



Mediastinal cysts : a 52-case retrospective study

Les kystes du médiastin : une étude rétrospective à propos de 52 cas

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ABSTRACT

Introduction: mediastinal cysts are rare lesions developed from mediastinal structures. They may be acquired like thoracic duct cysts or lymphangiomas or congenital like the bronchogenic cysts, enteric cysts or celomic cysts. These cysts are rare and may cause diagnostic challenges.

Aim : To assess the major characteristics of these cysts based on a single institution experience.

Methods: the authors performed a descriptive, retrospective study from January 2009 to March 2020 in a single institution. Cystic lesions taking birth from the mediastinum for which gross features, microscopic features were available were included.

Results: this study contained 52 mediastinal cysts that were completely resected and no patient presented complications after the surgical resection. The bronchogenic cysts were the most frequent and represented 57.69% of all lesions. Thymic cysts and pericardial cysts represented respectively 40.38% and 1.92% of the cases. The positive diagnosis was based on the microscopic exam. The final diagnosis was concordant with the radiologic findings in 15 cases reaching a rate of 28%.

Conclusion: the diagnosis of mediastinal cysts is based on the microscopic analysis of the cystic wall. Pericardial cysts may be suspected based on their characteristic location in the cardiophrenic angle, thymic cyst may be evoked based on their location in the thymic region and bronchogenic cysts are mainly located in the middle mediastinum. In spite of these most frequent locations, the cysts may be located in any part of the mediastinum and may be difficult to diagnose when the key diagnostic features are absent.

Key-words: bronchogenic cyst, celomic cyst, enteric cyst, mediastinal cyst

RÉSUMÉ

Introduction: les kystes du médiastin sont des lésions rares développées aux dépens des structures médiastinales. Ils peuvent être acquis comme les kystes du canal thoracique ou les lymphangiomes et les kystes congénitaux comme les kystes bronchogéniques, entériques ou coelomiques. Ces kystes sont rares et peuvent présenter des difficultés diagnostiques.

Objectif : Décrire les caractéristiques anatomo-cliniques des kystes du médiastin.

Méthodes: il s'agit d'une étude descriptive, rétrospective colligée sur une période allant de Janvier 2009 à Mars 2020 dans un hôpital universitaire.

Résultats: 52 kystes médiastinaux ont été inclus. Les kystes bronchogéniques étaient les plus fréquents représentant 57.69% des cas. Les kystes thymiques et péricardiques représentaient respectivement 40.38% et 1.92% des cas. Le diagnostic positif était basé sur l'examen anatomopathologique. Le diagnostic final était concordant avec les données radiologiques dans 15 cas soit 28% des cas.

Conclusion: le diagnostic des kystes du médiastin est basé sur l'examen anatomopathologique. Même si la localisation et l'aspect de certains kystes peuvent être évocateurs du diagnostic comme les kystes péricardiques situés au niveau de l'angle cardio-phrénique droit, ou les kystes bronchogéniques situés au niveau du médiastin moyen, leur localisation reste variable et l'examen anatomopathologique peut être délicat en l'absence d'aspect pathognomonique.

Mots clés: kystes bronchogéniques, kyste coelomique, kystes entériques, kystes médiastinaux

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INTRODUCTION

Mediastinal cysts are rare accounting for 12 to 30% of mediastinal masses. They consist in cystic lesions developed from mediastinal structures. Those cysts are subdivided into congenital cysts including bronchogenic cysts, esophageal cysts, gastroenteric cysts, celomic cysts (pericardial and mesothelial) and thymic cysts. Acquired cysts are less frequent and mainly represented by thoracic duct cysts and lymphangiomas. Congenital cysts are secondary to an abnormal branching of the tracheobronchial tree during embryonic development (bronchogenic cysts and esophageal cysts) or a celomic defect (celomic cysts) (1) mainly from embryonic origin, are benign and rare malformative lesions, gathering several varieties according to tissue origin. Diagnosis is mostly obtained thanks to tomodesitometry performance and sometimes by magnetic resonance imaging. It may be more difficult in some atypical topographies and in case of bulky MC. The most frequent, springing from division abnormality from embryonic foregut ("foregut cysts") in English literature. Mediastinal cysts are generally asymptomatic. Even, if radiologic findings may help to suspect the nature of the cysts, the positive diagnosis is based on microscopic exam. The mainstay treatment is based on the complete surgical excision of the lesions except for pericardial cysts, which require a narrow surveillance unless they become symptomatic. In that case, the complete surgical excision is also necessary. In symptomatic cysts, ultra-sound guided aspiration of the cyst content is useful in order to decrease the symptoms and to help the diagnosis, which is assessed by the analysis of the fluid and the nature of the cells. The authors' aim was to assess the major characteristics of these cysts based on a single institution experience

METHODS

The authors performed a descriptive retrospective study.
Inclusion criteria: the mediastinal cystic lesions diagnosed between January 2009 and March 2020 were included. Cystic lesions taking birth from the mediastinum for which gross features, microscopic features were available were included.
Non inclusion criteria: hematomas, hydatid cysts, cystic teratomas, cystic thymomas, cystic thymic carcinomas, cystic germ cell tumours or cystic lymphomas weren't included.
Exclusion criteria: mediastinal cystic lesions with incomplete data were excluded.
 The authors reviewed the hospital files of the patients, the clinical data, the radiological reports, surgical reported and all the histological slides.
Protection of personal data: the patient data were pseudo-anonymized and every case included was given a reference number.
Statistics: comparison of means was performed using the SPSS software 12.0.

RESULTS

Clinical characteristics: this study included 52 cases. Two cases were excluded because of the absence of clinical data. The mean age of the patients was 47.83 years, average from 3 to 83 years. Thirty men and 22 women were concerned. There was no significant difference in mean age between men and women ($p=0.52$).

Ten patients were asymptomatic and the mediastinal cyst was discovered incidentally in a chest-x-ray. The other patients presented respiratory symptoms including chest pain, thoracic pain, hemoptysis or dyspnea. The lesions were located in the anterior mediastinum in 34 cases (65%), the middle mediastinum in 8 cases (15%), the posterior mediastinum in 8 cases (15%) and the anterior and the middle mediastinum in 2 cases (4%). Clinical and radiologic features were suggestive of a bronchogenic cyst in 7 cases, thymic cyst in 9 cases, pericardial cyst in 3 cases and thymoma in 8 cases. In the other cases, no particular diagnosis was mentioned according to the clinical data and the radiologic. A complete surgical excision was performed in 52 cases and a biopsy was performed in 1 case of bronchogenic cyst. No patient presented particular complications.

Gross features: the mean size of the cysts accounted for 4.67 cm, average (0.5-11 cm). It accounted for 4.92 cm in bronchogenic cysts, 4.38 cm in thymic cysts, 3 cm in pericardial cysts. There was no significant difference in size between the different types of cysts. Gross features consisted in entire resected cysts in 45 cases. The cyst content was serous in 10 cases, mucoid in 30 cases and hemorrhagic in 5 cases. In 2 thymic cysts and 6 bronchogenic cysts, the cystic wall was fragmented because of adhesions. No suspicion of infection was mentioned.

Microscopic features: the final diagnosis of bronchogenic cyst was retained in 30 cases, thymic cyst in 21 cases and pericardial cyst in 1 case.

Bronchogenic cysts were characterized by a cystic wall lined with a respiratory epithelium in 26 cases and largely metaplastic epithelium in 1 case. The epithelium was largely denuded in 4 cases. The cystic wall contained cartilage in 15 cases, smooth muscle fibers in 18 cases and bronchial glands in 18 cases (figure 1). Fibro-inflammatory modifications were observed in 20 cases. The diagnosis was challenging in one case, because of the large ulceration of the epithelium, the absence of glands, cartilage or smooth muscle fibers. The diagnosis was assessed when focal foci lined with a respiratory epithelium were observed.

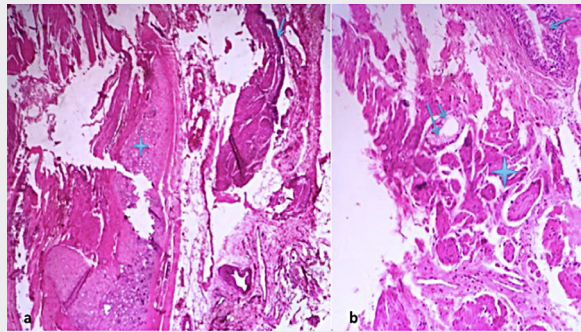


Figure 1. a/ microscopic features of bronchogenic cyst with a respiratory lining (arrow) and a cartilage (star) (HEx250), b/ respiratory epithelium lining the cyst (arrow) and presence of smooth muscle fibers (star) and bronchial glands (double arrow) within the cystic wall (HEx250).

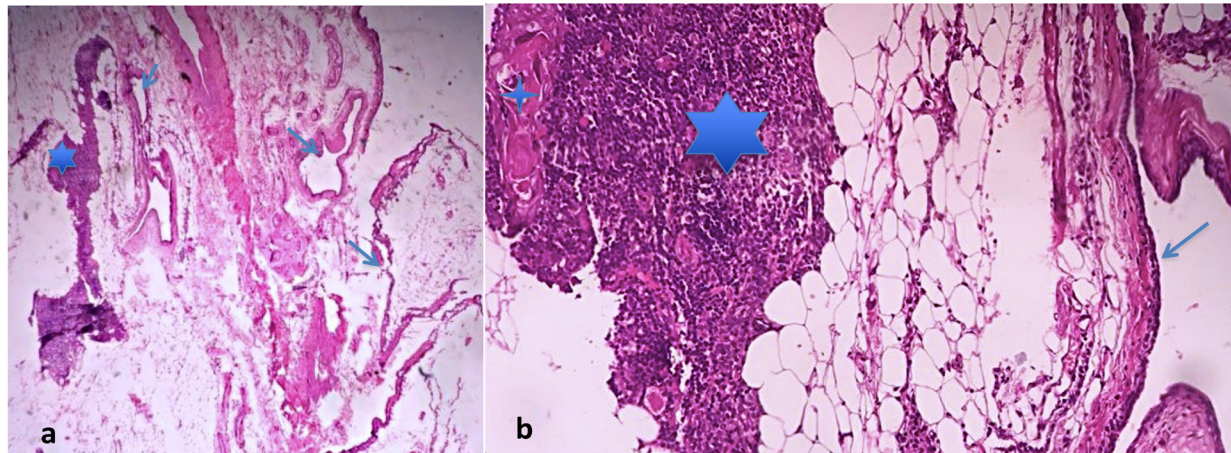


Figure 2. a/ microscopic features of a multilocular thymic cyst with multiple cavities (arrow) and remnant of thymic parenchyma (star) (HEx250), b/ mesothelial lining of the cyst (arrow) and thymic parenchyma (big star) containing Hassall's corpuscles (small star) (HEx400)

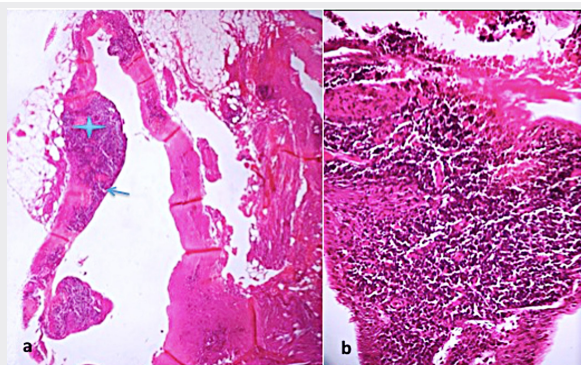


Figure 3. a/ microscopic findings of a pericardial cyst showing a largely denuded lining (arrow) and containing an abundant inflammatory infiltrate (star) (HEx250), b/ high magnification showing the dense inflammatory infiltrate (HEx400)

- The thymic cysts were mainly covered by a mesothelial lining in 8 cases, a columnar and ciliated epithelium in 10 cases and a largely denuded epithelium in 2 cases. All the cystic walls contained thymic parenchyma (figure 2). The diagnosis was challenging in 1 case because of the difficulty of highlighting remnants of thymic parenchyma.

Thymic cysts were classified as congenital in 19 cases and acquired in 1 case. One case wasn't classified because of the fragmentation of the cystic wall.

Pericardial cyst was characterized by a largely denuded epithelium and a thin fibrous wall with a dense inflammatory infiltrate (figure 3).

The diagnosis was based on the characteristic localization of the cyst in the right cardio-phrenic angle and the absence of the diagnostic features of the other mediastinal cysts.

- Correlation of radiologic and microscopic features: the clinical features and the microscopic characteristics of the different cysts are represented in table 1.

In bronchogenic cysts, the diagnosis was suspected by radiologic findings in 6 cases. The diagnosis of enteric cyst was suspected in 3 cases because of the esophageal location of the cysts. In thymic cysts, the diagnosis of pericardial cyst was suspected in 1 case based on the radiologic findings. The diagnosis of thymic cyst was suspected in 9 cases. The diagnosis of pericardial cyst was suspected by radiologists based on the characteristic localization of the cyst and the diagnosis was concordant with the microscopic features. The final diagnosis was concordant with the radiologic findings in 15 cases reaching a rate of 28%.

No patient presented complications after the surgical resection after a mean follow up period of 5 years.

Table 1: main clinical features and microscopic characteristics of the cysts.

	Mean	Standard deviation	95% confidence interval
Age	47.83	19.95	[42.27-53.38]
Age in men	49.80	20.32	[42.21-57.39]
Age in women	45.14	19.59	[36.45-53.82]
Gender			
	Number	Percentage	
Men	30	57.69	
Women	22	42.31	
Localization of cysts			
	Number	Percentage	
Anterior mediastinum	34	65.38	
Middle mediastinum	8	15.38	
Posterior mediastinum	8	15.38	
Anterior and middle mediastinum	2	3.85	
Diagnosis			
	Number	Percentage	
Bronchogenic cyst	30	57.69	
Thymic cyst	21	40.38	
Pericardial cyst	1	1.92	
Size			
	Mean	Standard deviation	95% confidence interval
Bronchogenic cyst	4.92	2.39	[4.03-5.81]
Thymic cyst	4.38	2.83	[3.09-5.67]
Pericardial cyst	3	-	-
All cysts	4.67	2.55	[3.95-5.38]

DISCUSSION

Our study contained 52 mediastinal cysts that were completely resected. The bronchogenic cysts were the most frequent and represented 57.69% of all lesions. Thymic cysts and pericardial cysts represented respectively 40.38% and 1.92% of the cases. The positive diagnosis was based on the microscopic exam. The final diagnosis was concordant with the radiologic findings in 15 cases reaching a rate of 28%. This rate was concordant with the concordant rates reported in other studies (2). The diagnosis was challenging mainly in 2 cases of

brochogenic cysts with a diffuse ulcerated epithelium. Bronchogenic cysts are the most frequent cysts reported in different studies. They belong to the foregut cysts group. Those cysts result from an abnormal migration of the foregut intestine and include bronchogenic cysts, esophageal cysts and gastroenteric cysts. Bronchogenic cysts account for 50 to 60% of the mediastinal cysts (1,3). They can be diagnosed at any age but are mainly reported in young adults and adolescents. The majority of the patients are asymptomatic or present symptoms related to the compression of the mediastinal structures. They are unilocular and rarely multilocular. The cyst wall is thin, with some smooth muscle cells and covered by a respiratory epithelium. This epithelium may be metaplastic. In this study, the most characteristic feature was the respiratory epithelium observed in 26 cases. The presence of bronchial glands and cartilage helps to assess the diagnosis. A degeneration into an adenocarcinoma has been rarely reported in the literature (2). Esophageal and gastroenteric cysts are less frequent than bronchogenic cysts. In our study, 2 cysts were located and intimately connected with the oesophagus but the microscopic exam concluded to a bronchogenic cyst. Esophageal cysts develop within the esophageal muscle layer (1,3,4). They are located into the posterior mediastinum. Their evolution is characterized by frequent complications consisting in perforation, haemorrhage or infection. They are spherical, unilocular and covered with a squamous epithelium with some foci of respiratory epithelium. The diagnosis is based on the discovery of esophageal glands circumscribed by a double layer of smooth muscle cells. Those cysts are differentiated from bronchogenic cysts by their location, the absence of cartilage and the presence of an individualized muscle layer. Gastroenteric cysts are rare and mainly observed in the posterior mediastinum with a fibrous connexion to the vertebrae. They are frequently associated to cervical and thoracic malformations. They are lined by a gastric epithelium (1,4). The latter induces ulceration and perforation. Adenocarcinomas have been reported and developed from a colonic mucosa (5).

The treatment of the foregut cysts is based on a complete surgical excision. Initially, only symptomatic cysts were resected but the indication concerned also asymptomatic cysts because of the frequency of complications reaching 25% in some studies (1) mainly from embryonic origin, are benign and rare malformative lesions, gathering several varieties according to tissue origin. Diagnosis is mostly obtained thanks to tomodensitometry performance and sometimes by magnetic resonance imaging. It may be more difficult in some atypical topographies and in case of bulky MC. The most frequent, springing from division abnormality from embryonic foregut ("foregut cysts" in English literature. According to the cysts' locations, surgical techniques of postero-lateral thoracotomy or mini-invasive surgery using video are used. In our study, only one pericardial cyst was observed. Those cysts account for 4 to

11% of mediastinal masses and 22 to 38% of mediastinal cysts (1). They can adhere to the pericardium (pericardial cyst) or develop from other parts of the mediastinum (mesothelial cyst). Patients are asymptomatic. Pericardial cysts are located into the right cardio-phrenic angle. They are unilocular with a thin wall. They are mainly lined with a unistratified epithelium of mesothelial cells. The thickness of the cystic wall and the absence of adherences induce a relative easy surgical excision. Those cysts are followed if asymptomatic and resected if symptomatic. Some authors advocate the surgical excision even in asymptomatic cysts because of the difficult narrow follow-up (1,3,6).

Thymic cysts can be congenital or acquired. They are generally asymptomatic. Uni or multilocular with a thin wall doubled by a thymic parenchyma (1,3,7). The presence of thymic parenchyma is the diagnostic key of the diagnosis. Rare squamous cell carcinoma developed in those cysts have been reported (8).

Most thymic cysts are congenital and unilocular. Acquired thymic cysts are multilocular lined with a squamous epithelium, columnar or cuboid with a thymic parenchyma. They are frequently associated to an inflammatory process, hemorrhage, necrosis, lymphoid infiltrate or granulomas. The diagnoses of thymoma or cystic teratoma or carcinoma may be evoked (1,3,9,10). In our study, we didn't find lymphangiomas or canal duct cysts. Those cysts are rare accounting for 0.7 to 4.5% of mediastinal tumors. They are generally congenital and can be observed in any territory of the cervicothoracic lymph nodes and the thoracic duct (11). They are lined by an endothelial epithelium with a thin wall (10). Other rare cysts with a non consensual pathogenesis like the mullerian cysts have been reported in the literature but haven't been described in this study (12). This study highlights a single institution experience and has the particularity to include many cases in comparison to the experiences published in the literature.

CONCLUSION

Mediastinal cysts are rare lesions whose treatment is based on surgical excision. The diagnosis is based on the microscopic analysis of the cystic wall or the cyst content when an aspiration has been performed. The diagnosis may be suspected based on the cysts' localization. Pericardial cysts may be suspected based on their characteristic location in the cardiophrenic angle and thymic cyst may be evoked based on their location in the thymic region. The most challenging cysts are also the most frequent and consist in bronchogenic cysts. These cysts may be located in any part of the mediastinum and may be difficult to diagnose when the key diagnostic features are absent.

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