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BREATHING DYSFUNCTION

Relationships between measures of dysfunctional breathing in a population with concerns about their breathing

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KEYWORDS

Hyperventilation;
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Summary *Background:* Dysfunctional breathing (DB) is implicated in physical and psychological health, however evaluation is hampered by lack of rigorous definition and clearly defined measures. Screening tools for DB include biochemical measures such as end-tidal CO₂, biomechanical measures such as assessments of breathing pattern, breathing symptom questionnaires and tests of breathing function such as breath holding time.

Aim: This study investigates whether screening tools for dysfunctional breathing measure distinct or associated aspects of breathing functionality.

Method: 84 self-referred or practitioner-referred individuals with concerns about their breathing were assessed using screening tools proposed to identify DB. Correlations between these measures were determined.

Results: Significant correlations were found within categories of measures however correlations between variables in different categories were generally not significant. No measures were found to correlate with carbon dioxide levels.

Conclusion: DB cannot be simply defined. For practical purposes DB is probably best characterised as a multi-dimensional construct with at least 3 dimensions, biochemical, biomechanical and breathing related symptoms. Comprehensive evaluation of breathing dysfunction should include measures of breathing symptoms, breathing pattern, resting CO₂ and also include functional measures such as breath holding time and response of breathing to physical and psychological challenges including stress testing with CO₂ monitoring.

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Introduction

Dysfunctional Breathing (DB) is commonly used to describe disturbances in breathing functionality that impacts on health (Dixhoorn, 1997, 2004; Morgan, 2002; Thomas, McKinley et al., 2005; Prys-Picard and Niven, 2008; Stanton, Vaughn et al., 2008). The definition of DB however is unclear and no gold standards exist to define it. Dysfunctional breathing includes hyperventilation or breathing in excess of metabolic needs but also refers to breathing pattern abnormalities, poor breathing control and presence of breathing symptoms (Dixhoorn, 1997; Morgan, 2002; Warburton and Jack, 2006). Scientists have until recently focused their attention on hyperventilation, which is defined as breathing in excess of metabolic requirements that results in depletion of carbon dioxide (Comroe, 1974). However the importance of hyperventilation and hypocapnia in producing all symptoms associated with DB is disputed (Hornsveld, Garsson et al., 1996; Hornsveld and Garsson, 1997). It has been proposed that DB symptoms may arise as a result of non biochemical breathing dysfunctions or have neurological causes (Howell, 1997). A broader definition of dysfunctional breathing, that considers the multiple functions of breathing may be a more useful way to characterise DB and to determine its prevalence and impact (Dixhoorn, 1997).

Maintenance of normal levels of blood gases such as carbon dioxide is an important, if not the key function of breathing; however breathing has other important functions. Breathing functions in posture and motor control (Lewitt, 1980; McGill, Sharratt et al., 1995; Hodges, Heijnen et al., 2001). It is a key influence on oscillating rhythms that are important for homeostasis, autonomic nervous system regulation and efficient interaction between body systems (Giardino, Lehrer et al., 2000; Bernardi, 2001; Song and Lehrer, 2003). Normal and precisely controlled breathing is also important for voice production and regulation of speech (MacLarnon and Hewitt, 1999; Sivasankar and Erickson, 2009).

Biomechanical, neurological and psychological aspects of breathing are not always tightly linked to biochemical parameters and their other relationships are complex and not adequately understood (Han, Stegen et al., 1996a,b). Attempts to tie the symptoms associated with dysfunctional breathing to only the biochemical dimension i.e., hyperventilation and hypocapnia have not been successful (Burton, 1993; Hornsveld and Garsson, 1997). Physical and psychological causes of breathing dysfunction are often interwoven and can be difficult to separate, however DB is thought to contribute to additional symptoms not adequately explained by the main presenting complaint (Han, Zhu et al., 2004). Research has shown that symptoms associated with dysfunctional breathing are strongly influenced by anxiety and other emotional states and in some cases the psychological influences are primary (Wientjes and Grossman, 1994; Han, Zhu et al., 2004). Other symptoms, particularly various qualities of dyspnea have been linked to breathing pattern abnormalities and poor neuromechanical coupling during breathing (O'Donnell, 2006; Prys-Picard and Niven, 2008). Muscular skeletal dysfunctions, speech and voice problems appear to be predominately linked with dysfunctions of

breathing pattern and neural control of respiration rather than to the body's carbon dioxide status (McGill, Sharratt et al., 1995; Gandevia, Butler et al., 2002).

The accumulation of studies showing the presence of breathing disturbances in highly symptomatic patients and results of research showing that patients with a range of symptoms and medical conditions improve after breathing therapy (Lum, 1975; Grossman et al., 1984; Tweedale, Rowbottom et al., 1994; Han, Stegen, et al., 1996a; Meuret, Rosenfield et al., 2009) lends weight to the importance of assessment and optimisation of breathing functionality in patient care. However, evaluation of dysfunctional breathing is currently hampered by lack of clear measurement guidelines. Measures used by practitioners as screening tools to identify dysfunctional breathing include biochemical measures such as end-tidal CO₂ (Hardonk and Beumer, 1979; McLaughlin, 2009), biomechanical measures such as assessments of breathing pattern (Prys-Picard, Kellett et al., 2004), breathing symptom questionnaires (Thomas, McKinley et al., 2005; Courtney and Greenwood, 2009) and tests of breathing function such as breath holding time (Courtney and Cohen, 2008).

Symptom questionnaires used to evaluate DB

The Nijmegen is the most commonly used questionnaire used to identify DB. (see Figure 1). The 16 item NQ was originally devised to test for HVS and includes 4 questions on respiratory symptoms and the other 12 items on peripheral and central neurovascular or general tension related symptoms (Dixhoorn and Duivenvoorden, 1985a,b). A questionnaire called the Self Evaluation of Breathing Questionnaire (SEBQ) has also been devised to specifically assess respiratory symptoms and breathing behaviours reported to be associated with DB. The SEBQ includes a larger number of respiratory items than the NQ and can differentiate 2 distinct dimensions of breathing discomfort "lack of air" probably related to chemoreceptor derived sensations and "perception of inappropriate or restricted breathing" probably related to the biomechanics of breathing and breathing perception (Courtney and Greenwood, 2009). Normal values for the SEBQ have not been established as yet and this questionnaire is only useful at present for assessing change in breathing symptoms in individuals after treatment. However, it does have potential as a screening tool for DB once further studies are done to validate this instrument. Normal values for the NQ in European studies are generally around a sum score of 10 (Han, Stegen et al., 1996a,b; Han, Stegen et al., 1998; Thomas, McKinley et al., 2005) whereas in China values are lower and average around 5 (Han, Zhu et al., 2004). In categorising individuals as having DB, cut-offs of both 20 and 22 have been found useful (Doorn, Folgering et al., 1982; Dixhoorn and Duivenvoorden, 1985a; Dixhoorn and Hoefman, 1985b).

Clinical measures of dysfunctional breathing pattern

Clinicians usually assess breathing pattern using observation and palpation and historically have used a range of techniques most of which have not been validated (Pryor and Prasad, 2002; Perri, 2007). One component of

Nijmegen Questionnaire

Please tick how often you suffer from the symptoms listed.

	Never 0	Rare 1	Sometimes 2	Often 3	Very often 4
Chest pain					
Feeling tense					
Blurred vision					
Dizzy spells					
Feeling confused					
Faster & 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					
Feelings of anxiety					

Figure 1 Nijmegen Questionnaire. A score of 23/64, or more, is suggestive of hyperventilation. (Vansteenkiste et al., 1991).

breathing pattern that is considered dysfunctional is chronic thoracic dominant breathing at rest. Recently a technique called the Manual Assessment of Respiratory Motion (MARM), which can quantify extent of thoracic dominant breathing as well as other aspects of breathing pattern, has been found to have high levels of inter-examiner reliability and to agree with measures made simultaneously with Respiratory Induction Plethysmography (Courtney, van Dixhoorn et al., 2008). Normal healthy individuals appear to have balanced breathing with relatively equal motion of upper rib cage to lower rib cage abdominal motion. Perfectly balanced breathing gives a MARM value of 0. Normal values for the MARM in this study of 12 yoga teachers and breathing therapy practitioners were around 6. MARM values above 30 can be considered dysfunctional, as they are at least 2 standard deviations above the mean values found in normal healthy individuals (Courtney, van Dixhoorn et al., 2008). Another aspect of breathing pattern considered dysfunctional is the presence of paradoxical or asynchronous breathing (Prys-Picard, Kellett et al., 2004). In paradoxical breathing the belly is drawn in and lower rib cage narrows rather than expands during inhalation. Practitioners generally assess presence of paradoxical breathing simply by asking the patient to breathe in gently, slightly deeply and into the belly while they observe the respiratory phase relationship of chest and belly motion. If the belly moves inward, decreasing its dimensions during inhalation, the breathing is considered to be paradoxical. This simple observation by the practitioner of chest and belly motion sometimes called the Hi Lo breathing assessment has been found to be reasonably accurate for determining different types of simulated breathing patterns including paradoxical breathing (Courtney and Reece, 2009).

Cabon dioxide levels and DB

Persistent low levels of resting carbon dioxide might be expected in individuals with dysfunctional breathing as

evidenced by chronic persistent hyperventilation. However there is considerable argument about what parameters constitute normal values of resting CO₂ and the usefulness of resting CO₂ as a means of identifying individuals with hyperventilation tendencies because the tendency to symptom producing hyperventilation can be intermittent rather than chronic and only become apparent in response to physical or psychological challenge testing (Hardonk and Beumer, 1979; Warburton and Jack, 2006). Some older texts state that levels of carbon dioxide below 37 mmHg indicate hyperventilation (Comroe, 1974) and more recent texts place normal CO₂ levels as above 35 mmHg (Levitsky, 1995). Gardener found that many individuals had CO₂ levels chronically below 35 mmHg with no apparent symptoms until levels were taken below 30 mmHg (Gardner, 1995). In fact he found that mean levels of CO₂ in healthy individuals were around 36.2 mmHg with 2 standard deviations below this level being 32.2 (Gardner, 1995). Regardless of arguments over CO₂ cut-offs it can be concluded that persistently low CO₂ and low CO₂ in response to challenge testing is an aspect of dysfunctional breathing worthy of measurement, particularly as end-tidal CO₂ which fairly accurately represents arterial CO₂ can be easily measured with modern capnometry equipment (McLaughlin, 2009).

Breath holding time and dysfunctional breathing

Breath holding ability is an aspect of breathing functionality that is commonly disturbed in individuals with tendencies to hyperventilation and to dysfunctional breathing (Jack, Darke et al., 1998; Warburton and Jack, 2006). Breath holding time in individuals with chronic idiopathic hyperventilation has been reported to be only 20 s, when held at the end of inhalation, in comparison to normal individuals whose breath holding time is around 60 s when performed according to the same instructions (Jack, Rossiter et al., 2004). Breath holding time differs markedly depending on how it is performed, being affected by whether the hold occurs after inhalation or exhalation and

by the size of the breath taken at the beginning of the breath hold (Mithoefer, 1965). One breath holding time protocol, which uses a somewhat standardized procedure and is used for evaluating and monitoring dysfunctional breathing, is the Buteyko Method technique of the Control Pause. The Control Pause is a post expiratory breath hold and is performed with 2 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). Control Pause levels of below 20 are proposed to indicate the presence of DB and to correlate with resting carbon dioxide levels (Stalmatski, 1999; Stark and Stark, 2002).

Little research has systematically investigated the relationships between biochemical, biomechanical and symptomatic measures of dysfunctional breathing commonly used by clinicians and therapists to evaluate their patients. The small amount of research that does exist tends to suggest that biochemical, symptomatic and breathing pattern aspects of breathing dysfunction do not necessarily co-exist in individuals suspected of having some type of DB. For example disturbances in breathing pattern are not always associated with chronically decreased baseline levels of CO₂ (Han, Stegen et al., 1996a,b; Pine, Coplan et al., 1998; Caldirola, 2004) and changes in breathing pattern and symptoms after breathing therapy may not be accompanied by changes in CO₂ (Han, Stegen, et al., 1996a). In a recent study Meuret found that changes in CO₂ mediate symptoms in patients with panic disorder (Meuret, Rosenfield et al., 2009), however other studies have found that general symptoms believed to be characteristic of dysfunctional breathing may be only mildly related to chronic CO₂ levels or even acute changes in CO₂ (Burton, 1993; Hornsveld and Garsson, 1997). It has been previously reported that Buteyko's Control Pause does not correlate with resting CO₂ levels (Courtney and Cohen, 2008). These observations could imply that breathing pattern, symptoms and carbon dioxide levels reflect distinct aspects of breathing functionality and that dysfunctional breathing might best be characterised as a complex condition with multiple dimensions.

While a combination of measurement tools is sometimes used to evaluate dysfunctional breathing and establish its prevalence in particular populations (Stanton, Vaughn et al., 2008), researchers and practitioners of different breathing therapies may for historical or convenience reasons evaluate only one aspect of breathing function. A number of studies have determined prevalence of DB on the basis of symptom questionnaires alone (Thomas, McKinley et al., 2001; Humphriss, Baguley et al., 2004; Thomas, McKinley et al., 2005) or have emphasized the measurement of carbon dioxide levels (McLaughlin, 2009) or breathing pattern (Perri and Halford, 2004). If measurement in one dimension of breathing functionality proves not to be highly correlated with measurement in other dimension, this may result in incorrect assumptions about prevalence. Research into the relationships between measure of DB will help to clarify what range of measures are needed for comprehensive evaluation of DB. This type of research would help to determine what minimum requirements are needed for comprehensive evaluation of the various aspects breathing dysfunction in the clinical

environment. It would also assist understanding of how best to characterise DB.

Aims of this study

1. To compare prevalence of DB on the basis of a range of measures
2. To evaluate relationships and correlations between various measures of DB

Given the difficulties and lack of consensus on gold standard definitions of DB, we have chosen a pragmatic study design and applied a range of clinically used measures of DB to a population with concerns about their breathing rather than attempting to test a population fitting any particular definition of DB. We believe this approach has greater external validity and applicability for practitioners as it more closely mimics what occurs in "real life" clinical situations.

Method

Participants

Participants were recruited from general practices and complementary medicine clinics in Sydney, Australia. Flyers and brochures were placed in waiting rooms and practitioners received a letter about the study that was described as an investigation into the measurement of "incorrect" and dysfunctional breathing and that it presented an opportunity for individuals to have their breathing assessed.

This attracted individuals with a range of mild medical conditions who had concerns or curiosity about their breathing or who wished to improve their breathing. They had not been comprehensively assessed for presence of dysfunctional breathing, however most subjects came to the study because they were referred or self referred for investigation of dysfunctional breathing. As people generally self refer or are referred for breathing therapies to improve a range of health problems as well as for health optimisation, this sample represents the types of individuals who might use breathing therapies or go to see a therapist for assessment of their breathing.

The 83 individuals who participated in this study were 29 males and 54 females, whose average age was 49 years. They were either healthy or suffered from mild medical conditions including mild asthma. Twenty-nine of these subjects were found to have abnormal spirometry defined by either Forced Expiratory Volume at 1 s (FEV₁) or Forced Vital Capacity (FVC) > 15% below predicted. Descriptive statistics and mean values for the various measures used can be seen in Table 1. Of the 83 participants, 63 (75.9%) had dysfunctional breathing according to at least one measure if the most conservative cut-off for this measure is applied. Number of participants with DB according to each of the individual measures are shown in Table 2.

Measures

Spirometry

Spirometry was performed using a laptop-based spirometer (Spirocard, QRS Diagnostics, Plymouth, MN). The variables

Table 1 Descriptive statistics for all variables ($n = 83$).

	M	SD	Min	Max
FEV ₁ (% predicted)	93	13	58	121
Av. SPO ₂ (% Hb Sat)	96	2	91	100
Av. ETCO ₂ (mmHg)	38	4	26	48
Respiration rate	16	4	7	26
MARM (% RC)	73	27	33	178
MARM Balance	19	18	-20	75
Av. BHT-DD (s)	26	12	11	68
Av. BHT-IRM (s)	30	12	13	72
Nijmegen Questionnaire	18	10	0	51
SEBQ	13	8	0	32

used were Forced Expiratory Volume in 1 s (FEV₁) and Forced Vital Capacity (FVC). Individuals with FEV₁ or FVC < 15% below predicted were classified as having abnormal spirometry.

Oxygen and end-tidal carbon dioxide measurement

End-tidal carbon dioxide (ETCO₂) levels were sampled with a two-pronged nasal canula, and readings were taken with a combined oxymeter and capnometer (BCI, Capnocheck, Waukesha, WI). The equipment was calibrated and checked for accuracy with a known gas mixture. ETCO₂, along with O₂ saturation (SPO₂), respiratory rate, and heart rate, were measured continuously for about 25 min while the person filled out various questionnaires including several that were not related to this study but were used for distraction purposes. The filling out of questionnaire was used to distract participants from excessive attention on their breathing, a source of error that tends to alter breathing rate, volume and pattern. They were advised not to speak and to breath nasally at all times. After excluding data from

the first 2 min to allow for the subject settling in, the average ETCO₂ was calculated and used in determining correlation coefficients with other variables. In classifying individuals as having DB based on low resting ETCO₂, 2 cut-offs were used, 35 mmHg and also 32 mmHg.

The manual assessment of respiratory motion

The Manual Assessment of Respiratory Motion (MARM) is a clinical tool used to assess breathing pattern that has been shown to have clinical utility and validity (Courtney, van Dixhoorn et al., 2008; Courtney and Reece, 2009). This palpatory technique permits the examiner to assess the relative contribution of upper thoracic to lower thoracic and abdominal compartments during breathing and calculate quantitative measures of the balance between these two compartments in breathing. A number of variables can be derived from the MARM procedure, and two of these were used in this study. The first of these is % rib cage motion (MARM % RC) and the second is MARM balance. The examiner using the MARM places their open hands over the subject's back at the region of the lower four to six ribs. The examiner's thumbs are about 1 inch from the spine and oriented vertically. The examiner's hands are spread so that the lower three fingers are oriented in a transverse direction. This hand placement makes it possible for the examiner to feel lateral and vertical motion of the rib cage and assess relative contribution from the upper rib cage and the lower rib cage/abdomen. The examiner draws a diagram with an upper line to represent extent of the upper rib cage and vertical motion and a lower line to represent extent of lower rib cage/abdomen motion. Calculations are then made for thoracic diaphragm "balance" and % rib cage motion (Courtney, van Dixhoorn et al., 2008). Mean measures for

Table 2 Individuals classified as having dysfunctional breathing according to a single measures.

Type of measure	Number in whole sample	% of whole sample ($n = 83$)	Number in abnormal spirometry group	% of abnormal spirometry group ($n = 29$)
<i>Spirometry measures</i>				
FEV ₁ or FVC < 15% below predicted	29	34.1%	29	100%
<i>Nijmegen Questionnaire (NQ)</i>				
NQ 20 or above	29	34.1%	11	37.9%
NQ 23 or above	23	27.1%	9	31%
SEBQ 11 or above	48	56.5%	21	72.4%
<i>Capnometry</i>				
ETCO ₂ -32 mmHg or below	8	9.4%	0	0
ETCO ₂ -below 35 mmHg	22	26%	5	17.2%
<i>Breath holding time</i>				
BHT-DD-20 or below	35	41.2%	21	72.4%
BHT-DD-30 or below	61	71.8%	25	86%
BHT-IRM-20 or below	14	16.5%	11	37.9%
BHT-IRM-30 or below	45	52.9%	23	79%
<i>Breathing pattern</i>				
MARM % rib cage >70%	36	42.4%	15	51.7%
MARM balance > 30	26	30.6%	11	37.9%
Paradoxical breathing	8	9.4%	3	10%

the MARM balance measure were found in this previous study to be around 6 (± 12 Cut-offs of 30 were used to classify individuals with DB on the basis of the MARM balance measure). For the MARM % RC measure, where normals had mean levels of 56 (± 8) a cut-off of 70 was used.

The Hi Lo breathing assessment

This technique involves simple observation by the practitioner of chest and belly motion. The Hi Lo breathing assessment has been found to be reasonably accurate for determining various types of breathing patterns including simulated paradoxical breathing (Courtney and Reece, 2009). In this study patients were asked to slowly and a little bit deeply "breathe into the belly" while the Hi Lo was used to assess the presence of paradoxical breathing.

During the Hi Lo, the examiners hands were placed on the anterior central upper chest and clavicular area (Hi) and the anterior upper abdomen (Lo). From this hand position the examiner determined whether abdominal motion was "paradoxical", i.e., whether it moved inward towards the spine, during inspiration despite the patients attempt to breathe into and expand their belly.

Breath holding time tests

Due to different views on the exact procedure for the Buteyko Control Pause, two breath holding tests were performed (Courtney and Cohen, 2008). In the first, participants held their breath until they experienced a definite sensation of discomfort or recognizable difficulty in holding the breath (BHT-DD). The second involved the time until the first involuntary movement of the respiratory muscles (BHT-IRM). Participants were instructed to sit quietly and breathe normally. They were then asked to breathe gently and at the end of a normal exhalation to pinch their noses and hold the breath. Of these 2 breath holding procedure, the BHT-IRM is likely to be the most reproducible and physiologically stable because involuntary motion of the respiratory muscles has been found to be a more consistent measure of breaking point of breath holding than subjective sensation of the urge to breath (Lin et al., 1974). Measurement was done with a stopwatch that measured to .01 of a second. This number was rounded to .1 of a second. All breath holding procedures were repeated three times. As the procedure did not require complex learning and all participants were able to master this procedure easily, the mean rather than the best result was used in calculating correlations. Prolongation of breath holding time, which can occur when subjects are asked to hold their breath to maximal breaking point (Heath, 1968), did not occur with either of breath holding procedure used in this study. We presume this was because subjects did not hold their breath "as long as they could" and instead followed instructions to only hold until the first muscular impulse (BHT-IRM) or intensification of dyspnea (BHT-DD).

Cut-offs of 20 and 30 were used to classify people as having dysfunctional breathing for both these 2 breath hold protocols. These cut-offs were based on Buteyko Method claims that BHT less than 30 indicated mild dysfunctional breathing (and correlated with resting ETCO_2 of 36 mmHg) and less than 20 indicated more severe dysfunctional

breathing (and correlated with resting ETCO_2 of approximately 32 mmHg) (Buteyko, 1990; Novozhilov, 2010).

The following series of questionnaires were administered and measurements taken.

The Nijmegen questionnaire

The Nijmegen Questionnaire (NQ) is a checklist of symptoms initially believed to reflect hyperventilation syndrome (HVS). It was first developed by van Doorn and colleagues who demonstrated that the test-retest reliability was valid ($r = .87$) (van Doorn, Folgering et al., 1982). The NQ has been demonstrated as able to identify patients (identified by clinicians on the basis of symptoms and observation of breathing behaviours) as suffering from HVS (Dixhoorn and Duivenvoorden, 1985a). Subsequent studies have also shown that the symptoms of the NQ are reproducible by voluntary hyperventilation (Vansteenkiste et al., 1991). In recent years, studies have used the NQ to identify DB as well as hyperventilation syndrome (Thomas, McKinley et al., 2001; Thomas, McKinley et al., 2005). According to van Dixhoorn the mean score for the healthy population is around 11.0 (SD 7.6) (Dixhoorn, 2008). Other studies have found mean NQ scores vary around 5–10 in healthy individuals without DB (Han, Stegen, et al., 1996a; Han, Stegen et al., 1998; Han, Zhu et al., 2004; Thomas, McKinley et al., 2005). Values for the Nijmegen Questionnaire greater than 23 are commonly used to signify DB (Dixhoorn and Duivenvoorden, 1985a). However in one study in a physiotherapy practice comparing patients with a clear musculoskeletal diagnosis with those with dysfunctional breathing, a cut-off score of 20 proved adequate to classify 88% of patients (Dixhoorn and Hoefman, 1985).

The self evaluation of breathing questionnaire (SEBQ)

This questionnaire was compiled from various sources. Its items were derived after considering symptoms proposed by Burton, Howell, Fried and other literature to be discriminative for DB, discussion with colleagues, relevant clinical experience of the author and from a public domain Internet questionnaire titled "How Good is your Breathing Test, Take our Free Breathing Test and See" (HGYB)(White, 2005). No item of the SEBQ was taken directly from any single source, most items were included because they were suggested by several sources and appeared plausible (Courtney and Greenwood, 2009) (Figure 2).

Procedure

The Human Research Ethics Committee of RMIT University approved the study. All data collection was completed within a single two-hour visit. The participants were given the series of questionnaires to fill out while attached to a capnometer and pulse oxymeter. While the participants were either reading or filling out the questionnaires the following respiratory parameters were measured, end-tidal CO_2 , oxygen saturation, respiratory rate and heart rate. These respiratory parameters were measured over approximately 25 min. A single examiner (RC) then assessed breathing pattern by performing the Manual Assessment of

Self Evaluation of Breathing Questionnaire

	0 Never or not true at all	1 occasionally a bit true	2 frequently mostly true	3 very frequently – very true
I get easily breathless on physical exertion out of proportion to my fitness	0	1	2	3
I get breathless even when resting	0	1	2	3
I get breathless when I am anxious	0	1	2	3
I get short of breath or very tired when reading out loud or talking a lot	0	1	2	3
I feel breathlessness in association with other physical symptoms	0	1	2	3
I feel that the air is stuffy, as if there is not enough air in the room.	0	1	2	3
I feel I cannot get a deep or satisfying breath	0	1	2	3
I can't catch my breath	0	1	2	3
My breathing feels stuck, restricted	0	1	2	3
I Feel that my ribcage is tight and can't expand.	0	1	2	3
My clothing often feels too tight or uncomfortable around my chest.	0	1	2	3
I sigh, yawn or gasp.	0	1	2	3
I find myself holding my breath at various times	0	1	2	3
I notice myself breathing shallowly using my upper chest and shoulders.	0	1	2	3
I notice myself breathing quickly.	0	1	2	3
I notice myself mouth breathing	0	1	2	3
I have trouble co-ordinating my breathing when I am speaking	0	1	2	3
I notice myself breathing irregularly.	0	1	2	3

Figure 2 Breathing self-evaluation questionnaire.

Respiratory Motion (MARM) (Courtney, van Dixhoorn et al., 2008). Following assessment of breathing pattern, breath holding times were tested. The final procedure performed was spirometry with participants asked to perform three forced respiratory manoeuvres and the best result used to minimize effects of technique error.

Results

As can be seen in Table 1, the mean levels for all dysfunctional breathing measures are not particularly high for this group, however means for the NQ, (18) and the

MARM (19) are higher and more dysfunctional than those found in studies of normal individuals. In individuals with normal breathing, mean values for sum scores of the NQ are around 10 (Han, Stegen, et al., 1996a; Han, Stegen et al., 1998; Thomas, McKinley et al., 2001) and for the MARM are around 6 (Courtney, van Dixhoorn et al., 2008).

The majority of individuals fit criteria of having dysfunctional breathing on the basis of at least one proposed DB measure. Of the 83 participants, 63 (75.9%) had dysfunctional breathing according to at least one measure using the most conservative cut-off criteria for measures such as the NQ, BHT and ETCO₂ that had 2 possible cut-offs. Using the less stringent cut-offs for the

Table 3 Descriptive statistics and differences between the normal and abnormal spirometry groups.

	Normal spirometry (n = 54)	Abnormal spirometry (n = 29)	Mean difference	P Values
Males	17	12		
Females	37	17		
Average age (years)	49(±13)	48 (±14)		
FEV ₁ (% predicted)	99(±10)	82(±11)	17	.0001
Av. ETCO ₂ (mmHg)	37(±4)	39(±4)	2	.01
Av. BHT-DD (s)	28(±12)	20(±8)	7.8	.0001
Av. BHT-IRM (s)	33(±11)	24(±10)	9	.0001
Av. SPO ₂ (% Hb Sat)	96(±2)	95(±2)	1	.04
Resp. Rate	16(±4)	17(±4)	1	.79
Nijmegen Questionnaire	17(±9)	19(±11)	2	.35
MARM (% RC)	70(±21)	77(±35)	7	.29
MARM Bal.	19(±16)	(19±18)	.9	.82
SEBQ-total	12(±7)	16(±8)	4.5	.008

NQ, (>20) and the two types of BHT (<30), 66 of the 83 participants (79.5%) might have been classified by some practitioners as having dysfunctional breathing. This is illustrated in Table 2.

There were 29 individuals who had FEV₁ or FVC < 15% below predicted. Mean values of the proposed DB measures were compared between the group with normal spirometry and those with normal spirometry; these are shown in Table 3. The group with abnormal spirometry had higher CO₂ level and lower O₂ levels, and therefore were not more prone to hyperventilation. NQ, MARM values and respiratory rates were approximately equal in normal and abnormal spirometry groups. The only increased signs of DB in the abnormal spirometry group were, shorter BHT and increased SEBQ scores.

Correlations between measures are shown in Table 4. As can be seen, significant correlations were found within the following categories of measures. 1. Symptom questionnaires; 2. Breath holding times; and, 3. Breathing pattern

measures. Scores for the two questionnaires, the NQ and the SEBQ, were strongly correlated. The two types of breath holding, BHT-DD and BHT-IRM, were also strongly correlated. The two MARM variables, MARM balance and % RC were also correlated.

Correlations between variables in different categories were generally not significant. The only significant correlation was between BHT-IRM and MARM % rib cage motion. And also FEV₁ levels and SEBQ sum scores were correlated.

Carbon dioxide, measured in this study by end-tidal CO₂, did not correlate significantly with symptom questionnaires, breathing pattern, PO₂ or FEV₁. A statistically significant, but weak, correlation was found with one type of breathing holding, BHT-DD. It should be noted that this correlation was negative, opposite to the expected direction.

There was a correlation between BHT-DD and SpO₂ levels.

The SEBQ was negatively correlated with FEV₁. FEV₁ also correlated with both types of breath holding time.

Table 4 Correlation matrix.

	Biochemical measures		Biomechanical measures			Breath holding time		Symptom questionnaires	
	PO ₂	ETCO ₂	RR	MARM % RC	MARM bal.	BHT-DD	BHT-IRM	NQ	SEBQ
FEV ₁	.10	-.08	-.09	-.19	-.08	.31	.31	-.133	-.264
	<i>p</i> = .38	<i>p</i> = .47	<i>p</i> = .44	<i>p</i> = .09	<i>p</i> = .43	<i>p</i> = .004	<i>p</i> = .005	<i>p</i> = .23	<i>p</i> = .02
PO ₂	1	-0.23	-.10	-.14	-.12	.24	.186	.11	.02
		<i>p</i> = .84	<i>p</i> = .10	<i>p</i> = .22	<i>p</i> = .26	<i>p</i> = .03	<i>p</i> = .09	<i>p</i> = .26	<i>p</i> = .80
ETCO ₂		1	-.14	-.02	-.136	-.241	-.198	-.12	.01
			<i>p</i> = .20	<i>p</i> = .82	<i>p</i> = .221	<i>p</i> = .03	<i>p</i> = .07	<i>p</i> = .27	<i>p</i> = .96
RR			1	.09	.07	-.18	-.16	-.20	-.14
				<i>p</i> = .40	<i>p</i> = .55	<i>p</i> = .11	<i>p</i> = .14	<i>p</i> = .08	<i>p</i> = .22
MARM% RC				1	.82	-.090	-.25	-.04	.11
					<i>p</i> = .0001	<i>p</i> = .42	<i>p</i> = .02	<i>p</i> = .70	<i>p</i> = .31
MARM bal.					1	-.07	-.204	.01	.14
						<i>p</i> = .54	<i>p</i> = .06	<i>p</i> = .90	<i>p</i> = .20
BHT-DD						1	.84	-.20	-.17
							<i>p</i> = .0001	<i>p</i> = .07	<i>p</i> = .12
BHT-IRM							1	-.18	-.20
								<i>p</i> = .10	<i>p</i> = .07
NQ								1	.75
									<i>p</i> = .0001

Discussion

Strict definitions of DB are not possible at present, but for practical purposes it is probably most useful not to think of DB as single entity limited to the biochemical dimension of breathing functionality (as in Hyperventilation Syndrome) but to consider breathing symptoms and breathing pattern as potentially separate aspects of DB which need to be measured in their own right. This study found that significant correlations exist only within categories of breathing measures but not between categories, showing there was no consistent linear relationship between categories of measures. The two symptoms questionnaires investigated in this study, the NQ and the SEBQ were found to correlate, as did the 2 measures of breath holding time and the 2 similar MARM measures of breathing pattern. Biochemical measures, oxygen and carbon dioxide, were not related. Therefore the clinician who wishes to do a comprehensive evaluation of breathing functionality should consider using a range of measurement tools, including breathing symptom questionnaires, breathing pattern evaluation and CO₂ measurement.

In this sample of people, which on average had only mild dysfunctional breathing, there was very little relationship between the categories of measures. The only significant correlation between categories found, was between one type of breath holding, (BHT-IRM) where breath was held until first involuntary motion of the respiratory muscles and extent of thoracic dominant breathing pattern, represented as percentage of upper rib cage contribution to breathing motion (MARM % rib cage). No measures were found to correlate with end-tidal CO₂. This indicates that individuals with dysfunctions in one aspect of breathing functionality do not necessarily have dysfunctions in other aspects, particularly if the breathing dysfunction is not severe.

The most well recognised form of dysfunctional breathing is hyperventilation which is strictly defined by a biochemical definition i.e., breathing in excess of metabolic requirements so that a depletion of carbon dioxide occurs. Despite its biochemical criteria, hyperventilation is sometimes presumed to exist on the basis of symptoms, on findings of abnormal breathing patterns or length of breath holding time (Lum, 1976a,b; Dixhoorn and Duivenvoorden, 1985a; Stark and Stark, 2002). This study suggests that dysfunctions of breathing pattern, shortened breath holding time and DB symptoms can exist without chronic hypocapnia. This is consistent with previously published studies that have also found that baseline CO₂ does not always relate to symptoms of dysfunctional breathing (Folgering and Colla, 1978; Vansteenkiste et al., 1991). However the literature also shows that individuals with high levels of hyperventilation type symptoms do have depressed levels of CO₂ when compared to symptom free controls (Gardner et al., 1986). So while it cannot be presumed that there is no association between symptoms of DB and resting CO₂, it should be recognised that the relationship is complex and can be influenced by other moderating factors such as anxiety. This was demonstrated by Wintjes and Grossman who showed that in a group of 83 healthy individuals CO₂ contributed only 4% of the variance in HV symptoms (Wientjes and Grossman, 1994). Our finding that dysfunctional breathing pattern is not

necessarily associated with low resting CO₂ levels is also consistent with the literature as other studies have also found that symptomatic individuals with disturbed breathing pattern often have normal levels of CO₂ (Han, Stegen et al., 1996a,b). Again it cannot be presumed that there is no association between breathing pattern and CO₂ levels, as many clinicians and researchers have observed abnormal breathing pattern in individuals with hyperventilation (Lum, 1976a,b). However it can be concluded that the level of resting CO₂ cannot be presumed from presence of symptoms commonly thought to be associated with dysfunctional breathing, from breath holding time or from breathing pattern. The clinician can only be certain of CO₂ levels if these are specifically measured. It should also be noted that measurement of resting CO₂ might not identify individuals who hyperventilate in response to psychological stress or physical exercise or whose CO₂ regulating capacities are compromised. This is done variously through capnometry combined with exercise or psychological challenge or the Hyperventilation Provocation Test (Hardonk and Beumer, 1979; Warburton and Jack, 2006).

In this study the MARM measures reflecting thoracic dominance in breathing shown a small degree of correlation with breath holding till first desire to breathe (BHT-DD). It seems reasonable to hypothesise that this may be because both these measures reflect respiratory drive, with increased respiratory drive increasing extent of thoracic breathing and decreasing breath holding time. It is also possible that thoracic breathing patterns themselves affect perception of dyspnea sensations related to the breaking point of breath holding.

This study did not show an association between general symptoms of dysfunctional breathing, as measured by the SEBQ or the NQ and breathing pattern. These findings are unexpected and not consistent with clinicians observations that individuals with high levels of symptoms associated with DB tend to have breathing pattern disturbances (Lum, 1976a,b; Howell, 1997). And that specific qualities of breathing discomfort or dyspnea are affected by patterns of respiratory muscle use (Simon and Schwartzstein, 1990; Altose, 1992; O'Donnell, 2006). The lack of relationship in this current study may be due to the fact that the sample used tended to represent individuals with only mild signs of dysfunctional breathing. Correlations not evident in this sample with mild DB might be stronger in individuals with more severe DB. Also relationships that do exist between breathing pattern and symptoms may be non-linear.

Another factor contributing to the poor correlation found between symptoms and actual physiological and biomechanical aspects of breathing, such as end-tidal CO₂ breathing pattern and FEV₁ might be the individuality of cognitive processes involved in recognizing, attending to, and then evaluating physical symptoms. Certain individuals show persistent tendencies to over-perceive or under-perceive symptoms related to breathing function (Teeter and Bleeker, 1998; Klein, Walders et al., 2004). Thus, while the findings of this study suggest that symptoms do not necessarily correlate with biochemical or biomechanical or breath holding measures across individuals, they do not diminish possibility or importance of relationships between symptoms and breathing functions within individuals.

Limitations of this study

Not all ways of measuring DB were assessed in this study. For example measurement of carbon dioxide at rest, as was undertaken during this study, will only reveal chronic persistent hyperventilation and may not be adequate for revealing which individuals are prone to intermittent hyperventilation in response to physical or psychological stress (Hardonk and Beumer, 1979).

The interpretation of these results is limited to their use as general screening tools, as the patient population used in this study was not representative of individuals with pronounced breathing dysfunction. Relationships not evident in this population might be evident in a sample more representative of individuals with hypocapnia, markedly disturbed breathing pattern or abnormal Nijmegen Questionnaire scores. Representation from patients in disease categories thought to be affected by dysfunctional breathing is needed to make more robust assumptions.

Future research

Further Research is required to investigate the presence of more complex relationships between the various dimensions of breathing dysfunction. Research should also target individuals with stronger evidence of breathing dysfunction or with specific ailments.

Conclusion

For practical purposes DB is probably best characterised as multi-dimensional. DB can occur in at least 3 dimensions: biochemical, breathing pattern and breathing related symptoms and these might not co-exist. Screening for DB with measures representing only one of these dimension may not lead to realistic estimations of the prevalence and impact of the various types of breathing dysfunctions. Comprehensive evaluation of breathing dysfunction should include measures of breathing symptoms, breathing pattern, resting CO₂ and also include functional measures such a breath holding time and response of breathing to physical and psychological challenges.

Conflict of interest statement

None of the authors of this manuscript shall derive any personal profit or gain, directly or indirectly, by reason of his or her authorship of this manuscript.

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