

A rare case of paediatric intramural esophageal bronchogenic cyst treated by VATS

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ABSTRACT

Over the years, the progress and the greater competence gained by the video-assisted thoracoscopic surgery (VATS) in the field of paediatrics have led this surgical option to be regarded as the first choice in the treatment of cystic mediastinal masses.

The case of a 12 year-old patient is reported below, who successfully underwent the thoracoscopic resection of a rare intramural bronchogenic cyst of the esophagus. Thanks to the thoracoscopic technique, the cyst was completely removed and the integrity of the muscular wall of the esophagus was restored, thus preserving the continuity of the esophageal mucosa.

The surgical intervention ended without any complication. This is the first case of an intramural bronchogenic cyst of the esophagus in a paediatric patient treated by VATS reported in literature.

1. BACKGROUND

Bronchogenic cysts are the most common mediastinal cystic masses (40/50%), although their actual incidence is uncertain, as they are often asymptomatic and incidentally diagnosed, as in the case described herein. They are usually related to the tracheobronchial tree and/or the esophagus because of their origin in the primitive gut (1).

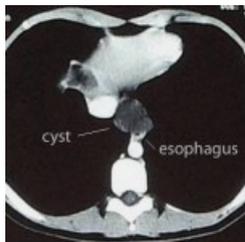
These masses include paraesophageal bronchogenic cysts, strictly adhering to the esophageal wall from which they may be separated by a cleavage plane. The cysts contained in the muscular wall of the esophagus, called intramural bronchogenic cysts, are much rarer and are in contact with the esophageal lumen only in exceptional cases (2,3).

The diagnosis of these masses in children is necessarily followed by a surgical treatment, considering not only the possible infectious complications, but also the potential increase in

volume resulting in the compression of the esophagus and/or the bronchial tree. Moreover, literature reports some cases of malignant degeneration also in asymptomatic patients (4)

2. CASE PRESENTATION

A 12 year-old girl was taken to the emergency room of our Hospital for observation due to “abdominal pain.” The pains were cramp-like and spread throughout the abdomen , with no sign of peritonitis and/or guarding.



A complete abdominal ultrasonography was performed, which showed the presence of an oval mass with a 4.9 X 3.1 cm. size, in close contact with the venae cavae and subject to an ambiguous diagnostic interpretation. The patient, therefore, underwent a CT angiography of the abdomen and chest, which confirmed the presence, in the distal esophagus, of an oval fluid mass, with peripheral enhancement after the injection of the contrast medium and without any evident cleavage plane with respect to the anterior wall of the esophagus (Fig.1). The thoracic MRI further indicated the presence of the mass in the distal esophagus, its size (3.5 cm transversal and 4.5 cm and longitudinal), thin and well-defined walls without any evident cleavage plane with respect to the esophageal wall as well as the signal of a cystic content (Fig.1). Finally, a contrast-examination of the esophagus showed a constant “semilune” footprint of the distal esophageal tract due to an extrinsic paraesophageal mass.

The patient then underwent a VATS with the diagnostic hypothesis of a paraesophageal bronchogenic or esophageal duplication cyst.

The intervention began with the orotracheal intubation with a Carlens tube, then the patient was positioned in the left lateral decubitus position. The procedure required an esophagoscopy which allowed for a better definition of the mass location and, above all, the lack of communication with the esophageal lumen. The esophagoscopy lasted for the whole duration of the intervention, thus ensuring a greater accuracy and safety during the thoracoscopic excision through a constant monitoring of the integrity of the esophageal mucosa.



Thanks to the presence of the Carlens tube, the procedure was mostly performed without CO2 insufflation, whose use has been limited only to a few steps and at a pressure of just 4 mmHg. The identification of the cyst, located in the anterolateral area of the distal esophagus, was easy thanks to the esophageal transillumination resulting from the endoscope. Once opened the mediastinal pleura, it was immediately evident that the cyst was intramural, as it was covered by the muscular wall of the esophagus. The complete enucleation has been carried out by

sectioning the muscle fibres and performing a blunt dissection then supported by a monopolar hook.(Fig.2) Once removed the mass and endoscopically controlled the integrity of the esophageal mucosa, the esophageal muscles were reconstructed by means of a 4-0 absorbable continuous suture and the overlying mediastinal pleura through a 5-0 absorbable continuous suture. The entire procedure lasted about 120 minutes and ended without complications. A thoracic drainage was placed through the 7th intercostal space along the posterior axillary line, and was removed after 24 hours.

3. DISCUSSION

The most common paraesophageal bronchogenic cysts are extramural, separated from the muscular wall of the esophagus by a sharp cleavage plane. In very rare cases, such as the one described in this paper, the mass can be contained within the esophageal muscular tract, in close contact with its mucosa (paraesophageal bronchogenic intramural cysts).

In this case, the real nature of the cyst was revealed by the histopathological examination, which demonstrated the presence of hyaline cartilage and respiratory epithelium within the mass enucleated from the esophageal wall. Although the diagnosis of the mass was accidental and the patient did not show dysphagia and/or any other symptom directly attributable to the presence of a cyst, the surgical intervention was chosen, in agreement with the studies reported in literature, considering not only the possible infectious complications, but also the potential increase in volume resulting in the compression of the esophagus and/or the bronchial tree, as well as some cases of malignant degeneration also in asymptomatic patients.

The nasogastric tube was kept for 7 days so as to favour the healing process while keeping the esophagus at rest. Liquids were administered through the tube starting from the recovery of the normal bowel function, followed by enteral nutrition supply until the 7th day. The removal of the tube was performed on the 8th day, liquids were administered orally, then a semi-liquid diet with the progressive addition of solid foods was followed in order to gradually resume the esophageal peristaltic activity and reduce the risk of developing an esophageal diverticulum in the surgical site.

The patient was discharged on the 12th day, without any complication and with a free diet. The radiological examination performed one month after the surgical intervention showed a regular peristaltic activity and a perfect recovery of the esophagus function without any alteration of its profile.

4. CONCLUSION

Paraesophageal bronchogenic intramural cyst was rare and had to be completely removed to prevent relapses. The combined use of VATS and esophagoscopy is the best choice in the treatment of such special cases. The intervention took place without any complication, and the postoperative course was excellent, thus showing that the VATS may be considered the best choice also in the treatment of rare cases. To date, in literature there are no cases of intramural bronchogenic cysts of the esophagus in paediatric patients treated through thoracoscopic surgery.

BIBLIOGRAPHY

1. Sarper A, Ayten A, Golbasi I, Demircan A, Isin E: Bronchogenic cyst. *Tex Heart Inst J* 2003, 30:105–108.

2. St-Georges R, Deslauriers J, Duranceau A, Vaillancourt R, Deschamps C, Beauchamp G, Page A, Brisson J: Clinical spectrum of bronchogenic cysts of the mediastinum and lung in the adult. *Ann Thorac Surg* 1991, 52:6–13.
3. Turkyilmaz A, Eroglu A, Subasi M, Findik G: Intramural esophageal bronchogenic cysts: a review of the literature. *Dis Esophagus* 2007, 20:461–465.
4. Limaiem F, Ayadi-Kaddour A, Djilani H, Kilani T, El Mezni F: Pulmonary and mediastinal bronchogenic cysts: a clinicopathologic study of 33 cases. *Lung* 2008, 186:55–61.