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Voluminous oesophageal bronchogenic cyst treated with thoracoscopic approach

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Voluminous oesophageal bronchogenic cyst treated with thoracoscopic approach

AIM: *Bronchogenic cysts are congenital lesions deriving from the primitive foregut, and are usually located in close relation to tracheobronchial tree or oesophagus. We report a case of an oesophageal bronchogenic cyst appearing at preoperative examinations as a benign fibromuscular tumour (leiomyoma).*
CASE REPORT: *A 62 years old male patient in good general conditions, was admitted to our Institution because of moderate dysphagia and upper post – prandial abdominal pain. Conventional imaging, endoscopy and echo endoscopy detected a parietal oesophageal wall mass looking like a solid formation, determining extrinsic compression and narrowing of the lumen.*
RESULTS: *The mass has been radically removed with thoracoscopic approach. Postoperative stay was uneventful and the patient was discharged three days after the operation. At histological examination the mass appeared as a cystic formation with fibromuscular wall and ciliated epithelium (so – called disembriogenetic bronchogenic cyst).*
CONCLUSION: *The case we have reported describes a very unusual case of a voluminous symptomatic intramural oesophageal disembriogenetic cyst whose characteristics had not been defined at preoperative examinations. Surgical removal of the mass has been achieved with a minimally invasive approach.*

KEY WORDS: Bronchogenic cyst, Oesophagus, Thoracoscopy

Introduction

Bronchogenic cysts are congenital lesions deriving from the primitive foregut, and are usually located in close relation to tracheobronchial tree or oesophagus [1].

Para – oesophageal bronchogenic cysts are usually closely adherent to oesophagus with or without a well defined borderline [2]; a communication with the lumen is rarely found [3].
We report a case of a symptomatic voluminous oesophageal bronchogenic cyst appearing at preoperative examinations as a benign fibromuscular tumour (leiomyoma), successfully treated with minimally invasive approach.

Case Report

A 62 years old male patient in good general conditions, was admitted to our Institution because of moderate dysphagia and upper post – prandial abdominal pain.

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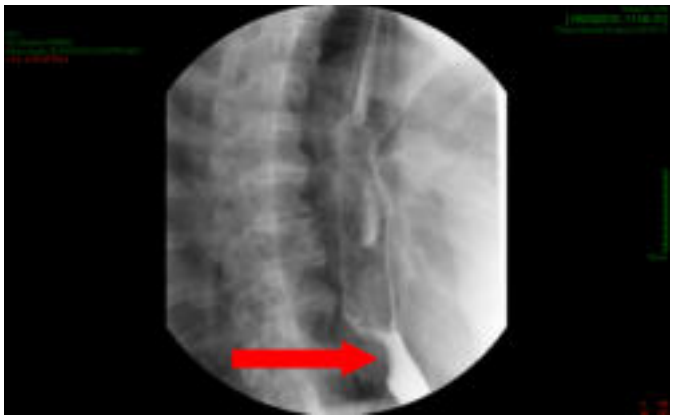

Fig. 1: Barium x ray: filling defect at lower thoracic oesophageal tract.

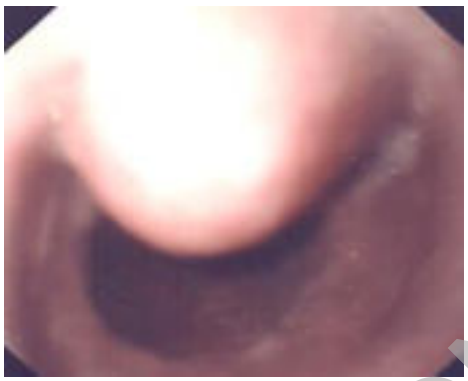

Fig. 2: Oesophagoscophy: extrinsic compression without mucosal alterations, narrowing the lumen.

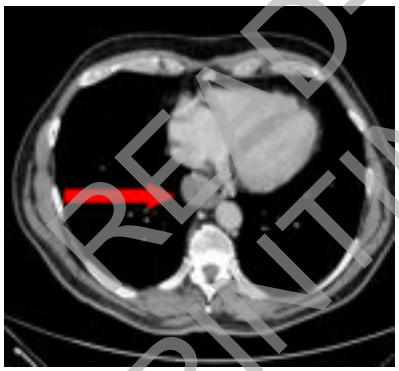

Fig. 3: CT scan: 60 X 40 millimeters oesophageal wall partially calcified solid formation.

Barium x ray showed 44 x 36 millimeters ovalar filling defect at lower thoracic oesophageal tract (Fig. 1).
Oesophagoscopy detected an extrinsic compression of the wall 33 cm from dental arch without any alteration of mucosa (Fig. 2); echoendoscopy showed the presence of an ovalar neoformation likely to originate from muscular oesophageal wall; no biopsy was performed.
At contrast enhanced CT scan, a 60 X 40 millimeters oesophageal wall partially calcified solid formation, looking like a leiomyoma, appeared at right side of lower thoracic tract, with lumen compression (Fig. 3).

Results

The patient underwent surgical radical removal of the mass with right thoracoscopic approach; the neoformation, appearing as soft in consistence, smooth surface, well delimited, bulging from the right side of oesophagus, has been isolated from muscular wall with mucosal exposure; miotomy has been sutured with self blocking absorbable continuous suture; mucosal integrity was checked with intraoperative endoscopy.
Alimentation was restarted 24 hours after the operation, thoracic drainage was removed in first postoperative day and the patient was discharged in third postoperative day.
Histological examination showed a cyst with fibromuscular wall, ciliated epithelium and very dense mucinous content, compatible with disembriogenetic bronchogenic cyst (Figs. 4, 5).

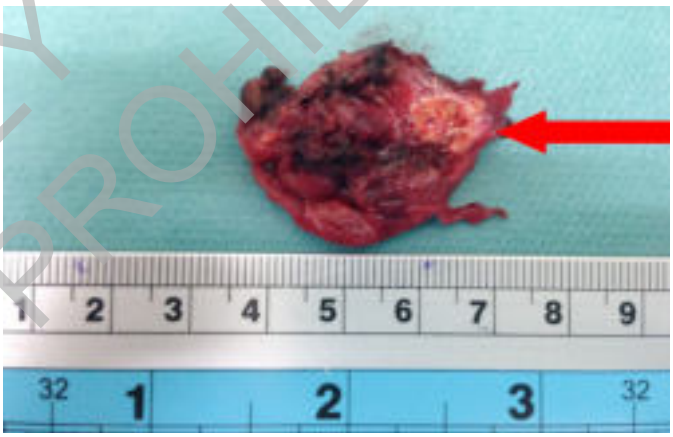

Fig. 4: Surgical specimen: cystic mass containing dense mucus and parietal calcifications (arrow).

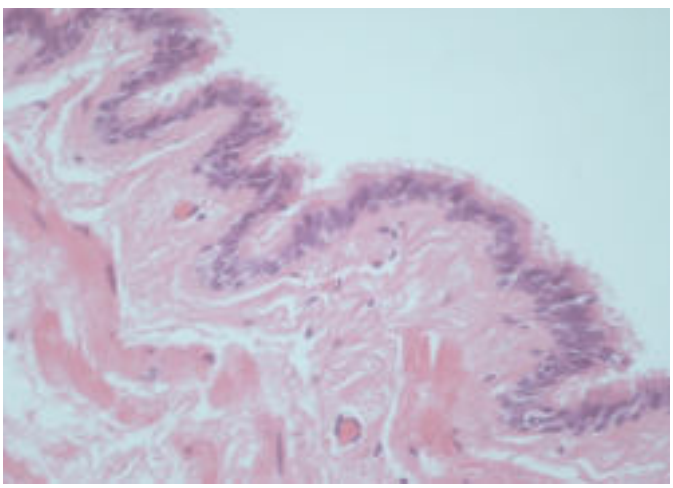

Fig. 5: Histological examination of the cyst: fibromuscular wall and ciliated epithelium.

Discussion

Bronchogenic cysts develop because of abnormal budding of branching of tracheobronchial tree within early stages of the gestation [4].
The most common locations of these lesions in mediastinum are para – tracheal, subcarinal, hilar, para – oesophagus, pericardium; less common locations are intra-pericardium, pre-sternal space, supraclavicular and subdiaphragmatic spaces, retroperitoneum and skin [5]; Literature also reports cases of ciliated hepatic foregut cysts [6].
About para-oesophageal bronchogenic cysts, the extramural type is the most common, but some of these lesions are located in the wall of the esophagus, with lumen mucosa intact [7]; connection with oesophageal lumen is very uncommon [8].
Preoperative diagnosis is usually difficult to obtain with classic imaging; endoscopic ultrasonography could be more helpful in distinguishing cystic from solid masses in the oesophageal wall context [9].
Bronchogenic cysts can become symptomatic because of structures compression and oesophageal lumen narrowing [10]; all suspected bronchogenic cysts should be removed in order to establish diagnosis, alleviate compression symptoms and prevent complications [11].
Literature review confirms the rarity of intramural oesophageal bronchogenic cysts; the majority of these lesions were located mediastinally and managed via open thoracotomy [12].

Conclusions

The case we have reported describes a very unusual case of a voluminous symptomatic intramural oesophageal disembriogenetic cyst; conventional preoperative imaging, endoscopy and also echo – endoscopy were not able to distinguish the cystic nature of the lesion versus a solid mass such as leiomyoma or gastrointestinal stromal tumour; surgical radical removal of the cyst has been successfully achieved with minimally invasive thoracoscopic approach.

Riassunto

INTRODUZIONE: Le cisti broncogene sono neoformazioni congenite derivanti dall'intestino primitivo che in genere si sviluppano in stretta contiguità con l'albero tracheo – bronchiale e l'esofago.
CASE REPORT: Descriviamo il caso di un paziente giunto alla nostra osservazione a seguito dell'insorgenza di moderata disfagia ed algie in regione epigastrica; la radiografia esofagea con bario aveva evidenziato un restringimento del lume esofageo a livello del terzo distale del tratto toracico attribuibile a compressione estrinseca; all'esame endoscopico, eco endoscopico ed alla TC con mezzo di contrasto è stata visualizzata una neoformazione parietale extramucosa avente caratteristiche simili ad un leiomioma.
RISULTATI: Il paziente è stato sottoposto ad asportazione chirurgica della massa esofagea con approccio toracoscopio. All'esame istologico definitivo la neoformazione è risultata una cisti broncogena.
CONCLUSIONI: Il caso che abbiamo illustrato descrive una voluminosa neoformazione parietale esofagea estremamente rara, sintomatica, della quale ne' l'endoscopia ne' la radiologia convenzionale ne' l'ecoendoscopia hanno evidenziato la natura cistica.
L'asportazione chirurgica radicale della suddetta massa è stata eseguita con approccio mini invasivo.

References

1. Wang W, Ni Y, Zhang L, Li X, Ke C, Lu Q, Cheng Q: *A case report of para-esophageal bronchogenic cyst with esophageal communication*. Journal of Cardiothoracic Surgery, 2012; 7:94.

2. St-Georges R, Deslauries J, Duranceau A, Vaillancour R, Deschamps C, Beauchamp G, Page A, Brisson J: *Clincal spectrum of bronchogenic cysts of the mediastinum and lung in the adult.* Ann Thorac Surg, 1991; 52:6-13.

3. Pages ON, Rubin S, Beahrel B: *Intra-esophageal rupture of a bronchogenic cyst*. Interact Cardiovasc Thorac Surg, 2005; 4:287-88.

4. Granato F, Voltolini L, Ghiribelli C, Luzzi L, Tenconi S, Gotti G: *Surgery for bronchogenic cysts: always easy?* Asian Cardiovasc Thorac Ann, 2009; **17**;467-71.

5. Li Z, Wang X, Yang E, Gao K, Huang L: *Gigantic intrapericaridal bronchogenic cyst*. Neth Heart J, 2011; 19:532-33.

6. Khoddami M, Aghdam MK, Alvandimanesh A: *Ciliated Hepatic Foregut Cysts: Two Case Reports in Children and review of the literature"* Case Rep Med. 2013; 372017.

7. Chafik A, Benjelloun A, Qassif H, El Fikri A, El Barni R, Zrara I: *Intramural esophageal bronchogenic cysts.* Asian Cardiovasc Thorac Ann, 2001; 19:69-71.

8. Knezevic J, Radovanovic N, Simic A, Micev M, Radojkovic S, Gerciz Z: *A paraesophageal bronchogenic cyst with esophageal communication*. Dis Esophagus, 1999; 12(4):321-32.

9. Lim LL, Ho KY, Goh PM: *Preoperative diagnosis of a paraesophagel bronchogenic cyst using endosonography.* Ann Thorac Surg, 2002; 73:633-35.

10. Nobuhara KK, Gorski YC, La Quaglia MP, Shamberger RC: *Bronchogenic cysts and esophageal duplications: common origins and treatment.* J Pediatr Surg, 1997; 32(10):1408-413.

11. Limaiem F, Ayadi-Kaddour A, Djilani H, Kilani T, El Mezni F: *Pulmonary and mediastinal bronchogenic cysts: A clinicopathological study of 33 cases*. Lung, 2008; 186:55-61.

12. Altieri MS, Zheng R, Pryor AD, Heimann A, Ahn S, Telem DA: *Esophageal bronchogenic cyst and review of the literature*. Surg Endosc, 2015 [Epub ahead of print].