



Abnormal breathing pattern features in chronic refractory cough

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Abnormal breathing features are common in patients with chronic refractory cough (CRC). Abnormal breathing features appear to be associated with laryngeal motor function but independent of laryngeal hypersensitivity that drives CRC and ILO. <https://bit.ly/4bh4dXT>

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Abstract

Background Chronic refractory cough (CRC) is a challenging condition that responds to speech pathology intervention. Clinical observation suggests abnormal breathing patterns occur in CRC and may be indirectly addressed as part of behavioural treatment, yet breathing pattern changes in CRC are poorly understood. The aims of this study were to 1) describe breathing patterns in patients with CRC, 2) compare breathing pattern features between patients with CRC and inducible laryngeal obstruction (ILO), and 3) estimate the effect of breathing pattern features on clinical measures of laryngeal sensory and motor dysfunction.

Methods This retrospective cross-sectional observational study included 634 patients with CRC or ILO. A file audit of speech pathology assessment data was undertaken. Analysis of self-reported laryngeal symptoms (*via* questionnaires) and clinical assessment of voice and breathing of those with CRC and ILO was conducted.

Results Most participants with CRC (73%) demonstrated at least one abnormal breathing pattern feature. The most common feature was thoracic breathing (69%) followed by oral breathing (33%). The type and prevalence of abnormal breathing patterns were similar between CRC and ILO. Abnormal breathing patterns were associated with reduced maximum phonation time (MPT); however, there was no association between these features and Newcastle Laryngeal Hypersensitivity Questionnaire scores.

Conclusions Abnormal breathing features are common in patients with CRC and are not significantly different from those occurring in ILO. There is some association between abnormal breathing features and MPT, suggesting impairment of laryngeal motor function. Conversely, there is no association between abnormal breathing features and laryngeal hypersensitivity.

Introduction

Chronic cough, defined as a cough lasting for >8 weeks, is common and has a negative impact on morbidity and healthcare utilisation [1]. Chronic cough is refractory to medical treatment in up to 42% of cases [2] and is termed chronic refractory cough (CRC) (also known as refractory chronic cough). Inducible laryngeal obstruction (ILO) is a related condition whereby the vocal folds involuntarily adduct during respiration leading to dyspnoea, throat tightness, cough, dysphonia and stridor. Clinical observation suggests that abnormal breathing patterns can occur in CRC and ILO, and that speech pathologists indirectly address breathing pattern changes in their behavioural treatment of these conditions [3, 4]. Despite this, there is a lack of clarity around breathing pattern abnormalities in the CRC population.

Breathing pattern changes have been reported as a significant cause of dyspnoea in conditions such as dysfunctional breathing. These abnormal breathing patterns include elevated respiratory rate/hyperventilation [5–8], a thoracic-dominant breathing pattern, accessory muscle usage [9, 10] and mouth breathing [11]. Behavioural treatment that addresses abnormal breathing patterns in dysfunctional breathing significantly improves quality of life [12–14]. Dysfunctional breathing has also been thought to drive dyspnoea in long COVID [15]. These studies indicate that a breathing pattern disorder can be a significant



primary problem in patients with unexplained dyspnoea, and that recognition and treatment of breathing pattern disorders can offer significant benefits to patients. This raises the question of the role of an abnormal breathing pattern in CRC, and how it should be incorporated into the assessment and management of patients with CRC.

The prevalence and characteristics of abnormal breathing patterns in CRC have not been reported, and the impact of these features on clinical outcomes is also unknown. In this study we examined the significance of abnormal breathing patterns in CRC and compared the results to a positive control group with ILO without cough, where breathing pattern and dyspnoea are both recognised and managed. The aims of this study were to 1) describe breathing patterns in patients with CRC, 2) compare breathing pattern features between patients with CRC and those with ILO without cough, and 3) estimate the effect of abnormal breathing pattern features on clinical measures of laryngeal sensory and motor dysfunction.

Method

The study was a retrospective cross-sectional observational study conducted in a tertiary referral hospital. A file audit was undertaken to analyse self-reported laryngeal sensory symptoms, sustained phonation and breathing patterns. The study was reviewed and approved by the Hunter New England Human Research Ethics Committee and a waiver of consent was approved (authorisation number AU202103-23).

Participants

The study included 542 participants with CRC and a comparison group of 92 with ILO without cough. Participants were 634 adult patients referred consecutively to the Speech Pathology Department at John Hunter Hospital (Newcastle, Australia) for behavioural treatment of CRC or ILO. Participants were recruited between October 2018 and June 2022.

Inclusion criteria included CRC or ILO. CRC was diagnosed by a respiratory physician, and defined as a cough persisting for >8 weeks and refractory to medical management. ILO was diagnosed *via* functional transnasal laryngoscopy using accepted diagnostic procedures [16, 17] as well as clinical features such as inspiratory dyspnoea, noisy breathing and throat tightness. Abnormal glottal closure during respiration was diagnosed when >25% closure of the vocal folds during inspiration and/or >50% closure during expiration was observed. Participants were not asked to breath-hold, and breath holding that occurred spontaneously during tidal respiration during functional transnasal laryngoscopy was considered abnormal.

Additional inclusion criteria for both groups included optimal assessment and treatment from an otolaryngologist or respiratory physician prior to referral to exclude asthma and other pulmonary disease as a reason for respiratory symptoms. If participants did have a coexisting diagnosis of asthma, their treatment needed to be optimised by a respiratory physician prior to referral.

Exclusion criteria included recent upper respiratory tract infection, untreated asthma, untreated rhinitis, untreated gastro-oesophageal reflux disease, significant psychological factors that prevented participation in the assessment or neurological impairment. None of the participants reported previously seeing a respiratory physiotherapist for assessment or management of their symptoms and all were treatment naïve. None of the participants had been referred specifically for assessment of dysfunctional breathing.

Measurements

Participants attended a single 60-min assessment visit with a qualified speech pathologist with experience in the assessment and treatment of patients with CRC and ILO. Assessment measures, outlined in table 1, were completed as per the outpatient speech pathology clinical procedures of the department, and included self-reported laryngeal symptoms (*via* questionnaires) and clinical assessment of voice and breathing completed while sitting. The parameters of breathing region (thoracic *versus* abdominal), breathing route and breathing rate were used to describe breathing pattern features in this study. Additional features that were noted included stridor or noisy breathing, breath holding, and poor coordination of respiration and phonation. Definitions of these features and criteria for assessment are reported in table 1.

Data analysis

Descriptive statistics included mean (standard deviation), median (interquartile range) and number (percentage) as appropriate. The prevalence of abnormal breathing pattern features between CRC and ILO groups was compared using Fisher's exact test. Linear regression was conducted to estimate the effect of breathing pattern features on maximum phonation time (MPT) and Newcastle Laryngeal Hypersensitivity Questionnaire scores. Analysis was conducted using Stata version 16 (StataCorp, College Station, TX, USA). The significance level was set at $p < 0.05$.

TABLE 1 Measurement of breathing characteristics and clinical measures

| Breathing feature | Definition |
|---|---|
| Clinical assessment | |
| Region (area of primary muscular engagement and movement while breathing, observed and noted by the speech pathologist) | Normal: normal relaxed diaphragmatic breathing with abdominal movement observed during tidal breathing Abnormal: clavicular and/or thoracic movement observed during tidal breathing |
| Route (primary route the patient uses to breathe, observed and noted by the speech pathologist) | Normal: nasal breathing route observed during clinical assessment Abnormal: oral or mixed (oral/nasal) breathing route observed during clinical assessment |
| Rate (number of breaths counted by the speech pathologist in 1 min of observation while sitting) | Normal: 8–14 breaths·min ⁻¹ Abnormal: anything outside 8–14 breaths·min ⁻¹ |
| Stridor (recorded by the speech pathologist if present or absent during the session) | Normal: absent Abnormal: present |
| Breath holding (obtained through observation of the patient in the session and recorded by the speech pathologist) | Normal: no breath holding observed during the clinical assessment Abnormal: breath holding observed during the clinical assessment |
| Coordination of respiration and phonation (reported on by the speech pathologist after observing the patient in conversation and during voice assessment tasks [#]) | Normal: adequate coordination of breathing and phonation onset as well as no breathing difficulties while speaking Abnormal: poor coordination and control of breathing and phonation when speaking, leading to observed breathlessness and a need to breathe more frequently within a spoken sentence |
| Questionnaires | |
| Newcastle Laryngeal Hypersensitivity Questionnaire [28] | Normal: score >17.1 |
| Leicester Cough Questionnaire [29] | Normal: score >17.68 [30] |
| Vocal Cord Dysfunction Questionnaire [31] | Normal: score ≤12 |
| Dyspnoea Index [32] | Normal: score >10 |
| Hospital Anxiety and Depression Scale [33] | Normal: score ≤7 |
| Instrumental assessment | |
| Maximum phonation time (recorded and analysed by the speech pathologist) | Normal: >15 s [34] |
| Forced expiratory volume in 1 s (% pred) | Normal: ≥80% predicted |
| Forced vital capacity (% pred) | Normal: ≥80% predicted |
| Forced expiratory ratio (% pred) | Normal: ≥70% predicted |
| [#] : observation of respiratory/phonatory coordination during conversation, paragraph reading, prolonged counting and vowel prolongation. | |

Results

Demographics

The study included 542 participants with CRC and 92 with ILO without cough. 87 of the CRC participants also had paradoxical vocal fold movement (PVFM) present on laryngoscopy; however, cough was their predominant symptom. The mean age was 58 years and the majority were female (77%). The median duration of symptoms was 24 months. Comorbidities were common and had been addressed prior to assessment. Most participants (61%) were never-smokers. The mean pulmonary function test results were in the normal range. Mean scores on patient-reported symptom questionnaires and MPT values were in the abnormal range (table 2). Hospital Anxiety and Depression Scale scores were in the normal range.

Breathing assessment

Most participants with CRC (73%) demonstrated at least one abnormal breathing pattern feature. Thoracic/upper chest breathing was the most common abnormality (69%), followed by oral/mixed breathing route (33%), and combined thoracic and oral breathing (29%) (table 3). Of the patients with chronic cough, stridor was present in 87 participants (16%), breath holding in 23 (4%), and poor coordination of respiration and phonation in 165 (31%). Breathing pattern features in participants with CRC were similar to the positive control group with ILO without cough (table 3). The CRC group did have a higher prevalence of abnormal rate and route combined than the ILO group (8% versus 2%; $p=0.048$).

Linear regression was used to estimate the effect of abnormal breathing pattern features on laryngeal motor and sensory function, assessed as MPT (table 4) and laryngeal hypersensitivity (table 5), respectively. MPT values were lower when several abnormal breathing pattern features (thoracic pattern, oral breathing, combined thoracic and oral breathing, combined oral breathing and rapid breathing rate, and combined thoracic pattern, oral breathing and rapid rate) were present (table 4). In contrast, there was no significant

TABLE 2 Participant demographics and clinical features

| | Total | CRC | ILO |
|--|----------|----------|---------|
| Subjects | 634 | 542 | 92 |
| Age (years) | 58±16 | 58±16 | 53±18 |
| Gender | | | |
| Female | 487 (77) | 418 (77) | 69 (75) |
| Male | 147 (23) | 124 (23) | 23 (25) |
| Comorbidities | | | |
| Gastro-oesophageal reflux disease | 406 (64) | 358 (66) | 48 (52) |
| Rhinitis/sinusitis | 299 (47) | 258 (48) | 41 (45) |
| Asthma | 218 (34) | 174 (32) | 44 (48) |
| Obstructive sleep apnoea | 108 (17) | 93 (17) | 15 (16) |
| Smoking | | | |
| Never-smoker | 388 (61) | 341 (63) | 47 (51) |
| Ex-smoker | 216 (34) | 178 (33) | 38 (41) |
| Current smoker | 30 (5) | 23 (4) | 7 (8) |
| Body mass index (kg·m⁻²) | 32±8 | 32±8 | 31±6 |
| Spirometry | | | |
| Forced expiratory volume in 1 s (% pred) | 87±20 | 87±19 | 84±21 |
| Forced vital capacity (% pred) | 95±17 | 96±17 | 93±20 |
| Forced expiratory ratio (% pred) | 77±15 | 78±15 | 75±17 |
| Questionnaire scores | | | |
| Newcastle Laryngeal Hypersensitivity Questionnaire | 14±3 | 14±3 | 15±3 |
| Leicester Cough Questionnaire | 14±4 | 13±4 | 18±3 |
| Vocal Cord Dysfunction Questionnaire | 40±9 | 40±8 | 40±11 |
| Upper airway Dyspnoea Index | 21±10 | 21±11 | 22±9 |
| Hospital Anxiety and Depression Scale: Anxiety | 7±4 | 7±4 | 9±5 |
| Hospital Anxiety and Depression Scale: Depression | 6±4 | 5±4 | 6±4 |
| Maximum phonation time (s) | 11±6 | 11±6 | 12±5 |

Data are presented as n, mean±SD or n (%). CRC: chronic refractory cough; ILO: inducible laryngeal obstruction.

association between any breathing pattern feature and Newcastle Laryngeal Hypersensitivity Questionnaire scores (table 5).

Discussion

This is the first study to describe and compare abnormal breathing pattern features in patients with CRC and ILO. Dyspnoea and breathing pattern changes are an established feature of ILO and are addressed in therapy. We observed a high proportion of abnormal breathing pattern features in CRC that was similar to ILO. This suggests that abnormal breathing pattern features are a relevant part of the profile in CRC. We also found an association between abnormal breathing pattern features and laryngeal motor function, demonstrated by reduced MPT values. MPT is a reliable indirect measure of glottal control and

TABLE 3 Comparison of dysfunctional breathing features between chronic refractory cough (CRC) and inducible laryngeal obstruction (ILO) groups

| | Total (n=634) | CRC (n=542) | ILO (n=92) | p-value [#] |
|----------------------------------|------------------|----------------|---------------|----------------------|
| Any abnormality | 458 (72) | 395 (73) | 63 (69) | 0.527 |
| Abnormal region | 432 (68) | 372 (69) | 60 (66) | 0.905 |
| Abnormal route | 202 (32) | 178 (33) | 24 (26) | 0.274 |
| Abnormal region and route | 181 (28) | 159 (29) | 22 (24) | 0.380 |
| Abnormal rate | 150 (24) | 128 (24) | 22 (24) | 0.895 |
| Abnormal region and rate | 145 (23) | 123 (23) | 22 (24) | 0.788 |
| Abnormal route and rate | 45 (7) | 43 (8) | 2 (2) | 0.048 |
| All abnormal | 44 (7) | 42 (8) | 2 (2) | 0.071 |

Data are presented as n (%), unless otherwise stated. #: p-value calculated using Fisher's exact test.

TABLE 4 Association between breathing pattern features and maximum phonation time values

| | Coefficient (95% CI) | p-value |
|---------------------------|----------------------|---------|
| Any abnormality | -1.1 (-2.2-0.1) | 0.070 |
| Abnormal region | -1.3 (-2.5- -0.1) | 0.034 |
| Abnormal route | -1.2 (-2.3- -0.2) | 0.020 |
| Abnormal rate | -0.3 (-1.4-0.9) | 0.640 |
| Abnormal region and route | -1.3 (-2.4- -0.3) | 0.013 |
| Abnormal region and rate | -0.4 (-1.6-0.7) | 0.445 |
| Abnormal route and rate | -2.1 (-3.9- -0.2) | 0.027 |
| All abnormality | -2.2 (-4.0- -0.3) | 0.021 |

management of aerodynamic forces of the lungs, and provides important information about the interaction between the respiratory and phonatory systems [18, 19]; therefore, reduced MPT may be due to insufficient lung volume to sustain a vowel, glottic insufficiency during phonation, the presence of laryngeal muscle tension or a combination of these. This result suggests that phonation is less efficient when abnormal breathing pattern features are present. In contrast to laryngeal motor function, our data showed no association between abnormal breathing pattern features and self-reported laryngeal sensory dysfunction. Overall, these results might suggest that abnormal breathing patterns are prevalent in CRC and may be related to laryngeal motor dysfunction, rather than self-reported laryngeal sensory dysfunction in CRC and ILO.

The aim of the study was to report observable breathing features in individuals with CRC and compare breathing features observed in those with CRC and ILO. The purpose of this study was not to diagnose dysfunctional breathing or breathing pattern disorder; however, the type and prevalence of abnormal breathing pattern features in the current study aligns with previous work in dysfunctional breathing and extends the observations to CRC. DE VOS *et al.* [20] reported dysfunctional breathing in 33 out of 34 participants referred to speech pathology for management of CRC. In an observational study, SELBY *et al.* [21] reported abnormal breathing patterns, defined as a score of ≥ 4 on the Brompton Breathing Pattern Assessment, in 41% of participants with ILO, which increased to 82% after provocation challenge. DENTON *et al.* [22] reported that in their difficult asthma cohort, a thoracic-dominant breathing pattern was most common and often overlapped with hyperventilation and mouth breathing. While we reported increased respiratory rate, our study did not specifically address hyperventilation syndrome as this was beyond the scope of this study.

The breathing pattern features reported in the current study are similar to descriptions of dysfunctional breathing in the literature, where severe and refractory dyspnoea is a common symptom. However, dysfunctional breathing can be difficult to define and validate with standardised diagnostic criteria, which are limited. The mechanisms driving dysfunctional breathing integrate biochemical, biomechanical, psychological, social and physiological aspects [12, 23]; however, how these components interact to result in the patient experience remains difficult to define. In patients with dysfunctional breathing, treatment of breathing pattern can improve quality of life [13]. Dysfunctional breathing appears to be a different phenomenon to CRC as it does not appear to involve laryngeal sensory dysfunction. The relationship between dysfunctional breathing and CRC is unclear, but it would seem that they are different diagnoses

TABLE 5 Association between breathing pattern features and Newcastle Laryngeal Hypersensitivity Questionnaire scores

| | Coefficient (95% CI) | p-value |
|---------------------------|----------------------|---------|
| Any abnormality | -0.7 (-0.7-0.5) | 0.821 |
| Abnormal region | 0.10 (-0.5-0.7) | 0.730 |
| Abnormal route | -0.3 (-0.8-0.3) | 0.359 |
| Abnormal rate | -0.1 (-0.7-0.5) | 0.680 |
| Abnormal region and route | -0.3 (-0.9-0.3) | 0.357 |
| Abnormal region and rate | -0.02 (-0.6-0.6) | 0.941 |
| Abnormal route and rate | -0.9 (-1.9-0.1) | 0.069 |
| All abnormal | -0.9 (-2.0-0.1) | 0.067 |

that share commonalities in symptoms and treatment [24]. Standardised assessment of laryngeal symptoms in patients with confirmed dysfunctional breathing both with and without ILO and CRC would be needed to better understand the overlap of symptoms, and the current study is unable to explore this relationship.

It is unclear whether the presence of abnormal breathing pattern features is a cause or result of laryngeal dysfunction. SELBY *et al.* [21] suggested that abnormal breathing pattern features can contribute to the sensation of tightness and dyspnoea that mimics ILO. DENTON *et al.* [22] suggested that abnormal breathing pattern features may be related to the use of maladaptive techniques involving suboptimal postures and learned responses. That is, an upper chest breathing pattern and increased breathing rate might be employed to compensate for the perception of breathlessness and mouth breathing might increase the sensation of airflow. Over time these patterns might become habitual. In contrast with this assumption, laryngeal abnormalities, including mass lesions and vocal fold palsy that partially occlude the airway, can change an individual's breathing pattern [25, 26]. It seems likely that the relationship between abnormal breathing patterns and laryngeal dysfunction is bidirectional and differs between patients.

The main clinical implication of this study is that patients with CRC are likely to demonstrate concomitant breathing pattern abnormalities, which should be noted during the clinical assessment. These patterns may impact laryngeal motor function during phonation but may be less likely to influence self-reported laryngeal sensory dysfunction. Therapy for CRC and ILO indirectly addresses breathing pattern abnormalities. For example, while PVFM release breathing focuses on optimal laryngeal postures during respiration, it simultaneously promotes nasal breathing, lower abdominal breathing and encourages a steady breathing rate and rhythm. It is a basic tenet of speech pathology treatment that sufficient breath is required for adequate performance of the vocal/laryngeal tasks required for therapy. Results suggest that an abnormal breathing pattern is associated with impaired laryngeal motor support. This provides a rationale for the current speech pathology approach to managing CRC where the breathing pattern abnormalities are identified and treated in order to support successful speech pathology management of CRC.

The limitations of this study are that it was a retrospective study utilising routinely collected clinical data from a single site. While this provides excellent validity, there is potential for misinterpretation of some of the fields and these results are unable to be generalised to the larger population. The assessment was conducted at a single time-point, hence reproducibility over time could not be assessed. There was no control group of healthy individuals in which to assess the presence of dysfunctional breathing and the clinical significance of a single breathing pattern feature, such as oral breathing. The measures used in the study were based on clinical assessment by speech pathologists with no instrumental analysis of breathing pattern (*e.g.* optoelectronic plethysmography). The Nijmegen Questionnaire was not included in the study as it was not part of routine clinical practice and, while it is commonly used in the assessment of individuals who present with features of dysfunctional breathing, there is controversy about the ability of this questionnaire to diagnose dysfunctional breathing [14].

Participants were seen by several speech pathologists in the department at the study site, all of whom have considerable experience in the assessment and management of CRC and ILO. The assessment procedures were standardised amongst the speech pathologists, and observational assessment of breathing pattern features is an inherent part of speech pathology assessment of patients with CRC and ILO. However, the retrospective nature of the study precluded assessment of inter-rater reliability. Furthermore, as this study was not designed to diagnose dysfunctional breathing, a standard classification system, such as the one outlined by BOULDING *et al.* [12] and BARKER and EVERARD [24], and known clinical features [27] were not included in the assessment.

Conclusions

Abnormal breathing pattern features are prevalent in patients referred to speech pathology for CRC and are similar to ILO. Abnormal breathing pattern features appear to be associated with laryngeal motor function and therefore might need to be addressed in treatment. In contrast, abnormal breathing patterns are independent of laryngeal hypersensitivity that drives CRC and ILO.

Provenance: Submitted article, peer reviewed.

Ethics statement: The study was reviewed and approved by the Hunter New England Human Research Ethics Committee and a waiver of consent was approved (authorisation number AU202103-23).

Conflicts of interest: All authors declare they have no competing interests.

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