Mediastinoscopy in the Treatment of Mediastinal Cysts

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ABSTRACT

Objective: Primary cysts constitute 25% of all masses in the mediastinum. Because radiological investigations are often inconclusive, many adults require mediastinoscopy, thoracotomy, video-assisted thoracic surgery, or computed tomography-guided transbronchial, transesophageal, or transcutaneous aspiration to confirm the cystic nature of these lesions. Minimally invasive procedures fail when the cyst contents are gelatinous and mucoid (failure to aspirate) or when the cyst wall continues to secrete fluid. Though Pursel reported mediastinoscopic extirpation of benign cysts 35 years ago, it remains a "therapeutic curiosity" with sporadic reports of its usage. We report 2 successful mediastinal cyst extirpations performed as outpatient procedures and review the literature with regards to its management.

Methods: A rigid, 8-mm mediastinoscope was inserted into the anterior mediastinum following the creation of a 2-cm suprasternal incision and dissection along the anterior surface of the trachea. After aspiration, cytology of the contents revealed their benign nature. Right paratracheal cysts in 2 adult males were successfully removed mediastinoscopically by blunt and sharp dissection.

Results: Histopathology revealed benign mesothelial cysts in both instances. Both patients had an uncomplicated procedure and were discharged within 23 hours. No other pathology was detected on mediastinoscopy, and follow-up at 3 months and 6 months has revealed no recurrence.

Conclusion: Mediastinoscopic cyst removal is a minimally invasive procedure with a very low morbidity and mortality rate. Morbidity, recovery, and discharge times are much less than those of more invasive procedures (video-assisted thoracic surgery / thoracotomy). We suggest that it should be the first-choice procedure for the excision of appropriately located benign mediastinal cysts.

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INTRODUCTION

The mediastinum is the extrapleural space within the thorax, lying between the pleural cavities. It is a mobile partition that extends superiorly to the thoracic inlet and the root of the neck and inferiorly to the diaphragm. Anteriorly, it extends to the sternum and posteriorly to the thoracic vertebrae. An imaginary plane from the sternal angle to the lower border of the fourth thoracic vertebra divides it into superior and inferior portions. The inferior mediastinum is subdivided into anterior mediastinum (between the pericardium and sternum), middle mediastinum (contains the heart and pericardium) and posterior mediastinum (between pericardium and vertebral column).[1] Important structures in the mediastinum include the thymus, trachea, heart within the pericardium, esophagus, large arteries and veins, thoracic duct, sympathetic trunk, and the phrenic nerves. Primary cysts constitute 25% of all masses in the mediastinum.[2] Mediastinal cysts can be differentiated into foregut cysts (bronchogenic, esophageal, gastroenteric, and nonspecific), pericardial cysts, thymic cysts, dermoid cysts (teratomatous cysts), and other rare variants (cystic mediastinal tuberculosis and cystic schwannoma).[3]

ETIOLOGY, PATHOLOGY, AND CLINICAL FEATURES OF MEDIASTINAL CYSTS

Foregut cysts are the most common mediastinal cysts with an incidence of nearly 50% of all cysts encountered. The gastroenteric type arises from adherence of the primitive endodermal tube to the notochord, with a traction diverticulum or a duplication cyst developing due to differential growth of the vertebral column (Neurenteric theory of Fallon). They are often associated with vertebral abnormalities like spina bifida or split vertebrae.[4–6] Esophageal cysts are characterized by a double layer of smooth muscle in their wall and arise from sequestration of isolated vacuoles, the normal fusion of which leads to canalization of the solid embryonic foregut tube.[7] At approximately the fourth week of embryonic life, the respiratory diverticulum appears as an outgrowth from the ventral wall of the

foregut. Two longitudinal ridges develop as the diverticulum grows caudally separating it from the foregut. The bronchial buds develop as 2 lateral outpocketings from this diverticulum. Bronchogenic cysts arise from abnormal budding of the bronchial tree[8,9] and are characterized by their ciliated epithelial lining with focal areas of hyaline cartilage, smooth muscle, and bronchial glands in their wall.[10–14] Maier[15] classified bronchogenic cysts based on location into paratracheal, carinal, hilar, paraesophageal, and miscellaneous and found that they were most common at the hilar location. Recent studies however have found them most commonly in the paratracheal or subcarinal regions.[16–18]

Foregut cysts occur equally in male and female subjects.[17] Cysts may be discovered incidentally in asymptomatic patients; however, a majority eventually cause aerodigestive compressive symptoms by enlargement secondary to bleeding, infection, or retained epithelial secretions. Airway obstruction and pneumonia are common presenting features in infants and children.[19,20] Rupture of a bronchogenic cyst into the bronchus,[21] pericardium,[22] and pleura has been reported. Neurologic symptoms may occur in patients who have intraspinal extension of neurenteric cysts.[23] Heterotopic, gastric, or pancreatic rests in esophageal duplication cysts may cause ulceration, hemorrhage, or rupture due to their secretions. A few reports of malignant transformation in foregut cysts[24–26] and bronchogenic cysts[27–30] exist. Due to the high incidence of complications, operative removal is the standard treatment for foregut cysts.[22]

Percardial cysts, also known as "spring water cysts," are thought to arise from persistent ventral parietal recesses of the pericardium and rarely communicate with the pericardial cavity.[31,32] They are uncommon asymptomatic lesions detected in the fourth or fifth decades of life. They are most commonly located in the right (70%) or left (22%) cardiophrenic angles.[33] They are unilocular masses composed of fibrous connective tissue and lined by a single layer of mesothelial cells. A majority of these cysts need only radiological and clinical follow-up with surgical resection being performed to exclude a foregut cyst or a cystic neoplasm.[34]

Thymic cysts are classified as unilocular or multilocular. While unilocular cysts are considered congenital (arising from the remnants of the thymopharyngeal ducts),[35] multilocular cysts are considered to form as a reaction to inflammation[36] and are reported in patients with HIV, congenital syphilis (Dubois' abscess), and in congenital rubella syndrome. Subscribing to this hypothesis are pathology reports that have shown thin-walled unilocular

cysts and pericystic fibrosis with inflammatory changes in the walls of multilocular cysts. Unilocular cysts cause symptoms when they enlarge, and surgical excision is recommended. Multilocular cysts can rarely transform into malignant lesions,[37] but surgery is usually needed due to compressive symptoms from increasing cyst size.

Dermoid cysts are benign teratomas. Malignant teratomas are less common and are usually solid with a cystic component. Components from all 3 germ layers are encountered in a dermoid cyst while malignant lesions exhibit fewer well-differentiated structures. The mediastinum is the second most common site after the ovary or testis as the primary location for a teratoma. Common symptoms include chest pain, productive cough, and hemoptysis. Complications include cystobronchial and cystocutaneous fistulae, compressive symptoms, lipoid granulomatosis, or pneumonia from discharge of cystic contents into the bronchial tree. Malignant transformation of benign teratomas has been reported.[38] Malignant teratomas often metastasize to bone. Ideal treatment for mediastinal teratomas is extirpation using a posterolateral thoracotomy approach.[39]

Miscellaneous cystic lesions encountered in the mediastinum include undifferentiated cysts, cystic schwannomas, cystic mediastinal tuberculous lymphadenitis, and lateral thoracic meningoceles.[3] Lateral thoracic meningoceles are the most common cause of posterior mediastinal masses in patients with neurofibromatosis.[31] They are usually followed up radiologically and clinically with surgical excision being indicated only if symptoms develop.[40] Undifferentiated cysts share in common the location and clinical features of a bronchogenic cyst and are unilocular. Indications for excision include compressive symptoms.

EVALUATION

Mediastinal cysts present as masses on plain radiographs performed for symptoms described above. In asymptomatic individuals, they are often detected incidentally on routine chest radiographs or barium swallows.[41] Air fluid levels on plain radiographs indicate communication of the cyst with a bronchus. A computed tomographic (CT) scan is however the most useful noninvasive investigation to confirm the cystic nature of a mediastinal lesion. A presumptive diagnosis of benign mediastinal cyst is based on the following CT findings: 1) a smooth, oval, or tubular mass with well-defined margins and no evidence of a thick and irregular wall; 2) homogenous CT attenuation usually but not invariably in the range of water density (0H to 20H); 3) no vascular enhancement; 4) no infiltration of adjacent mediastinal structures; and 5) character-

istic location in the paratracheal, carinal, subcarinal, or paraesophageal region.[42]

Because radiological investigations are often inconclusive, many adults require mediastinoscopy, thoracotomy, video-assisted thoracic surgery (VATS), or CT-guided transbronchial, transesophageal, or transcutaneous aspiration to confirm the cystic nature of these lesions. Although the latter procedures are minimally invasive, their long-term efficacy is debatable because the cyst wall is not removed and is likely to continue to secrete fluid, resulting in recurrence unless sclerosants can be injected into the cyst.[43] Further high-density mediastinal cysts, with high computed tomography coefficients (120H), containing gelatinous and highly mucoid contents cannot be aspirated successfully.[42] Pursel et al[44] nearly 35 years ago reported 2 cases of successful mediastinoscopic extirpation of benign mediastinal cysts. They offered this option as a "therapeutic curiosity" in subjects at poor risk for thoracotomy or in cases incidentally detected during mediastinoscopy. Since that time, sporadic reports have been made of successful mediastinoscopic removal of benign cysts as day surgery procedures.[45,46] Davis and colleagues[1] noted that nearly 65% of all mediastinal cysts presented in the middle or anterosuperior mediastinum, areas readily accessible to the mediastinoscope. However, many recent series continue to use this technique sparingly, preferring to perform thoracoscopic or surgical (thoracotomy) removal.[12,47,48] These are associated with prolonged hospital stay, (2.1 days average length of stay in Hazelrigg et al's series[48] after thoracoscopy; 12.4±12 days after muscle sparing or conventional thoracotomy incisions), and increased morbidity.[49] Ashbaugh[50] in his review of 9543 mediastinoscopies reported a morbidity of 1.5% and a mortality of 0.09%. We report 2 successful medastinoscopic cyst extirpations with follow-ups at 3 months and one year and recommend a more aggressive use of this procedure in the removal of anterosuperior and middle mediastinal benign cysts.

CASE REPORT 1

A 54-year-old man was noted to have a mediastinal mass revealed on a chest X-ray performed for an annual medical examination. A CT scan **(Figure 1)** showed a large right paratracheal mass (3x2x3 cm). The patient did not have a history of fever, chills, night sweats, weight loss, cough, or hemoptysis. His past medical history was significant for a stroke with some difficulty in communication thereof. He was a smoker (35 packs/year) and a retired army veteran. On physical examination, no significant
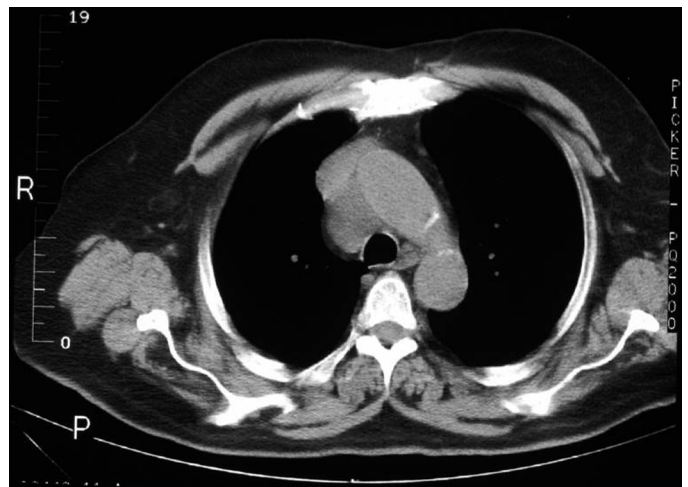

Figure 1. Preoperative computed tomographic scan shows right paratracheal mediastinal cyst in patient 1.

lymphadenopathy was found, and the lung fields were clear to auscultation bilaterally. Bronchoscopy and Wang needle aspiration of the mass was attempted initially with no success in establishing a diagnosis, and hence the patient was referred for surgical evaluation. An 8-mm rigid mediastinoscope was inserted into the anterior mediastinum following the creation of a 2-cm suprasternal skin crease incision and dissection along the anterior surface of the trachea. A large, soft ballotable mass was noted in the right paratracheal region, and aspiration with a 19-gauge needle produced 50 mL of clear fluid. The cyst was entered with the mediastinoscope, and the smooth nature of its wall was noted. The cyst wall was removed through the mediastinoscope by using blunt and sharp dissection. No other mediastinal abnormality was noted. The patient had an uncomplicated 4-hour stay in the hospital. The pathology report described the lesion as a benign mediastinal cyst. No recurrence has been noted at the third month follow-up. A CT scan performed at 1 year **(Figure 2)** showed no evidence of recurrence.

CASE REPORT 2

A 73-year-old man had been diagnosed with a mediastinal mass on routine chest x-rays performed at his diabetes clinic. On serial x-rays performed over a 3-year period, an increase was noted in the size of this lesion, and he was referred to the thoracic surgery clinic to exclude a neoplastic process. A CT scan **(Figure 3)** demonstrated a right paratracheal lesion. His past medical history was significant for a Greenfield filter placement for deep vein thrombosis with pulmonary emboli. He had significant ischemic

Figure 2. Follow-up computed tomographic scan at 12 months shows no recurrence.

Figure 4. Follow-up computed tomographic scan shows no evidence of recurrence.

Figure 3. Preoperative computed tomographic scan shows large right paratracheal cyst.

heart disease and had undergone PTCA 8 years earlier. He was a reformed smoker with a 30-pack/year history. On physical examination, no significant cervical or axillary adenopathy was found, and he had bilateral, normal vesicular breath sounds on auscultation. On mediastinoscopy, a cystic lesion was found and aspiration revealed 45 mL of clear fluid, cytology revealing a few small lymphocytes but no malignant cells. Subtotal excision of the cyst was performed by using blunt and sharp dissection. Histopathology showed a benign mesothelial cyst with a simple cuboidal epithelial lining. No other pathology was identified on mediastinoscopy. No recurrence was noted in 3 months of follow-up **(Figure 4)**.

DISCUSSION

Mediastinoscopic cyst removal is a minimally invasive method of cyst extirpation. Mediastinoscopy can be performed with a very low morbidity and mortality rate (less than 0.5% in several large series). Standard cervical mediastinoscopy (SCM) as performed by us allows access to the right and left paratracheal areas, the hilar areas, and the carina. These are the most common sites of mediastinal cysts. Malignant mediastinal cysts are extremely rare. Clinical features of infiltration and compression of surrounding structures and the radiological findings alluded to earlier should avoid the potential for a piecemeal extirpation of malignant cysts. Extended cervical mediastinoscopy (ECM) carried out in a prevascular plane in contrast to standard cervical mediastinoscopy (SCM), which is carried out in a pretracheal plane, carries with it a higher risk of recurrent laryngeal nerve and vascular injury. In more than 100 cases of mediastinoscopy performed by the senior author (JMA), we encountered only one vascular injury to the azygos vein. In patients with lesions in the aorto-pulmonary (AP) window, we perform an anterior mediastinotomy, as described by Chamberlain,[51] rather than ECM. This allows access to the pleural cavity and allows visualization of the lung at the same time. Cysts in this location are extremely rare. Bronchogenic cysts have been dealt with successfully by mediastinoscopic resections, other foregut cysts including esophageal duplication cysts, more common in the posterior mediastinum, are best dealt with by VATS. A review of recent English literature in the management of mediastinal cysts is shown in **Table 1**. Though sporadic reports occur in the literature of successful mediastinoscopic cyst removal, dating back

Table 1.
Management of Mediastinal Cysts: Majority Still Managed by Video-assisted Thoracic Surgery or Thoracotomy

Series	Year	Technique	N	Location in Mediastinum	Complication	Recurrence
Urschel[52]	1994	Mediastinoscopy	3	Middle	None	none
St. George[12]	1991	Open 65 Mediastinoscopy 1	66	Middle 23 Posterior 43	Vagal injury, Esophageal laceration, Bronchial laceration	
Ribet[47]	1995	Open 68 Mediastinoscopy 1	69	Middle 50 Anterior 19	1 thoracoscopy converted to open	none
Roviaro[53]	2000	VATS	9	Middle 5 Anterior 4	Bleeding	none
Hazelrigg[48]	1993	VATS	9	Anterior 1 Middle 1 Posterior 7	None	none
Zambudino[54]	2002	4 VATS 16 Open	20	Anterior 17 Middle 3	Phrenic nerve injury, hemothorax	none
Demmy[55]	1998	VATS	14	Middle 7 Posterior 7	Hemorrhage (converted to open)	1
Smythe[46]	1998	Mediastinoscopy	3	Anterior	None	None
Cohen[56]	1991	Open	45	Middle and Posterior	NA	NA
Cirino[57]	2000	VATS	11	Middle and Posterior	Air leak, Phrenic nerve injury, stellate ganglion injury, diaphragm injury	None
Martinod[58]	2000	VATS	20	Middle and Posterior	Bleeding Convert to open in 7	NA
Takeda[59]	2002	19 VATS 81 Open	105	All compartments	3/19 VATS converted to open	NA
Davis[1]	1987	NA	101	Anterior 7 Middle 60 Posterior 34	NA	NA

to the 1950s, it remains a second-choice procedure to VATS/thoracotomy. Morbidity, recovery times, and discharge times are all prolonged with more invasive procedures vis-à-vis mediastinoscopy. Pursel et al,[44] Ginsberg et al,[45] and Smythe et al[46] in different eras have performed mediastinoscopic cyst excision with success. It however remains an underperformed procedure for what is a fairly common condition (20% of all mediastinal masses). Because most mediastinal cysts occur in the anterosuperior/middle mediastinum, areas easily accessible to the mediastinoscope, we advocate that mediastinoscopy be attempted for excision of cysts at these locations with

more invasive procedures (VATS/thoracotomy) being used for failed mediastinoscopic removal; cysts not accessible to the mediastinoscope; or in situations where mediastinoscopy would be contraindicated (previous mediastinoscopic procedures or suspected malignant cysts with adhesion to surrounding structures). We agree with Smythe and colleagues[46] that total excision of the cyst wall might not be accomplished, but removal of greater than 90% of the wall will enable the surrounding tissues to absorb the small amount of fluid secreted by the "marsupialized" cyst wall remnant. It behooves the operating surgeon to perform cytology on the aspirated fluid to

exclude occult malignancy before attempting excision. Further, careful follow-up would be indicated to detect and treat a recurrence. All studies to date have not shown any recurrence in follow-ups over periods ranging from 3 months to 2 years.

Transcutaneous, transesophageal, and transbronchial aspiration have been attempted in the treatment of benign mediastinal cysts with varied success. While Kuhlman et al[42] treated 5 patients successfully with transbronchial/transesophageal aspiration, Van Beers et al[43] noted a symptomatic recurrence of a transbronchially aspirated cyst. If future reports from larger series confirm good results with low recurrence rates, one might adopt this technique to initially treat benign cysts, with mediastinoscopy being used to treat recurrences or failed aspirations due to viscid fluid collections. Until such time, mediastinoscopy should remain the first-choice procedure in the treatment of benign cysts.

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