

Personal view

Behavioural breathlessness

J B L Howell

The syndrome of symptoms caused by involuntary overbreathing is common, frequently overlooked, and often badly managed. Symptoms may not always be dominated by breathlessness, and may suggest such diagnoses as myocardial ischaemia, pulmonary emboli, hypoglycaemia, peripheral neuropathy, epilepsy; often no clear diagnosis is suggested. The presentation may therefore present formidable diagnostic problems.

Part of the reason for uncertainty about the diagnosis of the hyperventilation syndrome is that the condition has never been clearly defined. This was reinforced at the 4th International Symposium on Respiratory Psychophysiology in Southampton in 1984. Questionnaires were distributed to seek participants' views about the hyperventilation syndrome¹—for example, which features were considered essential for diagnosis and which features were commonly present but were not essential—and a definition of the syndrome was requested. There was no consensus. On the basis of the range of answers the following statement was subsequently proposed: "The hyperventilation syndrome is a syndrome characterised by a variety of somatic symptoms induced by physiologically inappropriate hyperventilation and usually reproduced in whole or in part by voluntary hyperventilation."¹ Regardless of whether this is an acceptable definition or not, it is of limited usefulness to the clinician. It indicates not which symptoms might lead to the suspicion of the hyperventilation syndrome but only how a diagnosis of the syndrome, once suspected, may be supported by the effects of voluntary overbreathing. Regrettably, suspicion of the hyperventilation syndrome is often raised by the absence of organic disease, rather than by the presence of positive features.

My aim in this paper is to describe my personal experience of the hyperventilation syndrome over the past 28 years and to discuss some studies which have provided positive criteria for diagnosis, a rational approach to treatment, and hints of the sort of neurological mechanisms that may play a part.

In 1961 I noted that among patients referred to the pulmonary function laboratory with disabling breathlessness some individuals had an FEV₁ that was either normal or only moderately reduced. The reasons for this disproportionate breathlessness were not apparent, but functional or psychogenic factors seemed most likely. Dr Bruce Burns, a psychiatrist, and I therefore designed a study that

we thought might shed light on the nature of the problems of these individuals.^{2,3} In outline, we compared a range of features shown by a group of patients with disproportionate breathlessness with those of a control group, the latter being patients who appeared to be appropriately breathless. The features to be compared were sought systematically by questionnaire and a personal interview with Dr Burns, and included a description of symptoms, premorbid personality, any psychiatric illness, and any notable events in their lives that had occurred in the three years before the assessment. We also carried out standard medical assessments, a routine physical examination, pulmonary function tests (spirometry, rebreathing carbon dioxide tension (PCO₂)), chest radiography, full blood count, and urine analysis. Electrocardiography and exercise tests were performed when indicated.

Over the next few months I identified 31 patients with disproportionate breathlessness from my outpatient clinics on the basis of dyspnoea grade III or more⁴ and the absence of adequate respiratory, cardiovascular, or other relevant disease. All had an FEV₁ of more than 1 litre. Follow up for at least three years did not lead to evidence that we had overlooked organic disease. Thirty one control patients with appropriate breathlessness associated with severe airflow obstruction (FEV₁ < 1.0 litre) were identified; all but four had chronic bronchitis.⁵ With their consent, they were referred for interview by Dr Burns. Initially we omitted to ensure that he was unaware of my categorisation of them, but this was corrected for the second half of the study. Examination of the results of all assessments provided no evidence that this omission had introduced bias in the earlier phase.

Some features of the two groups are shown in table 1. Disproportionately breathless patients

Table 1 Features of disproportionately and appropriately breathless patients referred from medical outpatient clinics*

	Disproportionate breathlessness (n = 31; 19 M)	Appropriate breathlessness (n = 31; 22 M)
Age (y)	48	58
Forced vital capacity (l)	3.1	2.2
FEV ₁ (l)	2.0	0.6
Paco ₂ (mm Hg)	39.6	50.6

*All values differed significantly (p < 0.01).
Conversion: Traditional to SI values—arterial carbon dioxide tension (Paco₂): 1 mm Hg ≈ 0.133 kPa.

Address for reprint requests:
Professor J B L Howell,
Medicine 1, Centre Block,
Southampton General
Hospital, Southampton
SO9 4XY.

Based on the presidential
address given to the British
Thoracic Society in 1988.

Table 2 Characteristics of breathlessness in the 31 disproportionately breathless (DB) and 31 appropriately breathless (AB) patients

	DB	AB
Poorly correlated with exertion	30	0
Symptoms of hyperventilation	29	5
Attacks at rest	28	6
Harder to breathe in than out	27	8
Fluctuating and recurrent	25	0
Fear of sudden death	25	1

were significantly younger and had much less airflow obstruction than the control group. They had a normal P_{aCO_2} on average at their first full assessment, though many were noted to have a low P_{aCO_2} on other occasions. Several of the control group had ventilatory failure.

Table 2 compares the main characteristics of breathlessness in the two groups. Disproportionately breathless patients are seen to be characterised by a high prevalence of: (1) episodic breathlessness at rest, usually associated with symptoms suggestive of hyperventilation (dizziness or lightheadedness, paraesthesiae, occasionally cramps)—they often feared that they were going to die during these attacks; (2) considerable variability of symptoms, often within a few hours or between days—they had good days and bad days and were often symptom free; (3) breathlessness poorly correlated with exercise—that is, they would be as breathless with mild, brief exertion as with severe exertion; (4) finding it harder to breathe in than out; (5) feeling hot and sweaty when breathless.

How often do we inquire whether breathlessness occurs at rest or not, or whether its severity depends on the degree of exertion? These are clearly discriminating features in identifying patients with disproportionate breathlessness.

Psychiatric assessment and premorbid personality

Of the 31 disproportionately breathless patients, seven were diagnosed as having anxiety reactions and 16 as being depressed (table 3). In those diagnosed as being depressed a general appearance of depression was frequently absent, this feature emerging only from specific questioning. The remaining eight patients were considered to have hysterical reactions, six having had premorbid hysterical personalities. A striking finding was the high prevalence of obsessional premorbid personalities, which characterised all those with anxiety reactions and three quarters of those with depression. Obsessionalism was mainly in the form of strong perfectionist traits—they were people who liked things to be right. For exam-

ple, there were housewives insisting that their house had to be tidy; one patient said she always closed all doors in her house because otherwise they would be open to different degrees and this was "untidy." Many men would insist on their work being perfect, and were irritated by fellow workers who displayed lower standards.

No psychiatric abnormalities were diagnosed in the control group. Four were judged to have hysterical personalities because of evidence of attention seeking (not significantly different from the number among the disproportionately breathless patients) and seven had obsessional personalities. Twenty were judged to have good social adjustment with normal personalities (compared with six of the disproportionately breathless group, a highly significant difference).

Precipitating factors

Personality is determined by genetic factors and early environment; it cannot be responsible for *initiating* reactions later in life, providing only an appropriate psychological background for reacting to more recent events. When we sought events in the preceding three years, the following were significantly different at the 1% level (figures represent numbers of subjects in disproportionately breathlessness/appropriately breathless groups): previous family history of depressive illness (10/0), bereavement or separation (22/3), current marital disharmony (18/3), gross secondary gain from the "illness" (15/1), living alone (9/1), previous surgical operation (13/2).

Three categories seemed to embrace most of these features: bereavement, resentment, and illness, especially uncertainty about whether they had a serious illness.

Bereavement

Bereavement was usually loss of a close relative associated with severe and prolonged grieving. One striking example was provided by a 40 year old man whose 8 year old son had wandered on to the railway line and been killed by a train. The patient had not realised the relevance of the bereavement as a possible precipitating factor until it was mentioned. Occasionally, the problem was separation rather than death—for example, removal of a foster child or emigration of a son or daughter.

Resentment

Resentment was usually a sense of injustice about the way in which the patient (or a close relative) had been treated—for example, failing to get promotion at work or being badly treated by a doctor. I recall a highly obsessional perfectionist who was extremely frustrated by his general practitioner, who never remembered who he was, never had the results of investigations to hand, and always had to retake the history. More recently, his wife had had a radical mastectomy and when he saw the result of this very mutilating operation he was angry and even more resentful with the medical profession. Furthermore, when he was investigated in hospital for symptoms due to the

Table 3 Psychiatric diagnoses and premorbid personality in the 31 disproportionately breathless patients

	Depression	Anxiety	Hysterical reaction
Psychiatric diagnosis	16	7	8
Premorbid personality*			
Obsessional	12†	7†	—
Hysterical	—	—	6

*Six patients were normal.
†p < 0.01.

hyperventilation syndrome no one explained their thinking. When a doctor inquired among other things about his sex life his suspicions were aroused and when, on being challenged, the doctor admitted that he was a psychiatrist the patient was enraged. The result was even greater resentment with our profession. The relevance of these events to his symptoms had never been appreciated by the patient.

Illness or uncertainty about the seriousness of an illness

Failure to receive reassurance about an insignificant but worrying symptom was sometimes the only initiating event; when symptoms remain unexplained fear of illness may become dominant. For example, a medical physicist engaged in radioisotope imaging experienced dizziness and wondered whether he might have a brain tumour. A neurologist found no abnormality but gave no convincing explanation for his symptoms. Symptoms persisted for four years yet he still had no idea of the cause of his symptoms.

On the basis of these and other results a profile of the individual liable to develop the hyperventilation syndrome emerges. Usually the patient

- is a perfectionist—that is, likes things to be “right”;
- may have become depressed or anxious or both, or less frequently may be using the symptoms consciously or unconsciously for some personal gain;
- has experienced one or more of the following factors: (a) bereavement or separation, and is still missing the person deeply; (b) resentment about the way in which he or she (or a close relative or friend) has been treated—for example, by employer, family, or doctors; (c) fear that he or she has some serious, usually lethal, illness; when doctors “reassure” patients that there is nothing wrong, they may induce fear that they are developing a “mental” illness;
- has experienced these characteristic symptoms: (a) episodic symptoms, occurring at rest for no apparent reason and usually when the patient is relaxed—reading or watching television—and associated with feeling hot and sweating; (b) symptoms associated with dizziness, paraesthesia (sometimes unilateral, usually on left), palpitation (patients frequently think they are going to die during the attack); (c) shortness of breath at rest or during exertion or both, not related to the severity of exertion; (d) during breathlessness greater difficulty in breathing in than breathing out.

There may also be objective features, including obvious overbreathing, poor exercise tolerance unrelated to the degree of exertion, and a grossly disorganised spirogram.²

Because an obsessional (perfectionist) personality is common and the factors described above are also common, we considered whether any other factors might lead to the development of the hyperventilation syndrome. The degree of sensitivity to hypocapnia seemed worthy of study.

Sensitivity to hypocapnia

The sensitivity of individuals to the effects of overbreathing and hypocapnia differs widely. Some are highly sensitive, developing symptoms rapidly within seconds, but a few appear to be virtually insensitive. It seemed possible that, if overbreathing occurred for any reason, individuals with high sensitivity to hypocapnia would be more likely to develop the clinical syndrome of hyperventilation syndrome. We argued that if they reacted to the alarming symptoms of hypocapnia with anxiety they might easily develop a vicious circle of overbreathing, more anxiety, and so on.

We therefore studied how quickly unselected patients attending outpatient clinics developed sensations on voluntary overbreathing and the relation between these induced sensations and their presenting symptoms. We also wished to find out how long a patient needed to overbreathe to induce symptoms, and whether depth and frequency of breathing had to be standardised. In testing for the hyperventilation syndrome, some clinicians instruct their patients to overbreathe at a fixed frequency for several minutes, but this can be distressing. It had long been my impression that 20 deep breaths were sufficient if symptoms were to be induced. In a group of randomly selected outpatients and normal volunteers the time taken to develop symptoms (*t*) was inversely related to the fall in end tidal PCO₂ (PETCO₂). Sensitivity to hypocapnia (SCO₂) was expressed as ΔPCO_2 (in mm Hg) $\times t/100$ (Grindrod and Howell, unpublished). In a group of patients diagnosed as having the hyperventilation syndrome the time taken for symptoms to develop for a given ΔPCO_2 was significantly shorter than normal—that is, SCO₂ was higher.

Because my diagnosis may have been influenced by knowledge of the results of this test we carried out another study to avoid this bias (Jones and Howell, in preparation). Patients referred to four medical outpatient clinics, three with a respiratory interest, were randomly invited to participate in the study while waiting to see the consultant; 205 (85%) patients accepted. They were asked to take 20 deep breaths (the 20 deep breaths test) through a mouthpiece and pneumotachograph, which enabled airflow and PETCO₂ to be recorded and to indicate the onset of any symptoms that might occur. They were then asked to describe their symptoms and to say whether they had experienced them before. Nine patients said that the deep breathing had reproduced the very symptoms for which they were attending and 24 said that they were part of their presenting complaints. The former were categorised as “probably” and the latter as “possibly” having the hyperventilation syndrome. They were then seen by the doctor to whom they had been referred, who had no knowledge of the results of these studies, and their diagnoses were later extracted from the notes. The results allowed us to test the consensus “definition” proposed at the International Symposium on Respiratory Psychophysiology in 1984. The definition would be supported if reproduction of symptoms by the 20 deep breaths test did

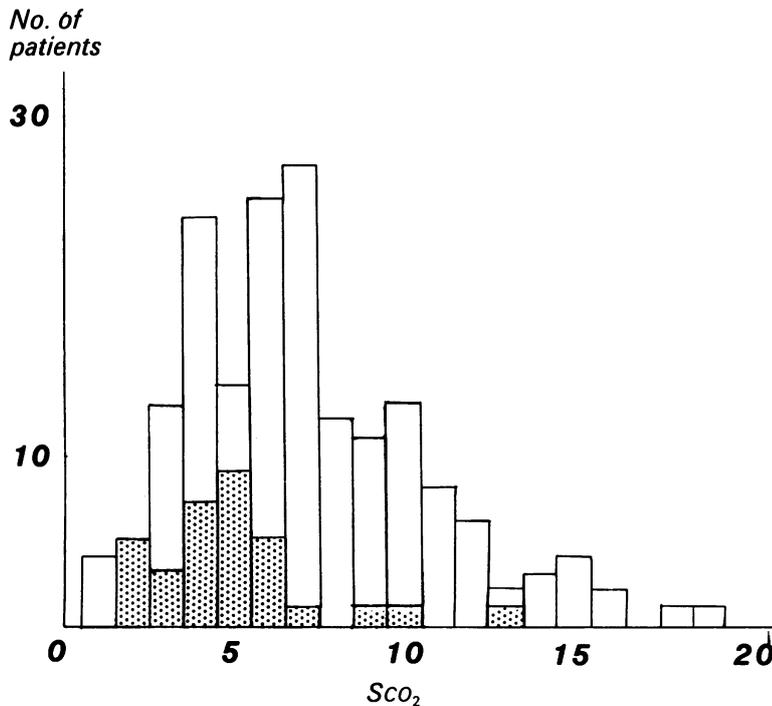


Figure 1 Frequency distribution of sensitivity to hypocapnia (SCO_2) in 205 outpatients. ($SCO_2 = \Delta PCO_2 \times t/100$, where PCO_2 is end tidal carbon dioxide tension in mm Hg and t is the time in seconds to the onset of onset of symptoms.) The mean was 7.2 in patients without the hyperventilation syndrome (\square) and 4.8 in those with the syndrome (\square) ($p < 0.001$).

identify patients who were subsequently diagnosed by clinicians as having the hyperventilation syndrome.

The mean fall in $PETCO_2$ was similar in the two groups—that is, in patients in whom the test produced and did not produce the symptoms for which they were attending—but the mean time to the onset of symptoms was significantly shorter in the possible and probable hyperventilation syndrome group (fig 1). There was no significant difference in age or sex distribution, or in the prevalence of “hyperventilation syndrome” between clinics. The clinical diagnoses made subsequently in the 33 “probable and possible” hyperventilation syndrome patients are shown in table 4.

Because there is no agreed “gold standard” for the diagnosis of hyperventilation syndrome we cannot say whether the categorisations made either through the 20 deep breaths test or by the consultants were correct. The 20 deep breaths test, however, identified a high proportion of subjects subsequently diagnosed as having hyperventilation syndrome or remaining undiagnosed. These results are consistent with the hypothesis that the hyperventilation syndrome is more likely to develop in those

Table 4 Clinicians' diagnoses in 33 probable and possible cases of the hyperventilation syndrome (HVS)

Clinicians' diagnosis	Possible HVS	Probable HVS
HVS	5	5
No diagnosis	8	3
Asthma	5	0
Allergy	3	0
Heart disease	1	1
Chronic obstructive airways disease	1	0
Pneumonia	1	0
Total	24	9

with high sensitivity to hypocapnia, in whom a short period of overbreathing from whatever cause induces symptoms, and hence may initiate a vicious circle or a conditioned response.

Ventilatory response to exercise

In addition to being breathless at rest, subjects with the hyperventilation syndrome are frequently very short of breath during exertion; rarely does shortness of breath occur only with exertion. Initially we wondered whether hyperventilation during exercise might provide a specific diagnostic test for the hyperventilation syndrome; but a few patients (<5%) diagnosed as having the hyperventilation syndrome on other grounds, including reproduction of symptoms by the 20 deep breaths test, were shown to have a normal ventilatory response to graded exercise. We therefore studied the ventilatory response to graded exercise on a bicycle ergometer before and after treatment in 12 patients diagnosed as having the hyperventilation syndrome on clinical criteria (Patel *et al*, in preparation).

After an initial period of observation at rest the work rate was increased in increments of 15 watts each minute to 75 watts. Heart rate (electrocardiograph) and ventilation were recorded. After a recovery interval of 15 minutes the exercise run was repeated. Immediately after this test the patients were treated for the hyperventilation syndrome as described later. About two weeks later they returned for reassessment. Similar exercise studies were again performed followed by an independent clinical assessment by me.

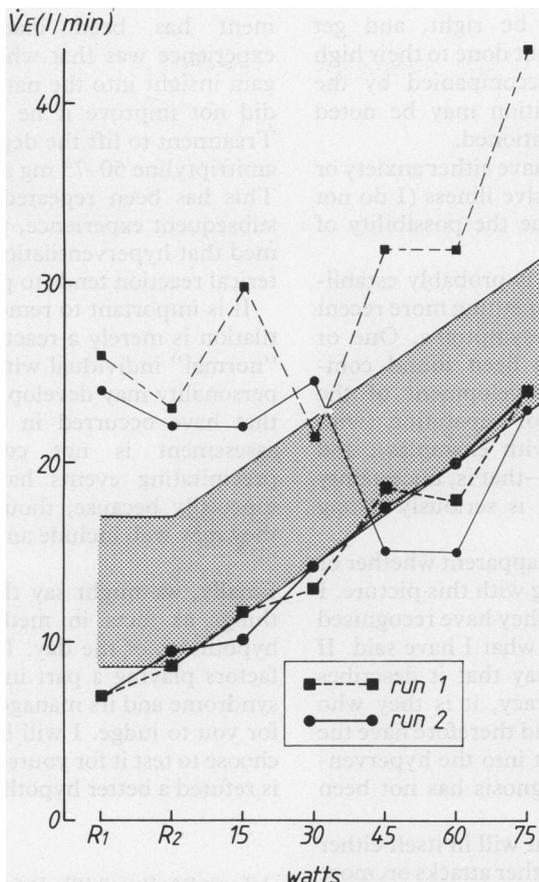
Before treatment individuals had highly variable results, but as a group they developed hyperventilation at intermediate and higher levels of exercise. The clinical responses to treatment were clearcut: five subjects improved symptomatically and seven showed no appreciable change, an unusually low rate of improvement. There was no difference in pretreatment mean ventilatory responses to exercise between those who did and those who did not improve subsequently.

The seven subjects who did not improve showed no significant change in their mean ventilatory responses at the different levels of exercise. By contrast, the five who improved with treatment showed a reduction in ventilation at all levels of exercise (see one example in fig 2). Individual responses varied, but the mean change in ventilatory response was due solely to a reduction in mean respiratory frequency of 4–7 breaths/min at each level of exercise (fig 3); mean tidal volumes remained unchanged. This may seem a small change to be associated with such an unequivocal reduction in symptoms, but tachypnoea is well known clinically to be associated with dyspnoea.

A model of the hyperventilation syndrome

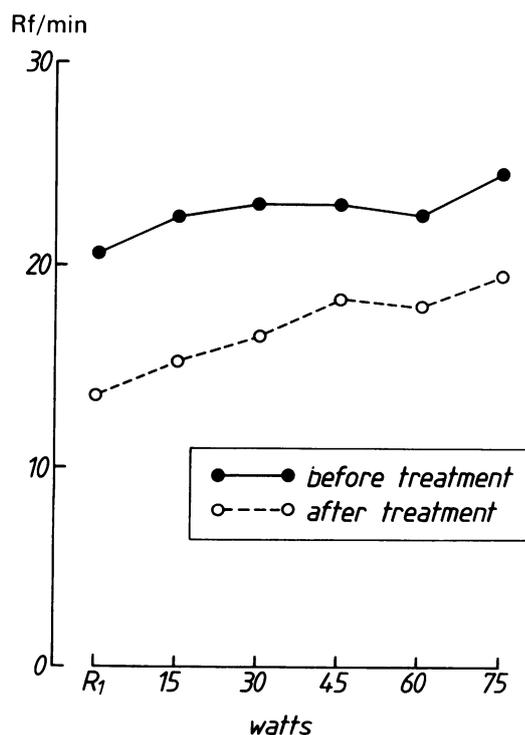
The conventional model of ventilatory control is based on the respiratory centre, with negative

Figure 2 Exercise ventilation in one subject who responded to treatment for the hyperventilation syndrome. Two studies were carried out, before and after treatment. The stippled area indicates the normal range of responses. R1 and R2—resting measurements.



feedback from chemoreceptors stabilising the PCO_2 , though it has always been accepted that higher centres are capable of influencing breathing. Plum⁶ made observations in rare individuals who after a cerebrovascular accident could not voluntarily control their breathing yet had normal carbon dioxide responsiveness, and inferred that a “behavioural pathway” exists that had been interrupted by the ischaemic damage. More recently, stimulation by magnetic coils on the scalp in normal

Figure 3 Mean breathing frequency response to exercise in five patients before and after improvement with treatment. R1—resting measurements. Frequencies are significantly different ($p < 0.02$) at rest and at 45, 60, and 75 watts.



subjects has produced evidence that cortical influences may have a role in the control of breathing under normal conditions as well as during voluntary breathing.⁷ It is a small step to suggesting that these behavioural cortical pathways are excessively activated by the emotional disturbances discussed above. Hence I have suggested “behavioural breathlessness” as an alternative title to “hyperventilation syndrome”.

Diagnosis and management of hyperventilation syndrome

Two approaches to treatment of the hyperventilation syndrome have been adopted. Lum and colleagues⁸ at Papworth Hospital have directed their attention to re-educating patients in relaxed and controlled breathing—that is, “behavioural” treatment of the behavioural pathway. My personal approach has avoided any attempts to influence breathing voluntarily (other than by relaxation) but has been directed at giving the patient insight into the mechanism of his symptoms and, I hope, breaking the vicious circle alluded to earlier, as follows.

It is usual during the consultation with the patient to suspect that the symptoms may be due to overbreathing rather than to organic disease. This may be due either to the way in which the symptoms are described or to the patient’s manner; in some cases failure to find sufficient evidence of organic disease raises the suspicion. I prefer not to discuss this possibility with the patient at this stage, but consider that the next crucially important step is the 20 deep breaths test—a convenient time is during physical examination. Thus I say, “Would you mind taking 20 deep breaths, and if you feel odd in any way stop and tell me what you feel?” If the result of this test is negative, the suspicion of hyperventilation syndrome is unlikely to be correct. But if the 20 deep breaths test gives a positive result—that is, if it reproduces the patient’s symptoms wholly or in major part—then the index of suspicion is considerably heightened.

From now on further support for the diagnosis and treatment proceed together. I prefer not to tell the patient that I believe his symptoms are due to overbreathing and are secondary to anxiety or tension or whatever it may be; the patient is likely to misinterpret my meaning, and suspect that I am implying that it is all due to “nerves” and resent the perceived implication. I avoid this potential misunderstanding by allowing the patient the opportunity to identify with the hyperventilation syndrome by explaining that:

1 He or she has the common and interesting problem of severe symptoms without any adequate abnormality to account for them. It is well known that symptoms may be so severe that many patients feel that they are going to die during the attacks. This, one hopes, shows real interest in and understanding of the severity of the symptoms.

2 In these circumstances studies have shown that such patients have many things in common. They are almost always perfection-

ist—they like things to be right, and get irritated when things are not done to their high standards. If they are accompanied by the spouse, a nod of recognition may be noted when perfectionism is mentioned.

3 Such patients often have either anxiety or more commonly a depressive illness (I do not find it profitable to pursue the possibility of hysterical reactions).

4 Because personality is probably established early in childhood something more recent must have triggered the symptoms. One or more circumstances have been found commonly to precede the development of the symptoms: bereavement or separation (with examples); resentment (with examples); and illness or “fear” of illness—that is, uncertainty about whether something is seriously wrong (with examples).

By this time it is usually apparent whether or not patients are identifying with this picture. I then ask directly whether they have recognised anything of themselves in what I have said. If they do, and they often say that it describes them with uncanny accuracy, it is they who have made the diagnosis and therefore have the crucially important insight into the hyperventilation reaction. The diagnosis has not been imposed.

I explain that this insight will in itself either remove the tendency to further attacks or, more likely, reduce their frequency; it will certainly remove the frightening element of the experience should attacks recur. This reassurance can be reinforced by offering the backup of a rebreathing bag if they find that they cannot control an attack, but this is rarely necessary.

I routinely review the patient after about six weeks in case uncertainties have recurred, and especially because failure to improve implies either that the diagnosis is wrong, that further explanation is needed, that depression has not been recognised, or that antidepressive treat-

ment has been inadequate. Our early experience was that whereas a patient might gain insight into the nature of the reaction he did not improve if he remained depressed.² Treatment to lift the depression (for example, amitriptyline 50–75 mg at night) was essential. This has been repeatedly reinforced by my subsequent experience, which has also confirmed that hyperventilation syndrome as a hysterical reaction tends to persist.

It is important to remember that hyperventilation is merely a reaction that an otherwise “normal” individual with a particular type of personality may develop in response to events that have occurred in his or her life. The assessment is not complete until these precipitating events have been identified—especially because, though this is not usual, they may well include an organic illness.

Finally, we might say that there are no such things as facts in medicine, only the best hypotheses of the day. I offer this account of factors playing a part in the hyperventilation syndrome and its management as a hypothesis for you to judge. I will have succeeded if you choose to test it for yourselves, because when it is refuted a better hypothesis must emerge.

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