Filip Olekšák, Peter Ďurdík, Ľubica Jakušová, Tomáš Turčan, Peter Bánovčin

Department of Children and Adolescents, Martin University Hospital, Slovakia

Young and exhausted

Introduction

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) is a complex, multisystem and often debilitating disorder of unknown aetiology [1]. It is a complicated disease characterised by at least six months (in paediatrics 3 months) of extreme fatigue that is not alleviated by rest and a group of other symptoms that are constant for a period of time [1].

Post-exertional malaise (PEM) and delayed recovery are core symptoms and the most useful when making a diagnosis [3]. PEM involves a constellation of substantially disabling signs and symptoms that occur in response to physical, mental, emotional, and spiritual overexertion [2]. In many people with chronic fatigue syndrome, the disorder begins suddenly, often after a flu-like infection or after an episode of physical or mental trauma. The diagnosis of CFS/ME relies on the typical clinical presentation and the exclusion of other causes of fatigue. A diagnosis can be made by taking a thorough history, examining the patient, and using blood tests to screen for other causes of fatigue [3]. Up until now there was no test to confirm the diagnosis of CFS/ME. The two-day cardiopulmonary exercise testing (CPET) is becoming a new diagnostic method that can be used in case of suspicion of CFS/ME in attempt to evaluate the presence of PEM [5]. CPET provides a wealth of data on the dynamic function and coordination of the heart, lungs and muscles, as well as on the efficiency of gas exchange between mitochondria and the surrounding air, even in patients complaining about PEM. In the last 2 years, there has been a boom in using CPET as a diagnostic tool for PEM as a core symptom of CFS. Results of this testing, used in centres for patients with CFS/ME, in small cohorts of adults, have been published. One study, applying single one-day CPET in patients with CFS/ME in paediatrics, was published in 2007 [7]. None of these reports and studies highlighted breathing pattern disturbances as a possible cause of the chronic fatigue itself, nor did they mention the occurrence of dysfunctional breathing in patients with CFS. Dysfunctional breathing (DB) is highly prevalent and is overlooked mainly in adolescents and often attributed to behavioural changes during adolescence [6]. Chronic fatigue might be a symptom of DB in adolescents. We present a case study supporting this claim.

Case report

In the case report, we present a 13-year-old patient with chronic debilitating fatigue who meets the criteria for CFS/ME. The patient and patient's parents reported 6 months of fatigue, which was not improved even after an adequate period of sleep and very low physical performance. According to the parents, the patient has difficulty concentrating, is morose most of the day and reports limb twitching and paraesthesia. The patient was examined in detail by a paediatrician (anamnestic unclear cause, resting tachycardia in the physical examination, laboratory tests within normal limits, serum minerals within normal limits), endocrinologist (normal hormonal profile for a given age, Tanner stage 3), infectologist (serology for typical viruses negative), psychologist (normal cognitive functions). For a history of resting tachycardia, the patient was examined by a cardiologist, where no cardiogenic cause

Address for correspondence: Peter Ďurdík, Department of Children and Adolescents, Martin University Hospital, Slovakia; Jessenius Faculty of Medicine in Martin, Comenius University in Bratislava, Slovakia; e-mail: peter.durdik@unm.sk
DOI: 10.5603/ARM.a2020.0196
Received: 05.08.2020

ISSN 2451–4934

Table 1. CPET data obtained on day 1, day 2 of the examination and after 3 months of rehabilitation

	CPET on day 1	CPET on day 2	CPET after 3 months
Exercise duration [min]	6:32	7:00	7:50
VO_2 peak [mL/kg/min]	34.5	36.5	36.7
WR peak [W/kg]	3.26	3.68	3.8
HR peak [1/min]	187	197	199
WR at anaerobic threshold [AT] [W/kg]	0.72	1.5	1.68
VO_2 at AT [ml/kg/min]	19.0	23.0	22.3
VE/VCO_2 [1]	34.1	34.6	27.3
Max Vt [mL]	1.18	1.33	2.15
Max respiratory rate [1/min]	72	81	41
EtCO2 resting [mm Hg]	25	23	36

CPET — cardiopulmonary exercise testing; VE — minute ventilation; WR — work rate; HR — heart rate; VT — tidal volume; EtCO2 — end-tidal carbon dioxide; VO_2peak — peak oxygen consumption; VE/VCO_2 — minute ventilation/carbon dioxide production

of fatigue was demonstrated, sinus tachycardia was present, and the patient was recommended head-up-tilt test, which showed the presence of postural orthostatic tachycardia. Due to the idiopathic nature of the difficulties and the excluded secondary cause, a two-day protocol examination by cardiopulmonary exercise testing was indicated. Written consent was taken and documented. Before the examination, we performed resting spirometry (FEV1 106% of predicted, FVC 101% of predicted), resting ECG (sinus tachycardia) and a shortened Schellong test with a tachycardic response to orthostasis immediately after standing up and after 3 minutes of standing. Subsequently, standard CPET using treadmill (Itam, Poland) with individualised protocol with progressive increase in workload until exhaustion and breath-by-breath analysis of exhaled gases (Geratherm, Germany) was performed. On the first day, basal values were determined, the patient subjectively tolerated the examination well, the exercise ended prematurely due to subjective fatigue and a feeling of lack of air. On the second day, under the same conditions, the CPET was repeated, during which the patient also terminated the exercise prematurely but a half minute later than on the first day. Using this methodology, the patient did not meet diagnostic criteria for PEM and subsequently CFS/ME (decrease in monitored values on the second day of the examinations) (Table 1, Figure 1G). On both days, during exercise, a bizarre pattern of respiration with malposition of the respiratory act to the large airways was observed by noticing the flow-volume loop. Analysis of the respiratory pattern identified an erratic respiratory pattern (Figure 1A) with low resting EtCO2 (Table 1) and tachypnoea at maximal workload with dominant ventilation of dead space (Figure 1B, 1C). The consequence of this pattern of respiration is a chronic state of hypocapnia and respiratory alkalosis, which is metabolically compensated in the patient. Fatigue and increased heart rate are expected clinical manifestations. Respiratory rehabilitation was recommended to the patient in order to fixate the correct breathing patterns (diaphragmatic breathing) and psychological guidance. The patient was subsequently retested 3 months after the start of physiotherapy and psychotherapy at the request of the parents. Retesting showed significant improvements in the monitored parameters (Table 1, Figure 1G) as well as in the clinical condition of the patient.

We observed changes in breathing pattern and subsequent normalisation of observed cardiopulmonary parameters during resting capnography and exercise test. The patient increased tidal volumes (Vt) with preserved inspiration capacity (IC) during exercise. Resting ETCO2 tracing showed normal values. The patient's overall fitness increased, an adequate resting respiratory pattern was present (Figure 1D), ventilatory efficiency was adjusted (VE/VCO2 in Table 1, Figure 1G), and the patient reported a subjective increase in energy. Prior to the examination, we performed the Schellong test on the patient, in which there were no signs of postural orthostatic tachycardia.

Discussion

Dysfunctional breathing is a condition of the airways characterised by an irregular breathing pattern and changes in the airways that cannot be

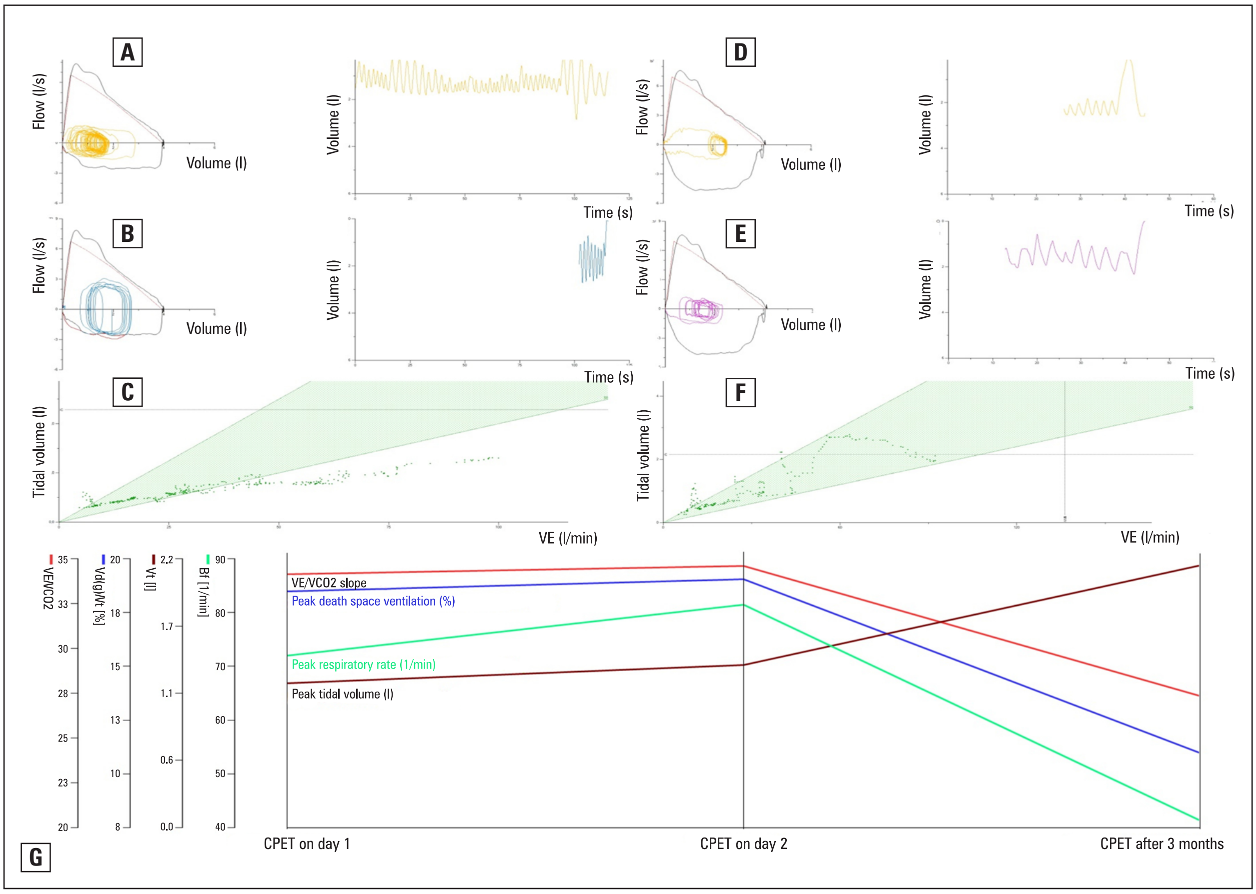

Figure 1. A. Resting flow volume loop, first day of examination. **B.** Flow volume loop at the top of exertion, first day of examination. **C.** Breathing pattern, first day of examination. **D.** Resting flow volume loop, examination after 3 months. **E.** Flow volume loop at the top of exertion, examination after 3 months. **F.** Breathing pattern, examination after 3 months

attributed to a specific diagnosis and that cause respiratory and non-respiratory problems [4]. It is not a disease process, but rather changes in respiratory patterns that disrupt normal respiratory processes. However, DB can coexist with diseases such as bronchial asthma or heart disease. The main symptom is shortness of breath or air hunger, associated with non-respiratory symptoms such as dizziness, palpitations, cervical spine pain or fatigue [6]. It also plays a role in chronic fatigue, neck and back pain, fibromyalgia, and some aspects of anxiety and depression [6].

The most common type of DB is hyperventilation syndrome, which is defined as respiration exceeding metabolic requirements, reducing blood carbon dioxide concentrations below normal values [4]. This changes the pH of the blood, increases the alkalinity and thus triggers a number of adaptive changes that cause symptoms. These conditions are non-somatic in nature and their treatment consists of respiratory rehabilitation with various techniques (diaphragmatic breathing, Feldenkrais method, Buteyko method, Pilates) and psychotherapy in order to control impulsive changes in the respiratory pattern in various situations [6]. Rehabilitation and physiotherapy seem to be efficient enough and should be encouraged in patients with DB.

Conclusions

CPET confirmed the presence of DB in the patient based on the low resting value of $EtCO_2$, the existence of a chaotic pattern of respiration during resting and exercise with the presence of tachypnoea (with very low ventilatory efficiency) in maximal exertion. Diagnosis of DB using CPET is one of the methods of DB diagnostics. Proper respiratory rehabilitation and psychological guidance resulted in the patient fixing the respiratory pattern and subsequently eliminating the primary cause of the examination — chronic fatigue. Patients with CFS/ME are a common paediatric problem. The current possibilities of diagnostics are enriched by the possibility of performing CPET, which can be a benefit in differential diagnostics as well as in confirming the diagnosis. Patients with CFS/ME and/or postural orthostatic tachycardia should be checked for the presence of DB as a treatable cause of clinical symptoms.

Conflict of interest

No conflict of interest to declare.

References:

1. Clayton EW. Beyond myalgic encephalomyelitis/chronic fatigue syndrome: an IOM report on redefining an illness. JAMA. 2015; 313(11): 1101–1102, doi: 10.1001/jama.2015.1346, indexed in Pubmed: 25668027.
2. Davenport TE, Lehnen M, Stevens SR, et al. Chronotropic intolerance: an overlooked determinant of symptoms and activity limitation in myalgic encephalomyelitis/chronic fatigue syndrome? Front Pediatr. 2019; 7: 82, doi: 10.3389/fped.2019.00082, indexed in Pubmed: 30968005.
3. NICE. National Institute for Health and Clinical Excellence (NICE) clinical guideline CG53. Chronic fatigue syndrome/myalgic encephalomyelitis (or encephalopathy): Diagnosis and management. 2007.
4. Boulding R, Stacey R, Niven R, et al. Dysfunctional breathing: a review of the literature and proposal for classification. Eur Respir Rev. 2016; 25(141): 287–294, doi: 10.1183/16000617.0088-2015, indexed in Pubmed: 27581828.
5. Stevens S, Snell C, Stevens J, et al. Cardiopulmonary exercise test methodology for assessing exertion intolerance in myalgic encephalomyelitis/chronic fatigue syndrome. Front Pediatr. 2018; 6: 242, doi: 10.3389/fped.2018.00242, indexed in Pubmed: 30234078.
6. Chaitow, L., Bradley, D., & Gilbert, C. Recognizing and Treating Breathing Disorders E-Book, 2014, Elsevier Health Sciences.
7. Takken T, Henneken T, van de Putte E, et al. Exercise testing in children and adolescents with chronic fatigue syndrome. International Journal of Sports Medicine. 2007; 28(7): 580–584, doi: 10.1055/s-2007-964888.