Usual and unusual locations of intrathoracic mesothelial cysts. Is endoscopic resection always possible?

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Received 7 April 2003; received in revised form 5 August 2003; accepted 6 August 2003

Abstract

Objective: Mesothelial intrathoracic cysts are congenital lesions classically located in the anterior cardiophrenic angle (pleuro-pericardial cysts). Locations elsewhere in the thorax are infrequent. The aim of the study was to describe a 10-year, single-institution experience with endoscopic management of mesothelial cysts by video-assisted thoracoscopy (VT) or video-assisted mediastinoscopy (VM), regardless of their location.

Methods: From January 1992 to December 2002, 13 patients (four males and nine females, mean age 49.9 years, range 22–75) underwent surgery for a mesothelial cyst. Information on past history, clinical and radiological presentation, indications for surgery, the surgical procedure and postoperative outcome were collected retrospectively and inserted in a dedicated database. A follow-up visit was performed on December 2002 in all of the patients.

Results: In five patients the cyst was in the right cardio-phrenic angle, in three cases it was in the left cardiophrenic angle. Five cysts were located in the mediastinum (right paratracheal space in two cases, anterior mediastinum in one case, paravertebral mediastinum in two cases). Mean lesion diameter was 7.5 cm (±4) × 5 cm (±2). Cyst density ranged between 1 and 10 Hounsfield units (HU) in 11 patients. It was respectively 38 and 52 UH in the other two patients. All patients were classed ASA 1 or 2 according to the guidelines of the American Society of Anesthesiologists (ASA). Indications for surgery included the presence of symptoms, uncertain diagnosis, practice of a particular sport or professional activity, and radiological evidence of compression of the superior vena cava (namely for the two paratracheal lesions). Eleven patients were operated on by VT. Two patients with a right paratracheal lesions were operated on by VM. Mean operating time was 60 ± 14 min (range 45–80). No postoperative complications were recorded. The mean postoperative stay was 4.3 ± 1.2 days (5 days for VT and 2.5 days for VM). Pathology studies confirmed the diagnosis of mesothelial cyst in all cases.

Conclusions: Mesothelial cysts have a heterogeneous distribution within the thorax, and nearly 40% are located elsewhere than in the cardiophrenic angle. Endoscopic resection by VT or VM can be proposed as the treatment of choice even for mesothelial cysts in unusual locations.

Chest computed tomography (CT) scans usually permit diagnosis by demonstrating the typical aspect of a fluid-filled cyst delimited by a thin capsule, without enhancement after contrast injection. Given their benign behaviour, the indication for surgical resection is usually limited to symptomatic cysts or cases for which the diagnosis remains uncertain.

Over the past decade, the development of video-assisted thoracoscopy (VT) has simplified the surgical treatment of such cysts in their classic paracardiac location by obviating the need for thoracotomy. Cysts in less frequent locations may still be amenable to endoscopic resection, by VT or

1. Introduction

Intrathoracic mesothelial cysts are congenital lesion due to an anomaly in the development of the pericardial coelom [1]. Although classically located in the anterior cardiophrenic angle (pleuropericardial cysts), on rare occasion they can be located elsewhere, in the paravertebral region, the anterior mediastinum [2] or the paratracheal region [3].
video-assisted mediastinoscopy (VM), but the feasibility
and results of endoscopic techniques in such situations have
only been reported in the form of case-reports.

The aim of the study was to report a 10-year,
single-institution experience with video-assisted endoscopic
resection of mesothelial cysts, with routine use of VT or VM
for paratracheal lesions.

2. Patients and methods

From January 1992 to December 2002, 306 patients
underwent surgery in the Thoracic Surgery Department of
the University Hospital of Nice for a mediastinal lesion.
Of these, 13 (4.2%) were operated on for a mesothelial cyst.
These 13 patients compose the study population. All lesions,
regardless of location, were explored by VT or
mediastinoscopy (video-assisted mediastinoscopy in the
last 5 years, VM) in order to assess the feasibility of
endoscopic resection. Information on age, gender, medical
history, clinical presentation, preoperative radiological
investigations, ASA score, indication for surgery, surgical
procedure, operating time, postoperative complications,
hospital stay and pathology findings were retrospectively
collected and inserted in a dedicated database. Quantitative
variables were expressed by the mean ± standard deviation.
All 13 patients were contacted for a follow-up visit and a
chest X-ray in December 2002.

3. Results

3.1. Clinical evaluation

The study population consisted of four males and nine
females, mean age 49.9 years (range 22–75). Analysis of
their medical history revealed that three patients were
smokers, one patient has arterial hypertension, and one
patient had undergone surgery for breast cancer.

The cyst was revealed by a chest X-ray in all patients.
In seven cases, the radiographic examination had been
prompted by the presence of symptoms. The most common
symptom was chest pain (five patients). In one patient pain
was associated with clinical signs of cyst infection.
In another case, pain was localized to the point of
herniation of the cyst at the level of the right anterior
chest wall (Fig. 1).

The preoperative work-up for all 13 patients consisted in
a chest X-ray and CT scan of the thorax. Five patients also
underwent magnetic resonance imaging (MRI). In two
cases, MRI was indicated because of the paravertebral
position of the cyst and results excluded the possibility of a
meningeal origin. In the other two patients MRI was
performed because of the abnormal density of the lesion on
CT scan. In the last case, MRI was performed in the patient
whose cyst herniated through the chest wall in order to
determine the relationships of the abdominal wall,
the intercostal space and the pleural cavity. Cyst aspiration
was not performed for diagnosis in any patient.

Chest CT scan accurately defined the position and
characteristics of the cysts. In five patients the cyst was
located in the right cardio-phrenic angle; in three others it
was in the left cardiophrenic angle. Five cysts were located in
the mediastinum (right paratracheal space in two cases,
anterior mediastinum in one case, paravertebral mediastinum
in two cases). The mean diameter of the lesions was 7.5 cm
(±4) × 5 cm (±2). Cysts density was between 1 and 10
Hounsfield units (HU) in 11 patients. It was respectively 38
and 52 HU in the remaining two patients. No enhancement
was recorded after contrast injection in any patient.

The decision to resect the cyst was made owing to the
presence of symptoms (seven patients), because the
diagnosis remained uncertain (three patients), or because
there were radiological signs of compression on the superior
vena cava by a paratracheal lesion (the two patients with
such lesions were both asymptomatic, Fig. 2). In two cases

Fig. 1. Cyst in the right cardiophrenic angle herniating through the anterior
insertion of the diaphragm.

Fig. 2. Right paratracheal cyst causing compression on the origin of the
superior vena cava (arrow).
the cyst was asymptomatic but the decision was based on the practice of a potentially traumatic sport or professional activity (karate, diving). All patients were classed ASA 1 or 2 according to the guidelines of the American Society of Anesthesiologists.

3.2. Surgery

Eleven patients were operated on by VT. Under general anaesthesia and selective intubation, the patient was turned in the lateral position with a roll underneath the scapula.

For cysts of the cardiophrenic angle or of the anterior mediastinum, the surgeon stood behind the patient. The port for the camera (10 mm) was inserted in the fifth intercostal space, below the tip of the scapula (trocar no. 1). Two additional 5-mm trocars were inserted in the seventh intercostal space posteriorly (trocar no. 2) and in the eighth intercostal space anteriorly (trocar no. 3). After visualization of the phrenic nerve, the cyst was aspirated to allow traction by forceps inserted through trocar no. 3. This manoeuvre allowed dissection of pericardial adherences using endoscopic scissors and cautery inserted through trocar no. 2. In one case, isolation of the cyst required ligation of the internal mammary artery by a clip. Upon completion of dissection, the cyst was easily removed from the thorax.

The technique used for paravertebral cysts was the same, but the surgeon operated standing in front of the patient. A 28-Charriere chest drain was left in the pleural cavity in the first seven cases of this series, positioned through trocar no. 3. A 15-Charriere drain was used for the last six patients. Drains were removed when the quantity of fluid collected over the previous 24 h was $\leq 100$ cc.

The two patients with a right paratracheal lesion were operated on solely by video-assisted mediastinoscopy using a Dahan/linder mediastinoscope (model INH 002756, Karl Storz Endoskope, Germany). The inferior mobile valve of the instrument can be locked in the open position, thereby improving the exposure of the mediastinal field. The mediastinoscope was connected to a camera (model INH 002756, Karl Storz Endoskope, Germany) which allowed the assistant to follow the operation. After general anaesthesia, the patient was placed in the supine position with a roll underneath the scapulas to allow maximal cervical extension. The surgical field comprised the neck and the whole thorax, in case conversion to sternotomy proved necessary. The surgeon stood at the head of the patient and the monitor was at the patient’s feet. A 2-cm cervicotomy was made above the sternal notch, the white line was opened and the pretracheal fascia entered. After digital exploration, the video-mediastinoscope was inserted in the pretracheal space and the valve was opened. At this point, the assistant held the mediastinoscope to permit bimanual manoeuvres by the surgeon.

In these two patients, the cyst extended from the right innominate vein to the pericardium, at the level of the pulmonary artery. The cyst was left intact in the first part of the operation to facilitate lateral dissection from the trachea and the mediastinal fat. Aspiration was then necessary in order to dissect, clip and cut adherences with the pericardium. After extraction of the lesion and control of hemostasis, the cervicotomy was closed without placement of a drain.

None of the 13 patients in our series required conversion to thoracotomy or sternotomy. Mean operating time was $60 \pm 14$ min (range 45–80). In patients managed by VT, the chest drain was removed after $2.2 \pm 0.8$ days (mean fluid volume drained $160 \pm 110$ ml). No postoperative complications were recorded. The mean postoperative stay was $4.3 \pm 1.2$ days (5 days for VT and 2.5 days for VM).

In 12 cases the cyst contained clear, water-like fluid. In one case the cyst material was purulent but sterile. All of the cysts were lined with flat, sometimes cuboid, mesothelial cells.

Follow-up information was available for all patients (mean observation time 57.7 months, range 4–125). In six cases, the observation time was longer than 6 years. No recurrence was recorded on clinical or chest X-ray examinations and preoperative symptoms never recurred in any patient.

4. Discussion

Mesothelial cysts represent 5–10% of mediastinal tumours and are usually diagnosed in the fourth to fifth decade. In our series they represented 4.2% of the mediastinal masses (13/306). As their classic anatomical location is in the cardio-phrenic angle, they are also referred to as pleuro-pericardial cysts. As reported by two old series [4,5], the classical position of mesothelial cysts is in the right (51–70%) and the left cardiophrenic angles (38–22%). The incidence of unusual locations in these series were between 8 and 11%. In the review of Stoller in 1985 [6], only 35 mesothelial cysts in an unusual location were identified. The advent of CT scan has improved the diagnosis and anatomical evaluation of these lesions. This probably explains why more recent series report a higher incidence of unusual locations (25%) [7], and more dimensional information (diameter ranging from 3 to 30 cm).

The different possible locations of mesothelial cysts are related to embryological reasons. Fusion of mesenchymal coelomic lacunae gives origin to the pleural and pericardial cavity on one side and the peritoneal cavity on the other, divided by the antero-posterior development of septum transversum. Incomplete fusion of a lacuna, often at the level of the pericardial coelom, can result in formation of a mesothelial cyst. Incomplete fusion or secondary migration of an isolated element can also occur at the level of parietal pleura, mediastinal pleura, or septum transversum and this...
can explain unusual locations in the chest wall, mediastinum and diaphragm [8].

Chest scan allows diagnosis in the vast majority of cases by showing a fluid-filled, thin-walled lesion in the classic cardiophrenic position. The fluid content of the cyst can sometimes be misinterpreted when the density is greater than 30 HU [9], as occurred in two cases of this series. The absence of enhancement after contrast injection is mandatory for radiological diagnosis of a mesothelial cyst. For mediastinal lesions, differential diagnosis of bronchogenic cysts and lymphangiomas can be very difficult. For paravertebral lesions, MRI is essential to rule out a cyst of meningeal origin. Whenever such a lesion is suspected, intraoperative evaluation by the neurosurgeon is necessary to avoid injury of the meningeal sac.

Mesothelial cysts can exceptionally herniate through the chest wall. One of the patients in our series had a cyst in the right cardiophrenic angle herniating outside the chest wall at the level of the common cartilage and it was palpable at clinical examination.

More than 50% of mesothelial cysts are asymptomatic, and incidental radiologic findings. However, their benign behaviour does not exclude the possibility of complications such as infection (one case in the present series), rupture [10], or hemorrhage with compression on contiguous structures [11,12]. Two cases of partial erosion of the right ventricular wall and the superior vena cava have been reported [13,14].

Certain authors have proposed transcutanous aspiration of mesothelial cysts, but adverse events and recurrence have been reported after this procedure [15]. Surgical resection is the ideal treatment of these lesions, and is classically performed by thoracotomy [16,17]. The feasibility of VT resection has previously been reported for typical pleuropedicardial cysts, and the safety and efficacy of the procedure have been demonstrated [18–20].

Surgical resection is widely accepted for the treatment of symptomatic cysts and lesions for which the diagnosis remains uncertain. In our opinion, even asymptomatic cysts deserve surgical evaluation in two circumstances: large lesions with a potential risk of compression on contiguous structures, and patients involved in particular activities that increase the risk of rupture [21,22]. Two patients in our series were asymptomatic, but they practiced sports associated with an elevated risk of rupture (karate athlete, professional diver).

In contrast, mere follow-up of a mesothelial cyst is justified when the patient is at higher risk for surgery or when all radiological criteria for diagnosis are fulfilled. During the study period, two patients were merely followed up for these reasons.

The anatomical characteristics of these lesions (well rounded, almost avascular) make them ideal for management by a minimally invasive procedure. Video-thoracoscopic resection is uniformly considered easy and without complications for cardiophrenic and paravertebral mediastinal cysts. For paratracheal lesions, the feasibility of resection by mediastinoscopy has already been reported [3,23]. The development of video-assisted mediastinoscopy has improved the possibility of this technique by allowing bimanual dissection of the mediastinum and insertion of several 5-mm instruments [24,25].

Analysis of the results of the present series suggest two main conclusions. Mesothelial cysts have a heterogeneous distribution within the thorax, with nearly 40% occurring in an unusual location. Despite this heterogeneity, endoscopic resection can be considered the treatment of choice. Even though no cases occurred in the present series, the possible need of conversion to thoracotomy or sternotomy must always be kept in mind and this possibility must be discussed preoperatively with the patient.

References