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Conservative surgical management of Boerhaave's syndrome: Experience of two tertiary referral centers

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ABSTRACT

Background: Surgery is generally proposed for Boerhaave's syndrome, spontaneous rupture of the esophagus. But diagnosis can be difficult, delaying appropriate management. The purpose of the present study was to evaluate outcome of conservative surgery for primary or T-tube repair performed in two tertiary referral centers.

Methods: From June 1985 to November 2010, among 53 patients presenting with Boerhaave's syndrome treated surgically, 39 underwent a conservative procedure. These patients were retrospectively divided into two groups by type of repair: primary suture (group 1, $n = 25$) or suture on a T-tube (group 2, $n = 14$). Patients in group 1 were further stratified into two subgroups depending on whether the primary suture was made with reinforcement (subgroup rS) or not (subgroup S).

Results: Length of stays in hospital and intensive care were shorter in patients in group 1 ($p = 0.037$), but after a shorter delay before therapeutic management ($p = 0.003$) compared with group 2. For the other variables studied, outcome was more favorable in group 1, but the differences were not significant. Comparing subgroups rS and S showed that the rate of persistent leakage was significantly lower after reinforced suture ($p = 0.021$).

Conclusions: These findings from the largest reported cohort of Boerhaave's syndrome patients undergoing conservative surgery showed that primary and T-tube repair provide at least equivalent results. Reinforced sutures appear to provide better outcomes by reducing postoperative leakage.

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1. Background

Spontaneous perforation of the esophagus occurs as a result of a sudden vomiting-induced increase in the internal esophageal pressure. Called Boerhaave's syndrome since the first description by Boerhaave in 1724,¹ spontaneous rupture of the esophagus accounts in only 10–35% of all perforations of the esophageal wall.^{2–4} Knowledge of this rare entity is of particular importance because of the high mortality, 20–40%,^{5–7} which is directly related to delay in diagnosis and therapeutic management, the main factors of poor outcome.^{5,8} Beyond 48 h, and for up to 5 days, mortality increases proportionally with therapeutic delay.^{4,9} Fatal outcome results from mediastinitis caused by chemical, enzymatic

and infectious processes.¹⁰ To date, studies in the literature have reported small series of Boerhaave's syndrome or grouped together different types of esophageal perforation. Analyses of such heterogeneous populations cannot provide the specific information required to establish appropriate strategies for Boerhaave's syndrome, explaining the lack of any real therapeutic consensus. The largest series published to date specifically devoted to Boerhaave's syndrome was reported by Griffin et al. in 2008 and included 51 patients.¹¹ Based on experience, most teams prefer a conservative surgical approach^{7,8,12,13} using primary repair as described in 1947 by Barrett¹⁴ or repair over a T-tube for drainage as proposed by Abbott et al.¹⁵

The purpose of this study was to report the surgical experience of two specialized tertiary referral centers with management of spontaneous rupture of the esophagus. Morbidity–mortality was the primary endpoint, comparing patients who underwent primary repair versus repair over a T-tube. Outcome after primary suture repair, with or without reinforcement, was the secondary endpoint.

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2. Methods

2.1. Patients

From June 1985 to November 2010, 53 patients were referred for treatment of Boerhaave's syndrome to two French centers (Rennes and Brest) specialized in esophageal surgery. Data were collected and analyzed retrospectively.

Diagnosis was established with contrast swallow, computed tomography scan or fibroscopy. Spontaneous perforation was defined as a full thickness tear of the esophageal wall not caused by an underlying disease or invasive procedure.¹⁶ Patients receiving conservative nonoperative care were excluded from this analysis (Fig. 1).

The followed data were collected: patient age and gender, time from symptom onset to therapeutic management, type of surgical management, length of hospital stay, length of stay in the intensive care unit, hospital mortality, persistent esophageal leakage, reoperation.

Hospital mortality was defined as death within 30 days post-surgery or during the initial hospital stay.

To analyze the impact of conservative surgical management on morbidity and mortality, patients were divided into two groups according to the type of procedure performed: primary repair suture (group 1); repair suture on a T-tube to control development of an esophago-pleuro-cutaneous fistula (group 2).

In the primary repair group, sutures were reinforced (subgroup rS) or not (subgroup S).

2.2. Surgery

Senior surgeons specialized in esophageal procedures performed all operations. A two-way approach was used in all cases: primary thoracotomy (side determined by the localization of the perforation) and laparotomy (for feeding jejunostomy).

All thoracic proceedings were performed by thoracotomy enabling debridement, decontamination and drainage of the mediastinal and pleural cavities. In the primary repair patients, the esophageal injury was sutured with or without reinforcement using a gastric patch or an absorbable mesh (group 1). In the other patients (group 2), the injury was repaired over a T-tube inserted through the perforation and drawn out to the skin at the end of the operation.

2.3. Outcomes

The main morbidity and mortality end points were compared between group 1 and group 2. The impact of reinforced sutures was a secondary end point.

2.4. Statistical analysis

Continuous variables were expressed as mean ± standard deviation (SD) or median for between-group comparisons with Student's *t*-test or Wilcoxon test. Discrete variables were expressed by number and percentage, and compared using the chi-squared test or Fisher's exact test, as appropriate. A *p* value of <0.05 was considered to be statistically significant.

3. Results

3.1. Demographic and operative data

During the study period, 53 patients were treated surgically for spontaneous perforation of the esophagus. Among these patients, 39 underwent conservative surgery and constituted the study cohort (Fig. 1). Mean age at diagnosis was 63.6 ± 13 years. There were 32 men (82.1%) and 7 women (17.9%) (sex ratio: 4.57).

Mean time from symptom onset to surgical management was 2 days (range 0–9 days). Mean length of hospital stay was 39 days (range 22–59 days), including 12 days (range 6–32 days) in the intensive care unit. Postoperative esophageal leakage developed in 14 patients (35.9%). Nine patients died (23.1%) and 16 (41%) required a revision procedure. The inaugural signs of esophageal perforation are summarized in Fig. 2. Pain in the lower thorax or epigastric region was the main sign (71.8%), followed by vomiting (43.6%). The classical triad described by Mackler¹⁷ was noted in two patients (5.1%).

3.2. Comparison between the group 1 and 2

Outcomes observed in groups 1 and 2 are presented in Table 1. The two groups were comparable for age and gender, but time to therapeutic management was significantly longer in group 2 (*p* = 0.003). In addition, more perforations were on the right side in group 2 (*p* = 0.047).

The duration of intensive care was significantly shorter in patients treated with primary suture repair (*p* = 0.037). There was no significant difference between the two groups for the other variables studied. Group 1 displayed trends for longer overall hospital stay (*p* = 0.39), higher mortality (*p* = 0.23), persistent leakage (*p* = 0.30) and revision surgery (*p* = 0.60).

3.3. Reinforced versus direct repair

The comparison between subgroups rS and S is presented in Table 2. The esophagus was repaired with a primary suture in 25

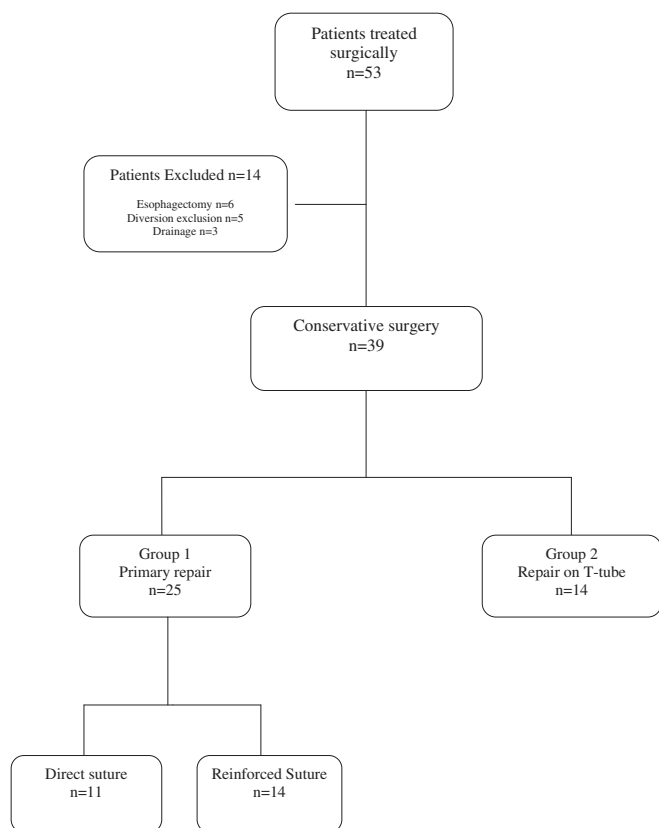


Fig. 1. Diagram chart.

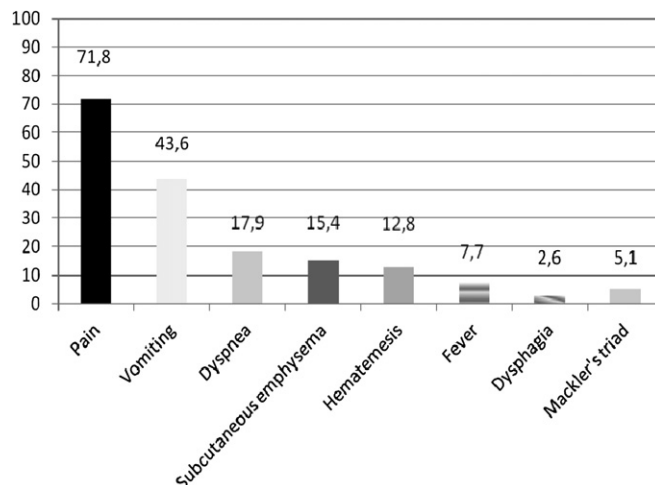


Fig. 2. Clinical signs leading to diagnosis.

Table 1
Patient characteristics and postoperative complications by study group.

Variable	Total n = 39	Group 1 primary repair n = 25(%)	Group 2 T-tube repair n = 14(%)	p
Gender				
Male	32(82.1)	20(80)	12(85.7)	1
Female	7(17.9)	5(20)	2(14.3)	
Age	63.6 ± 13	65.8 ± 14	59.7 ± 10	0.17
Localization of perforation				
Left	30(76.9)	22(88)	8(57.1)	0.047
Right	9(23.1)	3(12)	6(42.9)	
Time to treatment (days)	2[1; 2.75] (min:0 max:9)	1[0; 2] (min:0 max:6)	3.5[2; 8] (min:0 max:9)	0.003
Stay in intensive care (days)	12[6; 32]	8[4; 23]	19[9; 52]	0.04
Stay in hospital (days)	39[22; 59]	31[25; 52]	45[21; 74]	0.39
Death	9(23.1)	4(16)	5(35.7)	0.24
Revision surgery	16(41)	9(36)	7(50)	0.61
Persistent leakage	14(35.9)	7(28)	7(50)	0.30

patients (group 1), 14 with reinforcement (subgroup rS) and 11 without (subgroup S). The two subgroups were comparable for age, sex, time to therapeutic management and site of esophageal perforation. The rate of persistent postoperative leakage was statistically better in the rS subgroup (7.1% versus 54.5% in the S subgroup, $p = 0.021$). For the other variables studied (length of stay in hospital and in intensive care, mortality, reoperation), outcome was in favor of the rS subgroup but the differences did not reach statistical significance.

4. Discussion

Emergency therapeutic management is crucial after spontaneous perforation of the esophagus in order to prevent fatal outcome subsequent to infectious complications. Unfortunately, late diagnosis is common because of the non-specific clinical presentation, delaying referral to a specialized tertiary center. To our knowledge, the series reported here is the largest cohort of patients with Boerhaave's syndrome undergoing conservative surgery.

For conservative surgical treatment, the best outcome was obtained with primary suture repair. Compared with repair over

Table 2
Patient characteristics and postoperative complications by type of suture: direct versus reinforced.

Variable	Direct suture n = 11(%)	Reinforced suture n = 14(%)	p
Gender			
Male	9(81.8)	11(78.6)	1
Female	2(18.2)	3	
Age	65.6 ± 13	65.9 ± 16	0.97
Localization of perforation			
Left	9(81.8)	13(92.9)	0.57
Right	2(18.2)	1(7.1)	
Time to treatment (days)	1[0; 2] (min:0 max:3)	1[0; 2] (min:0 max:6)	0.96
Stay in intensive care (days)	7[3.8; 46] mean:25	9.5[4; 15] mean:12	0.68
Stay in hospital (days)	28[20.3; 62.5] mean:51	34[30; 41] mean:35	0.98
Death	3(27.3)	1(7.1%)	0.29
Revision surgery	6(54.5)	3(21.4)	0.12
Persistent leakage	6(54.5)	1(7.1%)	0.02

a T-tube to control fistula formation, primary repair was followed by a significantly shorter stay in the intensive care unit and a non-significantly shorter overall hospital stay. Lower rates of persistent leakage, revision surgery and mortality were also observed although the differences did not reach statistical significance. It must be noted however that the time from symptom onset to surgery was shorter in the primary suture patients (group 1) than in the other patients (group 2). This difference can be explained in part by the time frame of the present study. Up to the mid 1990s primary suture was only performed if the patient was seen within 24 h of the perforation. This attitude, generally advocated in the literature at that time,^{12,18,19} was later changed when new series showed that good outcome could be obtained with suturing even after 24 h.^{20–22} The data collected in the present series demonstrate the importance of primary suture as the first-intention strategy since outcomes were as least as good, and possibly superior, to those obtained with a T-tube to control fistula formation.

Controlled fistula formation can however be an attractive alternative for perforations involving substantial tissue loss compromising direct suture. In such extreme situations, the T-tube can enable outcomes similar to those obtained with primary repair, as was demonstrated earlier by Linden et al.²³ These conservative surgical options should always be attempted as the first intention treatment, since mortality can be controlled (23.1% in groups 1 + 2 and 20–40% in the literature).^{5–7}

An improvement in mortality and persistent leakage might be expected with a reinforced suture using a gastric patch²⁴ or an absorbable polyglactin910 mesh and fibrin glue.^{25,26} In our series, the rate of persistent leakage was significantly better after primary suture with reinforcement (7.1%) than without reinforcement (54.5%) ($p = 0.021$). Similarly, although the difference did not reach significance, mortality was lower with reinforced sutures (7.1% versus 27.3%). These differences can probably be explained by the frailty of sutures in a septic environment.

The challenge with Boerhaave's syndrome is to establish the diagnosis early enough to institute adequate treatment. The data in this series again recall that symptoms of spontaneous rupture of the esophagus are non-specific. The most common complaint was pain, generally in the thorax and/or the epigastric region, observed in 71.8% of patients. Vomiting occurred in 43.6%. Unfortunately patients with these symptoms are most often referred to an emergency cardiology unit, with the resulting delay in appropriate care. The classical triad described by Mackler,¹⁷ specific of Boerhaave's syndrome, was only found in 5.1% of patients. Very early diagnosis facilitates conservative treatment, at least initially, including the purely medical management as proposed by Michel et al.⁹ Another option is to insert a self-expanding stent,^{27,28} but with the risk of migration since there is no pre-existing stenosis. Freeman et al. recently reported an occlusion rate of 89% at 48 h in a series of 19 patients treated with stents for spontaneous perforations.²⁹ The limited data in the literature, the operator-dependent nature of the insertion process, and the requirement for a pleural decontamination limits the use of this option which should never retard surgery. Furthermore, as previously described by Scott et al., the thoracoscopic approach might allow more conservative proceeding.³⁰

Although we did not have the experience of this technique, it probably could improve the postoperative pain relief and ventilation.

5. Conclusion

The challenge with Boerhaave's syndrome is to establish diagnosis early enough for adequate management. Surgery may be the only option after late diagnosis. First-line treatment should be

conservative. Our findings show that outcome is approximately equivalent after primary or T-tube repair, irrespective of the delay before treatment. Best results are obtained with reinforced sutures. Despite the rarity of Boerhaave's syndrome, a prospective study should be conducted to compare primary repair versus T-tube repair.

Ethical approval

This study was undertaken after approval by our local institutional review board.

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Authors Contribution

LS conceived and designed study, undertook the analysis and interpretation of data, and wrote and finalized the manuscript. SD undertook the analysis and interpretation of data; MR provided the statistical analysis. BB analyzed and interpreted the data, and revised the manuscript. KB proof-read and revised the manuscript. JPB revised the manuscript; BM revised the manuscript and gave final approval.

Conflict of interest

No conflict of interest to declare.

References

- Derbes VJ, Mitchell Jr RE. Hermann Boerhaave's Atrocis, nec descripti prius, morbi historia, the first translation of the classic case report of rupture of the esophagus, with annotations. *Bull Med Libr Assoc* 1955;**43**:217–40.
- Brinster CJ, Singhal S, Lee L, Marshall MB, Kaiser LR, Kucharzuk JC. Evolving options in the management of esophageal perforation. *Ann Thorac Surg* 2004;**77**:1475–83.
- Abbas G, Schuchert MJ, Pettiford BL, Pennathur A, Landreneau J, Landreneau J, et al. Contemporaneous management of esophageal perforation. *Surgery* 2009;**146**:749–55.
- Vallbohmer D, Holscher AH, Holscher M, Bludau M, Gutschow C, Stippel D, et al. Options in the management of esophageal perforation: analysis over a 12-year period. *Dis Esophagus* 2010;**23**:185–90.
- Malledant Y, Tanguy M, Artus M, Cardin JL, Campion JP, Launois B, et al. Spontaneous rupture of the esophagus. *Ann Fr Anesth Reanim* 1986;**5**:128–33.
- Richardson JD. Management of esophageal perforations: the value of aggressive surgical treatment. *Am J Surg* 2005;**190**:161–5.
- Chirica M, Champault A, Dray X, Sulpice L, Munoz-Bongrand N, Sarfati E, et al. Esophageal perforations. *J Visc Surg* 2010;**147**:e117–28.
- Sung SW, Park JJ, Kim YT, Kim JH. Surgery in thoracic esophageal perforation: primary repair is feasible. *Dis Esophagus* 2002;**15**:204–9.
- Michel L, Grillo HC, Malt RA. Operative and nonoperative management of esophageal perforations. *Ann Surg* 1981;**194**:57–63.
- Saario I, Kostianen S, Salo J, Meurala H, Eerola S. Treatment of spontaneous rupture of the esophagus. *Acta Chir Scand* 1983;**149**:771–4.
- Griffin SM, Lamb PJ, Shenfine J, Richardson DL, Karat D, Hayes N. Spontaneous rupture of the oesophagus. *Br J Surg* 2008;**95**:1115–20.
- Hill AG, Tiu AT, Martin IG. Boerhaave's syndrome: 10 years experience and review of the literature. *ANZ J Surg* 2003;**73**:1008–10.
- Kollmar O, Lindemann W, Richter S, Steffen I, Pistorius G, Schilling MK. Boerhaave's syndrome: primary repair vs. esophageal resection – case reports and meta-analysis of the literature. *J Gastrointest Surg* 2003;**7**:726–34.
- Barrett NR. Report of a case of spontaneous perforation of the oesophagus successfully treated by operation. *Br J Surg* 1947;**35**:216–8.
- Abbott OA, Mansour KA, Logan Jr WD, Hatcher Jr CR, Symbas PN. Atraumatic so-called "spontaneous" rupture of the esophagus. A review of 47 personal cases with comments on a new method of surgical therapy. *J Thorac Cardiovasc Surg* 1970;**59**:67–83.
- Walker WS, Cameron EW, Walbaum PR. Diagnosis and management of spontaneous transmural rupture of the oesophagus (Boerhaave's syndrome). *Br J Surg* 1985;**72**:204–7.
- Mackler SA. Spontaneous rupture of the esophagus; an experimental and clinical study. *Surg Gynecol Obstet* 1952;**95**:345–56.
- Nesbitt JC, Sawyers JL. Surgical management of esophageal perforation. *Am Surg* 1987;**53**:183–91.
- Lawrence DR, Ohri SK, Moxon RE, Townsend ER, Fountain SW. Primary esophageal repair for Boerhaave's syndrome. *Ann Thorac Surg* 1999;**67**:818–20.
- Jougon J, Mc BT, Delcambre F, Minniti A, Velly JF. Primary esophageal repair for Boerhaave's syndrome whatever the free interval between perforation and treatment. *Eur J Cardiothorac Surg* 2004;**25**:475–9.
- Wang N, Razzouk AJ, Safavi A, Gan K, Van Arsdell GS, Burton PM, et al. Delayed primary repair of intrathoracic esophageal perforation: is it safe? *J Thorac Cardiovasc Surg* 1996;**111**:114–21.
- Zumbro GL, Anstadt MP, Mawulawde K, Bhimji S, Paliotta MA, Pai G. Surgical management of esophageal perforation: role of esophageal conservation in delayed perforation. *Am Surg* 2002;**68**:36–40.
- Linden PA, Bueno R, Mentzer SJ, Zellos L, Lebenthal A, Colson YL, et al. Modified T-tube repair of delayed esophageal perforation results in a low mortality rate similar to that seen with acute perforations. *Ann Thorac Surg* 2007;**83**:1129–33.
- Sannohe Y, Tanaka H, Inutsuka S, Yamashita Y, Futagami H. Onlay fundic patch method applied in spontaneous rupture of the esophagus: a case report. *Jpn J Surg* 1980;**10**:55–8.
- Bardaxoglou E, Campion JP, Landen S, Manganas D, Siriser F, Chareton B, et al. Oesophageal perforation: primary suture repair reinforced with absorbable mesh and fibrin glue. *Br J Surg* 1994;**81**:399.
- Bardaxoglou E, Manganas D, Meunier B, Landen S, Maddern GJ, Campion JP, et al. New approach to surgical management of early esophageal thoracic perforation: primary suture repair reinforced with absorbable mesh and fibrin glue. *World J Surg* 1997;**21**:618–21.
- Johnsson E, Lundell L, Liedman B. Sealing of esophageal perforation or ruptures with expandable metallic stents: a prospective controlled study on treatment efficacy and limitations. *Dis Esophagus* 2005;**18**:262–6.
- Fischer A, Thomsch O, Benz S, von DE, Baier P, Hopt UT. Nonoperative treatment of 15 benign esophageal perforations with self-expandable covered metal stents. *Ann Thorac Surg* 2006;**81**:467–72.
- Freeman RK, Van Woerkom JM, Vyverberg A, Ascoti AJ. Esophageal stent placement for the treatment of spontaneous esophageal perforations. *Ann Thorac Surg* 2009;**88**:194–8.
- Scott HJ, Rosin RD. Thoracoscopic repair of a transmural rupture of the oesophagus (Boerhaave syndrome). *J R Soc Med* 1995;**88**:414–5.