

Esophageal Intramural Pseudodiverticulosis

Juliana Bertassi Mazucato^{1*},  Fernando Carpentieri-Ferrarezi¹,  Emiliano de Carvalho Almodova²,  Luiza Cavalero de Lima¹ 

OPEN ACCESS

Citation:

Mazucato JB, Carpentieri-Ferrarezi F, Almodova EC, Cavalero de Lima L. Esophageal Intramural Pseudodiverticulosis. *Rev Colomb Gastroenterol*. 2022;37(1):78-82. <https://doi.org/10.22516/25007440.708>

¹ União das Faculdades dos Grandes Lagos (UNILAGO), São José do Rio Preto, Brazil.

² União das Faculdades dos Grandes Lagos (UNILAGO), Professor, São José do Rio Preto, Brazil.

Correspondence: Juliana Bertassi Mazucato.
ju.bertassi93@gmail.com

Received: 14/01/2021

Accepted: 09/12/2021



Abstract

A report of two cases of esophageal intramural pseudodiverticulosis, a very unusual disease, with other 240 cases reported in the entire world literature since 1960. Its etiology and pathogenesis are still not fully understood. However, it is believed that hypertrophy of the submucosal glands, with chronic inflammation, fibrosis, and consequent esophageal stenosis, causes dysphagia, which is the primary manifestation of esophageal intramural pseudodiverticulosis. The main diagnostic methods include the radiological examination of the esophagus with barium contrast (esophagogram) and esophagogastroduodenoscopy (EGD). Both reported cases were treated with endoscopic dilation, exemplifying the safety and efficacy of this therapeutic option for treating dysphagia in these individuals.

Keywords

Esophageal intramural pseudodiverticulosis, endoscopic dilation, dysphagia.

INTRODUCTION

Esophageal intramural pseudodiverticulosis (EIPD) is an extremely rare disease, first described in 1960 by Mendl *et al*. Since then, around 240 published cases have been collected worldwide until 2014⁽¹⁾. EIPD primarily affects men in their 60s⁽²⁾.

Its etiology and pathogenesis are not yet fully known. The main hypothesis to explain EIPD results from a hypertrophy of the submucosal glands with cystic dilation of the excretory ducts. During the illness course, inflammation of the submucosal glands can cause fibrosis of the esophageal wall, with consecutive stricture of the lumen. Esophageal stenosis leads to dietary impacts, malnutrition, and dysphagia, which is present in up to 80% of patients⁽²⁻⁵⁾.

Diagnosis is established by upper gastrointestinal endoscopy (UGE) and a radiological examination of the esophagus with barium contrast (esophagogram). Histological examination is essential to differentiate between benign and malignant stenosis. During UGE, numerous ostia can be visualized, measuring between 2 and 4 mm with whitish collections on the outside of the esophageal wall and stenosis. The esophagogram shows an esophageal stricture and small areas of contrast accumulation, corresponding to the pseudodiverticula⁽²⁾.

Conventional treatment is based on relieving symptoms. Dysphagia is treated by endoscopic dilation with Savary-Gilliard tubes. When there is gastroesophageal reflux and moniliasis, specific medications are used for their treatment. Most EIPD cases have a good evolution with this therapeutic scheme⁽³⁾. Esophagectomy was rarely necessary⁽⁶⁾.

MATERIALS, METHODS, AND RESULTS

Case 1

A 73-year-old female, native and resident of Barretos, SP, Brazil. A smoker who uses 60 packs per year. She suffered from progressive intermittent dysphagia for 17 years. During this period, she made a soft diet. She even had food impact treated by endoscopy. She sought medical attention when she had dysphagia for liquids, but the EIPD diagnosis took 1 year. During this period, she lost 35 kg and underwent a surgical gastrostomy. After diagnosis, she consulted for dilation with Savary-Gilliard tubes. The treatment was initiated with a 5.0 mm tube. After the second session, the 11 mm diameter was reached. At this point, she started eating solids, and the gastrostomy tube was performed. The treatment with dilatations was extended for 6 months, for a total of 5 sessions. She also received fluconazole to treat esophageal moniliasis, present in all performed endoscopies. Omeprazole was also administered for esophagitis due to gastroesophageal reflux disease (GERD), aggravated by a hiatal hernia. The patient had an excellent response to treatment and has remained asymptomatic for 3 years (**Table 1** and **Figure 1**).

Case 2

VJR is a 55 year old male, born in Valparaiso, SP, resident of Tucuruí, PA, Brazil. He suffered from dysphagia for 10 years. He ate only soft and liquid foods in this period. He had food impact 4 times and required endoscopic extrac-

Table 1. Clinical picture, endoscopic findings, and therapeutic response of reported cases

Personal data	Case 1	Case 2
Sex	Female	Male
Dysphagia time (years)	Yes (17 years)	Yes (10 years)
Impact	Yes	Yes
GERD esophagitis	Yes	Yes
Moniliasis	Yes	Yes
Smoker	Yes	Yes
Uses alcoholic beverages	No	Yes
Weight loss (kilos)	35	No
Weight gain after treatment	Yes	Yes
Esophageal stenosis	Yes	Yes
Treatment with esophageal dilation	Yes (up to 11 mm)	Yes (up to 12.8 mm)
Dysphagia return time	Asymptomatic 3 years ago	6 months

GERD: esophagitis due to gastroesophageal reflux disease. Source: Table prepared by the author

tion. When he sought medical help, he drank fluids for 3 months. He reported symptoms such as dysphagia, pyrosis, and moderate gastroesophageal reflux in chronic use of omeprazole. The patient claims to consume alcoholic

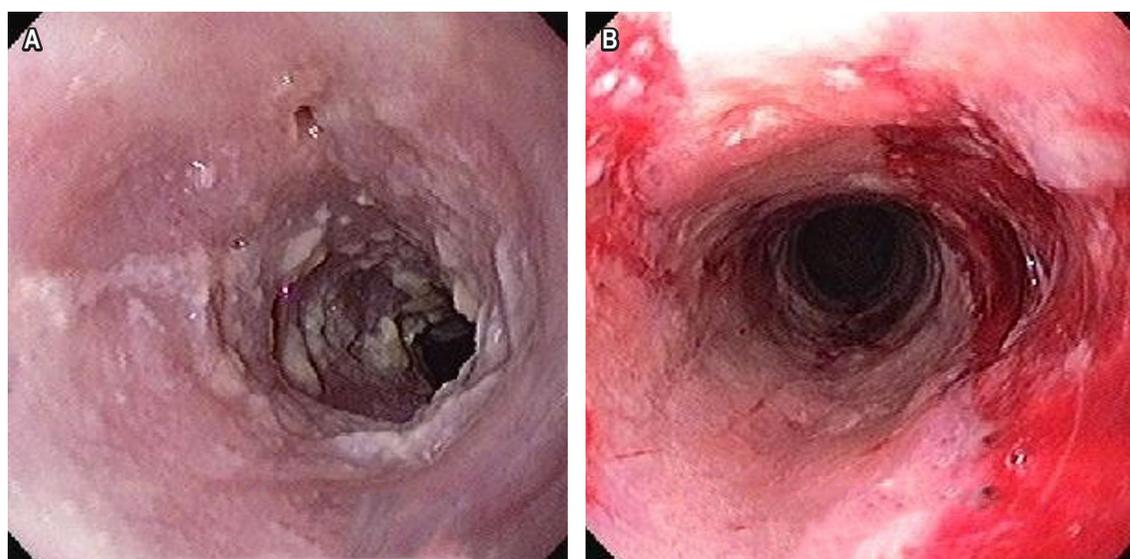


Figure 1. Endoscopic images of patient's EIPD from Case 1. **A.** Diagnostic image of EIPD for moniliasis. **B.** Image after endoscopic dilation. Source: personal file.

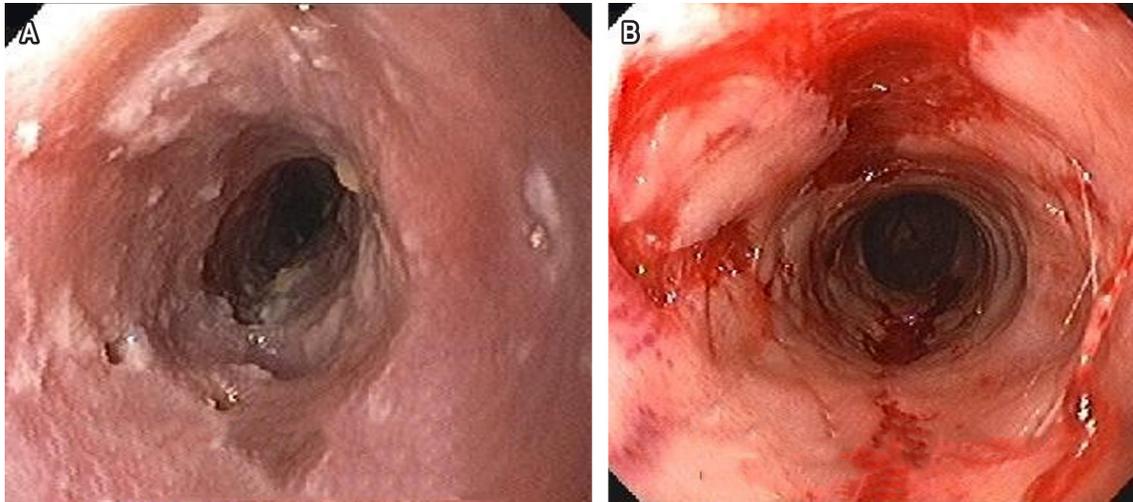


Figure 2. Endoscopic images of patient's EIDP from Case 2. **A.** Diagnostic image of EIDP for moniliasis. **B.** Image after endoscopic dilation. Source: personal file.

beverages at least once a month and is a smoker who uses 32 packs per year. EIDP was diagnosed 3 years ago through an upper gastrointestinal endoscopy when esophageal candidiasis was also found.

Initially, 3 sessions were held at 1-week intervals. Dilations began with 7.0 mm tubes. At the end of 3 weeks, they reached 12.8 mm with a complete reversal of dysphagia. Due to the distance between the state of Pará, where the patient resides, and the city of São José do Rio Preto, where he undergoes treatment, dilation sessions have been held every 6 months for the last 3 years. The patient reports remaining asymptomatic for 5 months until the dilation endoscopy presented dysphagia to solids in the last month. He also presents with moniliasis whenever digestive endoscopy is performed. From the beginning of the treatment, the patient gained 11 kilos (**Table 1** and **Figure 2**).

DISCUSSION

Dysphagia was the main symptom in the two cases reported in this study. It was also the main reason the individuals sought medical help. Similarly, more than 72% of patients in the following studies had dysphagia as their main symptom^(1-3,5-9).

Patients are over 70 years old in both reported cases, very similar to the data provided in most cases in the medical literature^(1-3,6,7,9-14). Except for patients with human immunodeficiency virus (HIV), who presented the condition at age 35 without evidence of moniliasis⁽⁸⁾, and at age 45 (**Table 2**)⁽⁵⁾.

Table 2. Summary of the main findings of the case reports in the medical literature

Datos personales	Número de casos	Porcentaje (%)
Years (average)	62 years	-
Woman	03	27.27
Man	08	72.72
Dysphagia	08	72.72
Impact	03	27.27
GERD esophagitis	07	63.63
Moniliasis	07	63.63
HIV	02	18.18
Smokes cigarettes	03	27.27
Uses alcoholic beverages	05	45.45
Esophageal stenosis	05	45.45
Treatment with esophageal dilation	07	63.63
No improvement after dilation	01	09.09
Dysphagia returned	00	00

GERD: gastroesophageal reflux esophagitis; HIV: human immunodeficiency virus. Source: Table prepared by the author.

In the total of the summarized studies in **Table 2**, a higher incidence of EIPD was demonstrated in men. However, in this report, one case was reported in a man and the other in a woman^(1,3,5,7-10,13).

Reflux esophagitis and fungal infection may accompany EIPD. In this study, both patients had GERD esophagitis and *C. albicans* infection. The authors^(2,6-10,12) reported cases of esophagitis due to GERD^(1-3,8,9,12,13), where a candid infection was found in the esophageal biopsy (**Table 2**).

Both patients in this study were treated with endoscopic esophageal dilation, medicines for esophagitis, and GERD moniliasis. Seven studies reported treatment with esophageal dilation and medication for existing pathologies in the literature^(1-3,5-7,9).

Case studies^(8,10,12,13) performed conservative treatment only with medication for existing pathologies. Only one of these cases⁽¹⁰⁾ showed no improvement in pseudodiverticula during the follow-up period (**Table 2**).

In this study's patients, improvement in dysphagia was felt after the first endoscopic dilation session. However, in case 2, there was a recurrence of dysphagia to solids 6 months after the last dilation session. This evolution is explained by the fact that this person lives 2200 km away from the healthcare facility where the treatment is performed, so he cannot carry out all consecutive dilations until a satisfactory esophageal diameter is established. The patient in case 1 received 5 consecutive sessions over 6 months and remains asymptomatic 3 years after the last endoscopic dilation session (**Table 2**).

CONCLUSION

Reports exemplify the difficulty of diagnosing EIPD and demonstrate that endoscopic dilation is a safe and effective option to treat dysphagia in these individuals.

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