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Case Report

Vocal Cord Dysfunction in Former World Trade Center (WTC) Rescue and Recovery Workers and Volunteers

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Background *Vocal cord dysfunction (VCD) is a condition characterized by paradoxical partial adduction of the vocal cords on inspiration. It has been associated with exposures to irritants, as well as with psychological illnesses and conditions. Workers who participated in the recovery of the WTC disaster site were exposed to a large amount of irritants as well as considerable psychological stressors. We describe the clinical characteristics of 10 symptomatic former WTC workers diagnosed with this condition, as well as the frequency of spirometric findings suggestive of variable extrathoracic obstruction.*

Methods *Workers who became symptomatic after their WTC work experience have been evaluated clinically by a multidisciplinary team at an academic medical center. The evaluation included history, physical examination, chest radiograph, blood tests, and pre- and post-bronchodilator spirometry in all patients. Additional evaluations and diagnostic tests included otolaryngological evaluation with flexible rhinolaryngoscopy and stroboscopy, gastroenterological and psychiatric evaluations. A randomly selected sample of 172 spirometry results were reviewed for evidence of inspiratory flow limitation.*

Results *Variable extrathoracic obstruction was found in 18.6% of the spirometries. Ten patients were diagnosed with VCD. In addition to symptoms suggestive of co-morbid conditions (particularly rhinitis and acid reflux disease), most of the 10 patients had (1) hoarseness, (2) dyspnea that was not associated with bronchial hyperreactivity, or (3) dyspnea associated with asthma, with either mild bronchial hyperreactivity and/or poor response to asthma treatment.*

Conclusions *VCD appears to be part of the spectrum of airway disorders caused by occupational exposures at the WTC disaster site. Further study of this association is warranted.* Am. J. Ind. Med. 51:161–165, 2008. © *2008 Wiley-Liss, Inc.*

KEY WORDS: occupational medicine; inhalation injury; vocal cord dysfunction; asthma; respiratory diseases; irritant exposures

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INTRODUCTION

Vocal cord dysfunction (VCD, also known as episodic paroxysmal laryngospasm or laryngeal dyskinesia, paradoxic vocal fold motion, Munchausen's, functional inspiratory, or psychogenic stridor, nonorganic functional or psychogenic upper airway obstruction, and factitious asthma) is a condition characterized by paradoxical partial inspiratory adduction of the vocal cords. It causes dyspnea and stridor, and can mimic (but also coexist with) asthma [Christopher et al., 1983; Martin et al., 1987; Thomas et al., 1999]. It has been associated with exposures to respiratory irritants, often in the occupational setting [Perkner et al., 1998], as well as with psychological illnesses and conditions [Selner et al., 1987; Brown et al., 1988].

Workers who participated in the rescue, recovery, and clean up of the WTC disaster site were exposed to a large amount of irritants (combustion products, alkaline cement dust, polyaromatic hydrocarbons, dust) [Lioy et al., 2002; Offenberg et al., 2003], as well as considerable psychological stressors. Screening data have demonstrated a substantial prevalence of physical [Prezant et al., 2002; Centers for Disease Control and Prevention, 2004b], and mental health symptoms [Centers for Disease Control and Prevention, 2004a] among these workers. A dedicated clinical program was created at our institution to provide individualized in-depth evaluations on former WTC workers and volunteers, with an all inclusive range of occupations. A frequent observation was the presence of inspiratory flow limitation in spirometry, which, together with the described exposures and consistent symptoms and clinical findings, prompted a diagnostic investigation for VCD in several patients. In this article, we describe the frequency of that spirometric abnormality, and the clinical characteristics of 10 symptomatic former WTC workers diagnosed with VCD.

METHODS

The WTC Health Effects Treatment Program was established in January 2003 by the Mount Sinai-I. J. Selikoff Center for Occupational and Environmental Medicine to provide clinical services to the former WTC rescue and clean up workers and volunteers. The evaluation included history (including occupational history), physical examination, chest radiograph, blood tests, and pre- and post-bronchodilator spirometry in all patients. Additionally, the following evaluations and diagnostic tests have been obtained based on standard clinical indications: (1) detailed pulmonary function tests (complete PFT and/or methacholine bronchoprovocation, 40% of the patients); (2) otolaryngological evaluation with flexible rhinolaryngoscopy (40% of the patients), with detailed assessment of laryngopharyngitis findings [Belafsky et al., 2001] and stroboscopy in a selected group of patients; (3) psychiatric evaluation (45% of the patients); (4) chest, and paranasal sinuses CT scans, and (5) esophagogastroduodenoscopy (EGD) and 24-hr pH monitoring examinations.

Due to the suspicion of VCD in this population, a randomly selected sample of spirometry results from 172 patients was reviewed for evidence of inspiratory flow limitation, suggestive of variable extrathoracic airflow obstruction. The criteria for inspiratory flow limitation included the appearance of the flow-volume curve (a flattened inspiratory loop) and a $FIF_{50\%}/FEF_{50\%}$ ratio of less than 1.0. A diagnosis of asthma required consistent clinical history and findings on examination, and evidence of bronchial hyperreactivity as assessed by methacholine bronchoprovocation test (a minimum decrease in FEV_1 of 20% with an inhaled concentration of methacholine not exceeding 10 mg/ml). Psychiatric diagnoses were confirmed by psychiatrists. Gastroesophageal reflux disease was diagnosed at three incremental levels: (1) clinically: requiring typical symptoms (heartburn, food regurgitation, dysphagia) with an average twice weekly frequency, reduced at least by 50% with a proton pump inhibitor medication; (2) findings by EGD; (3) a pH monitoring showing distal esophageal acid exposure (pH below 4) more than 4% of the time.

The Mount Sinai School of Medicine Institutional Review Board approved the protocol for this study, with an exemption from the requirement for informed consent.

RESULTS

Inspiratory flow limitation was identified in 18.6% (32/172) of the sampled spirometries by the appearance of the flow-volume curve (an example is illustrated in Fig. 1),

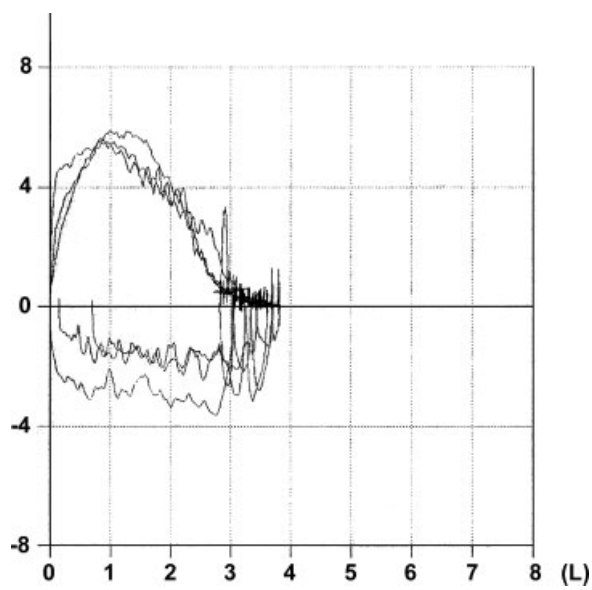

FIGURE 1. Spirometric flow-volume curve in one of the patients (no. 8), demonstrating a decreased inspiratory flow, suggestive of variable extrathoracic airway obstruction.

and a $FIF_{50\%}/FEF_{50\%}$ less than 1.0. In 10 patients, a diagnosis of VCD was established by flexible laryngoscopic examination, with the well described partial anterior adduction of the vocal folds during inspiration, with a posterior diamond-shaped glottic gap [Selner et al., 1987; Gallivan et al., 1996]. All patients had prominent laryngopharyngitis findings, suggestive of acid-reflux related disease [Belafsky et al., 2001]. Most patients (8/10) had spirometric evidence suggestive of inspiratory flow limitation and variable extrathoracic upper airway obstruction. The characteristics of the patients (including their comorbidities) are summarized in Table I. Their occupations at the WTC site included police officers (n = 2), firefighter, paramedic, volunteer, sanitation workers, building cleaner, administrator, photographer, and electrician. Five patients were lifetime nonsmokers, and five were former smokers [American Thoracic Society, 1978]. All 10 patients had symptoms suggestive of chronic rhinitis, asthma, and gastroesophageal reflux disease. Most of the 10 patients had (1) chronic intermittent, usually mild hoarseness, (2) dyspnea that was not associated with bronchial hyperreactivity (and thus, asthma, n = 6) or, (3) dyspnea that was associated with asthma (n = 4) with mild bronchial hyperreactivity, and/or poor response to asthma treatment. These characteristics prompted a search for VCD as an alternative or associated diagnosis. Of the nonasthmatic patients, one patient had open lung biopsy documented mild constrictive bronchiolitis (case No. 10).

DISCUSSION

We described 10 cases of VCD diagnosed in workers who developed chronic illnesses apparently as a consequence of their exposures at the WTC disaster site in the fall of 2001. As shown on Table I, these workers exhibited most of the most prevalent conditions among symptomatic WTC-exposed individuals: upper and lower airway disease, gastroesophageal reflux disease, and/or psychiatric diagnoses [de la Hoz et al., 2008].

VCD is a condition that has been well described in individuals exposed to large amounts of respiratory irritants [Perkner et al., 1998; Huggins et al., 2004; Galdi et al., 2005; Allan et al., 2006] and to psychological stressors [Selner et al., 1987; Brown et al., 1988], including war experiences [Craig et al., 1992; Das et al., 1999; Morris et al., 1999]. The diagnosis of VCD requires the exclusion of more organic laryngeal conditions, such as vocal cord paralysis or neoplasms. Symptoms of upper airway irritation, including rhinitis, sinusitis, and laryngopharyngitis were extremely prevalent in screening surveys of former WTC workers [Prezant et al., 2002], and our own clinical experience is corroborative [de la Hoz et al., 2008]. Acute inflammation can possibly explain laryngitis symptoms, but perpetuation of the inflammatory process and/or altered autonomic balance have been invoked as potential explanation for the chronicity of VCD following the inciting episode [Ayres and Gabbott, 2002]. Symptoms of gastroesophageal reflux disease have been very prevalent in symptomatic former WTC workers [de la Hoz et al., 2008], and GERD was diagnosed as described before in all 10 patients in this report. The reason for this association remains unclear. Since all the cases with VCD in this series have GERD and laryngopharyngeal reflux (LPR) disease [Koufman et al., 2002], it is quite likely that continued vocal cord acid exposure plays a role in causing and/or perpetuating VCD [Gallivan et al., 1996]. Finally, the substantial WTC-related psychopathology of so many of these patients is quite likely to play a role in the pathogenesis of VCD, as has been demonstrated in previous reports and in similar circumstances.

VCD is a very likely contributor to the symptom of dyspnea exhibited by all of these workers, not all of whom had irritant-induced asthma. Even those with asthma exhibited relatively mild bronchial hyperreactivity, with

TABLE I. Clinical Characteristics of 10 Symptomatic Former WTC Workers and Volunteers Diagnosed With Vocal Cord Dysfunction

	1	2	3	4	5	6	7	8	9	10
Age	43	56	50	54	39	70	35	60	37	36
Sex	F	M	M	M	F	F	M	M	F	M
WTC site arrival day	1	5	1	2	4	90	1	1	4	1
Exposure duration	6 weeks	20 weeks	16 weeks	10 weeks	23 weeks	12 weeks	10 weeks	3 weeks	9 weeks	8 weeks
IFL	−	+	+	+	−	+	+	+	+	+
Hoarseness	+	+	+	+	+	+	+	+	+	+
Asthma	+	+	−	+	−	−	+	−	−	−
Chronic rhinitis	Nonallergic	Nonallergic	Nonallergic	Allergic	Nonallergic	Nonallergic	Allergic	Nonallergic	Allergic	Nonallergic
GERD	Clinical	pH study	Clinical	EGD	EGD	Clinical	EGD	Clinical	Clinical	pH study
PTSD	Confirmed	Confirmed	—	Confirmed	—	Confirmed	—	—	—	Confirmed
Agoraphobia/panic	—	Confirmed	—	Confirmed	—	—	—	—	—	—

IFL, inspiratory flow limitation; GERD, gastroesophageal reflux disease; EGD, esophagogastroduodenoscopy; PTSD, post-traumatic stress disorder.

PC_{20} (provocative doses causing a decline of at least 20% in first–second vital capacity) at or near the clinical cutoff of 10 mg/ml. The majority of the reported cases had spirometric findings suggestive of variable extrathoracic airway obstruction, which we have found in approximately 18.6% of the spirometries in this patient population. One nonasthmatic VCD patient (case no. 10) had biopsy proven mild constrictive bronchiolitis, similar (but with less functional impairment) to what has been reported by others [Mann et al., 2005].

Although our level of suspicion for VCD was high for many of our patients, the diagnosis has been difficult to make. This is frequently reported in the literature, where it has been noted that the diagnostic yield can be low, even with careful laryngoscopic and/or stroboscopic examination, if the patients are not symptomatic at the time of that examination. As with 90% of our patient population, all of the cases reported here live at a considerable distance from our medical center, and were always examined during the intervals between their attacks. Methacholine challenge testing provoked VCD in only one of the patients (case no. 8).

Asthma practice guidelines have emphasized the importance of considering a diagnosis of VCD in patients with asthma symptoms who do not respond to sufficient or even large doses of medications, or who do not show bronchial hyperreactivity by appropriate testing [National Asthma Education and Prevention Program, 1997; Thomas et al., 1999]. Our experience with the former WTC workers underscores the need to consider this diagnosis into account in the setting of inhalation injuries, when substantial psychological comorbidities make it too easy for clinicians to dismiss reportedly severe symptoms as psychosomatic at best, or the result of malingering, at worst. Our patients' illness profile underscores the fact that WTC dust exposures affected the entire airway, form the nasal passages to the bronchioles. Future planning for medical response to disasters involving respiratory toxicants will have to include appropriate evaluation and management for the range of conditions that our patients presented, including VCD.

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