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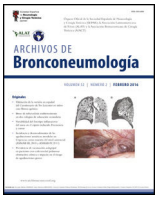

Scientific Letters

Short-term Outcome of Percutaneous Treatment of Pulmonary Sequestration in a Pediatric Hospital in the Andes: A Case Series☆

Resultados a corto plazo del tratamiento percutáneo de secuestro pulmonar en hospital pediátrico ubicado en la región andina: serie de casos

Pryce[1] described pulmonary sequestration (PS) as a spectrum of abnormalities in which part of the lung, with or without bronchial communication, receives aberrant systemic arterial vascularization. The extralobar type is covered by the pleura itself and drains into the systemic veins, while the intralobar type shares the pleura with the rest of the lung and drains into the pulmonary veins; it accounts for 0.15%–6.4% of all congenital pulmonary malformations. Treatment has conventionally been surgical, although nowadays a less invasive option is available in the form of percutaneous treatment.[2–4] Very few series of children with PS treated with embolization have been published, so we would like to report our experience in the percutaneous treatment of PS in a pediatric hospital in the Ecuadorian Andes.

This was a cross-sectional study with consecutive sampling performed between March 2014 and April 2016. Data in common (Table 1): single artery feeding PS; vascular access via the femoral artery. No complications occurred, except in the first patient. Check-up with tomography was performed in patients 1, 2, and 3. Informed consent was obtained for all patients.

In the first case, PS was suspected on radiograph, so we requested a tomography that confirmed a large PS. The procedure was complicated by reduced femoral pulse, treated with enoxaparin; the patient then developed systemic inflammatory response syndrome (SIRS) requiring intensive care. During follow-up, femoral pulses remained symmetric, pulmonary scintigraphy was normal, and the tomography showed full resolution of the PS.

In the other cases, PS was detected during catheterization. The second patient was catheterized due to suspected pulmonary hypertension on echocardiography. The third patient underwent catheterization due to poor progress after intervention for tetralogy of Fallot. Dilated bronchial arteries were detected and occluded with a 6 mm Amplatzer™ vascular plug (AVP)-II, and an AVP-I was implanted in the vessel feeding the PS. Residual flow was observed, so a Gianturco coil (GC) was implanted, completing occlusion of the aberrant artery. The fourth patient underwent catheterization after developing cyanosis. The last patient had a diagnosis of tetralogy of Fallot and collateral vessels on echocardiography, so catheterization was indicated, during which the diagnosis was ruled out.

The essential component for diagnosing PS is angio-MRI, angiotomography, or systemic artery angiogram of abnormal feeding of the pulmonary region. It is not yet clearly defined whether PS should be treated percutaneously or surgically, and the situation is even less clear if the patient is asymptomatic.[3]

Surgical treatment might require lobectomy, cause bleeding, infection or pneumothorax, and hospital stay is extended. Percutaneous treatment involves less risk of bleeding, is less incapacitating, and reduces length of hospital stay. However, complications, such as vascular events and SIRS, may occur, particularly in small children with large PS.

Several types of devices have been used for PS embolization[2,4]: we use AVP and GC because of their availability, safety and proven effectiveness in vascular occlusion, and their accessible, state-funded cost.

Patients were followed up with tomography 6 months after the procedure, and in all cases it was confirmed that the aberrant vessel

Table 1
Common Data in Patients With Pulmonary Sequestration.

Case	Age	PS Type	Arterial Diameter	Device	Aortic Origin of Feeding Artery	RF	Hospitalization	Associated Diseases	Follow-up (Months)
1	1.5 m	REB	1.5 mm	AVP-II 3 mm	Descending thoracic	No	8 days	No	24
2	2 years	RIB	3 mm	AVP-II 6 mm	Abdominal	No	1 day	Scimitar syndrome	20
3	8 m	RIB	5 mm	AVP-I 8 mm+GC 3 mm×4 mm	Abdominal	No	2 days	After intervention for tetralogy of Fallot	11
4	8 years	LIB	1.8 mm	GC 3 mm×4 mm	Abdominal	No	1 day	Trisomy 21+unbalanced CAVC+PAT banding	3
5	4 years	RIB	2 mm	AVP-II 3 mm	Abdominal	No	1 day	Tetralogy of Fallot	1

AVP (I and II): Amplatzer™ vascular plug (I and II); CAVC: complete atrioventricular canal; GC: Gianturco coils; LIB: left intralobar base; m: months; REB: right extralobar base; RF: residual flow; RIB: right intralobar base; PAT: pulmonary artery trunk; PS: pulmonary sequestration.

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had not rechanneled, the PS had resolved, and the residual lung had expanded. Resolution and expansion were particularly evident in the first patient, who had presented a very large PS; in this patient, the pulmonary scintigraphy also showed normal uptake in the right lung.

Although no significant differences have been identified between surgery and percutaneous intervention in terms of mortality, series with large numbers of patients undergoing surgery report 7–14 days of hospitalization, chest tube for 4 days, and lobectomy in most cases, particularly if the PS was intralobar.[3,5] We did not make a comparative study of the 2 techniques, but these comorbidities are avoided with the use of percutaneous treatment.

To our knowledge, this is the second report of percutaneous treatment for PS in children in South America,[4] and while endovascular was safe and effective in our series, it is still early to recommend it as an initial treatment choice, because more experience is required. However, thanks to the growing body of information about its effectiveness,[2–4] it can be considered as a first treatment option in places where resources are limited in terms of intensive care beds or the availability of pediatric surgeons trained in the correction of congenital pulmonary disorders.

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Endoscopic Cyanoacrylate for Persistent Air Leak From a Bronchopleural Fistula in a Patient With Idiopathic Pulmonary Fibrosis☆

Tratamiento endoscópico con cianoacrilato de fuga aérea persistente por fistula bronco-pleural en paciente con fibrosis pulmonar idiopática

Idiopathic pulmonary fibrosis (IPF) is a type of chronic fibrosing interstitial pneumonia of unknown cause with a radiological/histological pattern of usual interstitial pneumonia (UIP).[1] IPF can occur in combination with centrilobular and paraseptal emphysema in the upper lobes,[2] and 12% of patients present pneumothorax. Bronchopleural fistulae (BF), communications between the pleural space and the bronchial tree, can be a result of previous pleuroparenchymal changes. They represent a therapeutic challenge due to high associated morbidity, and the best approach is individualized treatment.[3]

We report the case of a 67-year-old man with a history of hydropneumothorax due to pulmonary contusion and spontaneous pneumothorax, diagnosed with IPF (combined fibrosis/emphysema syndrome subtype) according to ATS/ERS 2011 clinical, radiological and functional criteria,[1] treated with pirfenidone. He consulted due to chest pain, sudden onset dyspnea, with the use of accessory muscles of respiration. Examination showed loss of vesicular breath sounds in the right hemithorax, and the findings of a radiographic study were compatible with tension pneumothorax (Fig. 1A). After placement of a chest tube, the patient was transferred to the hospital ward, where a pleural space with persistent air leak was observed, despite 3 endothoracic drainage procedures (2 with fine caliber tube, 8 and 10 F, and another with thick caliber tube, 24 F) with no resolution of the pneumothorax.

Surgery was ruled out due to the high surgical risk posed by his parenchymal disease, so flexible fiberoptic bronchoscopy (FFB) was performed on day 21 of hospitalization, under deep sedation (patient in semi-sitting position), using a 24 G chest tube in the right hemithorax with water seal to identify the absence/presence of air leak. No endoscopic changes were found in the right bronchial tree. The lateral subsegmentary bronchus (SB) of the middle lobe bronchus was accessed, which on detailed examination of the results of a computed axial tomography (CAT) scan appeared to be the origin of the air leak. A Fogarty catheter® was then used to completely collapse the SB, revealing absence of air leak. Two ml of cyanoacrylate were then instilled into the bronchus, guided by telescopic catheter, with no immediate complications. On completion of the FFB, the air leak was intermittent and progressively resolving, and no pneumothorax was observed on the chest radiograph obtained before discharge, 16 days after the procedure (Fig. 1B).

No consensus guidelines are available on the appropriate treatment of these patients. Therapeutic options range from surgery to interventional FFB with the use of different glues, coils, and sealants.[3,4] The use of cyanoacrylate, a tissue glue widely used in clinical practice, initially seals the air leak and then subsequently induces an inflammatory response causing fibrosis and proliferation of the mucosa, which seals the leak permanently.

Other possible therapeutic options in this case could have included the use of silver nitrate (glue commonly used in rigid bronchoscopy for sealing fistulae with air leak at the surgical site), Watanabe® spigots (silicon cylinders with small rounded extensions that can be anchored in the bronchus and which rarely migrate), or endobronchial valves (removable, well tolerated, with few known complications, that do not rule out subsequent surgical intervention).[4,5] Nowadays, interventional FFB is proposed as an alternative treatment for many airway diseases that were conventionally the preserve of thoracic surgery.

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