Case Report

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Spontaneous regression of pulmonary herniation in 3 days: rare case report

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ABSTRACT

Pulmonary herniation can be defined as protrusion of pulmonary tissue and pleura from an abnormal opening at thoracic wall, diaphragm or mediastinum. Herniation is mainly localized at thoracic, cervical and diaphragmatic regions. Congenital herniation generally occurs as a result of costal agenesis/hypogenesis or absence of intercostal muscles. It is striking that up to 30% of acquired cases occur spontaneously. It is mostly seen in patients with weakness of thoracic wall or in case of acute increase in intrathoracic pressure such as severe coughs. Blunt or penetrating traumas resulting in rib fracture or separation of costal joint cause traumatic pulmonary herniation. In our case a 45-years old female presented to emergency department with height from fall and pain at right flank. On chest radiograph, there was rib fracture at higher level on right hemithorax. No marked pneumothorax was observed. On thorax CT scan, displaced rib fracture at right upper ribs, small amount of hemithorax and parenchymal contusion were observed but no pneumothorax was seen. In addition, right hemithorax, interruption at anterolateral thoracic wall at the level of lateral segment of middle lobe and pulmonary herniation (6x3 cm in size) at the same level were observed. Routine biochemical tests were normal in the patient. During 3-days follow-up, the pain at right flank relieved gradually. Such a large pulmonary hernia is usually treated surgically. However, as we have seen in our case, spontaneous regression can be seen unexpectedly.

Keywords: Pulmonary herniation, Spontaneous regression, Thorax CT

INTRODUCTION

Pulmonary herniation can be defined as protrusion of pulmonary tissue and pleura from an abnormal opening at thoracic wall, diaphragm or mediastinum.¹ Herniation is mainly localized at thoracic, cervical and diaphragmatic regions.² Pulmonary herniation is a rare phenomenon with about 300 cases reported since it was first defined in 1499.³ The currently used pulmonary hernia classification was reported by Morel Lavallee in 1845.³ Pulmonary hernias could be due to congenital and acquired (spontaneous, traumatic) causes.⁴ Congenital causes account up to 20% of all pulmonary herniation cases.⁵ Remaining cases are acquired and most of them are

traumatic in origin. Other causes of acquired herniation include inflammation, neoplastic processes, COPD accompanied by increased intrathoracic pressure due to postoperative cough or weight lifting, steroid use and cardiopulmonary resuscitation.^{1,6} Traumatic intercostal herniation is often associated to lesions of thinnest intercostal muscle adjacent to sternum at costochondral junction or rib fractures.⁷

Congenital herniation generally occurs as a result of costal agenesis/hypogenesis or absence of intercostal muscles. It is striking that up to 30% of acquired cases occur spontaneously.⁸ It is mostly seen in patients with weakness of thoracic wall or in case of acute increase in

intrathoracic pressure such as severe coughs. Blunt or penetrating traumas resulting in rib fracture or separation of costal joint cause traumatic pulmonary herniation. Particularly in children, blunt traumas (due to weakness of anterior and posterior intercostal muscles) and surgical intervention to thorax (some cardiac surgeries, thoracotomy, tube thoracostomy, VATS etc.) can cause pulmonary herniation.⁹

Reduction and surgical repair are methods used in the management of pulmonary herniation. Spontaneous regression can be observed in small herniations, but as in our case the large herniations quite exceptional.¹⁰ Here, we aimed to present a case which showed spontaneous regression within 3 days after large pulmonary herniation developed due to fall from height.

CASE REPORT

A 45-years old female presented to emergency department with height from fall and pain at right flank. On chest radiograph, there was rib fracture at higher level on right hemithorax. No marked pneumothorax was observed. On thorax CT scan, displaced rib fracture at right upper ribs, small amount of hemithorax and parenchymal contusion were observed but no pneumothorax was seen (Figures).



Figure 1: Pulmonary herniation in coronal slice CT.

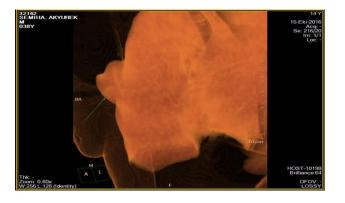


Figure 2: Pulmonary herniation in CT 3D image.



Figure 3: Pulmonary herniation in coronal slice CT intermediate window.

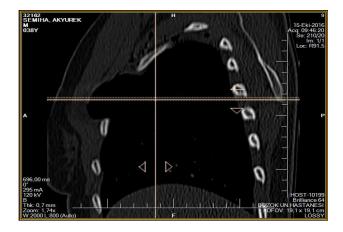


Figure 4: Pulmonary herniation in oblique sagittal slice CT.



Figure 5: Pulmonary herniation in axial CT parenchymal window.

In addition, right hemithorax, interruption at anterolateral thoracic wall at the level of lateral segment of middle lobe and pulmonary herniation (6x3 cm in size) at the same level were observed. Routine biochemical tests were normal in the patient.

During 3-days follow-up, the pain at right flank relieved gradually. No complication such as pneumothorax,

progressive chest pain, hemoptysis, or dyspnea was observed during clinical follow-up. Marked decrease in the size of herniation at the level of lateral-medial segment of middle lobe at right lung was observed on control CT scan. The patient was discharged by close monitoring given the likelihood of potential complications.



Figure 6: Regressed pulmonary herniation in axial slice CT.



Figure 7: Regressed pulmonary herniation in 3D CT image.

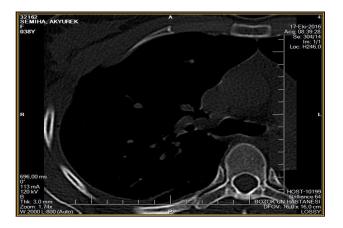


Figure 8: Regressed pulmonary herniation in axial slice CT intermediate window.



Figure 9: Regressed pulmonary herniation in axial slice CT parenchymal window.

DISCUSSION

Since symptoms of pulmonary herniation after chest wall traumas are associated with pneumothorax and/or hemothorax, they generally remain unrecognized. In settings other than chest trauma, pain at chest wall and palpable mass are most commonly encountered complaints. Occasionally, it could be observed that the mass enlarges with inspiration and reduces with expiration. In addition to above-mentioned complaints, pulmonary herniation should be suspected if persistent cough, history of chest trauma or chest surgery is present in the patient. The palpation and radiological detection of mass are important in the diagnosis. On plain radiographs, it is observed that herniated tissue is welldefined, radiolucent compared to lung tissue and at outside of pulmonary contours. CT scan is helpful in the definitive diagnosis.6

On contrary to above-mentioned discussion, a small proportion of pulmonary hernias has an asymptomatic clinical course and is diagnosed incidentally. It was reported that diagnosis was made 40 years after original injury in some cases.¹¹ Asymptomatic cases are managed with conservative approach including treatment of reasons aggravating cough and radiological follow-up while severe pain, enlargement in hernia and presence of incarceration findings are indications for surgical treatment. Although incarceration is rarely developed due to elasticity of lung tissue, dyspnea and strangulation may develop, compelling emergent surgical intervention.¹²

It was reported that pulmonary hernias were first described by Roland in 1499 and that primary classification was made by Morel Lavallee in 1845. Pulmonary herniation is most commonly seen at intercostal region (60-80%); followed by cervical region (35%), and less commonly at diaphragm and mediastinum. The patients present with swelling at chest wall, chest pain and painful mass at chest wall. Anamnesis, physical examination and radiological imaging are of important in the diagnosis. Symptoms has become more prominent valsalva maneuver and forced

expiration. On the chest radiograph, herniated tissue generally appears as well-defined and radiolucent tissue compared to lung at outside of pulmonary of contours. The definite tissue is made by CT scan. Detailed evaluation can be made by pathologically. In the treatment, conservative approach is recommended in small hernias while surgical treatment to prevent complications in greater hernias.¹³

Majority of cases at intercostal localization are spontaneous pulmonary herniation related to increased intrathoracic pressure resulting from heavy lifting or use of wind instruments. Rib fracture and/or cartilage fracture can accompany in such cases. In a case series including 16 cases, Brock reported that all patients were male and that one-half were smokers and obese.¹⁴ Traumatic intercostal pulmonary herniation occurs at costochondral where external intercostal muscles are lacking and internal intercostal muscles are thinner. There is an ongoing controversy about requirement of repair in such hernias. The increase in the size of defect and pain are potential findings of incarceration and primary indication for surgery. Defect is repaired by either primary closure or synthetic grafts based on size and condition of tissue.¹⁵

In acute traumatic cases, crepitation, swelling with fluctuation and subcutaneous emphysema could be observed in physical examination. The patients with hernias caused by intercostal muscle defect are generally more stable. The diagnosis can be readily made by considering anamnesis. It is attempted to visualize hernia by valsalva maneuver on plain radiographs or CT scan. However, it may not be possible to show hernia in cases with defect at limited area.

In such cases, reconstructive thoracic CT scan can be helpful. Potential complications include incarceration, hemorrhage, atelectasis and pneumonia. Elastic dressing can be considered as conservative treatment in cases not eligible for surgical treatment. However, it should be kept in mind that pulmonary dressing reduces pulmonary compliance and increases risk for atelectasis and infection by restricting respiratory movements. Thus, reduction and repair are recommended treatment in pulmonary herniation given potential complications.¹⁶

CONCLUSION

In our case, pulmonary herniation (6x3 cm in size) developed at anterolateral segment of middle lobe at right lung after trauma showed reduction on the day 3, regressing to 27x7 mm in size. Limited number of cases with substantial regression has been reported in the literature. In the clinical picture, the patient had only gradually decreasing flank pain with no dyspnea, hemoptysis and incarceration; thus, the patient was discharged by close monitoring. Such a large pulmonary hernia is usually treated surgically.

However, as we have seen in our case, spontaneous regression can be seen unexpectedly. We would like to point out that pulmonary hernia cases should be followed closely and should not be hastened for immediate surgical application because of the possibility of spontaneous regression.

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