

A 38-year-old hairdresser with irritant-associated vocal cord dysfunction

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SUMMARY

The present case study reports the first case of a 38-year-old hairdresser with irritant-associated vocal cord dysfunction (VCD) due to alkaline persulfate, who was referred on suspicion of occupational asthma. Several tests were performed, including specific inhalation challenge and upper airway endoscopy. During the specific inhalation challenge to alkaline persulfate, the patient experienced dysphonia and a non-significant decrease in forced

expiratory volume in 1 second on spirometry. Upper airway endoscopy was then performed and revealed VCD. A specific inhalation challenge test is therefore essential in cases of VCD to exclude possible concomitant occupational asthma.

KEY WORDS: irritant-associated vocal cord dysfunction; persulfate salts; occupational asthma

A 38-YEAR-OLD WOMAN, who had worked as a hairdresser for 20 years, was referred to our department with suspected occupational asthma. Her clinical history was unexceptional, apart from isolated, mainly night-time chronic cough at the age of 19. After treatment with omeprazole for gastro-oesophageal reflux without success for cough, she consulted a chest physician. The skin-prick test (SPT) with common respiratory allergens and the carbachol bronchial challenge test were performed, with negative results. The final diagnosis was that of psychogenic cough; the patient underwent no treatment for more than 15 years.

Two and a half years previously, as her hairdressing activity increased, she had presented with dyspnoea, chest tightness, noisy breathing and dysphonia in addition to the cough, particularly in association with bleaching operations. As she did not stop working, these symptoms gradually worsened. Peak expiratory flow (PEF) curves showed significant variability (21%) between working and non-working days, and her chest physician initiated treatment for asthma 1 year before our examination. Due to persistent symptoms, she was then referred to our department to complete the diagnostic procedure in November 2010. Total serum immunoglobulin E was normal. SPT was positive for *Dermatophagoides pteronyssinus* and birch pollen, but negative for latex, persulphates and quaternary ammonium compounds. Complete plethysmography was in the normal range, and β^2 -agonist challenge test was also negative. The patient underwent a

methacholine challenge test, during which she experienced cough and mild dysphonia. When interrupted at a dose of 2000 μ g, forced expiratory volume in 1 second (FEV₁) decreased by only 5% relative to baseline. The challenge test was considered to be negative; nevertheless, possible bronchial hyperresponsiveness could not be ruled out.

A specific inhalation challenge (SIC) to persulphate salts was then performed. Briefly, the patient prepared a bleaching substance (alkaline persulphate) 30 cm away from her face in a challenge booth with independent extraction. The patient was then consecutively exposed for 2, 3, 5 and 10 min, resulting in a cumulative exposure time of 20 min, unless significant respiratory symptom and/or significant change (variation of FEV₁ $\geq$ 20%) in FEV₁ occurred within 10 min at each step. The patient underwent spirometry and a clinical examination every hour up to 6 h after the end of the SIC. After the second persulphate salt exposure, the patient experienced pronounced dysphonia, cough and sore throat that she described as her usual symptoms during bleaching. The patient had no symptoms of nasal response. During the test, the FEV₁ decreased by only 4% relative to baseline. As cough occurred during the tests, inspiratory curves were not interpretable. All symptoms, including dysphonia, disappeared within 2 h. Six hours following the SIC, another methacholine challenge test was performed and the patient experienced cough and mild dysphonia again, as FEV₁ decreased by only 7% relative to baseline. Upper airway endoscopy performed

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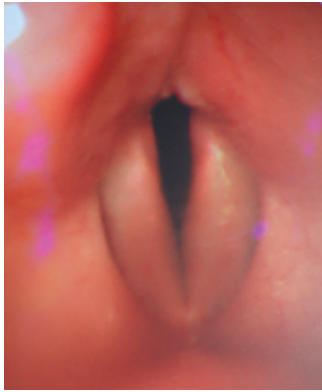


Figure Endoscopic view showing paradoxical adduction of the vocal cords with posterior glottic chink. This image can be viewed online in colour at <http://www.ingentaconnect.com/content/iatld/jitld/2012/00000016/00000001/art00025>

just before and after the SIC revealed vocal cord dysfunction (VCD), with oedema and paradoxical adduction of the vocal folds with posterior chink during inspiration (Figure). A diagnosis of irritant-associated VCD secondary to exposure to persulphate salts was made, and occupational asthma due to the same agent was formally excluded.

DISCUSSION

VCD has been defined as intermittent extrathoracic airway obstruction due to inappropriate movement of the vocal folds, mainly during inspiration.¹ Three forms have been described: psychogenic VCD, exercise VCD and irritant VCD.² Irritants include intrinsic factors (gastroesophageal reflux, etc.) and extrinsic factors (chemical or sensory irritants), as in our observation. Diagnosis is usually based on clinical symptoms of noisy breathing and dyspnoea, and is sometimes suggested by pulmonary function test anomalies. Confirmation, however, needs laryngoscopic evidence of vocal fold adduction.² The specific pathogenesis of VCD is unknown. The pathophysiology of irritant-associated VCD is hypothesised as an accentuation of the glottic closure reflex caused by various extrinsic stimuli that trigger closure as a protective response.³ It has also been reported that high histamine levels can trigger this syndrome even in normal persons³ or in the absence of proven allergy.⁴

Two main findings emerge from our case report. VCD is defined by the onset of symptoms after a single specific exposure to a respiratory irritant, onset of symptoms within 24 h after exposure, symptoms of wheezing, stridor, dyspnoea, cough or throat tightness, abnormal direct laryngoscopy and exclusion of other vocal cord diseases.⁵ The disease is usually associated with exposure to major irritants such as ammonia and organic solvents.⁵ Pre-existing VCD specifically aggravated by persulphate salts cannot be excluded in our case in view of the patient's clinical history. Functional dysphonia and psychogenic cough are most

characteristic of somatoform disorders seen in psychogenic VCD with conversion subcategory.⁶ However, the characteristic abnormalities seen on examination of the vocal folds during the SIC did not support this hypothesis according to Selner et al., who noted the importance of provocation challenges to triggering agents.⁷ To our knowledge, this is the first time that VCD has been associated with persulphate exposure. Our case also suggests that VCD can also be related to chronic exposure to mild respiratory irritants.

VCD is also a well known differential diagnosis of occupational asthma,^{1,2} although most chest physicians are not familiar with this diagnosis. The presence of dysphonia in this case was highly suggestive of VCD rather than occupational asthma.⁸ However, the diagnosis of occupational asthma is a complex procedure. Specific questionnaires, PEF curves and immunological tools have been shown to have only limited sensitivity and specificity for this diagnosis.⁸ A specific bronchial challenge test may be necessary to formally confirm or eliminate the diagnosis of occupational asthma.⁹ Clinicians should therefore refer their patients to more specialised centres, particularly in the presence of atypical clinical symptoms or an unsatisfactory course of asthma. The difficult diagnosis in our case illustrates the possible consequences for patients.

In conclusion, we describe here the first case of VCD associated with persulphate salt exposure. We propose that a negative bronchial specific challenge test could constitute a diagnostic criterion of VCD when occupational asthma is also suspected.

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R É S U M É

Les auteurs rapportent le premier cas de syndrome de dysfonctionnement des cordes vocales (VCD) liées aux persulfates alcalins chez une coiffeuse âgée de 38 ans, initialement adressée à notre consultation pour suspicion d'asthme professionnel. Plusieurs examens complémentaires ont été réalisés, dont un test de provocation bronchique réaliste aux persulfates alcalins et une endoscopie des voies aériennes supérieures. Pendant le test

de provocation bronchique, la patiente a présenté une dysphonie associée à une chute non significative du volume expiratoire maximal par seconde. L'endoscopie, réalisée après le test de provocation bronchique, a mis en évidence un VCD. Au total, dans les cas de VCD un test de provocation bronchique peut être nécessaire pour éliminer un éventuel asthme professionnel associé.

R E S U M E N

En el presente artículo se comunica el primer caso de un peluquero de 38 años de edad con disfunción de las cuerdas vocales (VCD), provocada por irritación con persulfato alcalino, a quien se remitió con presunción clínica de asma profesional. Se practicaron diversas pruebas, entre ellas una provocación por inhalación y una endoscopia de las vías aéreas superiores. Durante la provocación por inhalación de persulfato alcalino, el

paciente presentó disfonía y una disminución no significativa del volumen espiratorio forzado en el primer segundo en la espirometría. La endoscopia practicada después de la prueba de provocación reveló una VCD. En conclusión, en los casos de VCD es necesario practicar una prueba específica de provocación por inhalación, con el objeto de excluir la presencia eventual de un asma profesional concomitante.
