Medically Unexplained Dyspnea: Partly Moderated by Dysfunctional (Thoracic Dominant) Breathing Pattern

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Background. Dysfunctional breathing (DB) may contribute to disproportionate dyspnea and other medically unexplained symptoms. The extent of dysfunctional breathing is often evaluated using the Nijmegen Questionnaire (NQ) or by the presence of abnormal breathing patterns. The NQ was originally devised to evaluate one form of dysfunctional breathing - hyperventilation syndrome. However, the symptoms identified by the NQ are not primarily due to hypocapnia and may be due to other causes including breathing pattern dysfunction. Objectives. The relationships between breathing pattern abnormalities and the various categories of NQ symptoms including respiratory or dyspnea symptoms have not been investigated. This study investigates these relationships. Method. 62 patients with medically unexplained complaints, that seemed to be associated with tension and breathing dysfunction, were referred, or self-referred, for breathing and relaxation therapy. Dysfunctional breathing symptoms and breathing patterns were assessed at the beginning and end of treatments using the NQ for assessment of DB symptoms, and the Manual Assessment of Respiratory Motion (MARM) to quantify the extent of thoracic dominant breathing. Subscales for the NQ were created in 4 categories, tension, central neurovascular, peripheral neurovascular and dyspnea. Relationships between the NQ (sum scores and subscales) and the MARM were explored. Results. Mean NQ scores were elevated and mean MARM values for thoracic breathing were also elevated. There was a small correlation pre-treatment between MARM and NQ (r = 0.26, p = 0.05), but classification of subjects as normal/abnormal on both measurements agreed in 74% (p < 0.001) of patients. From the sub scores of NQ only the respiratory or ‘dyspnea’ items correlated with the MARM values. Dyspnea was only elevated for subjects with abnormal MARM. After treatment, both MARM and NQ returned to normal values (p < 0.0001). Changes in NQ were largest for subjects with abnormal MARM pre-treatment. There was a large interaction between the change in the NQ sub score dyspnea and initial MARM values. (p < 0.001).

Keywords dysfunctional breathing, breathing pattern, thoracic breathing, dyspnea, breathing therapy

INTRODUCTION

Dysfunctional breathing (DB) is thought to exist in patients with asthma although it is not well defined (1). DB is proposed to have several dimensions related to biomechanical, biochemical, and other psychophysiological functions of breathing (2). In the clinical and research setting, patients have been categorized as having DB on the basis of symptom scores from the Nijmegen Questionnaire (NQ) (3) and also on the basis of abnormalities of breathing pattern (4).

The causes of DB symptoms measured by the NQ are not well understood. Originally, NQ symptoms were thought to be primarily due to hyperventilation and hypocapnia, but recent findings have shown that this is not the case for all symptoms (5). Central and peripheral neurovascular symptoms, such as tingling and numbness, can be linked to abnormalities of CO2 levels (6); however, respiratory complaints appear to have a stronger relationship to breathing pattern (7, 8).

The NQ contains three subscales of symptoms that relate primarily to either respiratory, peripheral neurovascular or central neurovascular dimensions, and a fourth dimension that contains symptoms commonly associated with tension (9). These four symptom dimensions may arise from different pathophysiological and/or psychological causes.

Alterations in breathing pattern are often reported in individuals with functional breathing disorders (10, 11). Shallow upper chest or thoracic dominant breathing is one type of breathing pattern that has been observed in people with DB. Lum describes this breathing as “heaving of the upper sternum and lack of lateral costal expansion” (10). Thoracic dominant breathing is a normal response to certain conditions—change to more upright posture, sudden increases in the ventilation needs, rapid inspiration, and increased respiratory drive (12, 13). However, normal healthy individuals tend to have “balanced” breathing at rest with relatively equal contributions from upper chest/thoracic and lower chest/abdominal compartments (14). In patients without somatic disease, the presence of habitual upper chest/thoracic breathing is considered dysfunctional. This type of breathing pattern is commonly found in patients with respiratory and cardiac disease and is thought to be due to increased respiratory drive and subsequent increased neural output to inspiratory muscles, which can become shortened and hypertonic (15−17). Upper chest breathing might sometimes be helpful in patients with lung or heart disease but...
might also disadvantage the operating conditions of respiratory muscles, contribute to hyperinflation, and increase patient’s perception of respiratory difficulty and dyspnea (18, 19).

In asthmatics, DB in the form of hyperventilation or breathing pattern disorders may contribute to dyspnea and other symptoms. Asthmatics have been found to have higher scores on the NQ than non-asthmatics (3). Breathing retraining in asthmatics has been found to reduce NQ scores and to improve asthma control (20–22). The reason for the reduced symptoms in asthmatics after breathing therapy is not clear. It is proposed that breathing techniques might improve asthma because they increase carbon dioxide levels, which may be low due to the hyperventilation tendencies that exist in some asthmatics (23, 24). Some breathing therapies such as the Buteyko Breathing Technique and capnometry-assisted biofeedback specifically aim to raise CO2 levels as a means to improving asthma (22, 25). There is some evidence that normalization of CO2 levels by capnometry is beneficial in asthma (26), although it is unlikely that this is the sole mode of action of Buteyko Breathing Technique (27, 28). Dysfunctional patterns of breathing have also been proposed to contribute to exaggerated dyspnea in asthmatics (29), and the presence of abnormal breathing patterns is thought to provide one possible means of identifying asthmatics who might benefit from breathing therapy (4).

Instrumentation for measuring breathing pattern includes respiratory induction plethysmography (RIP) and more recently optical electrical plethysmography; however, clinicians often use observation and palpation (30, 31). One recently validated clinical tool is the Manual Assessment of Respiratory Motion (MARM) (14). The MARM has high inter-examiner reliability and was found to be able to evaluate more dimensions of rib cage motion than RIP. This is because the MARM includes the vertical motion of the upper rib cage in the calculation of thoracic dominance variables. The RIP, by contrast, only considers the lateral expansion of the upper thoracic relative to the lower thoracic/abdominal compartments.

To our knowledge, no studies have systematically explored the relationship between NQ and breathing pattern. In addition, although it has been shown that breathing therapy, which reduces patient’s symptoms, may also improve breathing pattern (32). The specific effects on thoracic breathing have not been investigated. These considerations have led to the questions addressed in this study:

- What types of relationships exist between breathing pattern (MARM) and DB symptoms (NQ) in patients before treatment?
- Do breathing pattern and the DB symptoms respond similarly to treatment?
- Does breathing pattern abnormality (thoracic dominance) affect the response of NQ scores to breathing therapy?

**Method**

**Participants**

The participants were consecutive patients receiving treatment with a method of breathing and relaxation therapy called whole body breathing (WBB) in the private practice of one of the authors (ELMA) from January 2008 to May 2009. There were 62 patients—16 men and 46 women, aged on average 39 (±16) years and treated for 5.9 (±1.5) individual sessions of 1 hour. Patients were referred by medical practitioners, psychologists, or self-referred. They suffered from a range of complaints, all classified as “functional” or “medically unexplained.” Patient data used in this study were conditional on the patient’s consent that their data, in anonymous form, could be used for research purposes.

**Materials/Measures**

**The Manual Assessment of Respiratory Motion.** The Manual Assessment of Respiratory Motion (MARM) was used to quantify the extent of thoracic dominance in breathing. The MARM procedure, which has been described previously (14), allows the examiner to record a graphic representation of their impression of the direction and relative dominance of upper rib cage motion to lower rib cage/abdomen motion. The MARM balance measure has been demonstrated to have good inter-examiner reliability and to be consistent with measurements performed using RIP (14). In this previous study, by Courtney and van Dixhoorn, balance values were on average 6 ± 12 and in another study of 16 normal individuals values were similar at 1.6 ± 15 (unpublished data). Thus, as a rather strict criterion for “thoracic dominance,” we choose a value of MARM balance of 30 as a cutoff as this represents a value more than 2 standard deviations from the theoretical optimal value of 0 and from the mean found in normal individuals.

**Nijmegen Questionnaire.** The NQ was originally developed as a symptom checklist to identify persons with hyperventilation syndrome (9). It 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. Test–retest reliability of NQ is excellent \((r = 0.87; p < .01)\). A cutoff 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 from those with a negative hyperventilation provocation test (33). However, because the provocation test is no longer a valid gold standard, we use a lower value of 20 as the cutoff criterion as a result of subsequent studies, which showed that this score was best able to differentiate hyperventilation patients from normals (34) (Dixhoorn and Courtney, NQ revisited—submitted for publication).

The 16 items correspond to the classical hyperventilation symptoms of dyspnea, dizziness, and paresthesias, in addition to symptoms of tension and anxiety. On the basis of principal components analysis (9), as well as the actual content, we calculated four subscores. We were
particularly interested in the four respiratory items and grouped them in one subscore “dyspnea” (range 0–16). The criterion value of abnormality for this subscore was calculated as 4/16 of the value of 20, which is equal to a cutoff value of 5. The items reflecting tingling, stiffness, and cold extremities were grouped in the subscore “Peripheral.” The items reflecting blurred vision and dizziness were grouped in the subscore “Central.” The items referring to anxiety, tension, palpitations, and chest pain were grouped in the subscore, labeled “tension.”

**The Intervention: Whole Body Breathing**

Whole Body Breathing (WBB) is a breathing and relaxation protocol. It is individualized so that modifications to the protocol are made according to the patients’ response. It includes a repertoire of approximately 50 instructions using movement, attention and breathing, manual techniques, and talking about the patient’s experiences (35). Central to the therapy is training in self-regulation of techniques, and talking about the patient’s experiences (35). By contrast, a thoracic dominant breathing pattern all subjects with high NQ dyspnea scores breathe thoracically. In the scatter plot (Figure 2), it is remarkable that only two subjects with normal MARM had high dyspnea scores. In the scatter plot (Figure 2), the upper left quadrant is practically empty, implying that all subjects with high NQ dyspnea scores breathe thoracically. By contrast, a thoracic dominant breathing pattern

**RESULTS**

**MARM and NQ Pretreatment**

At the beginning of treatment, mean NQ scores and MARM balance scores were substantially elevated and the majority of patients had NQ scores and MARM levels above the chosen cutoffs (Table 1). For the average NQ subscores, the score on tension was the only one with a value above the cutoff score of 5.

<p>| Table 1.—Pretreatment values for NQ total, NQ subscale scores, and MARM balance. |
|---|---|---|</p>
<table>
<thead>
<tr>
<th>n</th>
<th>M (SD)</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>MARM balance</td>
<td>62</td>
<td>44.27 (20.38)</td>
</tr>
<tr>
<td>NQ total</td>
<td>62</td>
<td>2277 (8.32)</td>
</tr>
<tr>
<td>NQ dyspnea</td>
<td>40</td>
<td>4.93 (3.61)</td>
</tr>
<tr>
<td>NQ peripheral</td>
<td>40</td>
<td>4.25 (2.60)</td>
</tr>
<tr>
<td>NQ central</td>
<td>40</td>
<td>3.90 (2.48)</td>
</tr>
<tr>
<td>NQ tension</td>
<td>40</td>
<td>7.00 (2.99)</td>
</tr>
</tbody>
</table>

Table 2 shows that classifications of normal or abnormal on both instruments were congruent for 46 (74%) individuals (Fisher’s exact test: $p = .009$). Some individuals had abnormal MARM ($n = 12$) and showed thoracic dominance in breathing pattern, but had normal NQ and few complaints. A minority of individuals ($n = 4$) had normal MARM and balanced breathing movement, but abnormal NQ and elevated complaints.

**Relationships Between MARM and NQ Scores**

At pretreatment there was a weak, but significant, correlation between total NQ scores and MARM balance scores in this clinical sample—$r (60) = 0.26, p = .04$. NQ subscale scores showed only significant correlation for dyspnea—$r (38) = 0.32, p = .04$. The score on tension approached significance—$r (38) = 0.30, p = .06$. Figure 1 shows that individuals with abnormal MARM did not have significantly different scores for NQ peripheral or NQ central subscale scores, but had significantly higher scores for tension ($p = .03$) and dyspnea ($p = .002$).

Classification as normal or abnormal on the MARM ‘balance’ score and the NQ subscore dyspnea was congruent for 65% of subjects: normal MARM and normal dyspnea: $n = 9$; abnormal MARM and abnormal dyspnea: $n = 17$; normal MARM and abnormal dyspnea: $n = 2$; abnormal MARM and normal dyspnea: $n = 12$. The association between MARM and dyspnea classifications was statistically significant (Fisher’s exact test: $p < .03$). It is remarkable that only two subjects with normal MARM had high dyspnea scores. In the scatter plot (Figure 2), the upper left quadrant is practically empty, implying that all subjects with high NQ dyspnea scores breathe thoracically. By contrast, a thoracic dominant breathing pattern

![FIGURE 1.—Comparison of NQ subscores according to MARM category.](image-url)
can be present without the presence of dyspnea symptoms (lower right quadrant).

**Normalization of MARM and NQ after Treatment**

The response of MARM and NQ to breathing and relaxation therapy was positive (Table 3): average MARM and NQ scores (both total and subscores) greatly improved after treatment by WBB \((p < .0001)\) and moved into the normal zone.

Cross tabulating normal and abnormal MARM and NQ scores, before and after treatment, showed that of 43 cases with initial abnormal MARM only 3 remained abnormal after WBB and no one became abnormal during treatment.

Of 34 cases with elevated initial NQ only 4 remained abnormal and in 1 subject NQ became elevated. After treatment, there was no participant with both elevated NQ and thoracic dominance. Thus, treatment was effective in normalizing DB.

The nature of the response to treatment was further studied by calculating correlations between the changes in the measurements. Changes in MARM balance were only correlated with changes in NQ dyspnea but not with changes in NQ total or other NQ subscale scores (Table 4). However, changes in NQ dyspnea also correlated with changes in NQ tension and NQ total, implying that dyspnea was also related to tension and possibly to other factors.

The final question was whether thoracic dominance moderates the effect of treatment on complaints. A two-factor, mixed-design ANOVA was conducted to compare NQ total scores between those with normal and abnormal MARM (between subjects factor) before therapy, at the beginning and at the end of therapy (within subjects factor). The ANOVA yielded three \(F\)-values that examined the main effect for the MARM group, the main effect for TIME, and the INTERACTION between the MARM group and TIME. The results are shown in Table 5.

For NQ total, main effects of both MARM group and TIME were highly significant and the interaction just reached significance. NQ scores were higher in the abnormal MARM group than the normal MARM group before therapy. After therapy, NQ total scores decreased in both groups, but more in the abnormal MARM group resulting in the groups having similar NQ total scores after therapy.
Both MARM and NQ scores returned to normal values. After therapy, no subject was classified as having DB on the basis of these two variables. However, it appeared that the breathing pattern only moderated dyspnea items and not other NQ subscales. By implication, treatment effect on all other NQ subscales was not due to the pattern of breathing movement but to other psychological or physiological processes. These processes, which are also targeted by WBB, include psychophysiological self-regulatory processes such as mental or physical relaxation, body awareness, and cognitive restructuring (36).

The single largest reduction of symptoms occurred in NQ subscale “tension” which includes items relating to tension, anxiety, heart palpitations, and chest pain. Reduction in these symptoms was not mediated by breathing pattern and it may have been due to other respiratory variables like CO2 level (37), or to various physical and psychological effects of relaxation (38, 39).

To our knowledge, this is the first study that documents the importance of thoracic dominant breathing pattern for the experience of medically unexplained dyspnea. It needs to be emphasized however that the association is found in patients without a somatic cause for dyspnea. The relationship of breathing pattern to dyspnea perception may be different in patients with respiratory or cardiovascular disease. In patients with chronic obstructive pulmonary disease presence of rapid and shallow pattern of breathing, a mode of breathing that is also usually thoracic dominant was found to correspond with extent of dyspnea (40). However, the authors of this study suggest that this mode of breathing is an adaptive response that might actually assist in reducing excessive respiratory drive. Preliminary data on 33 patients with chronic obstructive pulmonary disease showed they had thoracic dominant breathing and a higher score on MARM balance (27.8 ± 42.4) than normal individuals; however, there was no relationship with grading of disease severity (Gold1–4) (41). There are no MARM data yet on patients with asthma or cardiovascular disease. However, there is some evidence that abnormal breathing patterns occur in individuals with these diseases. In asthmatic patients, exaggerated dyspnea and unexplained complaints have been attributed to “dysfunctional breathing” (4, 8, 21, 42, 43), although only few of these studies include data on breathing pattern (8). In asthma, even mild broncho constriction has been found to produce shortening and hypertonicity of inspiratory muscles and these muscle changes would tend to favor thoracic dominant breathing (17, 18). Patients with heart failure often complain of dyspnea and this has been attributed in part to “inefficient pattern of rapid shallow respiration” (44). Assessment of MARM within these populations will be necessary to establish values of MARM balance, both before and after treatment. Possibly, MARM may be helpful to establish the pathway of therapy effect for various treatments, including medication, biofeedback, or exercise training (36). Few studies on breathing therapies for asthma have

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**Table 6.** NQ scores—before and after treatment according to MARM category.

<table>
<thead>
<tr>
<th>Category</th>
<th>Pretreatment, Mean (SD)</th>
<th>Posttreatment, Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal MARM</td>
<td>2.0 (2.1)</td>
<td>2.4 (1.6)</td>
</tr>
<tr>
<td>Abnormal MARM</td>
<td>6.0 (3.4)</td>
<td>3.5 (3.2)</td>
</tr>
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**Note:** Cutoffs for MARM balance, normal < 30, abnormal 30 and above.

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**Table 7.** Dyspnea scores—before and after treatment according to MARM category.

<table>
<thead>
<tr>
<th>Category</th>
<th>Pretreatment, Mean (SD)</th>
<th>Posttreatment, Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal MARM</td>
<td>17.3 (6.4)</td>
<td>10.1 (7.1)</td>
</tr>
<tr>
<td>Abnormal MARM</td>
<td>24.7 (8.1)</td>
<td>10.3 (4.7)</td>
</tr>
</tbody>
</table>

**Note:** Cutoffs for NQ balance, normal < 30, abnormal 30 and above.
investigated their effects on breathing pattern, and this may prove useful in refinement of the application and development of these types of therapies.

Implementation of MARM in the clinical setting may be hampered by the need for training and the time required to do the complete protocol. Test–retest reliability of the therapist in this study was not determined, but the very high correlation between pretest MARM and change in MARM indicates that reliability would be satisfactory. This issue will be important however in future studies that employ MARM.

An important limitation of this study is the absence of any longer-term follow-up to assess whether the changes in breathing pattern and complaints are stable and sustained. Previous studies using the MARM in a 2-year follow-up of myocardial infarction patients showed a stable effect of treatment and prompted later validation studies (45).

CONCLUSION

The majority of individuals with a large number of DB symptoms are also likely to have a thoracic dominant breathing pattern. Breathing pattern and symptoms can normalize after breathing therapy. The extent of thoracic dominant breathing seems an important moderator for dyspnea symptoms and an important component of treatment.

DECLARATION OF INTEREST

Dr. Jan van Dixhoorn, and Els L.M. Anthonissen are teachers of the Whole Body Breathing method. Also the Whole Body Breathing method was developed by Dr. Jan van Dixhoorn.

REFERENCES


