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A case report of achalasia and large epiphrenic diverticulum treated by peroral endoscopic myotomy combined with diverticuloseptotomy

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The combination of esophageal achalasia with epiphrenic diverticulum is extremely rare and mostly treated surgically. Several cases of successful endoscopic treatment were presented in the literature, but treatment strategy has not yet been defined.

Aim. To present a case of the rare combination as achalasia and giant esophageal epiphrenic diverticulum successfully treated endoscopically.

Case report. We present to your attention a case report of a 75-year-old woman with complete dysphagia, significant weight loss, who was diagnosed with symptomatic epiphrenic diverticulum secondary to achalasia. Upper gastrointestinal endoscopy showed signs of severe fibrosis in the submucosal layer of the diverticulum. She received peroral endoscopic myotomy combined with diverticuloseptotomy made from the same submucosal tunnel. The regression of symptoms from 10 to 2 points according to the Eckardt symptom score clinically showed the treatment success.

Conclusions. The presented endoscopic one-tunnel technique could be an effective miniinvasive option for large symptomatic epiphrenic diverticula associated with achalasia.

Ключові слова: дивертикул, ахалазія, ПОЕМ, стравохід.

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Клінічний випадок лікування ахалазії з великим епіфренальним дивертикулом шляхом ендоскопічної пероральної міотомії в поєднанні з дивертикулосептотомією

О. М. Кіосов, В. С. Ткачов, С. М. Гулевський, М. В. Коробов

Поєднання ахалазії стравоходу з епіфренальним дивертикулом діагностують вкрай рідко, лікування зазвичай хірургічне. У фаховій літературі описано певну кількість випадків успішного ендоскопічного лікування, але стратегія лікування досі не визначена.

Мета роботи – описати випадок рідкісної комбінації ахалазії з гігантським епіфренальним дивертикулом стравоходу, що успішно вилікуваний ендоскопічним шляхом.

Клінічний випадок. Наведено випадок повної дисфагії, значної втрати ваги в жінки віком 75 років, у якої діагностовано симптоматичний епіфренальний дивертикул на тлі ахалазії. Під час ендоскопії верхніх відділів шлунково-кишкового тракту виявили ознаки вираженого фіброзу підслизового шару дивертикулу. З метою лікування їй виконали пероральну ендоскопічну міотомію в поєднанні з дивертикулосептотомією, що здійснили з того самого підслизового тунелю. Регрес симптомів від 10 до 2 балів за шкалою симптомів Екардта свідчить про клінічний успіх лікування.

Висновки. Наведена ендоскопічна однотунельна техніка може бути ефективним варіантом мініінвазивного лікування симптоматичних епіфренальних дивертикулів, асоційованих з ахалазією.

Esophageal epiphrenic diverticula are extremely rare, and, in most cases, are pulsion i. e., false diverticula (a diverticular pouch histologically consists only of the mucosa and submucosa) [1]. Pulsion diverticula can be caused by prolonged intraluminal pressure of the food bolus on a weak area in the esophageal wall and are infrequently associated with achalasia and spastic motility disorders. While surgical treatment options for symptomatic patients include open versus minimally invasive approach, diverticulectomy versus diverticulopexy, conventional versus selective myotomy with the possible simultaneous anti-reflux procedure, alternative endoscopic treatments are represented by traditional peroral endoscopic myotomy (POEM), salvage POEM (S-POEM), and diverticular POEM (D-POEM) [2,3]. POEM is proven, effective and safe procedure for the treatment of achalasia [4], but a treatment strategy for epiphrenic diverticula associated with achalasia is still undefined due to its rarity. We present the case of giant epiphrenic diverticulum (ED), which presumably occurred secondary to achalasia, treated by POEM combined with D-POEM.

Aim

To present a case of the rare combination as achalasia and giant esophageal ED successfully treated endoscopically.

Case report

A 75-year-old woman was admitted to the Multidisciplinary Surgery Department with complaints of one-year history of dysphagia to solid food, odynophagia, halitosis, regurgitation, vomiting, general weakness, and unintentional weight loss of 40 kg. Over the past 3 months, the symptoms worsened up to dysphagia to liquids, chest pain and aspiration phenomena. The esophagography (barium swallow)

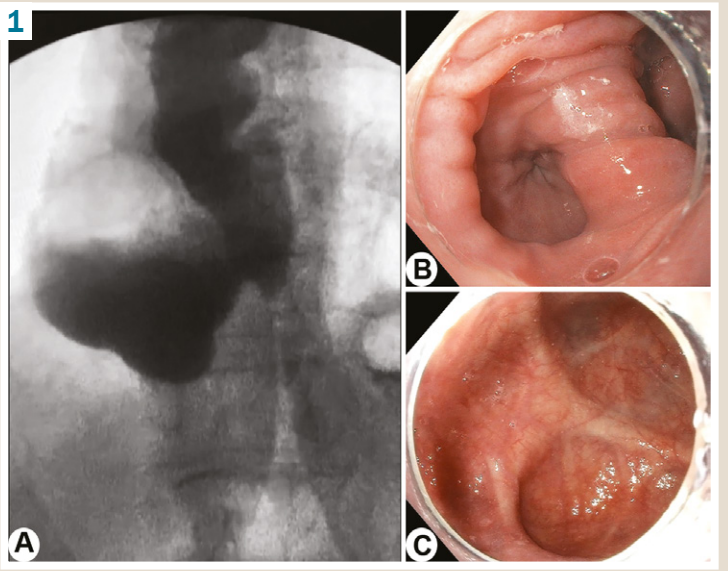

Fig. 1. A: Esophagoghraphy shows a diverticulum located in the distal 10 cm of the esophagus.
B: Upper endoscopy shows the thick diverticular septum, narrowed lumen of the lower esophageal sphincter (on the left) and ED (on the right).
C: Endoscopic signs of fibrosis in the diverticulum looks like pale strands in the submucosal layer.

Fig. 2. A: Diverticular septum visualized in the submucosal tunnel.
B: Myotomy of the circular muscle layer of the distal esophagus was performed.
C, D, E: Complete diverticuloseptotomy was performed to its bottom.
F: Mucosal incision was closed with 3 endoclips.
G: Postinterventional esophagography shows good passage of barium into the stomach.

showed achalasia with typical a bird-beak appearance and a large 7 × 6 cm right-sided ED with an air-fluid level in the distal third of the esophagus, esophageal dilation and tortuosity (*Fig. 1A*) with the absence of barium evacuation within 6 minutes. The findings were confirmed by upper gastrointestinal endoscopy (*Fig. 1B*) as well, that also revealed endoscopic signs of severe fibrosis in the submucosal layer of the diverticulum (*Fig. 1C*). On physical examination upon admission, her body mass index was 16.5 kg/m^2 and an Eckardt symptom score of 10.

The patient was scheduled to undergo endoscopic treatment, namely, the traditional POEM in combination with diverticuloseptotomy was performed. The operation was carried out under general anesthesia with endotracheal intubation, using CO_2 insufflation. The mucosal incision was done 26 cm from the incisors. Then, the submucosal tunnel was created, followed by 7 o'clock positioned, starting 3 cm below the tunnel entrance, 6 cm long myotomy of the esophagus, extended through the gastroesophageal junction on the 2 cm into the cardia (*Fig. 2B*). Subsequently, using the same submucosal tunnel, the muscular part of diverticular septum was dissected to its base (*Fig. 2A, C, D, E*). The septum bottom could be determined by reaching the connective tissue fibers of mediastinum on the esophageal side (*Fig. 2E*, on the left). 3 endoclips were applied for final closure (*Fig. 2F*). Immediately following the procedure, endoluminal passage of the endoscope to the stomach was effortlessly possible. The day after, postoperative esophagography showed a reduced size of the diverticulum, passage of contrast through the lower esophageal sphincter (LES) without delay and absence of leakage (*Fig. 2G*).

The mild asymptomatic carboxyperitoneum spontaneously disappeared on the third postoperative day. The patient was discharged on postoperative day 6 in a stable condition. The patient's clinical symptoms gradually decreased, and a regular diet was resumed. The Eckardt symptom score of 2 was 2 months after the treatment.

Discussion

Symptomatic epiphrenic diverticula are reasons for intervention and traditionally might be managed surgically [5]. Although it seems like the septum separating the diverticulum from the esophageal tract is main area of interest for endoscopic treatment [6], ED usually develops secondary to a primary esophageal motility disorder, such as achalasia. In patients with achalasia and ED, the underlying motility disorder cause the symptoms rather than the ED per se [7], as evidenced by the fact that a myotomy without ED resection results in symptom relief [8,9]. While surgical treatment in the past involved a lateral thoracotomy or thoracoscopic approach, the preferred treatment today is a less invasive laparoscopic procedure involving longitudinal transection of the diverticulum along with a cardiomyotomy and partial fundoplication. However, similar to laparoscopic Heller myotomy, which is used to treat spastic esophageal motility disorders such as type III achalasia or jackhammer esophagus, a laparoscopic transhiatal approach is not recommended for patients with large diverticula, significant distance between the neck of the diverticulum and the hiatus, or dense adhesions between the diverticulum and adjacent mediastinal tissues. These factors make dissection, stapling, and muscle layer approximation more challenging. Myotomy is typically performed on the opposite side of the diverticulotomy. Adverse events can include staple line leakage resulting in abscess formation and sepsis, particularly in cases when myotomy is not performed concurrently. These complications can cause significant morbidity and mortality rates of up to 33 % and 10 %, respectively [10].

One of the modern and relevant methods of achalasia treatment, that is not inferior to a surgical approach, is POEM, which was introduced by H. Inoue in 2010 [11] and today has gained immense popularity due to its effectiveness and less invasiveness, evolving into a mainstream treatment option for achalasia [12,13]. The procedure has been shown to be effective and safe for the treatment of esophageal motility disorders also in the presence of a large ED [10]. Furthermore, this technique has been found to be safe and achievable, even in older patients with pre-existing comorbid conditions or in those who are poor candidates for surgery, as well as in individuals after failed prior open surgery, and has demonstrated favorable short-term results [14].

Several endoscopic techniques are described for this case. During the traditional POEM, a submucosal endoscopic myotomy of the inner layer of the esophagus at the 6 or 12 o'clock position is performed, without regard as to the presence of ED. Meanwhile, S-POEM involves myotomy on the opposite esophageal side of diverticular neck, i. e., at 6–8 o'clock position or 1–2 o'clock position, according to the location of the diverticulum [15]. D-POEM entails performing a septotomy after creating submucosal tunneling twice on both sides of the ED [3]. If a patient is suffering from both symptomatic ED and achalasia, the choice between traditional POEM or S-POEM as an initial treatment will depend on factors such as the experience and familiarity of a team with abovementioned endoscopic procedures, as well as the complexity of the muscle fibers connecting the ED to the LES. If an operator chooses the D-POEM, the decision to conduct additional myotomy on the inner muscle beyond the diverticulum will depend on the underlying motility disorders and whether submucosal layer allows to make the tunnel wide enough for an endoscope to pass through [3].

In our patient, we performed standard POEM in conjunction with diverticuloseptotomy. The similar technique has been already described by Beyna et al. [10]. The choice of the technique was primarily determined by the size and position of ED, presence of large mucosal septum and endoscopic signs of extended submucosal fibrosis. Performing both myotomy and septotomy from the single submucosal tunnel could make the procedure less invasive, avoiding unnecessary tissue damage, less time consuming, easier, and more cost-effective. Due to its size and assumed adhesions to the surrounding tissues, the outpouching of the esophageal wall remained on the esophagography even on the next day after the operation. Nevertheless, the subjective clinical improvement was confirmed by regression of Eckardt symptom score to 2 from initial 10. In our opinion, the main goal of interventions for esophageal diverticulum is to relieve the symptoms in patients and improve their quality of life. As a result, the focus should not be solely on achieving complete removal of the diverticulum.

Conclusions

1. In conclusion, POEM combined with diverticuloseptotomy in a single endoscopic approach seems to be the effective and promising technique to control symptoms in case of large symptomatic epiphrenic diverticula associated with achalasia.

2. Since the majority of epiphrenic diverticula arise from underlying motility disorders, and myotomy is crucial for safe and effective treatment, this new and less invasive one-tunnel technique holds promise to replacing surgery for a large number of patients in the future.

Prospects for further research. Comparative studies of existing miniinvasive treatment options for ED associated with achalasia in order to define the method of choice.

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