Case report

Role of endovascular embolization in improving the quality of life in a patient suffering from complicated intralobar pulmonary sequestration — A case report

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ABSTRACT

Intralobar pulmonary sequestration is a rare congenital malformation characterized by the presence of nonfunctional parenchymal lung tissue, receiving systemic arterial blood supply and lacking normal communication with tracheobronchial tree. Recurrent pneumonia and massive hemoptysis are life threatening complications associated with it. Delay in the diagnosis and management can be fatal. We report here a case of intralobar pulmonary sequestration in a 18 year old female who presented with recurrent severe episodes of pneumonia and hemoptysis forcing her to drop out of school. The diagnosis was confirmed by CECT Thorax and CT Angiography. The patient was managed by minimally invasive endovascular treatment in the form of feeding artery embolization. She made a full recovery with satisfactory outcome. On subsequent follow up, there was no recurrence of symptoms and she is doing well socially and academically. The aim of this case report is to show feasibility and safety of embolization as a less-invasive management option for adult pulmonary sequestration complicated with hemoptysis and LRTIs and emphasize the importance of such minimally invasive technique to enhance the quality of life in such patients.

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1. Introduction

Pulmonary sequestration is defined as the presence of an area of non-functioning lung parenchyma without any connection to the tracheobronchial tree and the pulmonary arteries. It receives its vascular supply from a separate systemic artery. Pulmonary sequestration is classified as intralobar or extralobar types. Intralobar pulmonary sequestration is surrounded by the visceral pleura of adjacent normal lung, whereas extralobar pulmonary sequestration is surrounded by its own pleura. The venous drainage of intralobar pulmonary sequestration is commonly via the pulmonary veins, extralobar type usually having systemic venous return. Intralobar sequestrations occur almost exclusively in the lower lobes, slightly more frequently on the left. The lesion is often heterogeneous due to acute and chronic inflammation and bronchopneumonia with resultant bronchiectasis, fibrosis and cystic changes. Hemoptysis is a major complication and in this report we discuss how a minimally invasive technique plays a big role in management of such complication.

2. Case report

An 18 year old female presented at our hospital out patient department with recurrent hemoptysis and recurrent episodes of severe respiratory tract infections since childhood. Her deteriorating health resulted in decreased school attendance and negatively affected her scholastic performance ultimately forcing her to drop out of school. She had no other medical illnesses including no history of bleeding tendencies, arthralgia or skin rashes. Other past, medical, surgical, drug and family histories were unremarkable.

Upon examination, the patient had stable vitals. Her chest exam revealed normal expansion with equal air entry and vesicular breathing. Other physical findings were unremarkable. The routine hematological tests, ECG and echocardiography were normal. Chest X-ray showed a well defined consolidated area in the medial and basal portion of left lung (Fig. 1). On the basis of...
her symptoms and X-ray findings, she was prescribed a course of anti-tubercular drugs despite having negative sputum test results. There was no improvement in her condition after the anti-tubercular treatment and the episodes of hemoptysis continued. A contrast CT with CT Angiography revealed the presence of a smooth bordered consolidation in the left posterior basal para-vertebral region. (Figs. 2 and 3). A systemic feeding artery originating from left posterior lateral wall of the descending aorta was revealed. (Fig. 4, Fig. 5a and b). The consolidated segment did not have any obvious communication with the tracheobronchial tree, thus confirming the presence of intralobar pulmonary sequestration.

We decided to embolise the feeding artery through the right common femoral artery. She was admitted to our hospital for this endovascular procedure. Digital Subtraction Angiography (DSA) confirmed the presence of a feeding artery detected earlier by the CT Angiography (Fig. 6). Endovascular embolisation was done using a 5 Fr left coronary catheter and 500 microns size PVA particle under fluoroscopic guidance. Post embolization DSA revealed marked reduction of the abnormal vascular blush and occlusion of the feeding artery. (Figs. 7 and 8). There were no significant post embolization complications except for mild pain which subsided with administration of a simple analgesic. The patient was discharged 48 h after the procedure. The follow up CT done 3 months after the procedure showed persistent occlusion of the vascular supply. However only a small regression in the size of sequestrated lung tissue was noted (Fig. 9). At 6 month follow up, there have been no recurrences of symptoms and no episodes of hemoptysis. She was able to return to school and describes currently a good social and academic life.

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**Fig. 1.** Chest X-ray shows a triangular opacified area posterior medially in left lung.

**Fig. 2.** CT (mediastinal window, coronal) showing presence of a smooth bordered consolidation in the left posterior basal para-vertebral region.

**Fig. 3.** CT (lung window, sagittal) showing the sequestrated segment.

**Fig. 4.** CT angiogram showing systemic feeding artery originating from left posterior lateral wall of descending aorta.
3. Discussion

Pulmonary sequestration (PS), a rare congenital malformation, is an area of non-functioning lung tissue that is separate from the tracheobronchial tree embryologically and receives systemic arterial supply. The aberrant arterial supply may involve a single or multiple vessels. This condition may present in childhood or adulthood. Although patients may be clinically asymptomatic, complications such as severe hemoptysis or episodes of LRTIs may develop.

Bronchopulmonary sequestration accounts for only 0.15–6% of all congenital pulmonary malformations. It is found in 1–1.8% of all pulmonary resections [1]. PS is classified anatomically into intralobar pulmonary sequestration (ILS) and extralobar pulmonary sequestration (ELS) types based on pleural coverage. The ILS type forms 75–86% of all sequestration cases while ELS is about 14–25% [1]. They are more in the left side, with male predominance (4:1 ratio) is seen in the ELS type. ILS receives arterial supply through thoracic aorta (73%), abdominal aorta (21%) or intercostal arteries in 4% of cases. Venous drainage is mainly to pulmonary vein (95%) or azygous hemiazygous veins (5%) of cases. Associated anomalies are seen more with ELS (65%) and (6–12%)

Fig. 6. Pre-embolization digital subtraction angiography after selective catheterization of the left bronchial artery — showing single feeding artery later dividing into two branches to supply the sequestered portion resulting in blush on DSA.

Fig. 7. Mid embolization angiograph showing some reduction of blush.
with ILS [1]. In regard to presentation, ELS presents in early childhood while ILS in adulthood. A total of 30% are incidentally found and the symptomatic type is mainly ILS.

Imaging in suspected case of pulmonary sequestration is done with two principal objectives: to rule out other pathologies and to confirm the presence of an anomalous arterial supply. For a definitive diagnosis of pulmonary sequestration, the systemic arterial supply to the sequestered portion must be delineated to distinguish sequestration from other causes of the lung opacity, such as bronchiectasis, atelectasis and bronchial atresia [2]. The diagnosis relies mainly on identifying the feeding systemic artery. The gold standard remains angiography but with the improvement and development of imaging techniques, less-invasive techniques have proven to be equally effective and safer alternatives to angiography, including CT angiography and MRI. CT angiography is the most widely used, noninvasive diagnostic tool for the detection of pulmonary sequestration in the clinical setting [2]. CT and CT angiography cannot only delineate the origin and course of the anomalous systemic artery and venous drainage accurately, but also assess the abnormal lung parenchyma and the airways.

Surgical resection is the conventional standard treatment for symptomatic pulmonary sequestration. Recently, endovascular treatment with systemic feeding artery embolization has emerged as a less-invasive alternative to surgery. The sequestration process leads to friable vessels due to inflammation, atherosclerosis and even aneurysms. Embolization reduces the risk of hemorrhage especially intraoperatively [3]. Such an approach, has earlier proven to be safe with good outcome in infants and pediatric population but reports in the adult population are exceptional [4]. In our case report, the patient was a 18 year old girl successfully treated with particle embolization without the occurrence of complications. She is completely asymptomatic in 6 months of follow up and it has drastically improved the quality of life but only slight regression in size of the sequestered lung has been noted in follow up CT. So a higher number of cases and longer follow-up is needed to answer the question if regression may occur and, above all, if this has clinical significance [5].

Several reports demonstrated complete involution of the sequestrated segment in infants and pediatric population. Very few reports have described the use of such technique in the adult population. There is a possibility of recurrence due to incomplete occlusion of the feeding artery which is reported in 25–47% of pediatric population. The reasons for such occlusion failure is distal deployment to feeding artery, incomplete occlusion and development of collaterals. For larger feeding arteries, coils might migrate and occlude a non targeted artery [6]. Our patient was eligible for surgical treatment. Nonetheless, based on results with embolization in pediatric patients, we made the assumption that the same modality of treatment could be offered to her. She was informed that a less-invasive method was available in order to minimize complications and to shorten the hospital stay and that surgical treatment should have been considered in case of complete or partial failure of the endovascular treatment.

4. Conclusion

In children and adults with intralobular pulmonary sequestration, a minimally invasive procedure such as transcatheter particle embolization can reduce symptoms such as hemoptysis and improve quality of life. However, the failure to demonstrate regression in sequestered lung size has been noted following such procedures. Therefore, wider experience and longer follow up are needed to suggest embolization as a standalone treatment option and as an alternative to surgery.

5. Disclaimer

There was no commercial funding for the study. The authors have full control over all the data. The paper will not be published elsewhere in any language without the consent of the copyright owners.

Conflict of interests

None declared.
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