Case report

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LOBECTOMY OF THE CENTRAL LOBE AS A METHOD OF CHOICE IN THE TREATMENT OF LARGE ENDOBRONCHIAL MID-LOBES HAMARTOMA: CASE REPORT

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Pulmonary hamartoma is a rare benign tumor change in the lungs. Often, it is discovered randomly as a side finding on the chest x-ray in the form of asymptomatic, solitary, round tumor change, coin size, with "popcorn" calcifications. Although considered benign tumor with good prognosis, it differs diagnostically from carcinoid, tuberculosis, bronchogenic carcinoma, metastases and hydatid cysts. In histopathology diagnosis, the following are used: chest X-ray, computed tomography, bronchoscopy, fine needle aspiration cytology, surgical extirpation and histopathology verification. It is thought that symptomatic and large pulmonary hamartoma should be removed. Whenever possible, pulmonary hamartoma should be removed by minimally invasive-bronchoscopic procedure or by video-assisted thoracoscopic surgery. For classical surgical approach, lateral and anterolateral thoracotomy are more convenient, and lung hamartoma can be enucleated, wedge resectioned, removed by segmentectomy, lobectomy, and possibly by pulmectomy. The following case study describes the lobectomy of the central lobe through anetolateral thoracotomy as a method of choice to treat a large, symptomatic PH in the middle lobe, in a patient 21 years of age.

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Key words: *lung hamartoma, benign tumor, lobectomy of the central lobe*

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Introduction

Pulmonary hamartoma (PH) is a rare but most common benign tumor change in the lungs. It occurs in 0.025-0.32% of all lung neoplasms (1). PH accounts for 77% of all benign lung neoplasms and about 6% of all solitary lung nodules (2,3). It was first described by Albrecht in 1904 (4). It is more likely to occur in men with peak incidence in the sixth and seventh decades of life and male female ratio 2-4:1 (5). Pulmonary hamartoma is most often detected incidentally as an early finding of the chest Rtg in the form of a solitary, round, coin size tumor change (coin lesion) with "popcorn" calcifications (4). The presence of PH is usually asymptomatic. Rarely, depending on size and localization, PH can give symptoms in the form of cough, haemoptysis, chest pain, fever and signs of obstruction of the bronchial tree (6). Usually, these are solitary nodes ≤ 1-20 cm in the parenchyma of the lungs on the periphery, but they can be found both centrally and endobronchially (7). In 1.4% of cases PH was localized endobron - hially (8). Hamartomas are considered benign tumors with good prognosis and are often clinically monitored due to their slow growth (2). However, although it is traditionally considered that PH does not alter malignantly, there are some reports indicating the possibility of malignant alteration (4, 9).

Case study

Patient I.V. aged 21, was admitted to the Clinic of Lung Diseases Knez Selo due to the cough problems, high body temperature, general weakness and fatigue, pain in the muscles and joints. The symptoms started 7 days before the admission. He was treated clinically by Hemomycin tbl. when there was a slight drop in body temperature. On the chest x-ray P-A in the middle pulmonary field, an oval, soft

tissue change, diameter of about 8 cm was observed, with "popcorn" calcifications (Figure 1). The patient is a non-smoker and was not in contact with domestic animals. Immediately upon admission, a serological analysis was performed on Echin spp antibodies by an indirect method that was negative. Spirometrically, there is an easy-to-restrictive ventilatory disorder FEV1 85% (3.64 L), FVC 89% (4.49 L), FEV1%/FVC 81%. The lung multislice computed tomography (MSCT) discovered on the right, dominantly in the middle lobe, a change of 79x70x65 mm, extending from the right hilum to the pleura lateral wall, with which it is in wide contact, a highly heterodenous structure, with soft tissue attenuations and attenuations of calcium and fat, with postcontrast coloring of solid soft tissue parts, without adhesive growth; several subpleural micronodal changes were present on both sides (Figures 2 and 3). It was concluded that this was a hamartoma change (Ddg. of other etiology), and the observed subpleural micronodal changes were of non-specific characteristics. Bronchoscopically, mouthpiece for DB4 was completely frozen with a ball-like neo formation of a glossy, white, smooth surface ("membrane"), due to which transbronchial biopsy (BB) was not taken. It was concluded that the tumor change was in the middle lobe, of the neat environment and that it worked cystic and benign.

The patient was admitted to our facility for the operative treatment of the mentioned tumor change in the middle lobe. The patient underwent surgery. At first, video-assisted thoracoscopic surgery (VATS) was made to the right with the idea of hamartoma enucleation. After the introduction of the endocamera in the right pleural space, one finds an oval, white mass of about 8 cm in diameter, which engages almost the entire middle lobe. It was decided to perform the lobectomy of the central lobe. The approach was anterolateral thoracotomy through the 5th interrib space to the right. First, once again, the enucleation of the change was attempted. However, during the preparation of tumefact it was seen that the same was inseparably linked to the bronchial cartilage for the middle lobe (Figure 4). The preparation of tumor was done partly sharp, partly dull, with monopolar and bipolar instruments. Then, with the combined, front and back approach, the central lobe blood vessels were uncompressed, proximal double-bridged and cut. The middle lobe bronchus was first cut down more central from the level of the tumor growth, and then it was taken away by a bronchial splitter. A tumor with a bronchial stump and the rest of the central lobe tissue was sent to the histopathology examination (HP) (Figure 5). HP findings indicate that it is a 8x6x5 cm capsulated oval tumor, on a cross-section of multishape appearance, a brown, white-yellow color with the presence of car-tilaginous consistency and dense yellowish areas, as well as cystic formations up to 25 mm, partly filled with brown mass. HP diagnosis is hamartoma pul-monis.

Immediate postoperative procedure was neat and the wound has healed per primam. On the seventh postoperative day, the patient was released for home treatment. In a series of control x-ray examinations, the findings on the lungs are correct and correspond to the operation performed (Figure 6). Control check after six months has shown that the patient is subjectively well, there is no recurrence of tumor. The patient returned, all the way to the usual life activities.

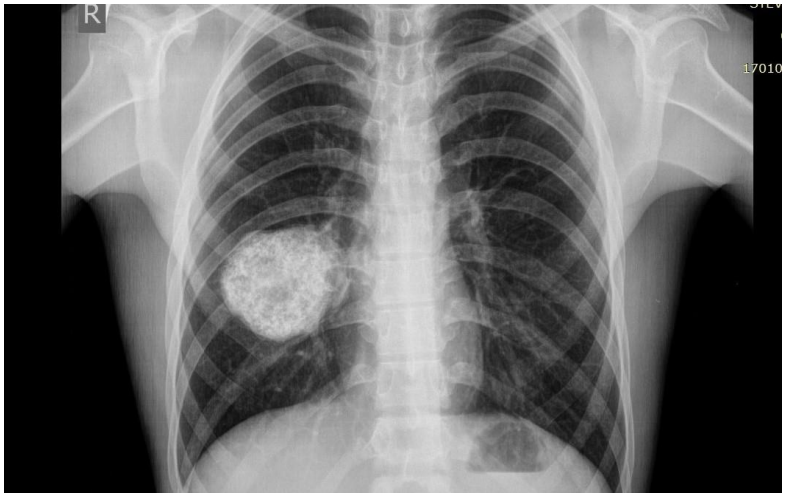

Figure 1. Rtg P-A, preoperatively, a large hamartoma in the middle lobe with "popcorn" calcifications

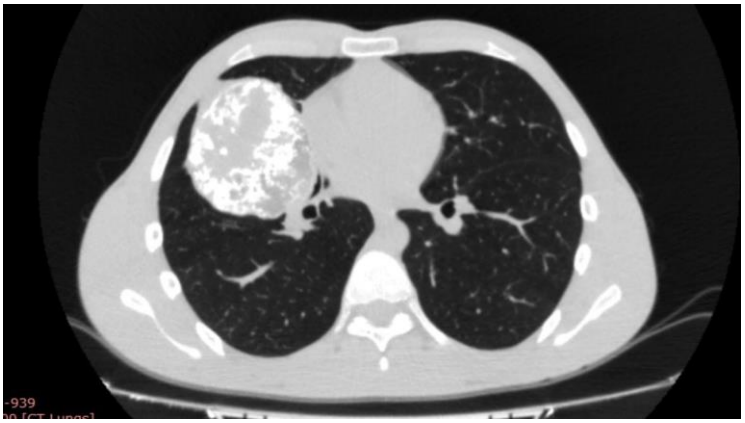

Figure 2. MSCT presentation of hamartoma in the middle lobe (axial cross-section)

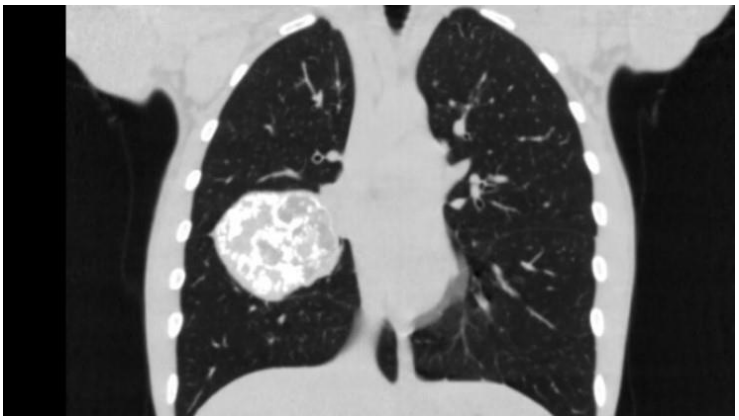

Figure 3. MSCT presentation of hamartoma in the middle lobe (coronal cross-section)

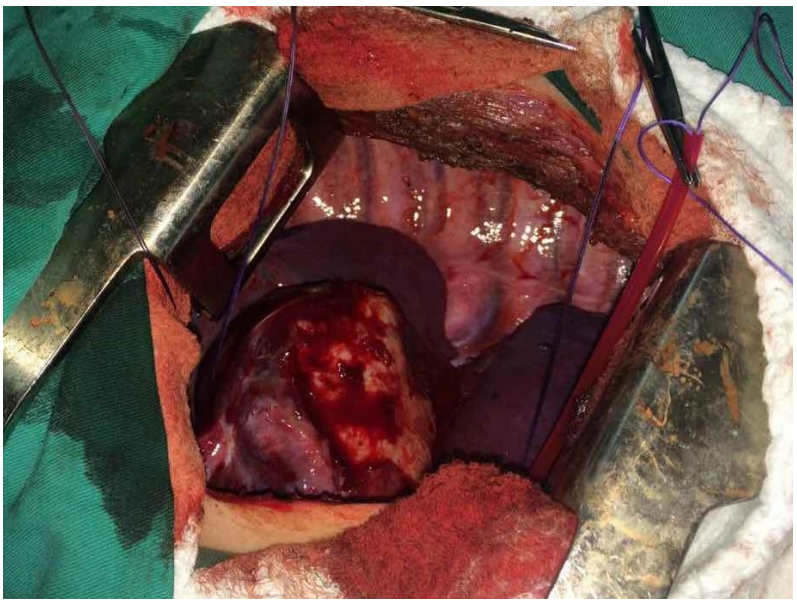

Figure 4. Intraoperative finding, a large hamartoma that engages almost the entire middle lobe

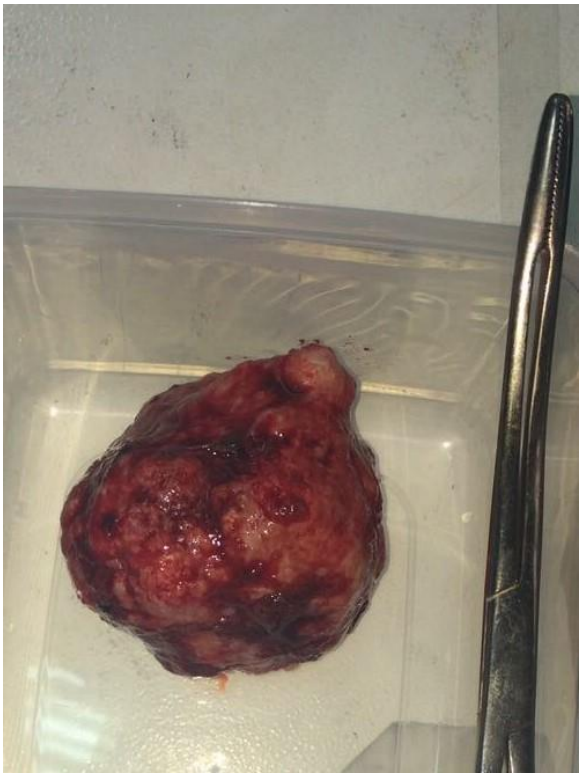

Figure 5. Enucleated hamartoma with a bronchial stump for the middle lobe

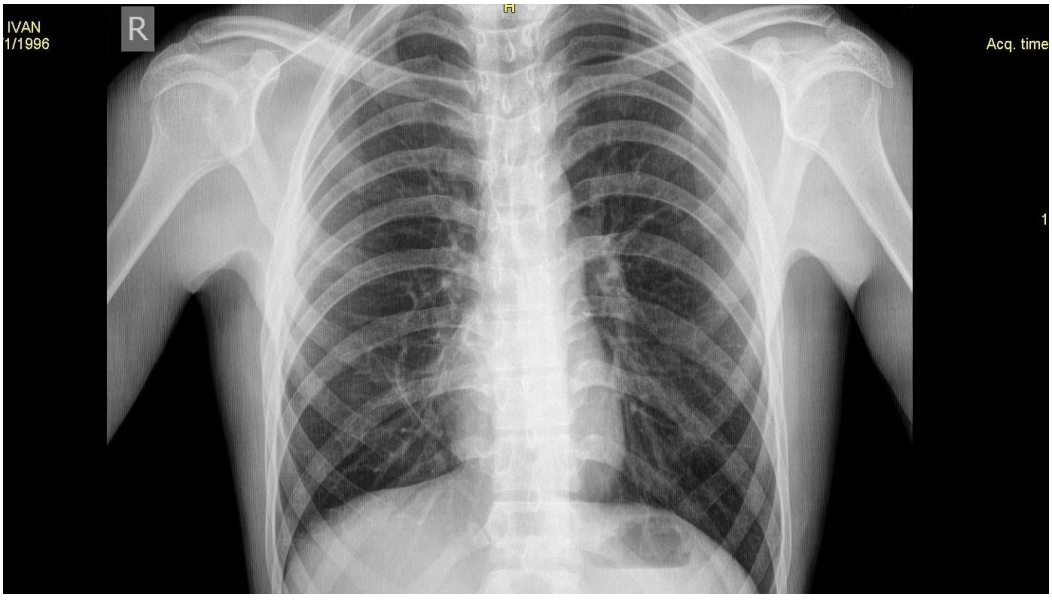

Figure 6. Rtg P-A three months after surgery, lung reexpansion is complete

Discussion

The pulmonary hamartoma is formed of embryonic remains, it is present in the fetal period, but is rarely detected before growing up (10). Although considered a benign tumor with a good prognosis, differentially diagnostically it is similar to carcinoid, tuberculosis, bronchogenic carcinoma, metastasis and hydatidic cyst (11). Hamartoma is typically of a smooth surface, a well-limited, moderately firm,

round or oval nodus of heterogeneous internal structure (2). The cytologic PH typically comprises a mixture of mesenchymal elements from fibromycose or cartilaginous connective tissue to mature cartilage and benign bronchial epithelial cells randomly alligned with-out necrosis in the background (5).

The pulmonary hamartoma usually occurs as a single nodus, but the occurrence of multiple, cystic and diffuse endotracheal hamartomas (12, 13) is possible. Multiple PHs have been described in Carney's triad: pulmonary chondromes, gastric epithelium leukemias, and functional extra-adrenal paragangliomas (11). There have been cases of malignant PH alterations that are extremely rare (4, 9, 14).

In PH diagnosis, the following are used: chest x-ray, computed tomography (CT), bronchoscopy, fine needle aspiration cytology (FNAC), surgical extirpation and HP verification. Positron-emission tomography (PET) is useful in the differentiation of malignant from benign solitary nodus in the lungs, but easy intake of 18F-FDG by PH was described, which makes it difficult to differentiate PH from the malignant nodular changes in the lungs (15).

Radiologically, the presence of "popcorn" calcification in PH is a pathognomonic radiological mark of hamartoma and is seen in 10-15% PH on the Rtg of the thorax, and according to some authors in up to 20-30% of cases (6,16).

Indications for removing PH and the way it will be removed are still controversial (16). It is believed that PH, which is symptomatic and large, should be removed (4). Surgical resection of tumefact in the lungs when diagnosis of PH is not confirmed is performed:

1) when it is difficult to distinguish between PH, metastasis, cancer, tuberculosis and other tumors of the lungs,

2) when patients with PH have respiratory problems due to endoluminal PH growth with postoperative complications,

3) when tumefact (PH) is rapidly increasing,

4) in patients who experience strong psychological pressure due to the presence of tumefact in the lungs of unknown origin and nature,

5) due to potential malignant alteration of PH (which is extremely rare, although described) (14, 16, 17).

In order to diagnose and treat the symptoms caused by PH, it can be removed bronchoscopically and surgically. It is desirable to remove PH, whenever possible, bronchoscopically (smaller, endoluminal PH) (18). Out of the surgical methods, VATS and conventional surgery are used (lateral and anterolateral thoracotomy are preferred) (17). Depending on the local finding, PH can be enucleated, wedge resectioned, removed by segmentectomy, lobectomy and, possibly, pulmectomy (in case of vast PH) (1). Hamartomas are moderately solid tumors and can be manually suppressed in the pulmonary parenchyma to the surface, after which a small incision on the visceral pleura is made, and the tumor is completely enucleated. Quite often a planned VATS procedure, due to the inability to identify intraparenchymal PH, has to be converted. In this case, it is desirable to make as small a cut on the chest (miitorocotomy) through which PH will be palpated and enucleated.

In the case presented PH was large and symptomatic. It was not possible to remove the same endobronchially. It was decided to remove PH surgically. First, VATS was made, and then the conversion into anterolateral torocotomy, with the idea to, with the smallest operative trauma per patient, remove hamartoma. Lobectomy of the central lobe was inevitable since hamartoma was large, it engaged almost the entire middle lobe and intimately grew with the bronchial tissue to the middle lobe with a break in his lumen.

Conclusion

Pulmonary hamartoma is a rare but most common benign tumor change in the lungs. Pulmonary hamartoma is most often detected incidentally as a side finding on the chest x-ray, in the form of asymptomatic, solitary, round tumor change, size of a coin, with "popcorn" calcifications. Differentially diagnostically lung hamartoma looks like carcinoid, tuberculosis, bronchogenic carcinoma, metastasis and hydatid cyst. In pulmonary hamartoma diagnostics chest x-ray, CT, bronchoscopy, FNAC, surgical extirpation and HP verification are used. Pulmonary hamartoma, which is symptomatic and large, should be removed. Whenever possible, pulmonary hamartoma should be removed minimally invasive-bronchoscopic or by VATS. For classic surgical approach, lateral and anterolateral thoracotomy are most convenient, and the lung hamartoma itself can be enucleated, wedge resected, removed by segmentectomy, lobectomy, and possibly by pulmectomy. In the presented case study, the lobectomy of the central lobe through anetolateral thoracotomy has been shown to be a method of choice in the treatment of a large, symptomatic lung hamartoma that engages the entire central lobe and has endoluminal growth in the bronchus for the middle lobe.

Disclosure: The authors declare no conflict of interest.

References

1. Shi H, Niu Z, Peng J, Yang Y, Chen L. Successful removal of a giant pulmonary hamartoma coexisting with an anomalous common pulmonary venous trunk. J Thorac Dis 2015; 7(3): E23-E27. [PubMed]
2. Itoga M, Kobayashi Y, Takeda M, Moritoki Y, Tamaki M, Nakazawa K, et al. A case of pulmonary hamartoma showing rapid growth. Case Rep Med 2013; 2013:231652. [PubMed]
3. Kishore M, Gupta P, Preeti, Deepak D. Pulmonary hamartoma mimicking malignancy: a cytopathological diagnosis. J Clin Diagn Res 2016; 10(11):ED06-ED07. [PubMed]
4. Umashankar T, Devadas AK, Ravichandra G, Yaranal PJ. Pulmonary hamartoma: Cytological study of a case and literature review. J Cytol 2012; 29(4):261-3. [CrossRef] [PubMed]
5. Liu Y, Chen G, Qiu X, Xu S, Wu Y, Liu R, et al. Intrapulmonary unicentric Castleman disease mimicking peripheral pulmonary malignancy. Thoracic Cancer 2014; 5:576-80. [CrossRef] [PubMed]
6. Thomas JW, Staerkel GA, Whitman GJ. Pulmonary hamartoma. Am J Roentgenol 1999; 172(6):1643. [CrossRef] [PubMed]
7. Joshi HM, Page RD. Giant pulmonary hamartoma causing acute right heart failure. Ann Thorac Surg 2014; 97(1):e21-2. [CrossRef] [PubMed]
8. Ucar N, Akpinar S, Aktas Z, Sipit T, Ozaydin E. Resection of endobronchial hamartoma causing recurrent hemoptysis by electrocautery and cryotherapy. Hippokratia 2014;18(4):355-6. [PubMed]
9. Lee BJ, Kim HR, Cheon GJ, Koh JS, Kim CH, Lee JC. Squamous cell carcinoma arising from pulmonary hamartoma. Clin Nucl Med 2011; 36(2):130-1. [CrossRef] [PubMed]
10. Hughes JH, Young NA, Wilbur DC, Renshaw AA, Mody DR. Fine-needle aspiration of pulmonary hamartoma: a common source of false-positive diagnoses in the College of American Pathologists Interlaboratory Comparison Program in Nongynecologic Cytology. Arch Pathol Lab Med 2005; 129(1):19-22. [PubMed]
11. Sodhi KS, Virmani V, Jindal SK, Khandelwal N. Pulmonary hamartoma. Ann Acad Med Singap 2009; 38(12):1110.
12. Miura K, Hori T, Yoshizawa K, Hamaguchi N, Morita J. Cystic pulmonary hamartoma. Ann Thorac Surg 1990; 49(5):828-9. [CrossRef] [PubMed]
13. Minami Y, Iijima T, Yamamoto T, Morishita Y, Terashima H, Onizuka M, et al. Diffuse pulmonary hamartoma: A case report. Pathol Res Pract 2005; 200(11-12):813-6. [CrossRef] [PubMed]
14. Basile A, Gregoris A, Antoci B, Romanelli M. Malignant change in a benign pulmonary hamartoma. Thorax 1989; 44(3):232-3. [CrossRef] [PubMed]
15. Wang W, Song J, Shi J, Hu H, Wu Y, Yan J, et al. Slight uptake of 18F-FDG on positron emission tomography in pulmonary hamartoma: a case report. Oncol Lett 2015; 10(1):430-32. [CrossRef] [PubMed]
16. Wang T, Liu Y. Outcomes of surgical treatments of pulmonary hamartoma. J Can Res Ther 2016; 12 (5): 116-9. [CrossRef] [PubMed]
17. Madan K, Sharma S, Singh N, Radhika S. Large pulmonary hamartoma with "popcorn" like calcification. Monaldi Arch Chest Dis 2011; 75(4):243-4. [PubMed]
18. Abdel Hady SM, Elbastawisy SE, Hassaballa AS, Elsayed HH. Is surgical resection superior to bronchoscopic resection in patients with symptomatic endobronchial hamartoma? Interact Cardiovasc Thorac Surg 2017; 24(5):778-2. [CrossRef] [PubMed]

Prikaz bolesnika

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LOBEKTOMIJA SREDNJEG REŽNJA KAO METODA IZBORA U LEČENJU VELIKOG ENDOBRONHIJALNOG HAMARTOMA SREDNJEG REŽNJA: PRIKAZ SLUČAJA

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Plućni hamartom je retka benigna tumorska promena u plućima. Često se otkrije slučajno, kao uzgredni nalaz na standardnom rendgenogramu grudnog koša u vidu asimptomatske, solitarne, okrugle tumorske promene veličine novčića sa „popcorn" kalcifikacijama. Iako se smatra za benigni tumor sa dobrom prognozom, diferencijalno dijagnostički liči na karcinoid, tuberkulozu, bronhogeni karcinom, metastaze i hidatidnu cistu. U dijagnostici plućnog hamartoma koriste se: standardni rendgenogram grudnog koša, kompjuterizovana tomografija, bronhoskopija, aspiraciona biopsija tankom iglom, hirurška ekstirpacija i histopatološka verifikacija. Smatra se da simptomatske i velike plućne hamartome treba ukloniti. Kad god je to moguće, plućni hamartom treba ukloniti minimalno invazivno, bronhoskopski ili video-asistiranom torakoskopskom hirurgijom. Za klasičan hirurški pristup najpodesnije su lateralna i anterolateralna torakotomija, a sam plućni hamartom se može enukleisati, klinasto resecirati, ukloniti segmentektomijom, lobektomijom i eventualno, pulmektomijom. Prikaz slučaja koji sledi opisuje lobektomiju srednjeg režnja kroz aneto-lateralnu torakotomiju kao metodu izbora u lečenju velikog, simptomatskog PH u srednjem režnju kod bolesnika starog 21 godinu.

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Ključne reči: *hamartom pluća, benigni tumor, lobektomija srednjeg režnja*