Incidental Diagnosis of Bronchial Atresia After Chest Trauma☆

Atresia bronquial diagnosticada incidentalmente tras traumatismo torácico

To the Editor,

Young adults with no prior disease often undergo their first computed tomography (CT) as a result of high-energy accidents. Incidental findings are difficult to interpret due to the urgent nature of the situation and lack of previous information. We report the case of a patient who was diagnosed with bronchial atresia after high-energy trauma in a traffic accident. A 35-year-old patient was admitted after a high-energy traffic accident. On arrival he presented: BP 100/80 mmHg, heart rate 101 bpm, respiratory rate 15 breaths/min, and hypoventilation of the right hemithorax. There were no signs of cardiac tamponade or hemoptysis. A chest-abdominal CT was performed that showed right pneumothorax, adrenal hematoma, liver contusion, multiple fractures of the right ribs and the right lumbar transverse processes, and a nodular lesion in the right hilum with air-fluid level that showed a dubious vascular communication in the left upper lobe with distal hyperinflation (Fig. 1A). A right chest tube was placed to treat the pneumothorax. Initially, the main hypothesis regarding the nodular lesion in the left lower lobe was that it was caused by vascular aneurysm or pneumatocele, enhanced due to extravasation of the intravenous contrast medium. Within a few hours, the team of thoracic radiology specialists considered the possibility of bronchial atresia, despite this being a rarer and more unusual diagnosis.

Fiberoptic bronchoscopy performed in the intensive care unit did not reveal any significant bronchial changes or signs of bleeding. Absolute bed rest and continuous hemodynamic monitoring were indicated. However, the patient became anemic, with initial hemoglobin levels falling from 13.4 g/dl to 7.6 g/dl, but with no signs of active bleeding or hemoptysis. Serial radiographic monitoring showed no changes, so a CT angiogram was repeated 1 week later, confirming the diagnosis of bronchial atresia, and ruling out the presence of vascular lesions (Fig. 1B). Anemia was attributed to the multiple fractures and contusions caused by his injuries. The patient was discharged after 11 days without complications. No new complications developed during the subsequent year of follow-up.

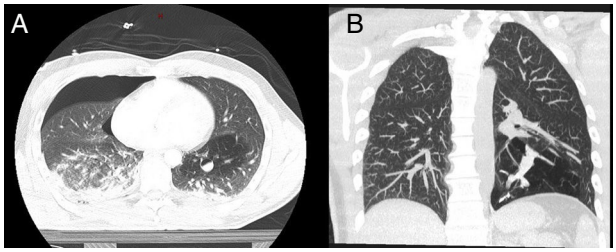

Fig. 1. (A) Air-fluid level in a pulmonary nodule with distal hyperinflation in the initial CT scan. (B) CT angiogram showing hyperinflation, intrabronchial mucoid impaction and hypovascularity of the left lower lobe.

Bronchial atresia is an uncommon malformation characterized by lobar, segmental, or subsegmental bronchial occlusion. Distal lung segments are hyperinflated, and the bronchial secretions are impacted (bronchocele). It generally affects young men, and usually occurs in the left upper lobe. The most common symptoms are fever, cough, hemoptysis, and dyspnea, although patients are mostly asymptomatic. Chest X-ray is significant for pulmonary hyperlucency (90%), and hilar nodular lesion (80%).[1] On chest CT, impacted secretions, pulmonary hyperinflation, and pulmonary hypovascularity should be pathognomonic. Pulmonary hyperinflation is due to collateral ventilation through the intraalveolar pores of Kohn, the bronchoalveolar Lambert's channels, and the interbronchiolar channels. Lipoid pneumonia may occur if collateral ventilation is insufficient.[2] Fiberoptic bronchoscopy is useful for the diagnosis of this entity, but the obliterated bronchus cannot be identified in half of all cases. Diagnosis in infancy is uncommon, and the condition may be confused with congenital cystic adenomatoid malformation, intralobar sequestration, or lobar emphysema.

Matsushima et al. described radiological findings in 8 adults and 1 child.[3] The most common differential diagnosis are vascular malformations, other malformations with impacted secretions, lung cancer, and bronchial adenoma.

Treatment is conservative when the patient is asymptomatic. Surgery is indicated in symptomatic patients, in cases of repeated pneumonia, in cases in which the presence of malignant lesions cannot be ruled out, and in those in which lipoid pneumonia prevails over hyperinflation.[4] Standard segmental pulmonary resection is the surgery of choice because a greater amount of the parenchyma can be preserved, as it must be remembered that this is a benign disease in young patients. The approach of choice is video-assisted thoracoscopy, although this procedure can be difficult due to the fact that the bronchocele normally occurs in the hilum, causing an inflammatory reaction that complicates the resection procedure. In lobar bronchial atresia, lobectomy can replace segmentectomy in order to achieve complete resection.

Our case was a trauma patient in whom the most likely initial differential diagnoses were pulmonary laceration due to an acceleration-deceleration injury, or vascular aneurysm.

No pulmonary contusion or alveolar consolidation were found in the initial CT scan, so lipoid pneumonia could be ruled out. Acquired bronchial obstruction, due to the presence of a foreign body or inflammatory disease was initially ruled out on fiberoptic bronchoscopy, and later confirmed by the absence of any type of neoplastic lesion after 1 year of follow.

In conclusion, bronchial atresia is a benign, uncommon malformation. It is usually asymptomatic and computed tomography helps to guide diagnosis. The differential diagnosis includes vascular malformations, pulmonary sequestration, and lesions with mucoid impaction. The treatment of choice is conservative, and pulmonary resection should be avoided and reserved for symptomatic patients only.

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Persistent Atelectasis in a Patient With Cystic Fibrosis: Are Antibiotics Always Needed?☆

Atelectasia persistente en paciente con fibrosis quística: ¿debemos tratarla siempre con antibioterapia?

To the Editor,

Plastic bronchitis (PB) is a rare, underdiagnosed entity,[1–7] characterized by the formation of obstructive bronchial plugs or cylinders of thick, tenacious mucus that cause the collapse of one or more lobes or even a whole lung.[1,4–7] These casts are often expectorated, but they might be discovered on bronchoscopy or appear in the bronchial tree during autopsy.[4,7] PB is also called fibrinous bronchitis, pseudomembranous bronchitis, or Hoffman's bronchitis.[4,6] It has been described in asthma, cystic fibrosis (CF), cyanotic congenital heart diseases, respiratory infections, bronchiectasis, allergic bronchopulmonary aspergillosis (ABPA), acute chest pain in sickle cell disease, alpha thalassemia, etc.[1,4–7] The pathogenesis of PB is not well understood.[1,4] There are probably 2 mechanisms involved in its development: (1) bronchial injury or changes in bronchial epithelial function due to inflammation or infection, as occurs, for example, in asthma, bronchiectasis, CF, sickle cell anemia, and (2) deterioration of pulmonary lymphatic drainage, as occurs in congenital heart disease.[4,6,8] We report a case of PB in a patient with CF, a situation rarely documented in the literature.

Our patient was a 15-year-old boy with a history of CF, chronically colonized with oxacillin-sensitive *Staphylococcus aureus*, *Haemophilus influenzae*, and *Pseudomonas aeruginosa*, who was admitted for fever of 39 °C, asthenia, anorexia, and chest pain in the left hemithorax. Posteroanterior and lateral chest X-ray showed left upper lobe atelectasis. He was admitted for the administration of intravenous antibiotic therapy with piperacillin–tazobactam and tobramycin. The study to rule out other possible causes of atelectasis associated with CF was significant for a total IgE level of 2500 IU/ml and *Aspergillus fumigatus* (AF)-specific IgE of 38.80 IU/ml, IgG, precipitins and skin prick positive for AF, so prednisone 60 mg every 24 h and voriconazole 200 mg every 12 h were added to the treatment. After 15 days of treatment, clinical but not radiological improvement was observed, so a lung CT was performed that revealed complete atelectasis of the left upper lobe and the presence of a hyperdense cast in the bronchial tree (Fig. 1A). Given these findings, a fiberoptic bronchoscopy (FB) was performed, which revealed an obstruction in the entrance to the bronchus of the left upper lobe, caused by a large mucous plug. Using 4.6 mm fiberoptic bronchoscopy with sustained aspiration, a bronchial cast measuring 7×1 cm (Fig. 1B) could be extracted, that was diagnosed as PB. One month after extraction of the bronchial cast, clinical and radiological improvement was confirmed, and the tapering and gradual withdrawal of corticosteroids began.

In CF, atelectasis occurs as a result of mucus plugs and severe parenchymal disease.[3] It is typically treated with IV antibiotics and intensification of respiratory physiotherapy.[3] If clinical and radiological improvement is not achieved, as occurred in our patient, complications such as PB must be considered. Several classifications of PB have been proposed: some are based on the histology of the cast,[1,4,9] distinguishing inflammatory PB from non-inflammatory PB, and others on the associated etiology,[1] which defines PB as caused by a specific disease, or as idiopathic, if the disease is unknown.[1] Madsen et al. recommended a classification based on the associated disease and the histology of the cast, if the etiology of the PB is unclear.[1,8] Basic treatment of PB is symptomatic, i.e., improvement of alveolar ventilation and mucociliary clearance, and reduction of inflammation and the bacterial or fungal load[1,4,6] in the case of PB caused by bronchial infection or ABPA. Topical treatment of the bronchial cast is not well defined, and no particular mucolytic agent is considered superior to others[1]; the cast may be difficult to remove with bronchial instillation of physiological saline or bronchoscopic suction.[1] Bronchodilators and mucolytics may disintegrate the secretions.[1,5] Recombinant human DNase (rhDNase) has been used to reduce the viscoelasticity of sputum in patients with CF.[3] In our case, rhDNase was instilled under direct vision bronchoscopy to treat the PB. The use of tissue plasminogen activator (0.7–1 mg/kg every 4 h) has been described in PB that develops after the Fontan procedure in children.[1,4,5,7,10] It has also been reported that inhaled heparin can be effective,[7,11] since its anti-inflammatory properties can help reduce mucin secretion, prevent activation of the fibrin tissue factor pathway, and reduce vascular filtration. Inhaled anticholinergics can reduce the formation of casts, and low-dose macrolides can decrease mucin production by inhibiting kinase 1 and 2 activation and reducing the severity of PB.[7,12,13] In patients with lymphatic abnormalities, the most effective therapy for PB is selective lymphatic embolization with magnetic resonance-guided lymphography.[6,7] The role

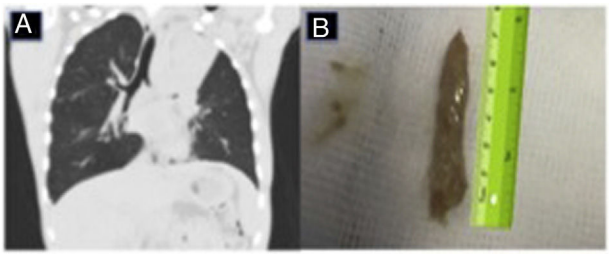

Fig. 1. (A) Complete atelectasis of left upper lobe, containing a hyperdense cast of the bronchial tree. (B) Bronchial cast extracted by fiberoptic bronchoscopy measuring 7×1 cm.

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