CLINICAL CASE

Intramural esophageal dissection due to pharyngeal abscess treated by endoscopic esophageal transection: A case report

Dissection œsophagienne intramurale en raison d’abcès pharyngeal traité par section transversale œsophagienne endoscopique : un cas clinique

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Summary Intramural esophageal dissection is a rare disorder characterized by extensive laceration between the mucosal and submucosal layers of the esophageal wall, but without perforation. The etiology of intramural dissection of the esophagus remains uncertain. Conservative management is usually considered adequate. Only one case of circumferential intramural esophageal dissection has been reported previously. We report here on a case due to an infectious cause (paralyseal abscess) that is also an unusual example of circumferential intramural esophageal dissection, which was then treated by endoscopic transection of the true internal esophageal wall and bougienage dilation.

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Résumé La dissection œsophagienne intramurale est une lésion rare caractérisée par une longue laceration entre les couches muqueuses et sous-muqueuses de la paroi œsophagienne sans perforation. L’étiologie de la dissection intramurale de l’œsophage demeure incertaine. Le traitement Conservateur est habituellement préconisé. Un cas unique de dissection œsophagienne intramurale circonférentielle a été rapporté précédemment. Nous rapportons un cas dû à une cause infectieuse (abcès pharyngé) qui est également le deuxième cas peu commun d’une dissection œsophagienne intramurale circonférentielle, traitée par transection endoscopique de la paroi luminaire vraie et dilatations aux bougies.

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Introduction

Intramural esophageal dissection (IED), a rare disorder, is the result of longitudinal separation of the submucosa and muscle layers of the esophagus, but without perforation. The disorder is seen predominantly in middle-aged or elderly women. The most common presenting symptoms are sudden severe retrosternal pain, hematemesis, odynophagia and dysphagia. The diagnosis is made by contrast esophagography, esophageal endoscopy or both. The etiology of IED is uncertain, and most reports of IED advocate conservative management. The majority of the previously reported cases of IED were partial, with only one case of circumferential IED reported previously [1]. Although endoscopic treatments of IED have been reported [2–6], their use in the management of circumferential IED has been reported only in a single case [1]. In the present report, we describe a second case of circumferential IED—and the first to be most likely due to a pharyngeal abscess. In this patient, dysphagia did not improve with conservative management, thereby necessitating endoscopic treatment, including transection (transverse resection) of the true lumen (internal esophageal mucosa/submucosa layers) and bougienage dilation.

Case report

A 32-year-old man with no previous medical or surgical history was admitted because of diabetes mellitus and severe acute pharyngitis. Rhinolaryngeal examination revealed edema of the larynx with epiglottal ulceration; cervical tomodensitometry confirmed that it was a non-collecting paryngeal abscess. The patient underwent antibiotic therapy, although the pathogen was not identified. The course of the disease showed regression of general signs, but persistence of dysphagia. Upper gastrointestinal endoscopy showed a congestive muscular lumen (false lumen) due to circumferential mucous separation that was virtually complete throughout the whole of the esophagus except for the last 6 cm, where dissection involved only two-thirds of the circumference. The true oesophageal wall appeared to be tubular and lateral (Fig. 1a), with the presence of abundant pus in the stomach (Fig. 1b). Esophagography did not show the classical ‘double-barreled’ esophagus with true and false lumen, but revealed an excentred thread-like narrowing along 8 cm of the lower esophagus, with dilatation upstream (Fig. 1c). The patient was kept on conservative treatment and triple antibiotherapy and was fed through jejunostomy.

Nine weeks later, the patient was readmitted because total enteral feeding had failed to bring about any improvement of dysphagia. Endoscopy revealed healing, with re-epithelialization of the esophageal muscular layer. In addition, it was possible at the same time to advance the scope into the stomach by way of a bidirectional mucosal bridge 20 cm from the incisor teeth via the luminal narrowing (center) into the true lumen and via the false lumen laterally (Fig. 2a). To resolve the dysphagia, endoscopic treatment was applied, consisting of endoscopic transection of the true internal lumen using endoscopic diathermy snare and hot biopsy (Olympus forceps) (Fig. 2b). The procedure was easily performed along most of the esophagus until the last 6 cm, where the procedure became more difficult because the dissection involved only two-thirds of the circumference. Two sessions were necessary with a 48-h interval. However, the procedure was successful, with no complications following the procedure.

One week later, the patient no longer complained of any discomfort. Endoscopic resection was total, with easy passage of the endoscope except for two mucous remnants at the gastro-oesophageal junction that were resected using the diathermy snare. Oral intake was then permitted.

Two months later, the patient complained of recurrence of dysphagia, and follow-up endoscopy examination revealed an esophageal stricture lying 35 cm from the incisor teeth, which was passable only by a pediatric videendoscope. Four sessions of endoscopic dilatation with bougienage were performed, using Savary—Gilliard bougies, which allowed easy passage of an adult-sized videendoscope. Six months later, clinical improvement, in particular, of dysphagia and oral feeding with recovery of weight loss was noted.

Discussion

IED was first reported by Marks and Keet [7] in 1968. The mechanism of IED is thought to be a sudden change in intraesophageal pressure. Predisposing factors include coagulation defects, endoscopic instrumentation and injection sclerotherapy for esophageal varices. However, in most cases, IED is the result of intramural hematoma [8]. Based on the reported cases, most patients with IED are women in the
Endoscopy shows the true lumen (center) and the false lumen (laterally) (a), endoscopic transection of the true internal lumen using hot biopsy (Olympus forceps) (b).

8th or 9th decade of life. However, younger patients have been reported and, occasionally, a male patient as well; the present case is the third report involving a young man [3,9].

The most common presenting symptoms include sudden onset chest pain, odynophagia, dysphagia and hematemesis. Thus, the age, gender and presenting manifestations in the present case were highly atypical. Also, the patient was not taking any medications that altered blood coagulation; his clotting profile was normal. Furthermore, in the present case, it is believed that the IED might have been related to infection because of the pharyngeal non-collecting abscess and the presence of abundant pus in the stomach. The pharyngeal abscess may have fused with and extended into the submucosal layer that led to separation of the submucosa from the muscular layer of the esophagus. Also, the majority of previously reported patients with IED had partial dissection, while only a single case of circumferential dissection has been previously reported [1].

In the present patient, the diagnosis was made by esophageal endoscopy. The IED was circumferential along the entire length of the esophagus except for the last 6 cm, where dissection involved only two-thirds of the circumference. In most reported cases, management was conservative and usually resulted in a favorable outcome. However, our patient was atypical not only in terms of age, gender, presentation and probable infectious etiology, but also because of persistent symptoms despite conservative management. Surgical intervention is usually not necessary for IED. Endoscopic treatments have been reported [2–6], but its use in the management of circumferential IED has been reported in only one case [1] that, for the first time, consisted of endoscopic transection of the true internal esophageal wall. In the present case, because dysphagia did not improve after a 9-week course of conservative treatment, we proceeded with several endoscopic treatments, including transection of the true internal esophageal wall. As reported in the first case of such transection, a stenosis occurred in our patient that required four sessions of endoscopic dilation by bougienage, using Savary—Gilliard bougies.

In conclusion, this was the first case report of an IED that was probably due to a laryngeal abscess that became fused with the esophageal wall, and the second report of a patient with circumferential IED who received a series of endoscopic treatments. In our experience, as in the single previously reported case, it is difficult to expect spontaneous healing with conservative treatment alone of circumferential IED, which appears to be a more severe variant of IED, given the lack of response to conservative treatment and the occurrence of stenosis after endoscopic treatment. Also, endoscopic treatments should be considered early in the therapeutic strategy.

Conflict of interest statement

The authors have not declared any conflict of interest.

References