© Copyright by Uniwersytet Opolski
e-ISSN 2544-1620

Medical Science Pulse 2022 (16) 1

Original papers

Published online: 3 March 2022

DOI: 10.5604/01.3001.0015.7878

WEDGE RESECTION OR ENUCLEATION IN PULMONARY HAMARTOMA?

Hülya Dirol[1 A-E]
• ORCID: 0000-0002-7712-6467

Hakan Keskin[2 C-G]
• ORCID: 0000-0002-5736-5954

[1] Akdeniz University Hospital, Chest Department, Antalya, Turkey

[2] Akdeniz University Hospital, Chest Surgeon, Antalya, Turkey

A – study design, B – data collection, C – statistical analysis, D – interpretation of data, E – manuscript preparation, F – literature review, G – sourcing of funding

ABSTRACT

Background: Wedge resection and enucleation are the most commonly preferred surgical modalities for the treatment of pulmonary hamartomas. There is a concern about the safety and efficacy of enucleation for a pulmonary hamartoma.

Aim of the study: To compare wedge resection and enucleation for the surgical treatment of pulmonary hamartomas.

Material and methods: We retrospectively analyzed the data from patients who were surgically diagnosed with pulmonary hamartoma between January 2014 and December 2019. We obtained data on clinical findings, radiological features, surgical modality, postoperative complications, and follow-up results from the hospital electronic database. We compared all these features in the patients who underwent wedge resection with those who received enucleation.

Results: A total of 18 patients with pulmonary hamartoma were analyzed. The mean age of the patients was 60.56±9.25 years and 10 (55.6%) were male. Twelve (66.7%) patients had no respiratory symptoms. Ten (55.6%) of the patients underwent wedge resection and 8 (54.4%) patients had enucleation. There were no significant differences in age, gender, symptom distribution, prolonged air leakage, hospital stay length, intensive care unit stay length, mortality, or relapse rate between the patients with wedge resection and enucleation. Nodule size, calcification, metabolic activity, central or peripheral location, and lobar distribution were also similar.

Conclusions: We observed that there were no significant differences in the postoperative complications, hospital stay length, mortality or relapse between the patients with wedge resection and enucleation. Minimal parenchymal damage and resection should be the aim of surgical interventions for benign lung diseases. Therefore, we believe that enucleation should be the first choice for the surgical treatment pulmonary hamartomas.

KEYWORDS: hamartoma, wedge, enucleation

Background

Hamartoma is a kind of malformation composed of an unorganized combination of different tissues like cartilage, adipose tissue, smooth muscle, and respiratory epithelial cells [1,2]. Congenital malformations, hyperplasia of normal structures, benign tumors of the cartilage, and responses to inflammation are some of the proposed etiologies [1]. However, the exact etiology and pathogenesis is unknown. Patients are usually asymptomatic and cases are often detected on routine lung scans [3]. Tru-cut or fine-needle aspiration biopsy may be enough for treatment in cases with compatible radiological and clinical findings. However, surgery is inevitable in symptomatic cases or if malignancy cannot be ruled out.

Wedge resection and enucleation are the most commonly preferred surgical modalities for the treatment of hamartomas. There is a concern about the safety and efficacy of enucleation of a pulmonary

This is an Open Access article distributed under the terms of the Creative Commons License Attribution-NonCommercial-ShareAlike 4.0 International (CC BY-NC-SA 4.0). License available: https://creativecommons.org/licenses/by-nc-sa/4.0/

hamartoma. The amount and duration of air leakage in enucleation may be higher due to cutting of the lung parenchyma and suturing it afterward. Moreover, recurrence may occur as the safety margin is not clear and residual cells may cause relapse. On the other hand, the main target in surgery for benign lung diseases should be the minimal resection that preserves the most lung tissue. Enucleation may be better than wedge resection in this regard. Which of these two methods should be prioritized is currently uncertain.

Aim of the study

The aim of this study is to compare wedge resection and enucleation for the surgical treatment of pulmonary hamartomas.

Material and methods

Study design

This is a retrospective, single-center study. In this study, we included patients with pulmonary hamartoma diagnosed surgically in our hospital between January 2015 and December 2019.

Setting

We obtained the data about clinical features (age, gender, symptom distribution), radiological findings (nodule diameter, location, calcifications, metabolic activity), surgical procedures, postoperative complications (prolonged air leak, hospital stay length, intensive care unit [ICU] stay length, mortality) from the hospital electronic database.

Data sources/measurement

We compared enucleation with wedge resection in terms of postoperative complications, hospital stay length, ICU stay length, mortality, and long-term recurrence.

We received ethical approval for the study from the Akdeniz University Faculty of Medicine Clinical Research Ethics Committee (decision no: KAEK-448, date: 23.06.2021). We conducted the study under the principles of the Declaration of Helsinki.

Statistical methods

The collected data were entered into IBM SPSS v. 23 and all statistical analyses were performed with this program. Continuous variables were expressed as standard deviations, means, and minimum and maximum values, and categorical variables were expressed as percentages and frequencies. For comparisons between groups, the chi-squared test was used for categorical variables, the Student's t-test for parametric variables, and the Mann–Whitney U test for nonparametric variables. The relationships between the parameters were determined by Pearson's correlation analysis. A p-value of <0.05 was considered statistically significant.

Results

Descriptive data

The data from 18 patients with hamartoma who underwent surgical intervention were analyzed. The mean age of the patients was 60.56±9.25 years and 10 (55.6%) were male (Table 1).

Table 1. The characteristics of the study group

Variables		Total (n=18)
Age (years)		60.56±9.25
Gender	Female	8
	Male	10
Symptoms	Asymptomatic	12
	Symptomatic	6
	Cough	6
	Sputum	4
	Chest pain	2
Characteristic Calcification	Yes	3
	No	15
Nodule diameter [mm]		16.94±3.56
Hospital stay [days]		5.28±2.23
Localization	Peripheral	14
	Central	4
	Left lower lobe	4
	Left upper lobe	3
	Right lower lobe	5
	Right middle lobe	1
	Right upper lobe	4
Follow-up time [months]		34.39±12.05
SUV max		2.12±0.81

n – number; mm – millimeters; SUV max – maximal standardized uptake value.

There were no respiratory symptoms in 66.7% (n=12) of the patients. All of these patients were detected incidentally during radiological evaluation for another reason. The most frequent symptoms in symptomatic patients were cough (33.3%, n=6) and

sputum (22.2%, n=4). Fifteen (83.3%) patients did not have characteristic calcifications. In 77.8% (n=14) of patients, the hamartoma was located peripherally. Considering the lobar distribution, the right lower lobe (27.8%, n=5) was the most common localization. The mean nodule diameter was 16.94±3.56 mm. Wedge resection was performed in 55.6% (n=10) of patients and enucleation was performed in 44.4% (n=8).

There were no significant differences in age, gender, and symptom distribution between the patients who underwent wedge resection and those who underwent enucleation (Table 2).

Table 2. The comparison of enucleation and wedge resection.

Variables		Wedge (n=10)	Enucleatıon (n=8)	p
Age (year)		59.40±11.46	62.00±4.97	0.293
Gender	Female	6	2	0.313
	Male	4	6	
Symptoms	Asymptomatıc	6	6	0.866
	Symptomatıc	4	2	
	Cough	4	2	0.502
	Sputum	4	0	0.092
	Chest paın	2	0	0.473
Characterıstıc Calcification	Yes	2	1	0.832
	No	8	7	
Nodule dıameter [mm]		16.50±3.20	17.50±3.90	0.294
Hospıtal stay [day]		5.80±2.60	4.62±1.98	0.203
Localızatıon	Perıpheral	8	6	0.752
	Central	2	2	
	Left lower lobe	2	2	0.752
	Left upper lobe	2	1	0.834
	Rıght lower lobe	4	1	0.444
	Rıght mıddle lobe	1	0	0.943
	Rıght upper lobe	1	3	0.752
Follow-up tıme [months]		34.8±9.91	33.88±14.68	0.444
SUV max		2.06±0.59	2.17±0.98	0.266

n – number; mm – millimeters; SUV max – maximal standardized uptake value.

Nodule size, calcification, metabolic activity, location, and lobar distribution were also similar. There were no significant differences in prolonged air leakage, hospital stay length, ICU stay length, and mortality between the patients who underwent wedge resection and those who underwent enucleation. The mean follow-up time was 34.8±9.91 months in the patients who underwent wedge resection and 33.88±14.68 months in the patients who underwent enucleation. No recurrence occurred during the follow-up in both groups.

Discussion

Key results

Wedge resection and enucleation are the main surgical treatment modalities for pulmonary hamartomas. In this study, we found that there were no significant differences in prolonged air leakage, hospital stay length, ICU stay length, mortality and relapse between wedge resection and enucleation. We suggest that enucleation is as effective and as safe as wedge resection in the surgical treatment of pulmonary hamartomas. Therefore, enucleation can be prioritized for functional lung tissue preservation.

Hamartoma, which was first described by the German pathologist Eugen Albrecht in 1904, is a malformation that results from the disorganized proliferation of at least two mature mesenchymal tissues. The name is derived from the Greek word hamartia meaning faulty and is used in this instance to refer to faulty tissue development [4]. Hamartomas can be seen in many different parts of the body, but one of the most common sites is the lung. Pulmonary hamartoma is also the most common benign tumor of the lung. It is more common in men and usually develops between the ages of 30 and 60 years [5]. The mean age of the patients in our study was 60.56±9.25 years, and our youngest patient was 36 years old, while the oldest was 76 years old. Often patients with hamartomas are asymptomatic and are identified on lung screening [3]. Similarly, about two-thirds of our patients were asymptomatic and the most common complaints of the symptomatic patients were cough and sputum.

Pulmonary hamartoma usually presents as a solitary pulmonary nodule, smaller than 3 cm in diameter. However, it can also be in the form of diffuse lesions, starting from the endobronchial tree and spreading to the pulmonary parenchyma [3,6]. In our study, they were all in the form of solitary nodules. The mean diameter was 16.94±3.56 mm, with the smallest nodule being 11 mm in diameter and the largest 24 mm. Some pulmonary hamartomas may grow, but this growth generally tends to be very slow. Some studies have reported that there is a relationship between lesion diameter and patient age. However, in our study, we did not find a relationship between nodule size and age.

Besides being solitary, the pulmonary hamartoma is typically smooth-circumscribed or slightly

lobulated [7]. Fat within the nodule is diagnostic, but this finding is found in only half of the patients. Popcorn or comma-shaped calcification is pathognomonic, but they are only present in 5–50% of the patients [8,9]. A previous study found no hamartomas with popcorn calcifications and the authors suggested that these calcifications could not be considered characteristic for hamartoma [10]. There was no fat density or popcorn calcification in any of our patients either.

Pulmonary hamartoma is usually located in the periphery of the lungs [11–14]. In our study, we found that the right lower lobe was the most common localization, but there was no significant difference between being in the right lung and left lung. In our study, they were mostly located in the periphery of the lung.

The presence of popcorn calcification and a high-density outer margin with lobulation may be enough for the radiological diagnosis of pulmonary hamartoma [15]. However, 20–30% of lung cancers present as a solitary pulmonary nodule. Moreover, some pulmonary hamartomas does not show typical radiological features. Thus, it may be impossible to differentiate them from primary or metastatic lung cancer without tissue sampling. A percutaneous biopsy is preferred for cytological evaluation in such cases. The positive predictive value of cytology in diagnosis is about 85%. However, it should be kept in mind that cytological diagnosis can be reliable only if the diagnosis is supported by radiological and clinical findings [16]. It should be noted that up to 22% of false-positive cytological results occur in carcinoid tumors, adenocarcinomas, or small cell carcinomas [17].

Pulmonary hamartoma generally does not cause mortality or morbidity unless they grow to a large extent or they are localized in the bronchi. In these cases, they can cause a mass effect and obstructive pneumonia or atelectasis [18]. As pulmonary hamartoma is benign and grows slowly, they are suggested to be followed without any need for surgery in cases with a definitive diagnosis without surgery [19]. However, in symptomatic cases or if malignancy cannot be ruled out, a surgical procedure is inevitable for both diagnosis and treatment.

The definitive curative treatment for pulmonary hamartoma is absolutely surgery. Lobectomy, and rarely pneumonectomy, may be necessary if they are localized in the deep parts of the lobe, if the distal lung tissue is non-functional or if they are multiple and giant [18,20]. In other cases, wedge resection or mass enucleation is preferred to protect the lung [21]. Minimal parenchymal damage and resection are recommended in the surgery of benign diseases detected in the lung. Enucleation in pulmonary hamartoma is a surgical modality that can decrease lung parenchymal loss when compared to wedge resection. In our study, we did not find a statistically significant difference between enucleation and wedge resection in the length of hospital stay, need for intensive care, postoperative air-leakage, mortality, and recurrence in long-term follow-up. Enucleation seems to be safe and effective in the surgical treatment of pulmonary hamartomas. Thus, it should the first choice among the surgical modalities for pulmonary hamartoma management in order to prevent lung parenchymal loss as much as possible.

Limitations

This study has several limitations. It is a single-center study and a limited number of cases were analyzed. Our follow-up period was 3 years on average, but pulmonary hamartomas grow very slowly. Thus, the follow-up period in our study may not be enough to rule out recurrence.

Conclusions

In conclusion, the main goal in the surgical treatment of pulmonary hamartoma should be the preservation of functional lung tissue as much as possible. Enucleation should be the first choice among surgical modalities. This procedure is as effective and as safe as wedge resection and, moreover, it is better than wedge resection in preserving lung parenchymal loss.

References

1. Wolfgang D. Radiology review manual. 3rd ed. Baltimore: Lippincott Williams & Wilkins; 1996.
2. Zheng RH, Chou DH, Shi MX. Hamartoma of the lung: an analysis of 24 cases. Zhonghua Jie He He Hu Xi Za Zhi 1994;17:86-7.
3. Hutter J, Reich-Weinberger S, Hutarew G, Stein HJ. Giant pulmonary hamartoma-a rare presentation of a common tumor. Ann Thorac Surg 2006;82:5-7.
4. Leiter Herrán F, Restrepo CS, Alvarez Gómez DI, Suby-Long T, Ocazionez D, Vargas D. Hamartomas from head to toe: an imaging overview. Br J Radiol 2017;90:20160607.
5. Hansen CP, Holtveg H, Francis D, Rasch L, Bertelsen S. Pulmonary hamartoma. J Thorac Cardiovasc Surg 1992; 104:674-8.
6. Minami Y, Iijima T, Yamamoto T, Morishita Y, Terashima H, Onizuka M, et al. Diffuse pulmonary hamartoma: a case report. Pathol Res Pract 2005;200:813-6.

7. Acar M, Özateş M, Ekici F, Şimşek M. Akciğer hamartomu. Türk Tanısal ve Girişimsel Radyoloji Dergisi 2001;7:373-5. (In Turkish).
8. Lu Z, Qian F, Chen S, Yu G. Pulmonary hamartoma resembling multiple metastases: a case report. Oncol Lett 2014;7:1885-88.
9. Radosavljevic V, Gardijan V, Brajkovic M, Andric Z. Lung hamartoma: diagnosis and treatment. Med Arch 2012;66:281-2.
10. Crouch JD, Keagy BA, Starek PJ, Delany DJ, Wilcox BR. A clinical review of patients undergoing resection for pulmonary hamartoma. Am Surg 1988;54:297-9.
11. Gjevre J, Myers J, Prakash U. Pulmonary hamartomas. Mayo Clin Proc 1996;71:14–20.
12. Salminen US. Pulmonary hamartoma. A clinical study of 77 cases in a 21-year period and review of literature. Eur J Cardiothorac Surg 1990;4:15-8.
13. Van den Bosch JM, Wagenaar SS, Corrin B, Elbers JR, Knaepen PJ, Westermann CJ. Mesenchymoma of the lung (so called hamartoma): a review of 154 parenchymal and endobronchial cases. Thorax 1987;42:790-3.
14. Wang T, Liu Y. Outcomes of surgical treatments of pulmonary hamartoma. J Cancer Res Ther Suppl 2016;12:116-9.
15. Madan K, Sharma S, Singh N, Radhika S. Large pulmonary hamartoma with "popcorn" like calcification. Monaldi Arch Chest Dis 2011; 75:243-4.
16. Hansen CP, Holtveg H, Francis D, Rasch L, Bertelsen S. Pulmonary hamartoma. J Thorac Cardiovasc Surg 1992;104:674-8.
17. 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 Inter laboratory comparison program in non-gynecologic cytology. Arch Pathol Lab Med 2005;129:19-22.
18. Kishore M, Gupta P, Preeti, Deepak D. Pulmonary hamartoma mimicking malignancy: a cytopathological diagnosis. J Clin Diagn Res 2016;10:6-7.
19. Ramirez-Chavez G, Celard P, Gamondes JP, Loire R, Pinet F, Cordier JF, et al. Pulmonary hamartoma. Apropos of 49 surgically treated cases. Sem Hop 1984;60:1181-4.
20. Radosavljevic V, Gardijan V, Brajkovic M, Andric Z. Lung hamartoma: diagnosis and treatment. Med Arch 2012;66:281-2.
21. Khan AN, Al-Jahdali HH, Irion KL, Arabi M, Koteyar SS. Solitary pulmonary nodule: a diagnostic algorithm in the light of current imaging technique. Avicenna J Med 2011;1:39-51.

Word count: 1927 • Tables: 2 • Figures: 0 • References: 21

Sources of funding:
The research was funded by the authors.

Conflicts of interests:
The authors report that there were no conflicts of interest.

Cite this article as:
Dirol H, Keskin H.
Wedge resection or enucleation in pulmonary hamartoma?
Med Sci Pulse 2022; 16(1): 33-37. DOI: 10.5604/01.3001.0015.7878.

Correspondence address:
Hülya Dirol
Dumlupınar Boulevard Akdeniz University Hospital 07059,
Campus, ANTALYA
E-mail: hulyadirol@akdeniz.edu.tr

Received: 14.12.2021
Reviewed: 23.02.2022
Accepted: 23.02.2022

Copyright of Medical Science Pulse is the property of Public Higher Medical Professional School in Opole and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.