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Case report

Serra doria procedure as an alternative treatment for end stage achalasia. A case report

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ABSTRACT

The standard of care for achalasia is laparoscopic Heller's cardiomyotomy. This procedure achieves satisfactory and long-standing results in over 85 % of patients. However, disease progression occurs in some patients leading to end-stage achalasia, occasionally requiring oesophagectomy. In a recent systematic review and meta-analysis of 1307 patients who underwent oesophagectomy for end-stage achalasia, the pooled prevalence of pneumonia, anastomotic leakage and mortality were 10 %, 7 % and 2 %, respectively. We present a Serra Doria procedure as an alternative 'esophagus-preserving' procedure in a 58-year-old female patient with end-stage achalasia. This advancement highlights the crucial role of personalized care and the ongoing research necessary to enhance outcomes for those suffering from this challenging condition.

1. Introduction

Achalasia is a primary motility disorder in which insufficient relaxation of the lower esophageal sphincter (LOS) and absent peristalsis result in stasis of ingested foods and subsequently, lead to esophageal symptoms of dysphagia, regurgitation, chest pain or weight loss.

Treatment can be considered for the purpose of reducing symptoms and consequently, improvement of quality of life. Heller's myotomy combined with partial fundoplication is considered the gold standard surgical management for achalasia, with satisfactory results in the majority of patients. However, disease progression occurs in some patients leading to end-stage achalasia, occasionally requiring oesophagectomy. In a recent systematic review and meta-analysis of 1307 patients who underwent oesophagectomy for end-stage achalasia, the pooled prevalence of pneumonia, anastomotic leakage and mortality were 10 %, 7 % and 2 %, respectively [1]. We present a Serra Doria procedure as an alternative 'esophagus-preserving' procedure in patients with end-stage achalasia.

2. Case report

A 58-year-old female patient without any particular pathological history presents with progressive dysphagia to solids that has been evolving for approximately 10 years. She did not consult a doctor during this period, but had taken a proton pump inhibitor irregularly.

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The history of the disease dates back to 1 month of his admission marked by the worsening of dysphagia which became mixed with solids and liquids associated with vomiting and retrosternal pain all evolving in a picture of alteration of the general condition as well as a weight loss of more than 15 kg in 3 months.

The physical examination shows a BMI of 17 kg/m^2 as well as arterial hypotension and signs of extracellular dehydration.

In biology we find a Metabolic Acidosis with K+ at 2.8 mmol/L, Anemia at 8 g/dL and Hypo Albuminemia at 25 g/L.

An esophagogastric fibroscopy was done after a nasogastric aspiration which revealed a dilated esophagus with a food stasis over its entire height, a Cardia in place at 40 cm, crossed with a sensation of jumping. An aspect that can thus fit with achalasia.

A high resolution esophageal manometry was indicated showing an absence of contractions and esophageal pressurization, a Residual Pressure integrated at 4s is at 36 mmHg, A Resting Pressure of the lower esophageal sphincter at 44 mmHg with absence of relaxation in 100 % of swallows.

He also claims obstruction of the esophagogastric junction in the 200 ml swallowing test.

All this fits with a pattern of ACHALASIA type I.

We thus completed with a thoraco-abdominal CT with digestive opacification (Fig. 1) which confirms the presence of esophageal dilatation extended from Killian's mouth to the cardia with stasis measuring 61 mm at most. This is associated with intense mucosal enhancement without any image of luminal stenosis or detectable extrinsic compression with passage of fluid intragastrically but not contributory since the dysphagic patient could not swallow a sufficient quantity of contrast product. What affirms an "End Stage" ACHALASIA.

Heller Cardiomyotomy should be avoided given the existence of several factors of failure in this patient notably an age greater than or equal to 40 years, a sigmoid esophagus, reported chest pain by the patient, esophagus' diameter exceeding 61mm and an achalasia pattern I.

We therefore turn to the non-conventional achalasia treatment type cardioplasty.

Open Serra-Doria operation was chosen as a surgical treatment in our case after nasogastric aspiration for 3 days to decompress the esophagus.

This technique consists in Grondhal cardioplasty (latero–lateral [LL] esophagogastrostomy between the lower esophagus in the last 6 cm above the cardia and gastric fundus close to the Hiss angle) associated to partial gastrectomy with Roux-en -Y gastrojejunal reconstruction.

The evolution was marked by simple follow-ups and she was discharged after 5 days post-operative. A checkup 4 months after surgery showed a clear improvement in symptoms, a stable weight and the patient was satisfied as he could again eat solid food without thoracic pain or regurgitation.

One month post operative Barium X ray showed improvement on the esophageal emptying (Fig. 2). At the same checkup, biology shows the absence of hydro-electrolytic disorders, an Albuminemia at 31 g/L, a Protein level at 59 g/L and a hemoglobinemia at 13 g/dL. Last clinical and biological check-up at 14 months post-op was satisfactory.

3. Discussion

The foundational concept of "decompensation" was originally articulated by Olsen [2] in the context of observations made at the Mayo Clinic concerning cardiospasm. Even as the motor dysfunction persisted without alteration, a discernible shift was noted from clinical to radiological stages. Within this transition, the esophagus demonstrated a pronounced loss of its contractile capacity, leading inexorably to its progressive dilation. This augmented esophageal dimension was consequently characterized as being in a state of silence, decompensation, and paralysis.

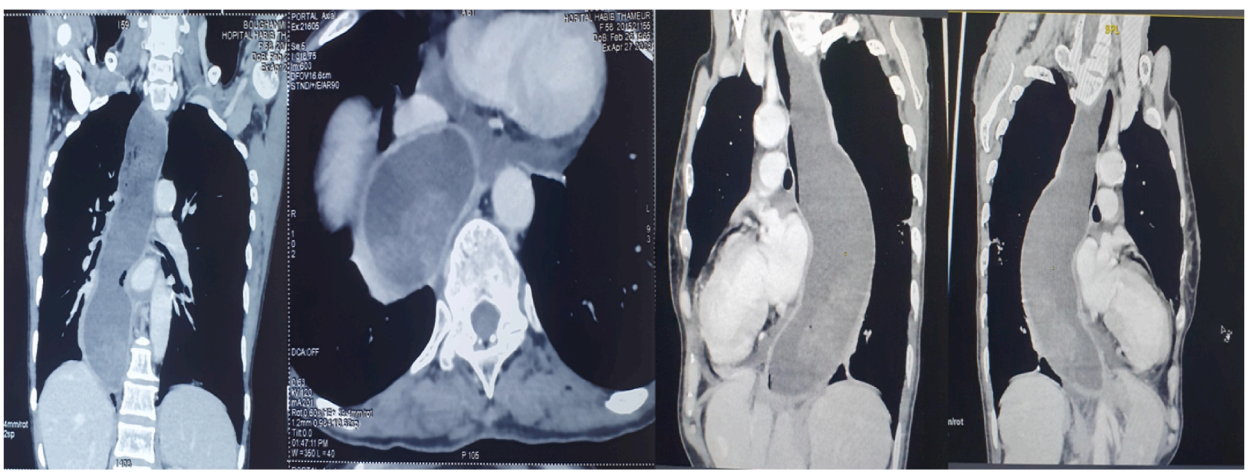

Fig. 1. Thoraco-abdominal CT with digestive opacification showed an esophageal dilatation with stasis measuring 61 mm at most, without any image of luminal stenosis or detectable extrinsic compression. Sigmoid shape esophagus.

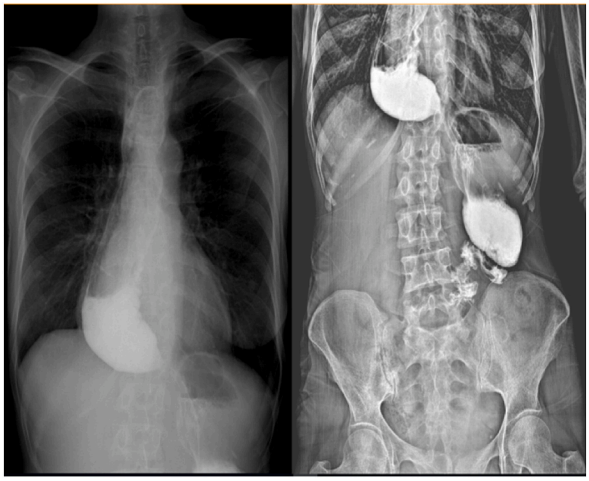

Fig. 2. Barium X ray showed improvement on the esophageal emptying.

In a distinct vein, Orringer [3] illuminated a unique perspective on this stage of esophageal achalasia. He delineated the tortuous megaesophagus as a mechanical impediment, drawing an analogy to a "sink-trap." Orringer [3] argued that merely incising the lower sphincter would not offer relief to the esophagus at this advanced disease stage.

Miller [4] echoed this sentiment, perceiving the extensively dilated esophagus as a severe manifestation of a condition that might necessitate surgical resection.

Building upon this foundation, Fekete [5] advanced the notion that the substantially enlarged and convoluted dolichomegaesophagus could be categorized as being in a condition termed "Esophageal Asystole." He emphasized the formidable challenges inherent in attempting to surgically rectify the pronounced angulation characteristic of such a sigmoid-like esophagus. Furthermore, Fekete [5] highlighted the elevated risk of suboptimal outcomes in patients subjected to the modified Heller myotomy.

Separately, Banbury [6] posited the definition of end-stage achalasia in a more encompassing manner. He described it as a state wherein debilitating dysphagia, regurgitation, and significant weight loss persisted despite aggressive therapeutic interventions directed at the primary disorder. Such a clinical presentation was frequently linked with pronounced esophageal dilatation and tortuosity.

In a contrasting viewpoint, Peters [7] discerned an end-stage esophagus in patients exhibiting a persistent or recurring pattern of dysphagia. This was particularly evident when combined with a dilated and convoluted esophagus, especially in cases where conventional treatments had proven to be inefficacious. Endoscopic assessments often unveiled signs such as retained food, a mucosal texture resembling "tree-bark," and/or a reflux stricture.

To encapsulate, an achalasic esophagus should be considered to have reached an "end stage" when substantial dilatation and retention manifest despite initial interventions. Moreover, the inability to achieve complete esophageal emptying, coupled with mucosal damage resulting from persistent reflux disease or even after multiple therapeutic interventions, should be integral to this

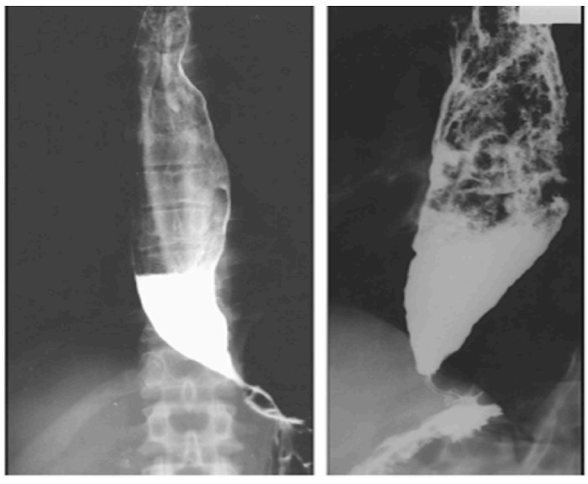

Fig. 3. Straight (St) type [7].

"end stage" definition.

Imaging serves as an indispensable tool in the comprehensive evaluation, diagnosis, and management of patients presenting with achalasia. The application of various imaging modalities offers crucial insights into the anatomical, functional, and potential pathological alterations associated with this disorder.

Barium Esophagography (Esophagogram) offers a morphological classification that facilitates the determination of more appropriate treatment strategies for individual cases. Over the years, this classification, further segmented into spindle, flask, and sigmoid types based on conventional descriptive criteria, has garnered widespread acceptance. However, it is noteworthy that there are no apparent pathophysiological distinctions between the spindle and flask types. Moreover, neither the medical practices in the United States nor those in Europe make a clear differentiation between these two classifications. Conversely, the extent of deviation and tortuosity observed in the upper esophagus in cases of the sigmoid type may significantly impact therapeutic outcomes. As a result, the current iteration of the descriptive criteria delineates achalasia into the following three distinct types [8]:

Straight (St) type (Fig. 3), characterized by a mild curvature of the esophageal longitudinal axis. This category encompasses the traditional spindle and flask types as previously described.

Sigmoid (Sg) type (Fig. 4), a notable curvature is observed along the longitudinal axis of the esophagus.

Advanced sigmoid (aSg) type (Fig. 5), a distinct rightward curvature of the esophagus, leading the organ to adopt an L-shaped trajectory.

When observing esophageal flexion(s), it is imperative to delineate a straight line aligning with the esophageal longitudinal axis.

In scenarios where two straight lines intersect, a singular angle is formed by their intersection. Conversely, the intersection of three straight lines yields two distinct angles.

The subsequent definitions are structured in ascending order based on the angle, α, resulting from the intersection of the straight lines (Fig. 6):

- For $\alpha \geq 135^{\circ}$, the condition is classified as the St type.
- For $90^{\circ} \leq \alpha < 135^{\circ}$, the condition is categorized as the Sg type.
- For $\alpha < 90^{\circ}$, the condition is designated as the aSg type.

Treatment of achalasia aims to reduce the pressure the pressure gradient across the lower esophageal sphincter and improve gravitational esophageal emptying in an effort to palliate symptoms and halt the natural history of the disease [9].

At present, pneumatic dilatation and Heller myotomy combined with an anti-refl ux procedure are the treatments of choice and have comparable success rates except for "end stage"achalasia: according to a paper published in the Lancet [10] positive prognostic factors after Heller myotomy are young age (<40 years), a Lower esophageal sphincter resting pressure greater than 30 mmHg and a straight esophagus with no tortuosities at its distal end as in sigmoid esophagus [10]. Heller myotomy patients with achalasia type II have the best outcome [10]. Wich is not the case for our patient.

Also, based on single center experience of thousand and One Laparoscopic Heller Myotomies for Esophageal Achalasia [10], the manometric pattern, presence of a sigmoid-shaped megaesophagus, chest pain, an age greater than or equal to 40 years, a sigmoid esophagus, an esophagus diameter exceeding 38mm and an achalasia pattern I predicted a poor outcome. These factors were independently associated with poor outcomes in both univariate and multivariate analysis. Wich is the case for ou patient.

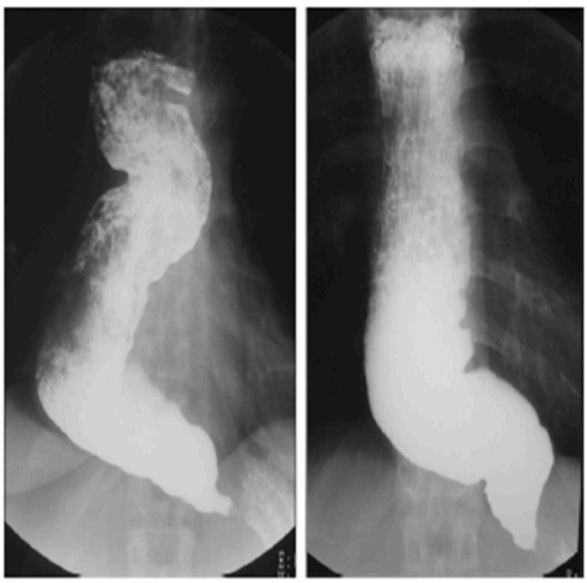

Fig. 4. Sigmoid (Sg) type [7].

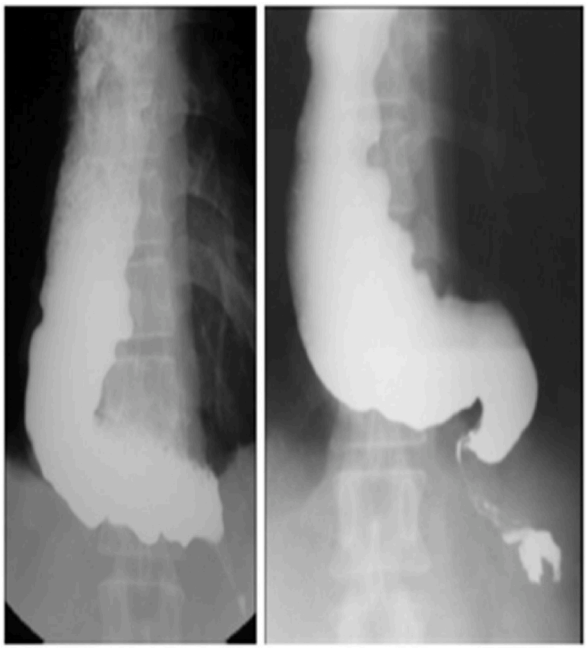

Fig. 5. Advanced sigmoid (aSg) type [7].

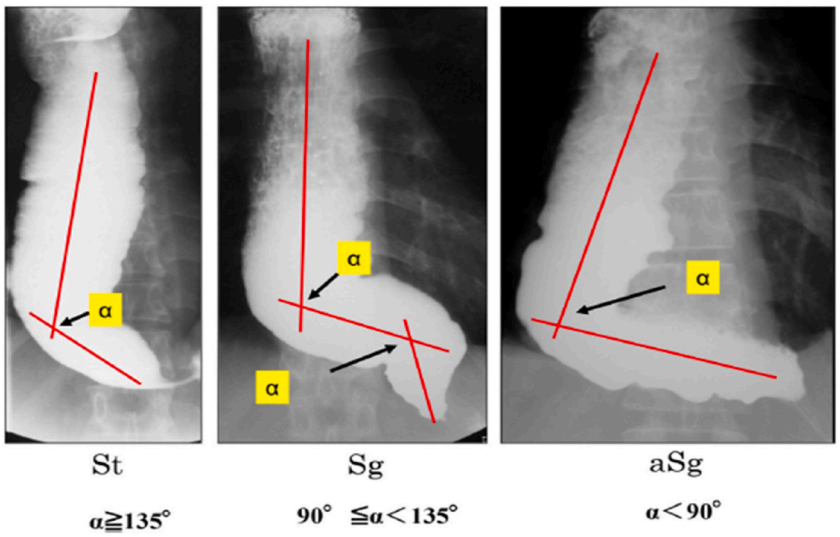

Fig. 6. Distinction among St, Sg and aSg types [7].

Concerning the oesophagectomy, it appears to be a procedure associated with high morbidity and mortality for a benign pathology.

For these patients we can propose "Non conventionnal achalsia treatments" as cardiectomy or cardioplasty [11]. Gastro-oesophageal junction resection or gastrooesophageal surgical modification aims to facilate esophageal emptying.

The Serra-Doria operation was proposed in 1970, This technique consists the following: annulling the LES obstacle through Grondhal's cardioplasty (esophagogastric LL anastomosis), preventing late stenosis; avoiding the formation of hydrochloric acid and blocking gastric stasis through subtotal gastrectomy (antrectomy) where the parietal cells producing hydrochloric acid are located; and finally to extirpate the bile, pancreatic, and duodenal refluxes through jejunojejunostomy by Roux-en-Y reconstruction [12]. This technique is still relevant in the age of robotic surgery [13]. In a study published by Aquino et al. [14], in which 32 patients underwent this technique, the resolution of symptoms in the medium and long term occurred in 81 % of the patients.

A study conducted at the Hospital das Clínicas of the Faculty of Medicine of the University of São Paulo, Brazil [15] involved twenty patients with achalasia who had previously undergone cardiomyotomie, aimed to evaluate the outcomes of reoperation using Serra-Dória procedure.

Authors concluded that inappropriate cardiomyotomy indications for severe megaesophagus or inadequate myotomy were primary reasons for early surgical procedure failure, evidenced by early dysphagia recurrence. The Serra-Dória procedure demonstrated immediate symptom improvement for severe reflux esophagitis in achalasia patients, without postoperative reflux evidence. Additionally, it showed a lower rate of postoperative complications compared to other methods, including the Heller procedure. The operation also improved regurgitation, particularly during late follow-ups, reduced stasis, and observed esophageal caliber regression in many patients.

4. Conclusion

The introduction of the Serra-Doria procedure represents a significant stride in treating end-stage achalasia, especially for recurrent cases after Heller's cardiomyotomy. By leveraging robotic technology for Grondhal's cardioplasty and partial gastrectomy with Roux-en-Y reconstruction, the procedure offers a minimally invasive solution. Patients have experienced considerable improvement in esophageal emptying and symptom relief, underscoring the Serra-Doria operation's efficacy as an innovative alternative specially at the presence of factors associated with heller procedure failure or a high risk operation patient. This advancement highlights the crucial role of personalized care and the ongoing research necessary to enhance outcomes for those suffering from this challenging condition.

Data availability statement

Data included in this study have not been deposited into a publicly available repository. In the case of science projects, data may be available on request Dr Haithem ZAAFOURI.

Ethics declarations

The participant gave written informed consent to the participation of this study and for the publication of their anonymised case details.

Guarantor statement

H-Z confirms full responsibility for the content of this manuscript.

Declaration of generative AI in scientific writing

Artificial intelligence has not been used in the writing process.

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Ethics committee approval

The study was approved by the Local Committee for Medical and Health Research Ethics in Habib Thameur Hospital.

CRediT authorship contribution statement

Haithem Zaafouri: Writing – review & editing, Writing – original draft, Project administration, Conceptualization. **Mona Cherif:** Methodology, Investigation, Data curation. **Nizar Khedhiri:** Visualization, Supervision, Formal analysis. **Meriam Sabbah:** Writing – original draft, Supervision. **Taha Sabri:** Writing – original draft, Visualization. **Anis Ben Maamer:** Validation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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