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Rare diseases of esophagus: Surgical treatment of cysts in adults. Case report

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Esophageal cysts (EC) are congenital, extremely rare malformation. Up to 80% of EC are diagnosed in childhood, therefore, we can find only few clinical observations of EC in adults in literature.

CASE PRESENTATION: During the period from October to December 2019, a successful surgical treatment of 2 patients (1 male and 1 female) with enterogenous and duplication cysts of esophagus was performed at the Clinic of Faculty Surgery at Sechenov University. In both cases thoracic tumors were incidental findings during routine health investigation.

CLINICAL DISCUSSION: Clinical manifestations of ECs are caused by compression or displacement of the adjacent anatomical structures, therefore, most often patients complain of dysphagia, vomiting, pain in the chest, which may be constant or occur during an act of breathing. There are also observations of neurological symptoms due to compression of the radicular nerves. The method of choice in the treatment of ECs is their surgical removal.

CONCLUSION: Patients with mediastinal tumors should be treated in specialized hospitals by experienced surgeons who can cope with an unexpected intraoperative finding and carry out the appropriate surgery.

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1. Introduction

Esophageal cysts (EC) are congenital, extremely rare malformation based on pathogenesis of embryogenesis abnormalities [1,2]. According to the world literature, 1 in 8.200 newborns is diagnosed with esophageal cysts, moreover, they are detected twice as often in boys than in girls [3]. Up to 80% of esophageal cysts are diagnosed in childhood, therefore, we can only find single clinical observations of EC in adults in literature [4]. According to the EC classifications, commonly there are duplication cysts and enterogenous cysts, both of which are regarded as the most prevalent [5]. Approximately 60% of esophageal cysts are localized in the lower thoracic region and 90% have no connection with the lumen of the organ [6,7]. During the period from October to December 2019, a successful surgical treatment of 2 patients (1 male and 1 female) with enterogenous and duplication cysts of esophagus was performed at the Clinic of

Faculty Surgery at Sechenov University. In view of the rarity of EC in adults, we are presenting our observations.

The work is reported in line with the SCARE 2020 Guidelines [8].

2. Presentation of case

Patient Ch., 56 years old male, underwent treatment in October 2019 with complaints of difficult passing of food through the esophagus.

On the thoracic CT with intravenous contrast, a round growth is detected at the level of Th 7–Th 11 on the front surface of the esophagus, of up to 30.0 × 32.0 × 56.0 mm in size, with even clear contours, deforming the lumen, with a density of up to 17.0 HU, accumulating the contrast drug up to 38 HU. On the esophagogastroduodenoscopy: in the lower third of the esophagus, 33–34 cm from the incisors, the esophagus is deformed along the posterior-left wall by a voluminous growth of about 3 cm in size, with a flat surface (Fig. 1). The growth is mobile and of soft-elastic consistency. On the endoscopic ultrasound: in the lower third of the esophagus, a significant non-epithelial growth is detected with a length of up to 5.0 cm, with a diameter of up to 3.5 cm, coming from the muscle layer of the wall, with a clear uneven contour, heterogeneous with cystic density, closely adjacent to the aorta along the medial wall. On the radiography of the esophagus and the stomach: in the

Abbreviations: CT, computed tomography; EC, esophageal cysts; HU, Hounsfield units; MRI, magnetic resonance imaging; RATS, robot-assisted thoracic surgery; VATS, video-assisted thoracic surgery.

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Fig. 1. Endoscopic photo. Esophagus cyst. The malformation almost completely covers the lumen of the esophagus, passable for the endoscope.

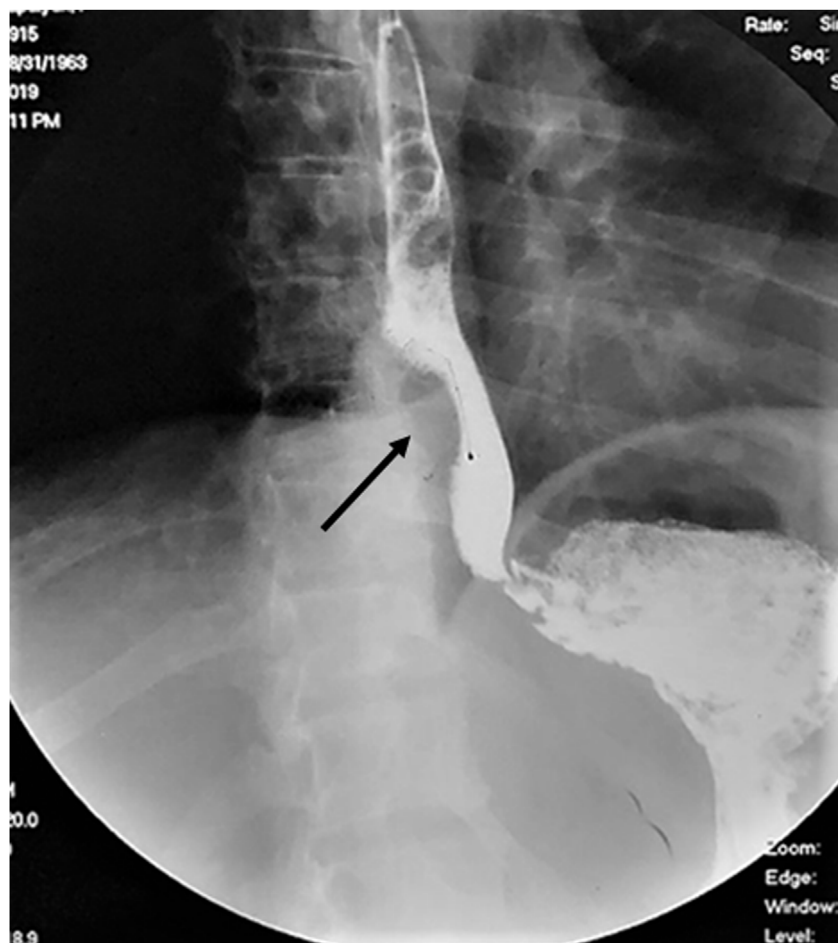


Fig. 2. The roentgenogram. Cyst of the lower thoracic esophagus (indicated by arrow).

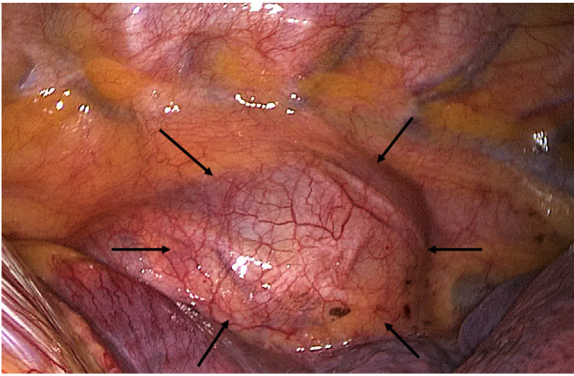


Fig. 3. Intraoperative photo. Esophageal cyst (indicated by arrows).

lower third of the esophagus in the supradiaphragmatic segment along the posterior-right contour, an oval-shaped marginal accumulation defect of about 3.5 cm long and with a clear contour was detected (Fig. 2).

Based on the clinical examinations, a preliminary diagnosis was made: leiomyoma of the lower thoracic esophagus. Second-degree dysphagia.

The patient underwent a video-assisted thoracoscopic surgery (2 thoracoports of 5 mm and 10 mm). The re-examination revealed: in the lower thoracic segment of the esophagus, 5 cm above the cardia, an intraparietal tumor was instrumentally palpated and was localized mainly in the posterior semicircle with a size of 5.0 × 4.5 × 3.5 cm (Fig. 3). After dissection of the muscle layer of the esophagus, a cyst with thin walls, not exceeding 1–2 mm was identified.

During the mobilization of the cyst, its lumen was opened and up to 90 mL of a jelly-like yellowish content was obtained. Further, the walls of the cyst were dissected in a blunt and sharp method, without opening the lumen of the esophagus. The muscle defect in the esophagus was stitched with a continuous suture on an atraumatic needle.

Intraoperative diagnosis: cyst of the lower thoracic esophagus.

The postoperative period proceeded without features and complications. The pleural drainage was removed on the 1st day after the surgery. According to the X-ray performed on the 3rd day after the surgery: upon taking 20 mL of water-soluble contrast orally, its free entry through the esophagus into the stomach was noted. No fluid exit beyond the organs was recorded.

The patient was able to eat independently through the mouth in full on the 4th day after the surgery and was discharged home on the 5th day after the surgery. According to the bacteriological research, the contents of the cyst were sterile. The results of the histological examination were consistent with duplication EC.

Female patient G., 29 years old, underwent treatment in December 2019. According to the anamnesis, she did not show characteristic complaints, however, the planned esophagogastroduodenoscopy performed at the place of residence, revealed narrowing of the esophagus at the level of the midthoracic section due to compression from the outside.

On the CT of the thorax: on the border of the upper and middle mediastinum, directly under the bifurcation of the trachea, an oval-shaped growth is detected, with clear, even contours, slightly accumulating the contrast along the contour. Its walls are thin, the contents do not accumulate the contrast drug. It is of moderately high density (+35–40 HU) 38.0 × 29.0 × 47.0 mm in size, slightly deforming the left main bronchus and moderately deforming the esophagus in the region of its middle third (Fig. 4).

According to the results of the instrumental examination, a preliminary diagnosis was made: a bronchogenic mediastinal cyst.

The patient underwent surgery. On the re-examination, in the mid-thoracic esophagus, immediately under the bifurcation of the trachea, an intraparietal tumor was detected, localized mainly in the right semicircle, closer to the posterior surface of the esophagus, of 4.0 × 3.0 × 4.5 cm in size (Fig. 5). After dissection of the muscle layer of the esophagus, a cyst with dense, 2–3 mm thick walls was identified. During the mobilization of the cyst, its lumen was opened and up to 70 mL of odorless, viscous, muddy yellow substance was obtained. Further, a blunt and sharp dissection of the cyst was performed, without opening the lumen of the esophagus. The muscle defect in the esophagus was stitched with a continuous suture on an atraumatic needle.

Intraoperative diagnosis: cyst of the middle thoracic esophagus.

The postoperative period proceeded without features and complications. The pleural drainage was removed on the 4th day after the surgery. On the radiography of the esophagus on the 5th day after the surgery, free passage of a water-soluble contrast medium through the esophagus was noted, no fluid exit beyond the organs was recorded. The results of the x-ray allowed to remove the nasogastric tube. The patient began to eat through the mouth in accordance with the esophageal diet on the 6th day after the surgery. On the 10th day she was discharged home in satisfactory



Fig. 4. CT scan. Esophageal cyst (indicated by arrow).

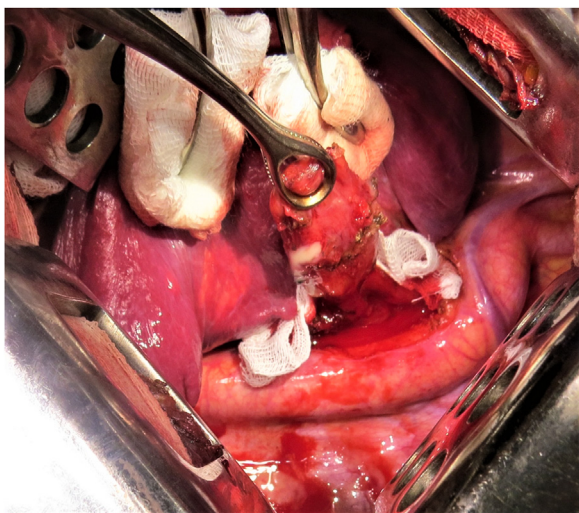


Fig. 5. Intraoperative photo. Esophagus cyst (taken by clamp).

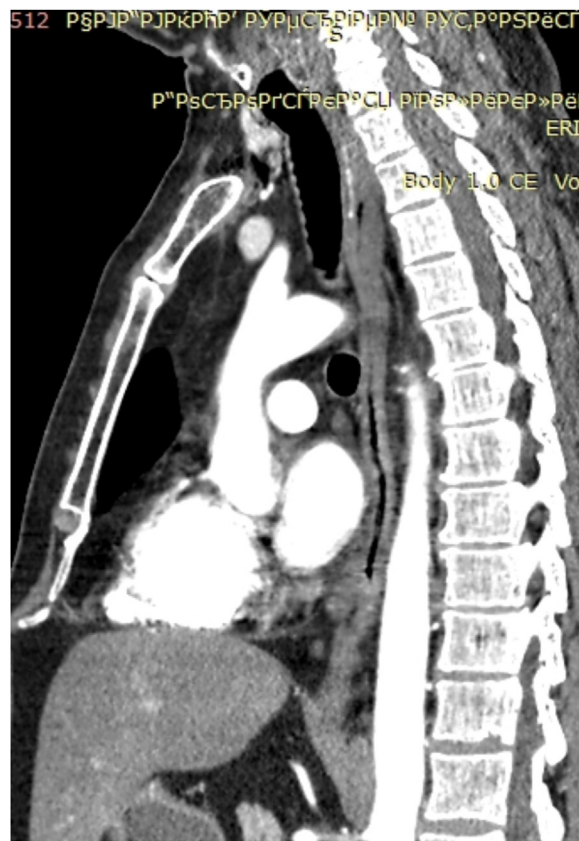


Fig. 7. CT scan of the 56 years old patient 9 month after thoracoscopic procedure.

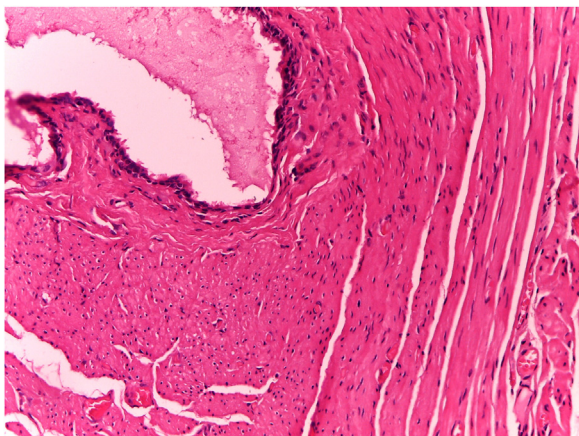


Fig. 6. Microphoto. Esophagus cyst. The cyst wall consists of two layers of smooth muscle tissue located at right angles, the inner surface is lined with greater extent. Foci of lymphoid infiltration are found in the subepithelial layer (there is no infiltration in this area). Stained with hematoxylin and eosin, x200.

condition under the supervision of a surgeon at the place of residence. On histological examination, the tumor corresponded to enterogenous EC (Fig. 6). Both patients were observed after 6–8 months after surgery and there were no recurrence and clinical symptoms of EC (Figs. 7 and 8).

3. Discussion

ECs were first described by Blasius in 1674. Later, in 1881 Roth presented simple epithelial (enterogenous) and duplication cysts [9]. Currently, there is no single commonly accepted classification of esophageal cysts. A number of authors use a classification based on the histological structure of the EC, suggested by Fallon in 1954. According to the latter, there are the following types of ECs: duplication, enterogenous, tracheobronchial enterogenous, posterior and neurogenic [10,11]. True duplication ECs are usually localized directly inside the organ wall and are lined from the inside with squamous or glandular epithelium. That's the EC structure we revealed during histological examination in our observation in the male patient. The female patient was diagnosed with enterogenous EC, which is by itself a completely isolated part of the digestive tract and can be lined with epithelium of any intestine section, including



Fig. 8. Image of the 56 years old patient chest 9 month after thoracoscopic procedure.

iliary. In the walls of these ECs, a double layer of muscle tissue is usually well defined (Fig. 6).

The etiology of EC has not been clearly studied, but it is believed that the development of both ECs is the result of impaired vacuolization and separation of the primary intestine at the 4th - 8th weeks of gestation [12,13]. ECs are most often localized along the right semicircle of the esophagus, due to rotation and elongation of the mediastinal organs during embryogenesis, as we were able to verify intraoperatively in both observations [13].

Clinical manifestations of ECs are caused by compression or displacement of the adjacent anatomical structures, therefore, most often patients complain of dysphagia, vomiting, pain in the chest, which may be constant or occur during an act of breathing. There are also observations of neurological symptoms due to compression of the radicular nerves [11,14,15]. Malignancy of ECs is rare [16]. ECs are usually randomly detected during endoscopic examination and/or endo-ultrasound, radiography, computed tomography or magnetic resonance imaging (CT or MRI).

In our observations, in both cases, ECs were randomly detected: on MRI in the male patient and on endoscopy of the esophagus and stomach in the female. However, when collecting the anamnesis, the male patient reported that over the past 5 years he had periodically had episodes of dysphagia, which is a characteristic symptom (in 20% of patients) for EC [16]. Despite the fact that at the preoperative stage the patients were examined as fully as possible using imaging methods of different resolutions, the preliminary diagnosis was not confirmed in either of the cases and the ECs turned out to be intraoperative findings.

The method of choice in the treatment of ECs is their surgical removal. Sauerbruch and Fick were among the first to report successful single-stage surgical intervention in EC cases in 1931 [11,17]. In case of simple enterogenous ECs, their enucleation is usually performed, and in case of duplication ECs, excision is performed. Traditionally, posterolateral thoracotomy is performed to remove an EC, but recently video thoracoscopic (VATS) and robotic thoracoscopic operations (RATS) have become common, and they provide the fastest patient recovery time and the best cosmetic effect [18]. The choice of surgical access in our observations was due to a preoperative diagnosis, so the female patient underwent surgery with thoracotomy.

4. Conclusion

Based on world literature and on our own experience, we can say that ECs are very rare diseases among adults. Despite this, it should be remembered that they comprise up to 10% of all diagnosed mediastinal neoplasms. It is significant that, despite the use of modern methods of instrumental diagnostics, it is not always possible to establish an accurate diagnosis before the surgery. Therefore, patients with mediastinal tumors should be treated in specialized hospitals by experienced surgeons who can cope with an unexpected intraoperative finding and carry out the appropriate surgery. The introduction of modern and minimally invasive technologies allows an adequate amount of surgical intervention to be performed, thus minimizing postoperative pain and the time spent by patients in the hospital, which positively affects their quality of life.

Conflict of interest

None of the authors have any conflict of interest to declare.

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Ethical approval

The study is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Sergey Osminin and Sergey Vetshev performed surgery, analyzed the literature and interpreted the patient's data regarding it. Vladimir Parshin, Roman Komarov, performed surgery and edited publication. Yuriy Strakhov performed the histological examination of the esophageal cysts. Ivan Ivashov participated to the manuscript editing to its final version, supervised the report and revised it critically. All authors read and approved the final manuscript to be published.

Registration of research studies

Not Applicable.

Guarantor

Sergey Osminin is the guarantor of submission and accepts full responsibility.

Provenance and peer review

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