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Endoultrasonography (EUS) examination of the esophagus in the diagnosis of esophageal duplication: a case report and a review of a literature

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Abstract. – Esophageal duplication cysts are a rare medical entity. In most cases they are located at the level of the distal esophagus. Although our case is not unique, we want to focus on it as a reflection on diagnostic methods. The aim of this article is to show through the report on a case of esophageal duplication treated by us, followed by a review of similar cases in the literature, the utility of EUS in the diagnosis of upper-diaphragmatic and not communicating esophageal duplication. We report a case of a 43 year-old woman. She came to our attention for heartburn and retrosternal sense of space. The patient underwent an endoultrasonography (EUS) examination of the esophagus. The framework put EUS diagnosis of cystic formation of the esophagus (esophageal duplication cysts likely). We demonstrate that only EUS has a correlation with the determination of the pre-operative diagnosis with a statistical significance (p <0.001). In the diagnosis of esophageal not communicating duplication cysts EUS is the most specific diagnostic exam.

Key Words:
Esophageal duplication, Esophageal duplication, Entheric cyst, Esophageal endoultrasonography.

Introduction

Enteric cysts above the diaphragm account for 20% of all intestinal cysts[1]. Although these cysts are commonly considered as benign, neoplastic degeneration has been reported[2,3]. Esophageal cysts can be intramural and completely separate from the esophagus, they can also communicate with the esophagus lumen.

Most duplication cysts are solitary; a thick and viscous fluid may fill most esophageal cysts, although necrotic debris, inflammatory cells, or old blood can accompany respectively ulceration, infection or hemorrhage. Usually, esophageal cysts that remain asymptomatic throughout childhood are incidentally discovered in adulthood. Many patients show a minimal to moderate amount of dysphagia[5], but more common symptoms would be attributable to esophageal perforation, hemorrhage or pain from ulceration[6]. Esophageal duplication cysts could be detected by chest radiographs who reveals a sharply defined, spherical or tubular mass that commonly displaces the trachea or the esophagus. Esophagography in not communicating duplication cysts shows a displacing of the esophagus. Esophagoscopy has little value in differentiating esophageal cysts from other posterior mediastinal masses because the esophageal mucosa near cyst is normal[7]. CT and MRI have been largely used to clarify obscure masses as well as to define their relation to surrounding structures and evaluate vertebral defects[8]. MRI has been considered the most accurate to define the lesion and to detect vertebral and intraspinal abnormalities[9].

In the second half of the nineties EUS has been increasingly used in the diagnosis of esophageal duplication[10].

The aim of this article is to show through the report on a case of esophageal duplication treated by us, followed by a review of similar cases in the literature, the utility of EUS in the diagnosis of upper-diaphragmatic and not communicating

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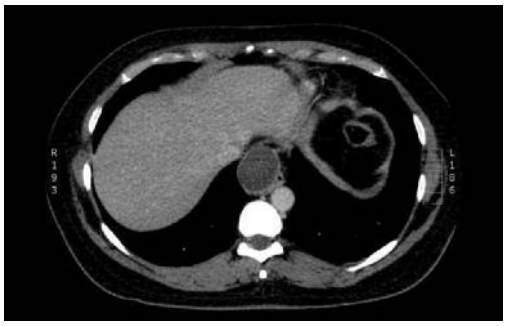

Figure 1. Coronal CT slide showing para-esophageal cystic lesion.

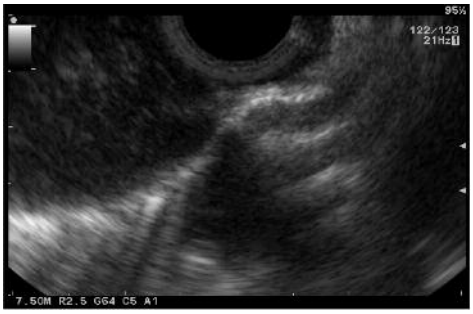

Figure 3. EUS image showing para-esophageal cystic lesion.

esophageal duplication. We decided to review all the literature concerning esophageal duplication cysts. We searched PubMed all articles by using as a keyword "esophageal duplication" and "oesophageal duplication."

Case Report

We report a case of a 43 year-old woman. She came to our attention for heartburn and retrosternal sense of space. An esophagoscopy was performed and it showed an ab-extrinsic compression of the lumen, with a length of about 2 cm, with normal mucosal appearance at the level of the distal third of the esophagus. For this reason the patient underwent a CT scan that showed formation of 42 mm x 40 mm x 47 mm pseudo-oval adherent to the distal esophagus (Figure 1 and 2). This formation caused a displacement of the esophagus slightly to the left and stenosis of the lumen. It did not show significant enhancement after contrast administration.

Later the patient underwent an endoultrasonography (EUS) examination of the esophagus (Figure 3 and 4). The examination was carried out with Olympus instrument and line-scan showed cystic lesion of the distal esophagus of about 4.5 cm in diameter, adherent to the esophageal wall, a hypoanechoic content. A fine needle aspiration biopsy of the lesion was performed with 19 G needle, with leakage of very dense mucous material. The framework put EUS diagnosis of cystic formation of the esophagus (esophageal duplication cysts likely).

The patient was, therefore, subjected to surgery to remove the lesion. After selective intubation and exclusion of right lung ventilation a right posterolateral thoracotomy at the seventh intercostal space has been performed. Incision of the mediastinal pleura and isolation of the esophagus with identification of the lesion were practiced. After incision of the adventitial tunica and of the longitudinal fibers of the muscularis mucosa the isolation of the cyst from esophagus was very difficult because of the inflammatory adhesions. After removing of the lesion the muscular plane was sutured. Histological examination confirmed the diagnosis of esophageal duplication cysts.

Four months later the patient is doing well without any signs of dysphagia.

Discussion

Esophageal duplication cysts are a rare medical entity. In most cases they are located at the

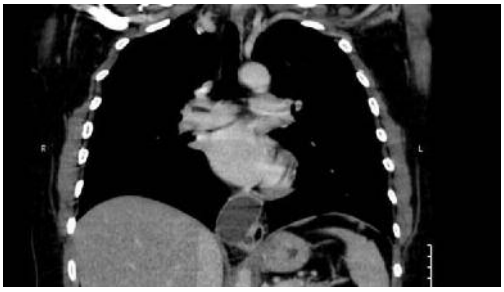

Figure 2. Frontal CT slide showing para-esophageal cystic lesion.

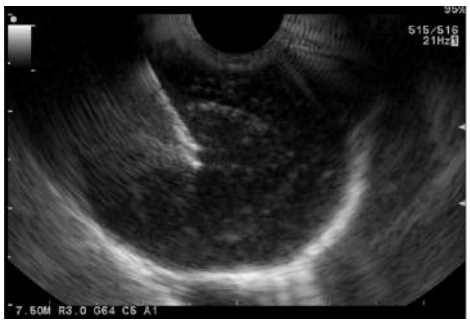

Figure 4. EUS image showing FNA of para-esophageal cystic lesion.

level of the distal esophagus. Many reported cases in the literature are related to esophageal duplication cyst, although studies with larger series, able to define the correct diagnostic and therapeutic iter, are still lacking. Our case is not unique, but we want to focus on it as a reflection on diagnostic methods.

We decided to review all the literature concerning esophageal duplication cysts. We searched PubMed all articles by using as a keyword "esophageal duplication" and "oesophageal duplication." Therefore, we selected 383 articles. Of these articles, we eliminated all those works written in a language different than English reducing our sample to 216. In order to make the sample as homogeneous as possible to our case, we decided to select only those reports describing similar cases. The exclusion criteria were, therefore:

- Sub-diaphragmatic esophageal duplication;
- Patients with age under 10 years-old;
- "Communicating" duplications;
- Cancerized esophageal duplication cysts;
- Cases previously published then 1995.

We decided to exclude cases of esophageal duplications located under the diaphragm, for the different diagnostic methods and a different surgical approach[11]. We eliminated the entire pediatric population suffering from congenital esophageal duplication cysts for controversial use of endoscopy in pediatric patients and EUS[12,13]. We also eliminated all cases of completely or partially "communicating" esophageal duplications for the different impact of endoscopy and esophagography in diagnostic for the specific morphology[14]. We eliminated cases of esophageal duplications presented as mediastinal neoplasms for different diagnostic and therapeutical approach[15,16]. We also decided to eliminate all those reports published prior to 1995 for the limited use of EUS in the instrumental diagnostic exams. Then we selected 31 articles[4,10,17-46]. The diagnostic tests used were chest x-ray, chest CT, MRI, Esophagografy and EUS. For this reason, we compared selected reports analyzing the different diagnostic techniques used before surgical treatment. Only in 20 cases the surgery was performed with a diagnosis of esophageal duplication. In the remaining 11 cases diagnosis was achieved in the post-operative histological analysis. We also analyzed the relationship between preoperative diagnostic technique and diagnosis by means of univariate analysis of the data. We demonstrate that only EUS has a correlation with the determination of the pre-operative diagnosis with a statistical significance ($p < 0.001$).

In the diagnosis of esophageal not communicating duplication cysts EUS is the most specific diagnostic exam. CT scan allows to obtain accurate informations to locate the lesion and about relations with surrounding structures but EUS makes possible to determinate the nature of the lesion itself.

The peculiarity of this exam is to precisely assess the splitting of the muscle lamina, containing anechoic fluid and smooth margins. The EUS is starting to be used also in operative treatment both in communicating esophageal duplication cysts[46] and in not communicating ones[18] with good results.

Conflict of Interest

The Authors declare that they have no conflict of interests.

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