

Intermittent Asphyxia Syndrome Caused by a Bronchial Cast in the Subglottic Region

J. Ferreres-Franco,^a J. Blanquer Olivas,^a E. Pastor Esplá,^b S. Borrás Pallé,^a G. Galán Gil,^c and A. Jordá Miñana^a

^aUnidad de Cuidados Intensivos, Hospital Clínico de Valencia, Valencia, Spain.

^bServicio de Neumología, Hospital San Joan, Alicante, Spain.

^cServicio de Cirugía Torácica, Hospital Clínico de Valencia, Valencia, Spain.

We report the case of an intensive care unit patient with acute respiratory failure caused by severe community-acquired pneumonia with empyema. She required orotracheal intubation and mechanical ventilation. Following extubation the patient's condition improved until the onset of several choking episodes caused by a dislodged laryngotracheal cast in the subglottic region. We discuss the differential diagnosis of upper airway obstructions and of the entities related to bronchial cast formation, in particular the clinical and pathophysiological features of plastic bronchitis and the treatment options available.

Key words: Choking. Intermittent airway obstruction. Bronchial cast. Plastic bronchitis.

Síndrome asfíctico intermitente provocado por molde bronquial en zona subglótica

Se presenta el caso de una paciente ingresada en la unidad de cuidados intensivos por insuficiencia respiratoria aguda secundaria a una neumonía comunitaria grave con empiema, que requirió intubación orotraqueal y ventilación mecánica. Tras su extubación cursó con buena evolución hasta presentar varios episodios de crisis asfícticas intermitentes secundarias a una obstrucción por el desplazamiento de un molde laringotraqueal en la zona subglótica. Se discute el diagnóstico diferencial de las obstrucciones respiratorias de la vía aérea superior, así como el de las entidades relacionadas con la formación de moldes bronquiales, con especial atención a las bronquitis plásticas, sus características clínicas, fisiopatológicas y posibilidades terapéuticas.

Palabras clave: Crisis asfíctica. Obstrucción intermitente de la vía aérea. Molde bronquial. Bronquitis plástica.

Introduction

A bronchial cast is a complication of uncertain pathogenesis and is often associated with diseases involving mucus hypersecretion. The organization of the mucus into the shape of the tracheobronchial tree, also known as plastic bronchitis, is a rare clinical entity that occasionally leads to acute respiratory failure.¹

We report the case of a patient with a laryngotracheal cast that gave rise to choking episodes caused by intermittent glottic obstruction. The patient had previously been ventilated and extubated following a severe community-acquired pneumonia. We establish the rare nature of the case and discuss the differential diagnosis of upper airway obstructions and the entities related to

bronchial cast formation. In particular, we discuss plastic bronchitis, its clinical and pathophysiological features, and treatment options.

Case Description

The patient was a 50-year old woman who was admitted to the intensive care unit because of acute hypoxemic respiratory failure within the context of severe community-acquired pneumonia. She was a 50-pack-year smoker with an otherwise unremarkable medical history. The week before she was admitted she had experienced pain that radiated from the left side of the chest and upper abdominal region to her arm, of 48 hours' duration. In the following days she developed progressive dyspnea on minimal exertion, nausea, and vomiting in spite of antibiotic treatment with fosfomycin. She felt feverish intermittently and the pleuritic pain in the left side of her chest persisted. She then came to our emergency department with a temperature of 37.5°C, tachypnea (25 breaths/min), and tachycardia (120 beats/min). Auscultation revealed an overall decrease in vesicular breath sounds in the left side of the chest with rhonchi and diffuse wheezes during

Correspondence: Dr. J. Ferreres-Franco.
C/ Tres de Abril de 1979, 46023 Valencia, España.
E-mail address: ferreres@ono.com

Manuscript received March 7, 2005. Accepted for publication March 15, 2005.

expiration. The chest x-ray showed a shadow on the left side but no mediastinal shift. Blood gas analysis (fraction of inspired oxygen, 0.21) revealed a pH of 7.38, PaCO₂ of 46 mm Hg, PaO₂ of 42 mm Hg, and HCO₃ concentration of 26 mEq/L. Further findings were leukocytosis with left shift and a sodium level of 113 mEq/L. Given the clinical situation and blood gas findings, it was decided to admit the patient to the intensive care unit, where oxygen therapy and treatment with bronchodilators, corticosteroids, and empirical antibiotics (cefotaxime and levofloxacin) were started. Orotracheal intubation and mechanical ventilation were provided for 6 days. Results for pneumococcal antigenuria and urine *Legionella* species, blood cultures, urine cultures, and respiratory serology were negative. The chest sonogram revealed a loculated pleural effusion, and *Streptococcus milleri* sensitive to the antibiotics prescribed was isolated in fluid obtained by diagnostic thoracentesis. A pleural drain was placed, and 2 L of purulent fluid was removed. Urokinase for intrapleural fibrinolysis was administered to increase the drain output. The presence of a mucus plug or intrabronchial masses was ruled out by fiberoptic bronchoscopy. The patient was weaned from mechanical ventilation without complications. Her clinical condition was very good, but 48 hours after extubation several intermittent choking episodes began with coughing attacks followed by severe dyspnea and tachypnea. There was use of accessory muscles and absence of vesicular murmurs on both sides. Oxygen saturation fell below 50%. Sometimes the choking was accompanied by laryngeal stridor, and the episodes occasionally responded to the administration of bronchodilators, corticosteroids, and subcutaneous adrenaline. During one of such episodes it was decided to reintubate and ventilate the patient. After sedation, good oxygenation was achieved with ventilation by airbag mask with a Guedel tube. Fiberoptic bronchoscopy was performed through the tube as intermittent upper airway obstruction related to the previous orotracheal intubation was suspected. A 6-cm-long mucus cylinder plugging the subglottic and tracheal regions was observed (Figure 1). The plug was removed with an aspirator as its size prevented it from being extracted through the fiberoptic bronchoscopy tube. Pathology revealed that the fragment was fibrinous with secondary calcification and was composed of mucoid material (Figure 2). After removal of the bronchial cast, the patient experienced no other episodes of dyspnea. She was discharged to the respiratory medicine ward and later to home with no further problems.

Discussion

At first our patient's episodes of asphyxia were thought to be related to severe asthma given that the clinical signs were intermittent, hypoventilation was marked in both lung fields, and the patient initially responded to bronchodilators, corticosteroids, and subcutaneous adrenaline. However, the clinical picture consisting of improvement between attacks, with no evident residual wheezing after the severe hypoventilation episode, as well as the presence of stridor in some episodes and no history of asthma, all in a patient who had undergone several days of orotracheal intubation, led us to reconsider various causes of upper airway obstruction and to perform diagnostic fiberoptic bronchoscopy. Although stridor was not observed in some of the choking episodes, its absence was perhaps



Figure 1. A 6-cm-long pale yellow mucoid cylinder with a rubbery/plastic-like consistency.

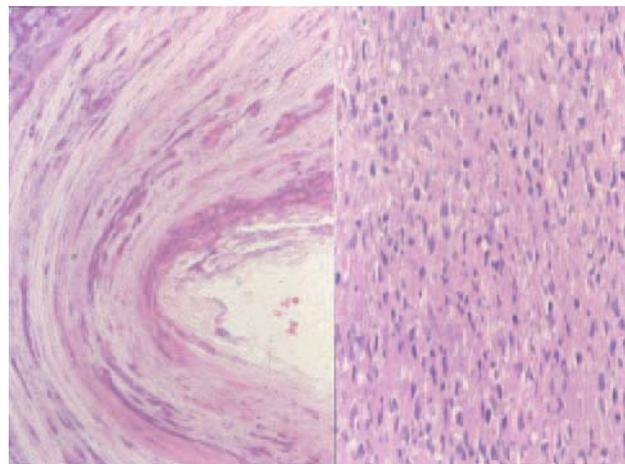


Figure 2. The cast under the microscope: histological samples show a structure composed of mucoid material with organized fibrous networks and inflamed cells with a lamellar arrangement (left). The sample is polymorphonuclear cell predominant with some eosinophils. No Charcot-Leyden crystals or Curschmann spirals are evident (right).

due to the intermittent nature of complete obstruction of the glottic lumen. The improvement observed on various occasions after treatment with inhaled β -2 agonists could have been caused by the shifting of the mucus cast as it was pushed down into the laryngotracheal lumen by the propellant. Later it would have risen to the glottis with the onset of coughing. Foreign body obstructions, especially reported in children, are clinically similar,¹ although intermittent presentation is rare.² In a case with clinical features similar to ours, Garvin³ reported a subglottic obstruction by a mucus plug in an asthmatic patient

TABLE
Entities Related to Intermittent Airway Obstruction

Angioneurotic edema
Tracheomalacia
Tonsillar hypertrophy
Thyroglossal duct cyst
Vocal cord polyp
Laryngeal spasms secondary to vocal cord dysfunction
Mediastinal masses
Arthritis of the arytenoids
Foreign bodies

who was taking antihistamines; he suggested that the mucus plug migration from the peripheral airways to the central airway was the result of increased viscosity of the mucus secondary to antihistamine use. However, unlike in the case we report, there was no history of intubation and no further reported episodes. Lung embolism may also be considered in the differential diagnosis as it can have a clinical presentation similar to that of impacted mucus plugs.⁴

The most common cause of upper airway obstruction after endotracheal intubation is glottal edema, which appears immediately after extubating a patient. It is not intermittent, and a typical finding is stridor. It was unlikely that tracheal stenosis or the formation of granuloma was the underlying cause of our patient's condition because endotracheal intubation was of short duration, and the intermittent choking attacks began only 48 hours after extubation. Glottal obstruction by tracheal membranes after short-term intubation has been reported,^{5,6} but because of the rare nature of our patient's clinical picture and the intermittency of the attacks, such an obstruction was not probable.

The most common cause of upper airway obstruction in the general population is probably angioneurotic edema,⁷ which gives rise to intermittent episodes. Other less frequent entities that may manifest acutely or intermittently are listed in the Table. All of these causes were reasonably ruled out for our patient after fiberoptic bronchoscopy.

This case may be considered as a type of plastic bronchitis given that the cast had molded to the shape of the laryngotracheal zone. Plastic bronchitis is usually associated with diseases that involve bronchial hypersecretion such as allergic bronchopulmonary aspergillosis and asthma (the most common entities), bronchiectasias, cystic fibrosis (mucoviscidosis), etc. Plastic bronchitis has also been reported in pulmonary processes, as in the case of our patient.⁸ When it cannot be related to any of these entities, it is considered idiopathic.^{9,10} Casts may form at any point in the tracheobronchial tree, but the literature yields many more cases of plastic bronchitis, with striking examples of casts bearing perfectly defined bronchial branches.¹¹ Our review of the literature revealed no cases of a tracheal cast intermittently obstructing the glottic lumen, however. Sanerkin et al¹² hypothesized that underlying such entities are common immunological and allergic processes affecting the bronchial mucosa, leading to hypersecretion of thick mucus. Possible contributing factors could be poor hydration and the use of antihistamines.³ Tracheal trauma caused by prolonged intubation¹³ or by transtracheal oxygen catheters¹⁴ could be another such factor since the use of oxygen at high concentrations leads to tracheobronchitis that, along with endotracheal tube inflation, may give rise to loss of ciliary epithelium and impaired mucus flow.¹⁵

Bronchial casts characteristic of plastic bronchitis are usually pale yellow and have a plastic consistency.^{8,10} The casts' fibrous mucoid material with cell debris

favors molding to the shape of the tracheobronchial tree. The typically lamellar arrangement of a cast can be seen under the microscope.

In our patient, removal of the cast through the bronchoscopic channel was not possible because of the size of the cast. A conventional aspiration probe was therefore chosen. The mucus cast was grasped by suction so that both the probe and the cast were removed en bloc. If the cast had adhered more strongly to the endothelium, it would probably have been necessary to use forceps to grasp it and remove it in pieces through a rigid bronchoscope.

Plastic bronchitis is managed by humidification of the secretions, prescription of antibiotics if there are underlying infections, and administration of β -2 adrenergic agents. Corticosteroid use is also recommended in some cases because of the possibility of an underlying immunoallergic process.

In conclusion, if a patient experiences choking caused by intermittent airway obstruction, as airway clearance and oxygenation is performed, differential diagnosis should include the possibility of a bronchial cast plugging the subglottic region. This diagnosis should be added to the extensive range of possibilities reported in the literature.

REFERENCES

- Noizet O, Leclerc F, Leteurtre S, Bricchet A, Pouessel G, Dorkenoo A, et al. Bronchitis mimicking foreign body aspiration that needs a specific diagnostic procedure. *Intensive Care Med.* 2003;29:329-31.
- Blaschke U, Cheng EY. Foreign body in upper airway. Unsuspected cause of obstruction. *Postgrad Med.* 1989;86:235-7.
- Garvin JM. Nearly fatal partial obstruction of the upper airway. *Postgrad Med.* 1990;87:81-4.
- Bray ST, Johnstone WH, Dee PM, Pope TL Jr, Teates CD, Tegtmeier CJ. The "mucous plug syndrome." A pulmonary embolism mimic. *Clin Nucl Med.* 1984;9:513-8.
- Lewis S, Jardine A. An unusual case of postextubation stridor. *Anaesthesia.* 2003;58:601-2.
- Harbison J, Collins D, Lynch V, McNicholas WT. Acute stridor due to an upper tracheal membrane following endotracheal intubation. *Eur Respir J.* 1999;14:1238.
- Fraser RS, Muller NL, Colman N, Pare PD. Upper airway obstruction. In: Fraser RS, Muller NL, Colman N, Pare PD, editors. *Fraser and Pare's diagnosis of diseases of the chest.* 4th ed. Philadelphia: WB Saunders Co.; 1999. p. 2021-5.
- Kao NL, Richmond GW. Cough productive of casts. *Ann Allergy Asthma Immunol.* 1996;76:231-3.
- Park JY, Elshami AA, Kang DS, Jung TH. Plastic bronchitis. *Eur Respir J.* 1996;9:612-4.
- Jett JR, Tazelaar HD, Keim LW, Ingrassia TS III. Plastic bronchitis: an old disease revisited. *Mayo Clin Proc.* 1991;66:305-11.
- Studer S, Terry P. Bronchial casts. *N Engl J Med.* 2002;346:981.
- Sanerkin NG, Seal RM, Leopold JG. Plastic bronchitis, mucoid impaction of the bronchi and allergic broncho-pulmonary aspergillosis, and their relationship to bronchial asthma. *Ann Allergy.* 1966;24:586-94.
- Mah MW, Jones RL, Man GC, Skippen PW, York EL. Immediate postextubation upper airway obstruction by a mucous cast. *Crit Care Med.* 1990;18:797-8.
- Burton GG, Wagshul FA, Henderson D, Kime SW. Fatal airway obstruction caused by a mucous ball from a transtracheal oxygen catheter. *Chest.* 1991;99:1520-3.
- Cohen SP, Anderson PL. Mucoid impaction following nasal intubation in a child with an upper respiratory infection. *J Clin Anesth.* 1998;10:327-30.