

complex of the tube, the bronchoscope and the grasped prosthesis were withdrawn until the prosthesis was brought to the level of the oropharynx. The prosthesis was extracted using Magill forceps together with the tube and bronchoscope. Immediately following the extraction, a new endotracheal tube was inserted. The patient tolerated this procedure well; she was weaned from mechanical ventilation the following day and discharged from the hospital 10 days later.

Foreign body (FB) aspiration is a common problem necessitating prompt recognition and early treatment to minimize the potentially serious and sometimes fatal consequences.<sup>1</sup> Aspiration of a very large FB can constitute a technical problem, since its extraction may require an invasive procedure, such as rigid bronchoscopy, tracheotomy or thoracotomy, especially in an intubated patient. The use of a flexible bronchoscope decreases the risks associated with those procedures<sup>5,6</sup> and has a high success rate (89.6%) in the extraction of FBs; thus, it should normally be considered as the initial approach. There are certain situations, however, in which a rigid bronchoscope should be considered, for example, with FBs that are impacted in extensive granulation tissue or excessive tissue scarring, asphyxiating FBs, FBs that cannot be gripped with flexible forceps as a consequence of their large size, sharpness and smooth margins, as well as failed attempts with a flexible one. In these cases, rigid bronchoscopy remains the procedure of choice.<sup>6</sup> Nevertheless, in our case due to the patient's condition, and even though the FB was very large, we decided to begin immediately with a flexible bronchoscope.

Bronchoscopic procedures are not commonly performed in unstable patients, especially immediately following myocardial infarction, as it can result in fatal consequences. Pre-procedural planning is required and should include the whole care team: the bronchoscopist, assistants, nurses and anesthesiologist. Planning should involve discussion of the FB's characteristics, location, and the patient's comorbidities. Proper ventilation must be arranged for during the procedure, and a back-up plan developed to account for the potential that the foreign body may become lodged within the retro pharynx and completely occlude the airway.<sup>2</sup> This scenario is more common when the FB is very large. In this case, the FB was indeed very large; however, the patient was also unstable, and so the normal pre-planning process was impossible and immediate action was required.

We describe the technique we used to successfully remove a large dental prosthesis in a mechanically ventilated patient, by extracting the FB with the endotracheal tube followed by an immediate re-intubation. Case reports on extractions of very large FBs are scarce. Sampan-Singh et al.,<sup>7</sup> described extraction of a 3-unit dental prosthesis via direct laryngoscopy and tracheotomy. Tu et al.,<sup>8</sup> described an approach using wire loop snares together with a flexible bronchoscope for the removal of a 4-unit dental prosthesis.

Our patient aspirated a 7-unit dental prosthesis, which we believe represents the largest reported FB extraction by flexible bronchoscopy.

This case represents an emergent exceptional approach of extracting large foreign bodies from the tracheobronchial tree in an unstable patient, with relatively minimal temporary compromise of the airway, using only a flexible bronchoscope. The advantages of flexible over rigid bronchoscopes have been described; they provide applicable, cost-effective, safer and faster method of extraction of FBs, and thus should be considered first in experienced hands, even in cases in which rigid bronchoscopy is apparently indicated. Although reports on FB extraction are common, our case, which describes the removal of a very large prosthesis from the bronchial tree in a high-risk patient, is unique.

## Bibliografía

- Passali D, Lauriello M, Bellussi L, Passali GC, Passali FM, Gregori D. Foreign body inhalation in children: an update. *Acta Otorhinolaryngol Ital.* 2010;30:27–32.
- Hewlett JC, Rickman OB, Lentz RJ, Prakash UB, Maldonado F. Foreign body aspiration in adult airways: therapeutic approach. *J Thorac Dis.* 2017;9:3398–409.
- Köse A, Kostak D, Aramagan E, Durak A, Seçkin NS, Dönmez SS, et al. Tracheobronchial foreign body aspiration: dental prosthesis. *Case Rep Pulmonol.* 2014;2014:1–4.
- Batra H, Yarmus L. Indications and complications of rigid bronchoscopy. *Expert Rev Respir Med.* 2018;12:509–20.
- Cunanan OS. The flexible fiberoptic bronchoscope in foreign body removal. Experience in 300 cases. *Chest.* 1978;73:725–6.
- Sehgal IS, Dhooria S, Ram B, Singh N, Aggarwal AN, Gupta D, et al. Foreign body inhalation in the adult population: experience of 25,998 bronchoscopies and systematic review of the literature. *Respir Care.* 2015;60:1438–48.
- Bist S, Lutheria M, Arora P, Kumar L. Missing aspirated impacted denture requiring tracheotomy for removal. *Iran J Otorhinolaryngol.* 2017;29:359–63.
- Tu CY, Chen HJ, Chen W, Liu YH, Chen CH. A feasible approach for extraction of dental prostheses from the airway by flexible bronchoscopy in concert with wire loop snares. *Laryngoscope.* 2007;117:1280–2.

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## Ischaemic Heart Disease Induced by Intralobar Pulmonary Sequestration



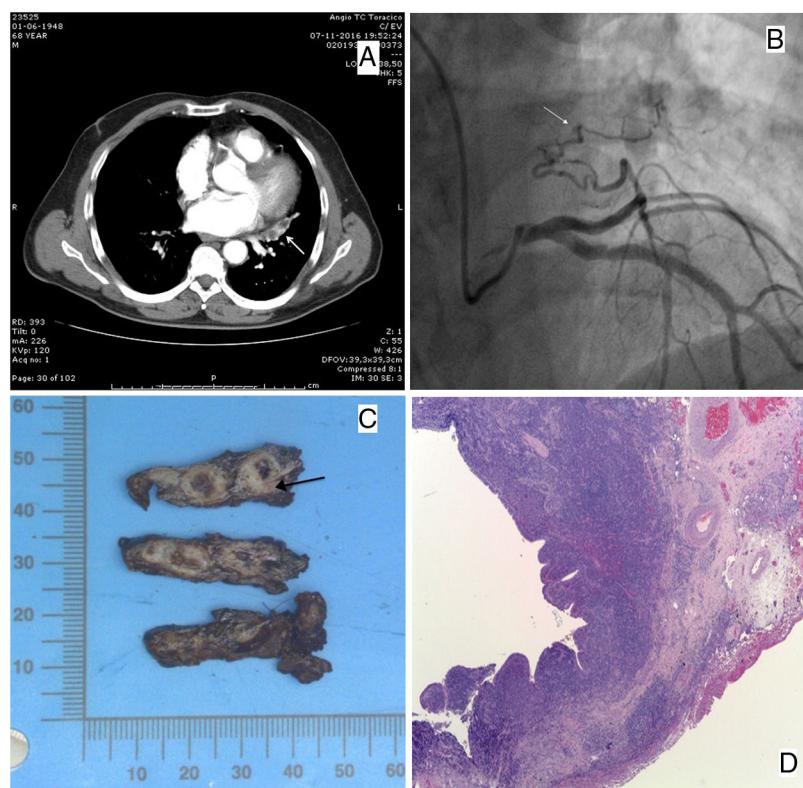
### Isquemia miocárdica inducida por secuestro pulmonar intralobar

Dear Editor,

Pulmonary sequestration (PS) represents a rare congenital malformation (0.15–6.45% of all pulmonary malformations) usually supplied by a systemic artery, frequently merging from the aorta or one of its branches.<sup>1</sup> Vascularization originating from the coronary

circulation is extremely rare with less than 20 cases reported – mostly intralobar sequestrations (presence of independent visceral pleural encasing) supplied either by the right coronary or circumflex artery. Diagnosis can be incidental (e.g. abnormal density on chest radiograph) or in the context of ischemic heart disease due to a coronary steal effect, although arrhythmia has also been described.<sup>2–5</sup>

A 68-year-old male with exertion-related chest pain and a recent cardiac stress test suggestive of ischemia (but no confirmation on myocardial scintigraphy), presented in the Emergency Department with a 3-day epigastric pain irradiating to the left hemithorax associated with nausea and dizziness. No remarkable



**Fig. 1.** (A) Axial CT in the plane of the aortic valve after contrast revealing a left paracardiac fusiform opacity with non-enhancing areas inside, compatible with bronchial impactions and millimetric vessels surrounding the lesion on the peripheral inferior margin. (B) Coronary catheterization revealing a branch of the left circumflex artery supplying an extracardiac structure in the left lung (arrow). (C) Macroscopy of the surgical resection showing cystic areas surrounded by fibrosis; a systemic artery is present near the base (arrow). (D) (H&E, 25×) Cystic bronchial-like structures, with a respiratory tract epithelial lining, with multifocal erosions and a marked chronic inflammation and fibrosis; intimal and medial hyperplasia in muscular pulmonary arteries.

alterations were found on physical examination. The electrocardiogram revealed a sinus rhythm with a slight ST segment depression (<1 mm V3–V5); serial measurements of high-sensitivity troponin I were elevated (until T12 with a maximum 2825 ng/L). Considering the severe thoracic pain and the difference in blood pressure readings between both arms a thoracic CT-angiography was performed to exclude aortic dissection or pulmonary embolism [despite the stronger possibility of a myocardial infarction (MI)] – a left paracardiac fusiform opacity with non-enhancing areas inside, in the plane of the aortic valve was revealed after contrast administration, with surrounding millimetric vessels in the inferior margin (Fig. 1A); no changes were observed in the adjacent lung parenchyma.

The patient was hospitalized with the diagnosis of MI (Killip class I) and had no recurrence of symptoms nor rhythm changes on monitorization. Echocardiogram revealed septal and lower wall hypokinesis with preserved left ventricular ejection fraction (62%). Heart catheterization showed an aberrant branch arising from the left circumflex artery (LCA) supplying an extra-cardiac structure on the left lung (Fig. 1B) without other significant hemodynamic stenoses.

The possibility of ischemic heart events due to a steal phenomenon by an anomalous coronary artery arose. A MR angiography (MRA) revealed a 41 × 20 mm mass in the left lower lobe in close contact with the left oblique fissure and with the mediastinum, with arterial vascularization from the LCA, raising the suspicion of an intralobar PS with coronary irrigation. Despite the possibility of occluding the supplying artery through a transcatheter procedure, considering the risk of infection/necrosis (and eventually malignancy), the patient underwent left lateral thoracotomy – an intralobar PS with supply from an aberrant branch from the

left circumflex artery was identified and the lesion was excised. Macroscopic examination showed cystic areas surrounded by fibrosis (Fig. 1C). Histology revealed cystic bronchial-like structures surrounded by respiratory epithelium with fibrosis and chronic inflammation (Fig. 1D). Myocardial scintigraphy after surgery showed no signs of myocardial ischemia. The patient remained asymptomatic ever since.

A PS supplied by a coronary artery (PSsCA) can theoretically cause symptoms of ischemic heart disease (IHD) through a mechanism of blood steal (even in the absence of significant stenotic coronary vessels) as reported by Nakayama et al.<sup>6</sup> In the few cases published, most patients presented with symptoms of IHD on exertion or even while resting, which prompted a cardiac catheterization. In two cases manifestations included frequent episodes of ventricular tachycardia and of bradycardia (due to sick sinus syndrome) – the former treated with radio-frequency ablation and angioplasty, the latter received a pacemaker.<sup>4,5</sup> A history of recurrent respiratory infections is not uncommon, usually beginning at a very young age (more often with intralobar sequestrations).<sup>7–9</sup> Hemoptysis as a PS manifestation has been reported (with massive hemoptysis being uncommon but a potentially serious event) and can result from structural changes (like bronchiectasis) or even pulmonary hypertension.<sup>3,10</sup>

Chest CT provides the best display of the airways and parenchymal abnormalities in PS – they most commonly appear as a homogeneous or inhomogeneous mass, with or without cystic changes and less frequently as multiple small cystic lesions or a large cavitary lesions with air-fluid level.<sup>11</sup> Identification of the aberrant artery is crucial, either for diagnosis (as PS can mimic a malignant tumor) and for preoperative assessment considering the risk of accidental incision and hemorrhage; lack of visualization

may happen with smaller size vessels (<1 mm) or with an unfavorable orientation.<sup>11,12</sup> Multidetector CT angiography usually reveals both the arterial supply and the venous drainage, making this a diagnostic procedure of choice.<sup>11–13</sup> MRI also has the ability to demonstrate the precise anatomic localization as well as the arterial and venous course. However cystic or emphysematous changes close to the sequestration may not be well delineated and respiratory artifacts can cause low spatial resolution – breath-hold contrast-enhanced MRA can overcome the last one and be as adequate as a CT angiography for vascular characterization.<sup>13</sup> Chest radiography has not the diagnostic value of the previous image techniques but abnormal findings can motivate further investigation.<sup>11</sup> In the case described (as in most cases with PSsCA) the patient presented with signs and symptoms suggestive of ischemic heart disease and the abnormal irrigation was firstly revealed during heart catheterization. The MRA, together with the CT and the cardiac catheterization findings, supported the possibility of an artery originating from the LCA area.

Imaging differential diagnosis generally includes lung cancer, pulmonary cysts or mediastinal tumors. Regarding PSsCA in particular coronary-bronchial artery fistulas (CBF) are another differential diagnosis to consider. These are congenital anastomoses usually found incidentally during invasive coronary angiography and are often associated with bronchiectasis.<sup>14</sup> Most CBF are clinically silent but can become hemodynamically significant in association with a variety of cardiovascular diseases such as cardiomyopathies or supravalvular aortic stenosis. Chest pain and dyspnea related to steal-phenomenon and hemoptysis are the most common symptoms.<sup>14</sup> Diagnosis can be achieved using the same image modalities as for PSsCA.<sup>14</sup>

Surgical resection is recommended in symptomatic patients although coil embolization of the feeder artery (during angioplasty) is also an option. In the case presented, surgery was preferred considering the symptoms, the risk of infection and also of cancer – a few cases of malignant neoplasms being involved in or near sequestered segments have been reported.<sup>15</sup>

## Bibliografía

- Halkic N, Cuénoud PF, Corthésy ME, Ksontini R, Boumghar M. Pulmonary sequestration: a review of 26 cases. *Eur J Cardiothorac Surg.* 1998;14:127–33.
  - Marinos T, Bitzikas G, Madesis A, Galanos O. Sequestered hypoplastic pulmonary lobe supplied by the circumflex coronary artery in a patient with coronary artery disease: a case report. *Heart Surg Forum.* 2006;9:E565–7. <http://dx.doi.org/10.1532/HSF98.20051039>.
  - Lo W, Hemli JM, Brady PW. Bronchopulmonary sequestration supplied by the coronary circulation associated with a right-sided aortic arch. *Heart Lung Circ.* 2004;13:92–6. <http://dx.doi.org/10.1016/j.hlc.2004.01.028>.
  - Rao DS, Barik R. Rare presentation of intralobar pulmonary sequestration associated with repeated episodes of ventricular tachycardia. *World J Cardiol.* 2016;8:432–5.
  - Matsunaka T, Hara Y, Okayama H, Kodama K, Shigematsu Y, Kohara K, et al. Pulmonary sequestration receiving arterial supply from the right coronary artery. A case report. *Angiology.* 1997;48:827–31. <http://dx.doi.org/10.1177/000331979704800910>.
  - Nakayama Y, Masakuni K, Minami K, Miwako I, Kato Y. Pulmonary sequestration with myocardial ischemia caused by vasospasm and steal. *Ann Thorac Surg.* 2000;70:304–5.
  - Van Langenhove G, Convens C, Seynaeve P, Van de Heuvel P, Van den Branden F, Stockman D, et al. Intralobar pulmonary sequestration supplied by the right coronary artery. *Catheter Cardiovasc Interv.* 2000;47:218–20.
  - Fernandez-Vega A, Vilacosta I, Vedia OA, Pedraja I, Vivas D, Martinez-Vives P. A 'kidnapper' left circumflex coronary artery. *Eur Heart J.* 2016;37:919.
  - Silverman ME, White CS, Ziskind AA. Pulmonary sequestration receiving arterial supply from the left circumflex coronary artery. *Chest.* 1994;106:948–9.
  - Gompelmann D, Eberhardt R, Heußel C-P, Hoffmann H, Dienemann H, Schuhmann M, et al. Lung sequestration: a rare cause for pulmonary symptoms in adulthood. *Respiration.* 2011;82:445–50. <http://dx.doi.org/10.1159/000323562>.
  - Ko SF, Ng SH, Lee TY, Wan YL, Liang CD, Lin JW, et al. Noninvasive imaging of bronchopulmonary sequestration. *AJR Am J Roentgenol.* 2000;175:1005–12.
  - Lin CH, Chuang CY, Hsia JV, Lee MC, Shai SE, Yang SS, et al. Pulmonary sequestration-differences in diagnosis and treatment in a single institution. *J Chin Med Assoc.* 2013;76:385–9.
  - Abbey P, Das CJ, Pangtey GS, Seith A, Dutta R, Kumar A. Imaging in bronchopulmonary sequestration. *J Med Imaging Radiat Oncol.* 2009;53:22–31.
  - Said SAM, Oortman RM, Hofstra J-H, Verhorst PMJ, Slart RHJA, Eerens F, et al. Coronary artery-bronchial artery fistulas: report of two Dutch cases with a review of the literature. *Neth Heart J.* 2014;22. <http://dx.doi.org/10.1007/s12471-014-0518-z>.
  - Hertzberg C, Daon E, Kramer J. Intralobar pulmonary sequestration in adults: three case reports. *J Thorac Dis.* 2012;4:516–9.
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