CASE REPORT

Intralobar pulmonary sequestration in right lower lobe with secondary infection in an adult male

Amita Yadav *, Rajendra Mohan Mathur, Sanjeev Devgarha, Viju Joseph Abraham, Anula Sisodia, Gaurav Goyal

Department of Cardiothoracic and Vascular Surgery, Sawai Man Singh Hospital, Jaipur, Rajasthan, India

Received 21 September 2013; accepted 10 October 2013
Available online 31 October 2013

KEYWORDS
Pulmonary sequestration; Intralobar; Right lower lobe

Abstract Pulmonary sequestration is a rare congenital bronchopulmonary foregut malformation in which a segment of lung parenchyma is not connected to the tracheobronchial tree. This abnormal segment receives blood supply from the systemic circulation. Varying clinical techniques have been utilized to manage this disease process including surgical intervention, endovascular procedures and operative approaches. The most common presentation of this entity is involvement of the left lower lobe. We present a rare case of intralobar pulmonary sequestration in an adult male involving the right lower lobe with secondary infection.

Case report

A 24 year old male patient was admitted with complaints of chronic dry cough, shortness of breath and right sided chest pain on deep respiration since birth. He had a past history of pulmonary tuberculosis for which he received anti-
tuberculous treatment for two and half months. Treatment was however subsequently stopped due to an unfavorable reaction. On examination his vital signs were stable. Chest and systemic examination at the time of presentation were unremarkable.

Laboratory values were normal with the exception of mild leucocytosis. Contrast enhanced tomography and CT angiography of thorax showed a right lower lobe cavitating lesion with air fluid levels and systemic arterial supply through anomalous arteries from the upper thoracic aorta, proximal abdominal aorta and right bronchial artery (Figs. 1 and 2). Venous drainage was into the left atrium through the right inferior pulmonary vein, findings suggestive of intralobar sequestration of the right lower lobe. Plain films of the chest did not show an active disease process within the thorax. Pulmonary function test was within normal limits.

The patient was taken up for posterolateral thoracotomy. Mobilization of the inferior pulmonary ligament revealed the aberrant arterial supply. Suture ligation with transfixation of...
aberrant artery, venous drainage and bronchial artery was done. The remainder of the lobectomy was performed in the usual fashion. The patient had a relatively uncomplicated clinical course and was discharged on hospital day four in a satisfactory condition. Pathologic review of the resected lung segment was pertinent for chronic and granulomatous inflammation along with interstitial fibrosis. The patient is on regular follow up and is doing well.

Discussion

Intralobar pulmonary sequestration is a relatively rare congenital anomaly with few reports of initial diagnosis occurring during adulthood. Patients can present with an incidental pulmonary lesion on imaging and be otherwise asymptomatic. More commonly however, they may present with pleural effusions or recurrent pneumonia. Few reports have mentioned more severe sequelae such as overlying aspergillosis and even fatal hemoptysis [1–3]. Pulmonary sequestration was mainly located in the left lower lobe (71.53%). Majority (83.95%) of pulmonary sequestration was intralobar, which shared common visceral pleura with the adjacent lung tissue. Extralobar sequestration accounted for 16.05%, which was wrapped by independent visceral pleura [4]. For these reasons, sequestration has traditionally been treated by definitive resection of the affected lung segment. Computed tomography will typically suffice in most adult cases with some debate still held over the need for angiography [5]. In our experience, the contrast CT with angiography was used to make the diagnosis and delineate anatomic features for operative planning. For prenatal diagnosis, ultrasonography has become a useful tool, typically revealing a homogenous, echodense and well-defined mass [6]. Despite its increasing utility, attempts to discriminate sequestration from other congenital malformations can prove to be difficult with ultrasound alone.

Lung cancer associated with sequestration in adults deserves special mention. There have been few reports of malignant neoplasms being involved in or near sequestered segments. In cases with simultaneous involvement, resection is obviously the mainstay of treatment. In contrast, if a patient has limited pulmonary reserve, it is recommended that the neoplasm be resected with the remainder of the lung and sequestered segment be left intact to preserve as much lung function as possible [7].

Definitive treatment involves resection of the affected lung segment. There are several key elements that should be considered: (I) a preoperative course of antibiotics in the setting of a pneumonia exacerbation can be beneficial by limiting the inflammation found at the time of surgery, (II) accurate preoperative identification of the arterial blood supply is crucial since inadvertent injury of these systemic vessels can have a fatal consequence, and (III) great care should be given to securing the systemic arterial branches at the time of operation, which can be quite large in diameter. The extent of resection is aimed at preserving as much normal lung tissue as possible thus warranting a sequestrectomy when feasible. This is more applicable when a diagnosis occurs during childhood due to the ability of further lung development in retained normal tissue. A lobectomy is appropriate in scenarios when it is difficult to distinguish sequestered tissue from functioning parenchyma [8].

Two alternative approaches should be noted. Exclusion of the aberrant arterial supply via an endovascular approach utilizing various occlusion devices. This carries the downside of retaining the un-aerated pulmonary parenchymal tissue that is still subject to recurrent infection [9]. The other option is resection via minimal-access procedures such as VATS lobectomy [10]. The advantages of this approach must be weighed against potential difficulty in controlling the systemic arterial branches.
Pulmonary sequestration is a rare entity especially in the adult population. There can be a vast array of manifestations ranging from asymptomatic patients to those that present with sequelae, such as recurrent pulmonary infections or hemothysis. In this case report, the patient had involvement of the right lower lobe and history of pulmonary tuberculosis which makes this case rare and unique. Early surgical resection should continue to be the standard of care in both adolescent and adult patients.

Conflict of interest

None declared.

References

[1] B. Savic, F.J. Bertel, W. Tholen, et al, Lung sequestration: report of seven cases and review of 540 published cases, Thorax 34 (1979) 96–101.
[2] N. Halkic, P.F. Cuénoud, M.E. Corthésy, et al, Pulmonary sequestration: a review of 26 cases, Eur. J. Cardiothorac. Surg. 14 (1998) 127–133.
[3] H. Morikawa, T. Tanaka, M. Hamaji, et al, A case of aspergillosis associated with intralobar pulmonary sequestration, Asian Cardiovasc. Thorac. Ann. 19 (2011) 66–68.
[4] Yong. Wei, Fan. Li, Pulmonary sequestration: a retrospective analysis of 2625 cases in China, Eur. J. Cardiothorac. Surg. 40 (2011) 39–42.
[5] J. Ikemoto, S. Murayama, J.D. Godwin, S.L. Done, J.A. Verschakelen, Bronchopulmonary sequestration, CT Radiol. 176 (1990) 375–379.
[6] M.S. West, J.S. Donaldson, A. Shkolnik, Pulmonary sequestration: diagnosis by ultrasound, J. Ultrasound Med. 8 (1989) 125–129.
[7] T. Okamoto, D. Masuya, T. Nakashima, et al, Successful treatment for lung cancer associated with pulmonary sequestration, Ann. Thorac. Surg. 80 (2005) 2344–2346.
[8] L.M. Marine, F.E. Valdes, R.M. Mertens, et al, Endovascular treatment of symptomatic pulmonary sequestration, Ann. Vasc. Surg. 25 (696) (2011) e5–e11.
[9] D. Gonzalez, J. Garcia, E. Fieira, et al, Video-assisted thoracoscopic lobectomy in the treatment of intralobar pulmonary sequestration, Interact. Cardiovasc. Thorac. Surg. 12 (2011) 77–79.
[10] J. Somja, L. De Leval, J. Boniver, et al, Intrapulmonary lung sequestration diagnosed in an adult, Rev. Med. Liege 66 (2011) 7–12.