



## Original Article

## Benign Primary Cystic Lesions of Mediastinum in Adult: The Clinical Spectrum and Surgical Treatment

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## ABSTRACT

**Background and objective:** The mediastinal cysts form a group of heterogeneous and uncommon benign lesions of neoplastic, congenital, or inflammatory conditions. The forgoing controversy is how to manage them; surgical removal or observation. We reviewed our experience including some rare conditions, emphasizing the clinical spectrum and surgical treatment.

**Patientes and methods:** This is a retrospective review between 2000 and 2007 included 34 cases of primary mediastinal cystic lesions. Clinical features, imaging techniques, surgical operation, morbidity, mortality and follow-up were analyzed.

**Results:** There were 18 females (53%) and 16 males (47%), with a mean age  $\pm$  standard deviation of 45.3  $\pm$  14.1 years (range: 22-74). Most of cysts were congenital (94%), except patients with hydatid disease (6%). 24% of cysts (n = 8) were detected in anterior mediastinum. Rest of them (n = 26) were located in visceral mediastinum. Patients usually were symptomatic (61%). Chest pain and discomfort was most common symptom, others were dyspnea, cough and hemoptysis, respectively. Cysts excision was performed in all cases with an uneventful recovery and with no recurrence in long term follow up.

**Conclusions:** Asymptomatic mediastinal cysts are not rare. Surgery is a reliable method of treatment of mediastinal cysts with acceptable mortality and morbidity.

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## Lesiones quísticas primarias y benignas del mediastino en el adulto: espectro clínico y tratamiento quirúrgico

## RESUMEN

**Introducción:** Los quistes mediastínicos forman un grupo de lesiones benignas heterogéneas y poco comunes de naturaleza neoplásica, congénita e inflamatoria. Continúa la controversia sobre cómo tratarlos, ya sea mediante extracción quirúrgica u observación. Examinamos nuestra experiencia en este campo, que incluye algunas situaciones infrecuentes, haciendo hincapié en el espectro clínico y el tratamiento quirúrgico.

**Pacientes y métodos:** Se ha realizado un estudio retrospectivo de 34 pacientes atendidos entre 2000 y 2007 por lesiones quísticas primarias del mediastino. Se analizaron las características clínicas, las técnicas de imagen empleadas, el tipo de cirugía, la morbilidad, la mortalidad y el seguimiento.

**Resultados:** El grupo estuvo constituido por 18 mujeres (53%) y 16 varones (47%), con una media de edad ( $\pm$  desviación estándar) de 45,3  $\pm$  14,1 años (rango: 22-74). La mayoría de los quistes eran congénitos (94%), excepto en los pacientes con enfermedad hidatídica (6%). El 24% de los quistes (n = 8) se detectó en el mediastino anterior y el resto (n = 26) se localizó en el mediastino visceral. La mayoría de los pacientes (61%) presentaba síntomas. El dolor y el malestar torácicos fueron los síntomas más frecuentes, seguidos de la disnea, la tos y la hemoptisis. En todos los casos se procedió a la escisión del quiste, con la recuperación sin incidentes de los pacientes y sin recurrencias en el seguimiento a largo plazo.

**Conclusiones:** Los quistes mediastínicos asintomáticos no son infrecuentes. La cirugía es un tratamiento seguro para los quistes mediastínicos, con una mortalidad y morbilidad aceptables.

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## Palabras clave:

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## Introduction

Mediastinal cysts are heterogeneous group of neoplastic, congenital, and inflammatory conditions and account for 20% to 32% of all primary mediastinal masses<sup>1</sup>. They are relatively infrequent in daily practice and significant proportions of these lesions are found incidentally through routine investigations for other indications<sup>2</sup>. They include different pathologic entities with overlapping clinical and radiologic features. These lesions are encountered both in child and adult population. Classification is based on etiology<sup>2,3</sup>.

Classification is somewhat controversial and some minor exceptions and acceptions depend on authors' appreciation. The controversy regarding on these cysts is whether to manage them with observation or surgical removal. We sought to elucidate the clinical spectrum and characteristic of mediastinal cystic lesions. This is a retrospective study reviewing the clinical characteristics, diagnostic and therapeutic modalities for the primary spectrum of cystic lesions of the mediastinum.

## Patients and Methods

A comprehensive review was undertaken between 2000 and 2007. Over this 7-year period, 34 patients were diagnosed with cystic masses of the mediastinum, all of whom subsequently underwent surgical intervention. Clinical presentation, radiologic findings and mode of surgical procedures were recorded for all patients. Radiological evaluation was primary step. Deformed cystic structures because of infection, intracystic hemorrhage or rupture were excluded and radiologically intact cystic structures were evaluated. Histopathological diagnosis was established. We include all primary cysts of mediastinum with primary hydatid disease of mediastinum that is not true cyst but has indistinguishable radiologic and clinical features with primary cysts of mediastinum. We exclude tumoral cystic degeneration, mediastinal abscess, pancreatic pseudocysts, all had no true epithelial lining or with irregular cystic wall thicker than 3 mm.

Statistica v5.1 (StatSoft, Inc., Tulsa, OK, USA) software programme and Fisher's exact test or Pearson  $\chi^2$  test were used in statistical analysis and  $P < 0.05$  was accepted meaningful.

## Results

Between 2000 and 2007, 105 patients were referred our department for surgical management of mediastinal masses. 34 of them were mediastinal cysts and comprising 33% of total mediastinal tumors. There were 18 females (53%) and 16 males (47%), ranging age 22 to 74 years. The mean age was  $45.33 \pm 14.19$  years. The mean age of patients with anterior and visceral mediastinal cysts were  $51.00 \pm 14.08$  and  $44.63 \pm 15.20$  years.

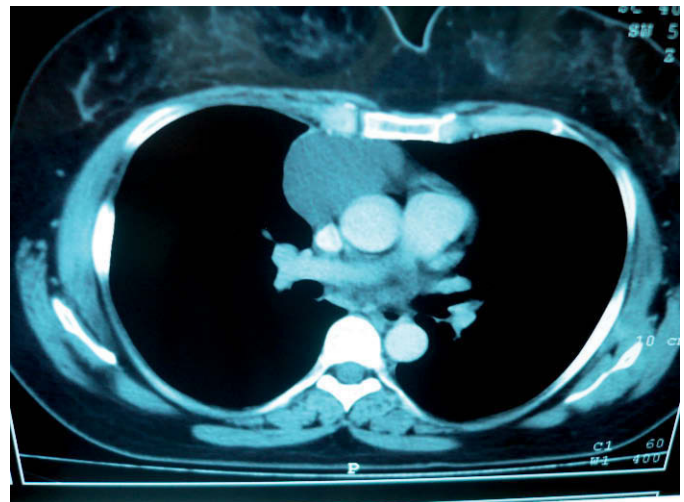
Except patients with hydatid disease (6%), majority of cysts were congenital (94%). There were 14 bronchogenic cysts (41%), 7 pericardial cysts (21%), 4 thymic cysts (12%), 3 teratomas (9%), 3 pleural cysts (9%), 2 hydatid cysts (6%), 1 simple thyroid cyst (3%).

Overall, 21 patients (61%) with mediastinal cysts were symptomatic. Asymptomatic patients (39%) were found incidentally on chest radiography. Chest pain and discomfort were most common symptoms (41%). The other presenting symptoms were dyspnea in 3, cough in one, hemoptysis in one patient respectively. Other rare mode of presentation was fatigue in a patient and in another one, interestingly headache that had no unreasonable explanation. The size of the cysts those were located at anterior mediastinum was  $7.2 \pm 2.17$  cm and 88% of them were symptomatic. These were  $6.76 \pm 4.5$  cm and 54% in cysts those were located at visceral mediastinum. There was no correlation between size and symptoms ( $P = 0.380$ ). The clinical characteristics of mediastinal cysts are shown in Table 1.

**Table 1**

General Features and Clinical Characteristics of Mediastinal Cysts

	Anterior (n = 8)	Visceral (n = 26)
Age	51.00 ± 14.08	44.63 ± 15.20
Gender M/F	3/5	13/13
Size	7.200 ± 2.167 (range 5-10 cm.)	6.760 ± 4.52 (range 2-25 cm)
Symptoms	7 (88%)	14 (54%)
Lesions		
Thymic	4	
Teratoma	3	
Simple thyroid cyst	1	
Bronchogenic	14	
Pericardial	7	
Pleural	3	
Hydatid	2	
Surgical approach		
Sternotomy	6	
Thoracotomy	2	25
Mediastinoscopy		1



**Figure 1.** A pericardial cyst of right side, originated in visceral mediastinum and extended into anterior compartment of mediastinum.

## Localization

Regardless of pathology, 76% of cysts ( $n = 26$ ) were detected in visceral mediastinum. Others ( $n = 8$ ) were located in anterior compartment. There was no cyst in posterior mediastinum. Clinical manifestations were more prominent in anterior mediastinal cysts which 88% of them had symptoms. Cysts in visceral compartment were quite silent, only 55% of them were symptomatic. The localization of mediastinal cysts are shown in Table 1.

Fourteen (48%) of 29 bronchogenic cysts resected surgically during the trial period was located at mediastinum. Pericardial cysts were 50% of cysts those were mesothelial originated mediastinal cysts. 57% of them were asymptomatic. We detected 4 (57%) on the right and 3 (43%) on the left side (Figure 1). General characteristics of bronchogenic and pericardial cysts were shown in Table 2.

Rarely seen cysts, thymic, pleural, hydatid cysts, teratoma and thyroid were diagnosed in 13 (38%). In our series, thymic cysts ( $n = 4$ ) represent 13% of all and 29% of mesothelial cysts and 50% of all anterior mediastinal cysts (Figure 2). General features of less common cysts were shown in Table 3.

**Table 2**  
General Features of Bronchogenic and Pericardial Cysts\*

	Bronchogenic Cysts (n = 14) (n = 7)	Pericardial Cysts
Age	41.357 ± 14.736	44.85 ± 15.826
Range age	25-73 22-67	Sex M/F 6/8 4/3
Size	6.15 ± 2.34 cm (2-12 cm)	8.142 ± 5.814 cm (4-20 cm)
Symptomatic, n	7	2
Asymptomatic, n	7	5

\*Numbers were available for statistical calculations.



**Figure 2.** Unilocular thymic cyst caused widening of anterior mediastinum.

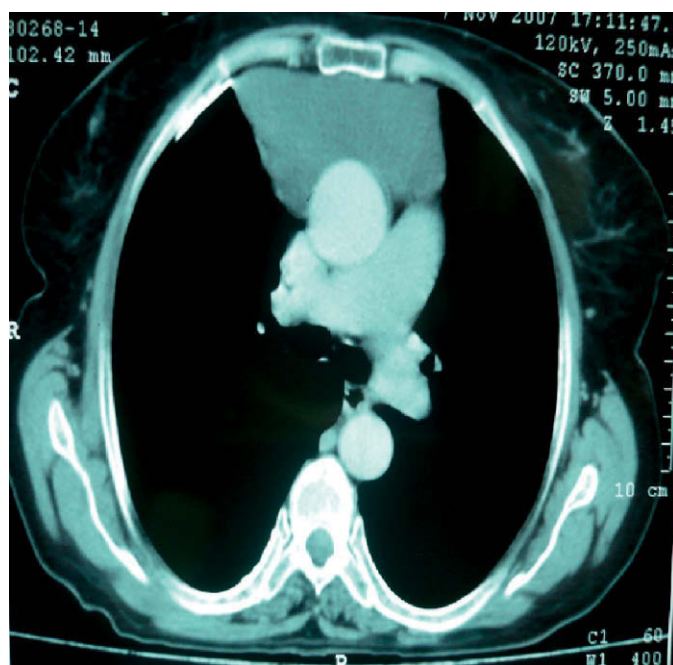
**Table 3**  
General Features of Less Common Cysts

	Thymic	Pleural	Teratoma	Hydatid	Thyroid
Age, range	45-67	42-70	30-43	26-54	48
Sex F/M	3/1	2/1	2/1	2/0	0/1
Size (range), cm	5-6	2-25	8	5-6	9
Symptomatic, n	3	1	3	2	1
Asymptomatic, n	1	2	-	-	-

In visceral compartment both sexes were equal in number (13 female and 13 male), but female predominancy was in anterior compartment. However, there was no relation between gender and localization ( $P = 0.276$ ).

#### Surgical treatment and outcome

Surgical procedure was chosen with regard to the tumor size, location and extension into surrounding structures. Surgical indications were to achieve exact diagnosis, to remove pressure to the vital mediastinal organs and to avoid possible complications like malign transformation. Excisions were performed via 6 median sternotomy, 27 thoracotomy and one mediastinoscopy. Video-assisted thoracoscopic surgery was not performed. There was no major morbidity or mortality after surgical removal of mediastinal cysts. All patients had uneventful recovery. All the cysts were intact. Except a multilocular thymic cyst, all the cysts were unilocular and had congenital structure with thin cystic wall and serous fluid (Figure 3). The follow-up period was  $23.1 \pm 15.4$  (9-70) months. There was no recurrence for any particular cyst.



**Figure 3.** A congenital thymic cyst with thin cystic wall and contains serous fluid.

#### Discussion

Mediastinal cystic structures accounts for 31% of all mediastinal masses in this serial. Other series reported different rates<sup>3</sup>. Kirwan et al<sup>4</sup> reported congenital mediastinal cysts representing approximately 10% of all mediastinal masses in adult population. However, it is difficult to calculate the exact prevalence of the cysts, since some aged patients have lesions that remain forever silent. The overall frequency of cysts in our series was in the limits of the literature<sup>5</sup>. The higher ratio of cysts may be related to the exclusion of cystic lesions with malignant degeneration and esophageal cysts, in our review.

Majority of lesions in this serial were found in middle mediastinum (76%), because of diversity of tissues other than anterior or posterior mediastinum. The current literature also supported that finding<sup>6</sup>. The remainder of them was in anterior compartment (24%) and no cyst was present in posterior compartment.

Symptoms are nonspecific and mostly due to compression to the adjacent structures. Dysphagia, weight loss, anorexia and non productive cough are suggestive of underlying malignancy<sup>2</sup>. In this serial, 61% of patients with mediastinal cysts were symptomatic. Chest pain was the most common presenting complaint regardless of cystic pathology. This may be result of irritation or inflammation of pleura.

Whatever the etiology is, these masses have similar imaging appearances. Extensive degeneration of solid masses may lead cyst formation that radiologically indistinguishable from true cysts<sup>7</sup>. Conventional chest radiograms are primary step in evaluation and usually demonstrate them as a sharply marginated, round or oval area of increased opacity<sup>7,8</sup>. Magnetic resonance imaging tends to be the current imaging modality of choice for establishing diagnosis and determine cyst location<sup>9</sup>. Magnetic resonance imaging was not performed routinely in this serial, because conventional methods and computed tomography helped us to define cystic characteristics and to plan the surgical procedure. Magnetic resonance imaging is helpful especially in differential diagnosis of other mediastinal masses and in assessment of the chance of complete resection in some cases.



The morbidity of cysts regardless of nature is usually result of an enlarging lesion on adjacent structures such as the heart, great vessels or tracheobronchial tree. And also infection of cyst, intracystic hemorrhage, spontaneous rupture, airway obstruction hemodynamic compromise including cardiac tamponade and recurrence after resection are other reported morbidities<sup>10-13</sup>. Malign degeneration of these cysts were rarely reported<sup>2,5,14</sup>. In this serial, there was no complication related with morbidity and mortality, due to the absence of complicated cysts in any patients.

Mediastinal cysts may be formed by various clinical entities; bronchogenic, mesothelial, hydatid cyst in endemic areas. Bronchogenic cysts constitute 5-10% of all mediastinal tumors and 50-60% of all mediastinal cystic pathologies<sup>15-18</sup>. They can be found at any location of body<sup>19,20</sup>. We only included mediastinally located cysts constituted 48% of all bronchogenic cysts (total of 29 bronchogenic cysts). In the series of Takeda et al<sup>2</sup>, 48% of cystic mediastinal lesions were bronchogenic cysts. Bronchogenic cysts are sometimes found in association with other congenital pulmonary malformations such as sequestration and lobar emphysema<sup>21</sup>, but we did not meet such a relation.

Mesothelial cysts include pericardial, pleural and thymic cysts<sup>3</sup>. Thymic cysts are found in anterior mediastinum, where pericardial and pleural ones are usually present in visceral compartment. We detected that 41% of all cysts were mesothelial in origine. Unlike literature findings, most of mesothelial cysts were found in visceral mediastinum<sup>6</sup>. Pericardial pathology was most common etiology in mesothelial cystic masses (50%), thymic (29%) and pleural (21%) cystic masses are others in our serial, respectively. Most of pericardial cysts arise in anterior cardiophrenic angle and are frequently on the right side and pedunculated<sup>1,3</sup>. We found 4 (57%) on the right and 3 (43%) on the left side. In the reported series dyspnea and chest wall discomfort are most frequent symptoms, but patients are usually asymptomatic<sup>1,7,22,23</sup>. Different from the findings in literature, 57% of our patients were symptomatic.

Pleural cysts have common pathologic, radiologic and embryologic features with other mesothelial cysts. Some authors advocate watchful waiting for small cysts and asymptomatic cases<sup>24</sup>. We believe that surgical removal and histopathological confirmation is reasonable. Thymic cysts constitute majority of mesothelial cystic lesions of mediastinum and can be found at any anatomic level between neck and diaphragm<sup>6</sup>. They are one of largest group after bronchogenic cysts in most of series. Takeda et al<sup>2</sup> reported thymic cysts were second large group of cyst with 28.6% of the mediastinal cysts. Uniloculated cysts are usually congenital with thin cystic wall and contain serous fluid. Multiloculated cysts with thick wall contain gelatinous fluid and though to be acquired in origin<sup>8,23</sup>. There is debate in treatment of unilocular thymic cysts. Some believe all cysts should be removed for definite diagnosis and to prevent future complications<sup>24</sup>. However, multilocular cysts must be excised because of risk of malign degeneration<sup>1,25</sup>. In our series, primary thymic cysts represent 13% of all and 29% of mesothelial cysts and 50% of all anterior mediastinal cysts. There were 3 cases of teratoma that were located in anterior mediastinum. Clinical and radiological findings were similar to other cystic masses of mediastinum.

Primary mediastinal cysts constitute 0.1% of all hydatid cystic disease. Hydatid disease of true mediastinal involvement is very rare but we encountered 2 cases. Both of them were intact, therefore symptoms were referable to their mass effect. Simple excision was performed for both of them.

There is considerable debate on preferred therapeutic approach on mediastinal cysts. Some authors support waiting or conservative approach via computed tomography guided transbronchial, transeophageal or percutaneous needle aspiration<sup>26</sup> on asymptomatic cases, but both are controversial and the latter has recurrence risk. Some investigators advocate application of sclerosing

agents following aspiration to degenerate epithelial debris. But results are not always satisfactory<sup>11,26,27</sup>. The traditional approach to mediastinal cysts, especially with atypical radiologic features or being symptomatic, is surgical excision for definitive diagnosis and treatment<sup>26,27</sup>.

Short and long term follow up results after surgical intervention of mediastinal cysts are satisfactory. Surgical intervention should be cases depended. Less invasive methods, such as video-assisted thoracic surgery (VATS) are popularized recently. Pericystic dense adhesions, communication of the cysts with tracheobronchial or esophageal structures, difficulties in control of bleeding, failure to eradicate all of cystic wall remnants are some of known difficulties during VATS<sup>21</sup>. And moreover, the size, location depth and relation with major vascular, tracheobronchial or esophageal structures of cysts inevitably may result in conversion to thoracotomy. While difficulties in removal and possible complications and recurrence are similar to those of classical posterolateral thoracotomy, VATS offers clear postoperative advantages<sup>28</sup>. There is also encouraging results for thoracoscopic excision of mediastinal bronchogenic cysts in selected patients<sup>29</sup>. We preferred thoracotomy and sternotomy for almost all cases. With its postoperative advantages, VATS may be preferred in suitable patients and at experienced departments. Rules of complete and safe resection should be followed in operative procedures and choice of procedure should be done with them. Asymptomatic patients of mediastinal cysts are not rare. Surgery is most reliable method for definite diagnosis and treatment with acceptable mortality and morbidity.

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