ORIGINAL ARTICLE

MEDIASTINAL BRONCHOGENIC CYSTS RESECTION IN ADULTS: RESULTS OF VATS AND THORACOTOMY PROCEDURES

El Hassane Kabiri^{1,2}, Massine El Hammoumi¹, Mounia Griguihi³, Mohamed Bhairis¹, Souheil Boubia^{3,4}, Mohammed Ridai^{3,4}, Meryem Kabiri^{2,5}

Department of Thoracic Surgery - Mohammed V Military Teaching Hospital - Rabat, Morocco
Faculté de Médecine et de Pharmacie- Université Mohammed V- Rabat, Morocco
Department of Thoracic Surgery - Ibn Rochd CHU - Casablanca, Morocco
Faculté de Médecine et de Pharmacie - Université Hassan II - Casablanca, Morocco
Department of Pediatrics- Rabat Children Hospital - Rabat, Morocco

* Corresponding author: hassankabiri @yahoo.com

Abstract

Background: The objectives of our study are to evaluate our surgical experience of mediastinal bronchogenic cyst (MBC) and to determine the results of resection by video-assisted thoracic surgery (VATS) and posterolateral thoracotomy (PLT).

Methods: The demographic characteristics, clinical and radiological features intraoperative data, outcomes and follow-up information were reviewed and analyzed from 38 patients who underwent resection of a MBC between 2008 and 2019.

Results: cystectomy was performed for thirty eight patients included in the present study. Seventeen of them, benefited from VATS cystectomy (VATS group) with 1 conversion to thoracotomy (5.9%) and 21 underwent PLT cystectomy (PLT group).

In our series 27 (71.1%) were male. Their average age was 42.6 years. While 09 patients (23.7%) had no symptoms preoperatively, 29 patients (76.3%) were symptomatic.

There were no operative deaths and 3 patients (7.9%) presented postoperative complications. The average duration of hospital stay was 3.2 days for patients who had VATS, but 5.8 days for those who had thoracotomy. Long-term follow-up (range, 1 to 8 years) showed no late complications and no recurrence.

Conclusion: VATS and PLT are main approaches for the surgical resection of MBCs VATS is a safe procedure, with less pain and time spent at the hospital. Early surgical procedures of MBCs may be recommended to prevent complications. Surgical adhesions are unfavorable conditions to thoracoscopic treatment.

Key Words: bronchogenic cyst, mediastinum, video-assisted thoracic surgery (VATS); thoracotomy

INTRODUCTION

Bronchogenic cysts are rare congenital malformations of bronchial development from the diverticulum of the primitive foregut or the tracheobronchial tree in the embryonic period between the 26th and 40th days of gestation^{1, 2}. It may develop in the mediastinum, or intrapulmonary parenchyma but rarely in extra-thoracic sites ^{1, 2}. They represent 15 to 20% of all primary mediastinal malformations^{3, 4}, 7% of all mediastinal tumors and 33% of mediastinal cysts. These

incidences of mediastinal bronchogenic cysts (MBCs) aren't a reflection of the reality because most patients are asymptomatic^{4, 5}.

Standard treatment for mediastinal bronchogenic cysts is resection through thoracotomy. However, over the last decade thoracoscopy and VATS are now the best approaches.

Thus, the data of patients operated for MBC were retrospectively reviewed and compared the results of thoracotomy and VATS.



Table 1 Comparison of the groups according to the demographic characteristics of the 38 patients

Variables		Thoracotom y (n=21)	VATS (n=17)	p value
		55.3%	44.7%	
Gender	Male	16	11	
	Female	05	06	
Age (years)		45.7± 2.5	41.8±1.9	0.054
		(31-69)	(18-53)	
Side	Right	14	11	0.084
	Left	07	06	
Symptom	No	04	05	
	Yes	17	12	
	Cough	12	07	0.075
Symptoms	Chest Pain	08	06	
	Dyspnea	05	01	
	Dysphagia	02	00	
	Hoarseness	01	00	
Cyst diameter (cm)	<5	08	14	0.063
	>5	13	03	
Anatomical location	Anterior mediastinum	01	01	
	Middle	14	11	
	Posterior mediastinum	06	05	
Cyst diameter (cm)		5.8± 2.1	4.3±1.9	0.048
		(4.5-8.3)	(2.6-5.2)	
		,	,	
Maier classification	Type I(paratracheal)	03	02	
	Type II(carinal)	10	03	
	Type III(Hilar)	06	01	
	Type IV(paraesophageal)	02	07	
	Type V(Miscellaneous)	00	04	

MATERIAL AND METHODS

This a retrospective study of patient who underwent resection of MBC from 1st January 2008 to 31th December 2019 (12 years). 38 patients were operated by thoracic surgeons from 2 General thoracic surgery centers in

Morocco.

General data (age, gender, clinical history, symptoms), radiological findings in chest-X-ray, CT or/and MRI of the chest (site, size and adherences to vital structures), surgical techniques, intraoperative and postoperative complications,



hospital stay, mortality and follow-up were obtained from medical records. The indications for VATS were small cyst size, and no previous lung disease predictive of adhesion. Patients were followed up one month after surgery, three months later and every year for 5 years. After 5 years of follow-up the risk of recurrences is considered very low.

We express values in the text and tables as mean value. Analysis was performed by the software Statistical Package for the Social Sciences (SPSS Statistics, version 15, SPSS Inc.) system. Parametric independent t-test was used for group differences. A variable was statistically significant when p value was less than 0.05.

RESULTS

Our study included 27 men and 11 women with 42.6 ± 3.5 years average age (18 to 69 year-olds)

Symptoms were noted in 29 patients, including one patient with acute dyspnea secondary to cyst complicated by intra pleural rupture. Discovery of the cyst was fortuitous in 9 patients through chest radiological exams.

Preoperative radiological data included chest-x-ray and computed tomography scan (CT) in all cases. Magnetic Resonance Imaging (MRI) in 08 cases. Bronchoscopy was done only in 3 cases.

On chest- X radiographs the cysts were described as homogeneous water-density opacity in the mediastinum in 36 cases, with air-fluid levels in 1 case and left pleural effusion in 1 case.

In the chest CT scan data, the localization of the cysts was in the middle mediastinum in 25 cases (65.8%) (Right hilar 7; subcarinal 13; paratracheal 5), in the posterior mediastinum (paraesophageal) in 11 cases (28.9%), postero–superior mediastinum in 2 (5.3%) of the patients and the antero-superior mediastinum in 02 cases. The cyst's average diameter was 4.8 cm (range: 2.6 –8.3 cm). Also, there was no change after the contrast injection.

Eight patients had CT and MRI scans. On T1-weighted images, 05 cysts were little hyper-intense while the other 03 cysts were iso-intense. On T2-weighted MRI, 02 cysts were much hypo-intense and 06 cysts were isointense. There is no enhancement after injection of gadolinium in MRI scan.

The preoperative CT scan was performed to precise the cyst size, location and connections to neighboring organs or tissues to minimize the per-operative difficulties and to avoid injuries intra-operatively.

Using the Maier's classification⁶ the location is in five sites, paratracheal, paraesophageal, carinal, hilar and miscellaneous (table 1)

Patients were operated through posterolateral thoracotomy (n=21) or video-assisted VATS (n=17).

Complete removal of the cyst was performed in 33 patients (86.8%) while incomplete resection in only 5 patients (13.2%) because of important adhesions to vital

structures and the dissection was very dangerous, so a part of the cyst wall was electrocuted and left in place (table 2). In one case, a wedge resection for pulmonary parenchyma destruction by compression of the cyst was done.

The average operating time was 105 min from 75 to 125 min in thoracotomy group and 75 min from 65 to 90 min in VATS group. Simple cysts with small size without connections to vital structures was chosen for VATS procedures. Intraoperative complications occurred in 01 patient (3.2%) who had a chylothorax requiring a ligation with clip of the thoracic duct.

No operative related deaths and postoperative complications included 03 patients with pulmonary

Table 2

Perioperative outcomes

Variables	Thoracotomy (n=21)	VATS (n=17)	p value
Approach			
Thoracoscopy	_	16	
Conversion	-	01	
Thoracotomy	21	-	
Mediastinal Adhesions			
No	0	14	0.045
Yes	18	01	
Additional procedures:			
Adhesiolysis	16	05	
Bronchial suture	01	_	
Azygos vein resection	02	-	
Pericardial resection	02	_	
Resection			
Complete	18	15	
Incomplete	03	02	
Operation time (minutes)	105±5 (75-125)	75±6 (65-90)	0.026
Postoperative complications	03	00	
Pleural effusion	01		
Pneumonia	01		
Wound infection	01		
Drainage time (days)	3.4±1.2 (2-6)	1.7 ±0.9 (1-3)	0.045
Post-operative Stay (days)	5.8± 1 (3-8)	3.2±0.5 (2-4)	0.041
Follow-up period (months)	58±6 (34-96)	44±8 (12-66)	0.066

infection (n=1), one with wound infection and one with pleural effusion in a patient who had intraoperative chylothorax in group of the thoracotomy (table 2).

Chest tubes were left in place for an average of 1.7 days in group of VATS and 3.4 in the thoracotomy group. Chest drain removal was possible when the chest X ray is normal with volume drainage lower than 200cc. the hospital stay was longer in the thoracotomy group (5.8 days) than VATS group (3.2 days).

Pathologic exam confirmed the diagnosis of bronchogenic cyst in all cases. All lesions were benign.

No late complications and no recurrence of the cyst were noted in the follow-up period from 12 to 96 months (average: 52 months) in 15 cases (88.2% of VATS Group) and 14patients (66.67%) of thoracotomy group. The other patients were lost for follow-up particularly during Covid 19 pandemy.

DISCUSSION

MBCs are rare benign bronchopulmonary congenital malformations of the primitive foregut that are thought to emerge from the ventral foregut during the early stages of gestation (days 26-40)^{6, 7}. MBCs account for 40-50% of all mediastinal cysts. The incidence of MBCs has increased in recently due to the spread and frequent radiological examinations^{1, 8, 9}.

According to the literature, the majority of patients with MBC are assymptomatic (50-79%), but there are no specific symptoms for the diagnosis of MBC preoperatively. Moreover, symptoms are always secondary to the size of the cyst and compression of the mediastinal structure ³.

The most common clinical signs are cough, chest pain, dyspnea, dysphagia and hoarseness ⁸. Occasionally, there are symptoms related to complications, including compression, pleural and/or pericardial rupture, infection, or intracystic hemorrhage. In our study, 29 (76.3%) patients were symptomatic on admission. The most important imaging methods (thoracic CT and MRI) allowed the detection of all mediastinal cystic lesions with precise location, size and especially adhesions to mediastinal vital organs. The content of the cyst: fluid, hemorrhagic, viscous or infected and sometimes mucoid like a solid tumor can be explored by CT scan and MRI. Therefore, the importance of MRI is to differentiate MBC from other mediastinal malignancies ^{1, 10}.

Recently, ultrasound and MRI have enabled prenatal diagnosis ^{1, 10, 11}. Fluorodeoxyglucose (FDG) positron emission tomography (PET) is not indicated in mediastinal cysts but when the mediastinal bronchogenic cyst is infected, a focal increase in FDG uptake mimicking malignancy is possible ¹².

The treatment of mediastinal bronchogenic cysts is surgical resection but the timing of this procedure is still debated, especially for asymptomatic patients ^{1, 10} but some authors have suggested surgery for all patients

because the spontaneous evolution of asymptomatic cysts is unclear with possibilities of complications such as compression of pulmonary structures with dyspnea and/or chest pain, compression of vascular structures such as vena cava syndrome, fistulization in the bronchial tree, infections or hemorrhage ^{1,11}. The possibility of life-threatening cardiac tamponade is extremely rare, following intrapericardic rupture of the bronchogenic cyst ¹⁶⁻¹⁸. The risk of malignant transformation has been reported in the literature but is exceptional (less than 0.7%) ^{1, 2, 11-16}. Spontaneous involution of mediastinal bronchogenic cysts does not occur according to the literature unlike intrapulmonary forms that fistulate into the airways and spill their contents ¹⁷.

Some authors suggest early surgical resection when the diagnosis of MBC is suspected ¹. Fievet et al ¹¹ propose early surgery 6th to 12th month of life because surgery allows confirmation of the diagnosis and prevention of complications ¹⁶, but others indicate surgery only if malignancy is suspected ^{1, 3}.

Over the past decade, posterolateral thoracotomy cystectomy has been the main surgical procedure for the treatment of MBC. The emergence of video thoracoscopic surgery (VATS) and robotic surgery has been increasingly used

The safety of VATS in MBC has been discussed by many authors ^{1, 3, 11}. The choice of technique is related to the location and particularly size of the cysts. When the cyst is small, it is easy to remove completely, but in larger forms, the bronchogenic cyst wall may adhere to vital structures and thus be more difficult to remove completely. The same is true for complicated cysts with inflammatory reactions or adhesions ³.

Some authors have compared the advantages of VATS for adults and/or children undergoing excision of a CBM and conventional operative methods and concluded that the VATS procedure decreases the duration of chest drainage. It is associated with shorter operative time, shorter postoperative hospitalization, less intraoperative blood loss, and rapid return to activity and/or labor. It is associated with less intraoperative blood loss, shorter operative time, shorter postoperative hospitalization, and early return to life activity 1, 18. VATS didn't increase the risk of surgical complications such incomplete removal, or long stay in intensive care unit 1, 11. Fievet et al ¹¹ compared symptomatic vs. asymptomatic patients (adults and children) and concluded that there are more intraoperative and postoperative complications in symptomatic group in adult and only in this group for children's. Technically surgery was more difficult, with necessary additional procedures (bronchial or esophageal suture, huge adhesiolysis, Azygos vein or thoracic duct ligation, muscle or pericardial flaps resection).

Late recurrences after incomplete removal of all the cyst wall have been described some years later^{1,19}, thus, complete cystectomy should be purposed as



often as possible except when severe adhesion to vital structures.

Some authors' preconized electrocautery in the remaining cyst wall to prevent local recurrences 1; we used these procedures.

Robotic thoracic surgery 28 and endobronchial ultrasound (EBUS) with transbronchial needle aspiration (TPNA) ^{11, 21} are still in development. Maturu et al ²¹ reports 32 patients treated with TPNA without mortality and no recurrences but more complications 16.1% (n; 5) (infection in 4 and bradycardia in1patient) with a short follow-up (14 months).

Aravena et al ²² concluded that diagnostic and therapeutic EBUS-TBNA for mediastinal cysts (Bronchogenic cyst: 4) is limited with successful long-term treatment occurring in only 5.5% of the patients (all mediastinal cysts types) and surgical resection is the treatment of choice.

CONCLUSION

Posterolateral thoracotomy (PLT) and VATS are the approaches of choice for the treatment of MBCs knowing that VATS approach is easier and safer than PLT according to the literature. In our study despite the smaller sample of case series and initial selection of VATS or PLT, VATS provided satisfactory results.

Surgical treatment of MBC can be recommended early, before patients become symptomatic, to prevent intraoperative difficulties and postoperative complications.

Patients who have undergone incomplete surgical resection should have long-term follow-up because of the risk of recurrence.

REFERENCES

- 1. Guo C, Mei J, Liu C et al. Video-assisted thoracic surgery compared with posterolateral thoracotomy for mediastinal bronchogenic cysts in adult patientsJThorac Dis 2016 Sep; 8(9):2504-2511.
- 2. Granato F, Voltolini L, Ghiribelli C et al. Surgery for bronchogenic cysts: always easy? Asian CardiovascThorac Ann 2009; 17:467-71.
- 3. Jung HS, Kim DK, Lee GD et al. Video-assisted thoracic surgery for bronchogenic cysts: is this the surgical approach of choice?InteractCardiovascThoracSurg 2014; 19(5):824-9.
- 4. De Giacomo T, Diso D, Anile M et al. Thoracoscopic resection of mediastinal bronchogenic cysts in adults. Eur J Cardiothorac Surg. 2009; 36(2):357-9.
- 5. Panchanatheeswaran K, Dutta R, Singh KI et al. Eleven year experience in thoracoscopic excision of bronchogenic cyst. Asian CardiovascThorac Ann 2012; 20:570-4.
- 6. Maier HC. Bronchogenic cysts of the mediastinum. Ann

- Surg 1948; 127: 476-502.
- 7. Wang X, Li Y, Chen K et al. Clinical characteristics and management of primary mediastinal cysts: A single-center experience. Thorac Cancer 2020; 11(9):2449-6.
- 8. Wang X, Chen K, LiY et al. The Video-Assisted Thoracic Surgery for Mediastinal Bronchogenic Cysts: A Single-Center Experience. World J Surg 2018; 42(11):3638-45.
- 9. Aker C, Sezen CB, Dogru MV et al. Prognostic Factors and Long-Term Results in Patients Who Underwent Videothoracoscopic Bronchogenic Cyst Excision. Ann ThoracCardiovascSurg 2021; 27(4):225-9.
- 10. Fievet L, D'Journo XB, Guys JM etal.Bronchogenic cyst: best time for surgery? Ann Thorac Surg. 2012; 94(5):1695-9.
- Fievet L, Gossot D, de Lesquen H et al. Resection of bronchogenic cysts in symptomatic versus asymptomatic patients: an outcome analysis. Ann Thorac Surg2020; S0003-4975(20)30969-3.
- Zhang J, Ni Y, Dong A. Infected Mediastinal Bronchogenic Cyst Mimicking Malignancy on FDG PET/CT.Clin-Nucl Med 2020;45(2):172-173
- 13. Han SJ, Cho HJ, Kang MW et al. A Life-Threatening Bronchogenic Cyst.Korean J ThoracCardiovascSurg 2018; 51(1):69-71.
- 14. Lateef N, Kuniyoshi J, Latif A et al. Cardiac tamponade as a complication of bronchogenic cyst.Proc (BaylUniv Med Cent) 2020; 34(1):172-174.
- Yildiz H, Reichwein R, Poncelet A et al. An unusual case of cardiac tamponnade: Bronchogenic cyst infection due to Salmonella bredeney. J Infect Chemother. 2019; 25(2):151-153.
- Fiorelli A, Rambaldi P, Accardo M et al. Malignant transformation of bronchogenic cyst revealed by 99mTc-MIBI-SPECT. Asian CardiovascThorac Ann 2012; 20(3): 347-9.
- 17. Shin MS, Buchalter SE, Ho KJ. Intrapulmonary bronchogenic cyst: spontaneous dissolution? Ala Med 1990; 59(10):20-2.
- 18. Tölg C, Abelin K, Laudenbach V et al. Open vs thorascopic surgical management of bronchogenic cysts. SurgEndosc 2005; 19:77-80.
- 19. Hasegawa T, Murayama F, Endo S, et al. Recurrent bronchogenic cyst 15 years after incomplete excision. Interact CardiovascThoracSurg 2003; 2: 685–7.
- 20. Chen K, Zhang X, Jin R et al. Robot-assisted thoracoscopic surgery for mediastinal masses: a single-institution experienceJThorac Dis 2020; 12(2):105-113.
- Maturu VN, Dhooria S, Agarwal R. Efficacy and Safety of Transbronchial Needle Aspiration in Diagnosis and Treatment of Mediastinal Bronchogenic Cysts: Systematic Review of Case Reports. JBronchology IntervPulmonol. 2015; 22(3):195-203
- 22. Aravena C, Patel J, Goyal A et al. Role of Endobronchial Ultrasound-guided Transbronchial Needle Aspiration in the Diagnosis and Management of Mediastinal Cyst. JBronchologyIntervPulmonol. 2020; 27(2):142-146.

