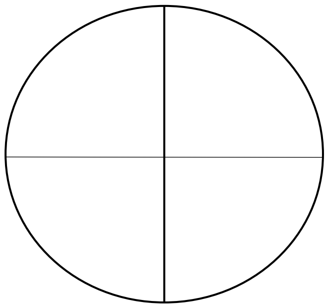
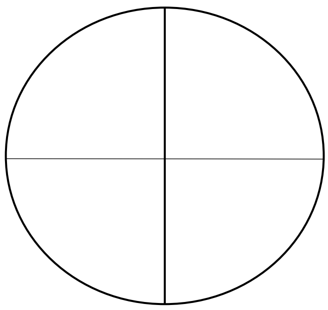
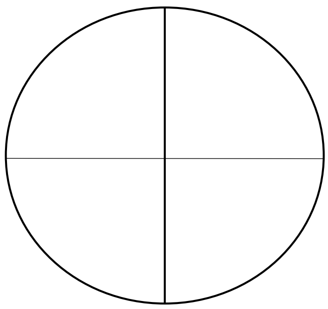
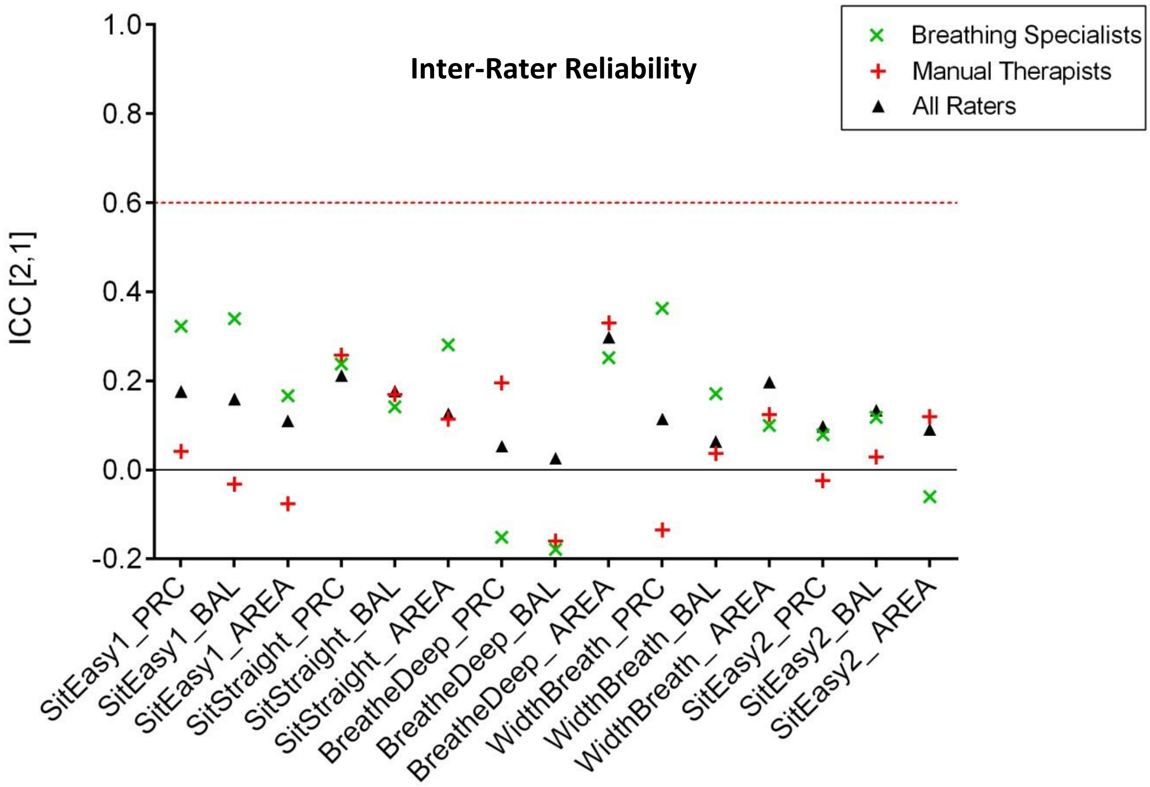
Appendix G: MARM Notation and Testing Protocol

Sit easy *Feet apart, in front of the knees, Hands flat on upper thighs, look straight ahead, rock fwd/back until sitting fully on the sitting bones, bit slump, but don't hang back*		Balance: Area: %RC:
Sit up straight *Sit up straight, less slump* *Keep looking straight ahead*		Balance: Area: %RC:
Breathe deep *Breathe slowly in, and slow out, breathe deep*		Balance: Area: %RC:
Notice width breathing *Participant to focus on pushing rater's hands on back sideways with inhalation, make sure Pt senses sideways expansion/back becoming wider.*		Balance: Area: %RC:
Sit easy *Same as first assessment.*		Balance: Area: %RC:

Appendix H: Inter-Rater Reliability, All Variables



Appendix I: Intra-Rater Reliability, All Variables