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RESEARCH ARTICLE

Asymptomatic Tremendous Lung Shadow in a Young Man: A Case Report

Yue Li, Xiao Fang Zheng, Yan Zhou, Li Min Wang*

Nanjing Medical University, Hangzhou, China.

***Corresponding Author: Li Min Wang**

Abstract

The study is devoted to a unique asymptomatic case of pulmonary sequestration with no infection detected and offers an appropriate treatment solution.The study was carried out in the Surgery Department of the polyclinic No.47011661433010211A1001, Hangzhou, China, in 2019. The patient diagnosed with pulmonary sequestration was 26 years old. No associated symptoms were detected. CT Angiography (CTA) was applied to diagnose pulmonary sequestration, whereas Digital Subtraction Angiography (DSA) was conducted before operation in order to localize abnormal vessels. CT Angiography (CTA) suggested thickening and tortuous regional arteries in the right lower lung, with obvious increasing in branches. The 12th thoracic level aorta branched out into the right lower lobe of the lung, the vein drained back into the pulmonary vein. The surgical treatment of pulmonary sequestration was performed with the embolic thread and involved embolization with an embosphere of 0.5 mm diameter and a spring coil. The post-surgical angiography revealed the disappearance of distal blood flow, indicating success of the therapy. After the intervention, the patient developed a recurrent fever. After the oral administration of low-dose glucocorticoids, the patient's body temperature dropped to the normal level. The final CT scan of the chest conducted a year after treatment showed shadow absorption.The embolization therapy turned out to be effective and hence is suitable for the treatment of pulmonary sequestration. It was established that lung malformations may manifest as an asymptomatic condition. This finding expands what we know about the pulmonary sequestration.

Keywords: *Pulmonary sequestration; Interventional embolization, Angiography, Clinical case.*

Introduction

Pulmonary sequestration (PS) is a rare congenital pulmonary vascular malformation [1]. This term was first used by Pryce in 1946, whereas the phenomenon of pulmonary sequestration was first described by Rokitansky in 1856, who defined it as a cystically altered accessory lung adjusted to the normal lung. It is a cystic/solid mass composed of the lung tissue supplied by abnormal circulation. This part of the lung tissue can communicate with the bronchus, causing recurrent localized infections; when not connected, no respiratory symptoms will occur.

Abnormal lung tissue does not have ventilatory function because of systemic circulating blood supply. In this case, the patient's right lower lung presented asymptomatic tremendous lung shadow, without obvious positive signs, and the property of the occupation was unknown [2]. Pulmonary sequestration refers to a medical condition wherein a piece of lung tissue is separated anatomically and functionally from the normally connected lung. The blood supply of PS is normally derived from thoracic and abdominal aorta, rarely from the intercostal arteries [3]. Finding tissue zones that receive normal and abnormal blood supply is not hard because of their visible boundaries. The sequestered mass sometimes involves bronchial structures, which just as pulmonary vessels, are disconnected [4].

In inflammatory conditions, the infection may extend to the normal lung if a small bronchial communication retains. Some studies note the presence of a bronchial communication between the sequenced mass and the normal lung in all clinical cases [5]. Furthermore, the presence of bronchial communication can lead

to diseases such as bronchiectasis. Pulmonary sequestration is normally found in 0.8% to 2.0% of patients with lung diseases [6]. Pulmonary sequestration may be latent and diagnosed expediently upon screening. It lacks prominent clinical characteristics, barring heart failure and profuse hemoptysis, which can become massive, in adult cases [7].

Although non-functioning tissues are normally found in the chest cavity, rare reports show sequestration in the abdominal cavity [8]. Pulmonary sequestrations may be found in newborns on prenatal ultrasound [9].

Such cases account for 25% of all PS reports [10]. PS diagnoses in 60% of patients are made during their first three months of life [11]. Other patients are diagnosed with lung malformation later in adulthood due to its latent nature. There is a gender predisposition: boys are 4 times more likely to have pulmonary sequestration than girls [12].

Pulmonary sequestration is often mistaken for other lung pathologies [13].According to other sources; half of the newborns lack clinical symptoms, for X-ray findings soon after birth are not conclusive [14]. Therefore, the contrast enhanced computed tomography is recognized as a technique that is most effective for the diagnosis of pulmonary sequestration [15]. The bulk of available research is devoted to the clinical cases of pulmonary sequestration with infection [16] and infection-induced supporative symptoms. Some cases even involved concomitant adenocarcinoma [1].

This study reports a unique asymptomatic case with no infection detected. The relevance of the study lies in the fact that it discusses a latent type of pulmonary sequestration found in an adult. The more clinical cases are discussed the better the understanding of pulmonary sequestration. Furthermore, an effective treatment strategy was offered, which serves an alternative surgical technique.

The study aims to explore the clinical case of asymptomatic pulmonary sequestration and decide on an appropriate treatment. The objectives of the study are to examine the patient's history; to get acquainted with the symptoms; and to decide what kind of therapy to provide given the previous inappropriate treatment decisions.

Materials and Methods

Materials

The study was carried out in the Surgery Department of the polyclinic No. 47011661433010211A1001, Hangzhou (China), in 2019. First, the patient underwent the standard antibiotic treatment for infection (pulmonary sequestration was not diagnosed). By the end of the therapy, no improvement was detected and the patient underwent re-examination to be diagnosed with the pulmonary sequestration. The patient received the outpatient care for a month and was assigned to a surgical intervention according to the proposed technique.

The patient was offered a confidentiality agreement before participation in the study. All procedures meet the generally accepted moral and ethical standards. The object of the study is pulmonary sequestration, whereas the subject is a patient with the unconventional case of lung malformation (latent, without bacterial infection and inflammation).

Methods

The instrumental examination involved measuring the body temperature, heart rate, and blood pressure, as well as assessing breath sounds (wheezing and other noises). Instrumental diagnostics was carried out once a day during the patient's stay in the hospital and 1 time per month for 1 year after discharge. The laboratory examination included the following blood tests: the complete blood count to determine the total number of blood cells; coagulation factor test; hepatorenal function test, CEA test; and HIV antibody test. CT Angiography (CTA) was applied to diagnose pulmonary sequestration, whereas Digital Subtraction Angiography (DSA) was conducted before operation in order to localize abnormal vessels.

The surgical treatment of pulmonary sequestration was performed with the embolic thread and involved embolization with a 0.5ml embosphere and a spring coil. The post-surgical angiography was performed to determine if any improvements took place.A year after the operation, the patient underwent his final CT angiography.

Results

Case Report

A male, aged 26, was found to have tremendous long shadow half a month ago, without cough, expectoration, dyspnea, chest pain, chills, rigors and fever, etc. (Fig.1).The patient stated that he had not been examined before and had no discomfort.

Due to asymptomatic pulmonary shadow, the local hospital suggested that the patient should complete the relevant examination, but the patient refused to continue the examination and took antibiotic for half a month. Half a month after oral administration of the drug, there was no significant change in the shadow of chest CT, but the patient still had no obvious symptoms (Fig.2, Fig.3).

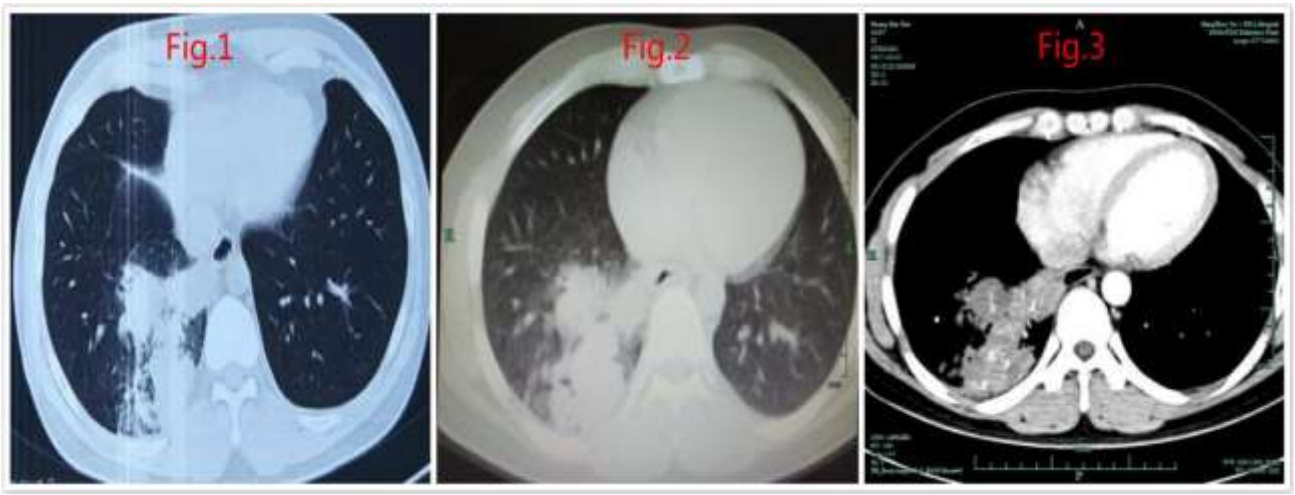

Figure 1: First chest CT half a month ago; **Figure 2-3.** Second chest CT after half a month

Therapy

Physical examination: Temperature 36.9 ℃, heart rate 84/min, respiratory rate 21/min, blood pressure 138/71 mmHg, breath sounds low in lower right lung, no dry and wet rales; rhythm of the heart is neat, without pathologic murmur. Laboratory inspection: No prominent abnormality was found in the blood cell count, coagulation function, hepatorenal function, CEA and HIV antibodies. CT Angiography (CTA) suggested thickening and tortuous regional arteries in the right lower lung, with obvious increasing in branches. The 12th thoracic level aorta branched out into the right lower lobe of the lung, the vein drained back into the pulmonary vein (Fig.4-6).

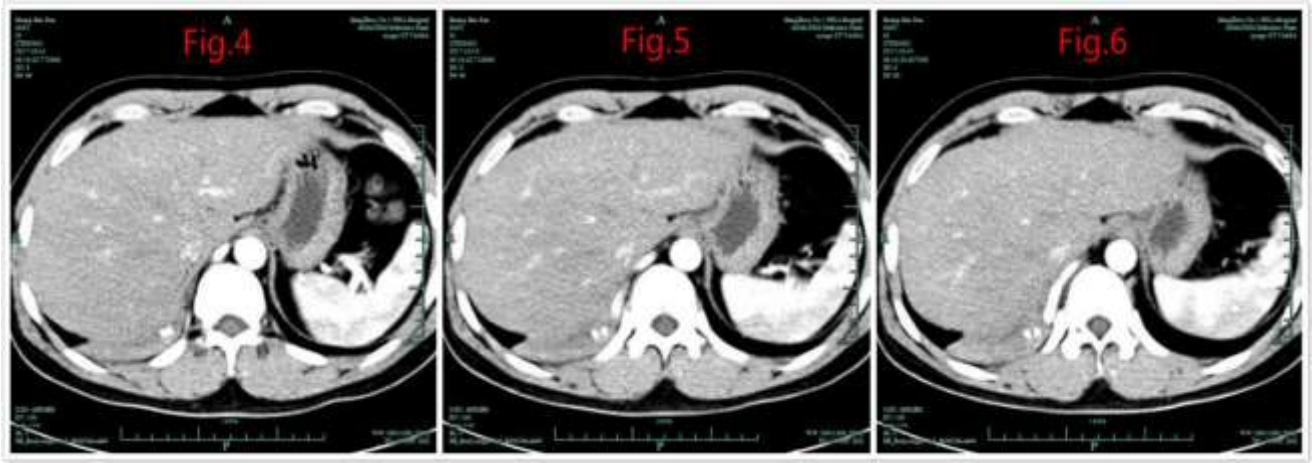

Figure 4-6: the branches of the blood supply to the lower lobe of the right lung can be seen in CTA

Preliminary diagnosis was pulmonary sequestration (PS).

No significant surgical contraindication was found after preoperative evaluation. Digital subtraction angiography (DSA) was conducted in the catheter room, abnormal blood vessels and venous drainage were found, supporting the diagnosis of PS (Fig.7). After confirming the location of the catheter, used silk thread to embolization first, then used embosphere with diameter about 0.5ml, and finally used spring coil to embolization.

Angiography after embolization indicated the disappearance of distal blood flow, indicating that the embolization was successful. After the embolization, recurrent fever occurred in the patient, and the absorption of pulmonary necrosis was considered. After oral administration of low doses of glucocorticoids, body temperature drops to normal levels. One year later, reexamination of chest CT showed that shadow absorption was obvious (Fig.8-9).

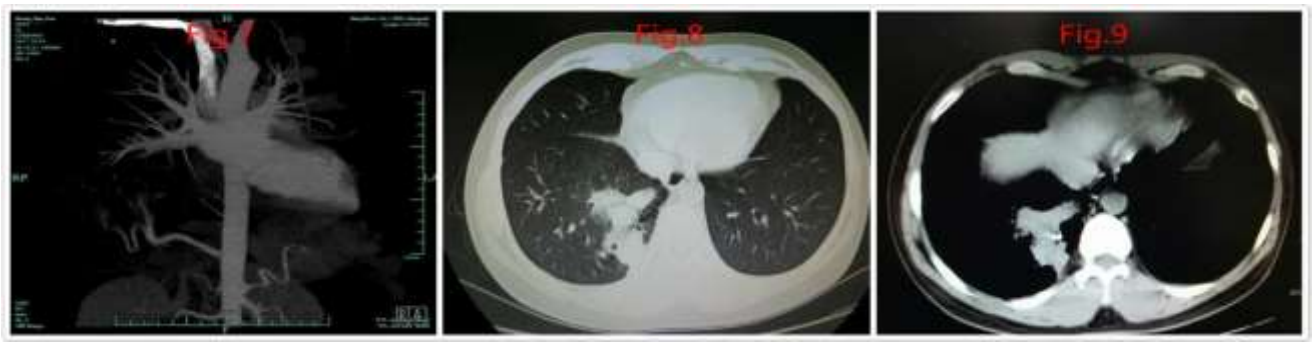

Figure 7: DSA; Figure 8-9. The chest CT lesion was smaller than before

Discussion

Pulmonary sequestration is a rare abnormal pulmonary development, accounting for 0.15-6.40% of all lung malformations [17]. The aetiology of these lesions is under much debate. In the past, it was believed that abnormal lung tissues were caused by vascular traction, that is, in the process of lung development, the blood vessels connecting the original aorta and the original lung were not degenerated, and some lung tissues were separated from the lung by traction. The high blood pressure of abnormal blood supply leads to cystic and fibrous changes in this part of lung tissue, resulting in PS.But now there is growing evidence to support the idea that pulmonary sequestration is caused by primordial ventral lung buds forming during embryonic development [18].

Classification

Pulmonary sequestration is divided into two types: Extralobar pulmonary sequestration (ELS) and Intralobar pulmonary sequestration (ILS).Among this, ILS accounted for 75% and ELS accounted for 25% [19]. ILS shares visceral pleura with normal lung tissues [19], and exists in normal lung lobes, which can communicate with the normal bronchus, thus presenting respiratory symptoms. An EL is contained within aindependent pleural covering, thereby maintaining an anatomical boundary between itself and the surrounding lung tissue. Most ELS occur in the thoracic cavity, and a few occur under the diaphragm. Some ELS can be accompanied by other malformations, among which the most common is congenital diaphragmatic hernia, and others include pulmonary dysplasia, congenital cystic adenomatoid malformation, congenital emphysema, esophageal malformation, bronchial esophageal fistula and cardiovascular malformations [20].

Supplying Vessels

The main artery of abnormal blood supply is thoracic aorta and abdominal aorta, followed by intercostal artery, subclavian artery and internal thoracic artery [21].The blood supply of a few patients is from coronary artery and can induce coronary ischemic events [22, 23].There is also report of pulmonary sequestration of superior mesenteric artery blood supply [24]. One study suggested that 82.6 percent of patients had an abnormal lung tissue supplied by a single vessel, and 17.4 percent had two or more supply vessels [25]. The venous reflux of ILS mainly returns to the left atrium through pulmonary vein, and few enter the systemic circulation. In ELS, venous reflux enters the right atrium through the systemic circulation veins [19].

Clinical Symptoms

Clinical symptoms are not specific, and some patients may have no prominent symptoms. Because of repeated coughing, expectoration, fever and dyspnea, ILS can be misdiagnosed as respiratory tract infection and lower lung pneumonia. A few patients may have pleural effusion, hemoptysis, severe back pain and no other symptoms. There were also reports of PS with only CA-199 and no clinical symptoms [26].

ELS usually does not show repeated upper respiratory tract infection, and 25% of the patients, due to the accompanying other malformations, show feeding difficulties, respiratory distress, slow growth and so on in infancy [20].When there is left to right shunt, patients can present precardiac murmur and heart failure.

Imaging Diagnosis

Imaging is the key to diagnosis and classification of diseases. At present, the commonly used include color doppler, digital subtraction angiography (DSA), chest CT plain scan/enhancement, CT angiography, MR angiography (MRA) and so on. Color doppler is helpful in the diagnosis of PS close to the diaphragm or liver, and PS presents higher echo than normal lung parenchyma. It is a non-invasive and safe examination method, but lacks specificity, and can be used in the diagnosis of early PS in utero.

DSA can clearly identify the abnormal blood supply artery of PS, and it can be used for interventional therapy at the same time of diagnosis, which is the gold standard for the diagnosis of PS. However, the disadvantages of DSA include trauma, large amount of radiation, long time, relatively expensive and hospitalization. On chest CT, PS presents as solid, cystic mass. Solid PS presents as a round or oval mass with clear edges. Cystic PS presents as single or multiple cysts with thin walls and visible liquid level. If infection occurs, the cystic wall shows thickening and the edges become blurred [27].

In a retrospective study [28], the manifestations of pulmonary sequestration were: mass lesions (37.2%), cystic lesions (32.6%), pneumonia lesions (16.3%), vacuous lesions (9.3%) and bronchiectasis (4.6%) [28]. CT angiography (CTA) can provide multi-planar, three -dimensional reconstruction of image data; accurately display the number of abnormal blood vessels, the location of the issue, measurement of their diameter, and the location of the relationship with large blood vessels.

Specific information such as abnormal pulmonary parenchyma, tracheal and peripheral emphysema can also be displayed, which is useful for diagnosis and surgical resection/ prevascular evaluation. In the report of Long et al [28]. CTA accurately showed abnormal blood supply arteries (100%) in all 43 patients, confirming the accuracy of CTA diagnosis.

A comparative study confirmed that CTA and DSA had no difference in diagnostic ability of PS. CTA could replace DSA as a tool for preoperative diagnosis of PS [29]. Compared with CTA and DSA, MRA has a larger field of vision and higher spatial resolution, especially in displaying distorted blood vessels and the movement in lung [30]. In addition, MRA is radiation-free and does not require iodine contrast agent injection. It is non-invasive blood vessel imaging and has more prominent diagnostic advantages than CT and MR scanning.

Treatment

Currently, there is no clear guideline for the treatment of PS, and appropriate treatment should be selected according to the patient's age, clinical manifestations, lesion location, blood supply vessels and venous reflux. In a retrospective study of newborns, by comparing initial and subsequent follow-up CT scans, the percentage reduction in PS volume (PDV) and the percentage reduction in the diameter of donor arteries (PDD) were calculated to assess the probability of ELS spontaneous regression.

In 4 years, the proportion of patients reaching PDV> 50% and PDD> 50% was 93% and 73.3% respectively [31]. This study suggests that neonatal PS has a certain self-limiting, asymptomatic children, can consider clinical observation, assess the risk of surgery, if necessary, surgery. At present, common treatment methods include surgical resection and interventional arterial embolization.

Surgical resection should be based on the location of the lesion, blood supply and venous return. After lobectomy, most patients can return to normal life without recurrence. The comparative study by Wang et al [13]. Showed that there was no significant difference in operative time, hospital stay and perioperative complications between thoracoscopic surgery and thoracotomy. However, the amount of bleeding in the thoracotomy group was more than that in the thoracoscopy group, suggesting that minimally invasive thoracoscopy could basically replace the old-fashioned thoracotomy. Interventional embolization is usually performed via femoral vein access to the aorta for angiography.

Before embolization, the blood vessels of the patients need to be clearly defined before embolization, so as to avoid omission.

At present, it is believed that since PS is a congenital disease, there is no collateral circulation after embolization; unlike the presence of angiogenic factors in tumors [32]. The commonly used embolic agents include spring coil, anhydrous ethanol, polyvinyl alcohol particles, etc.

During embolization, patients' vital signs should be paid close attention to, and symptoms such as chest pain and dyspnea should be focused to avoid ectopic embolization. Li [33] reported 13 patients with PS treated by arterial embolization, the success rate was 100%, and no obvious complications and recurrence occurred after operation, suggesting that interventional therapy is effective and safe.

In this case, the patient had a tremendous shadow of the right lower lung without any symptoms. CTA showed that the thoracic aorta branches to the right lower lobe of the lung and nourished the abnormal lung tissue. The imaging was consistent with the diagnosis of pulmonary sequestration. DSA showed abnormal arterial blood supply and venous drainage, which was diagnosed as pulmonary sequestration (PS).However, it is not clear whether it was ILS or ELS. Considering that the patient's abnormal lung tissue was tremendous, although no symptoms were occured at present, there are many risks in the future.

It was suggested that the patient should be treated by surgery or interventional embolization. Because the patient was young, the surgical resection was traumatic and the recovery was slow, interventional embolization was the most suitable treatment. Angiography after embolization indicated the disappearance of distal blood flow, indicating that the embolization was successful. One year later, reexamination of chest CT showed that shadow absorption was obvious.

The current study showed that pulmonary sequestration can exist without infection and subsequent inflammation. Moreover, the patient was not aware of his diagnosis until examination at the 26 year of age. The embolization therapy decided upon for this case proved 100% effective, although other studies report fatal consequences from its use [34, 39]. This suggests that the treatment in point may be assigned in non-inflammatory infection-free cases of pulmonary sequestration. Findings show that PS is not necessarily clinically prominent, which expands the knowledge about this abnormality.

Conclusion

Pulmonary sequestration is rare in clinic and often misdiagnosed and missed. The most common is ILS, ELS is relatively rare. Due to the risk of massive hemoptysis, adults should be treated immediately at the once of diagnosis. The embolization therapy proved effective. Pulmonary sequestration is not necessarily symptomatic or accompanied by bacterial infection.

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