Surgical Management of Spontaneous Esophageal Rupture: An Evaluation

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Abstract

Spontaneous esophageal rupture is rare. The distal esophagus ruptures upon a sudden vomiting-induced increase in the internal esophageal pressure. We evaluated surgical management of spontaneous esophageal rupture by examining clinical details and outcomes of 10 patients treated for the disorder at our hospital between 1987 and 2014.

Mean age of the patients was 58.1 years (range, 41–75 years). The sex ratio (M/F) was 9/1. Diagnosis was achieved by chest computed tomography (CT) or chest CT and esophagography. The rupture occurred in the left lower thoracic esophagus (n=8), left middle thoracic esophagus (n=1), or right lower thoracic esophagus (n=1). Mean time from symptom onset to surgery was 36.2 hours (range, 5–96 hours).

Patients were divided between those treated within 24 hours of onset (early treatment group, n=5) and those treated 24 hours or more after onset (late treatment group, n=5). Treatment comprised primary repair in 7 patients and esophagectomy in 3. Postoperative complications occurred in 6 patients: surgical site infection (n=4), anastomosis leakage (n=3), intrapleural abscess (n=1), sepsis (n=1), and pneumonia (n=1). Mean postoperative hospital stay was 78.5 days (range, 22–228 days). There was no in-hospital mortality.

Postoperative complications were somewhat more frequent in the late treatment group (n=4, 80%) than in the early treatment group (n=3, 60%), and postoperative hospital stay was longer in this group (114.2 vs. 42.8 days). Outcomes in terms of postoperative complications and hospital stay were good in the early treatment group. In the late treatment group, severe intrathoracic contamination in 3 patients necessitated life-saving esophagectomy. When primary repair was performed, the incidence of anastomotic leakage was lowest when both layers were sutured, perforation sites were reinforced, and nutritional support was provided.

Early diagnosis and treatment are essential for spontaneous esophageal rupture, and we believe that selecting the appropriate surgical technique increases survival.

Key words

spontaneous esophageal rupture, surgical management

Introduction

Spontaneous esophageal rupture occurs only rarely. It generally occurs as a rupture of the distal esophagus due to a sudden vomiting-induced increase in the internal esophageal pressure. Delayed diagnosis is associated with substantial morbidity and can be life-threatening. Therefore, early diagnosis and treatment are important. Prompt surgical repair with drainage is the standard treatment¹–³. Conservative therapy can be applied only when symptoms are mild and mediastinitis is localized⁴. We evaluated the sur-
gical management of spontaneous esophageal rupture by examining clinical details and outcomes in patients we have treated.

**Patients and Methods**

Our study group comprised 10 patients treated surgically for spontaneous esophageal rupture. All were treated at our hospital between 1987 and 2014. Age and sex of patients, symptoms, cause of rupture, location, time from symptom onset to surgery, operative procedures, postoperative complications, hospital stay, and outcomes were examined.

We divided patients into an early treatment group (n=5), those who were treated surgically at less than 24 hours after symptom onset, and a late treatment group (n=5), those who were treated surgically at 24 hours or more after symptom onset. In addition, we divided the patients who underwent surgical perforation site repair into 2 groups: those in whom anastomotic leakage developed and those in whom no anastomotic leakage developed, and we compared the 2 groups to identify risk factors for anastomotic leakage.

This study was approved by the Ethics Committee at St. Marianna University School of Medicine (Approval No 3149).

**Statistical analyses**

Values are shown as mean±SD and/or range. Between-group differences in categorical variables were analyzed by Fischer’s exact test, and between-group differences in continuous variables were analyzed by Student’s t-test. A p value of <0.050 was considered statistically significant.

**Results**

The 10 patients who underwent surgical treatment of spontaneous esophageal rupture and details of their cases are shown in Table 1, and the case details are summarized in Table 2. Patients’ mean age was 58.1 years (range, 41–75 years), and the sex ratio (M/F) was 9/1. Eight of the 10 patients presented with chest and back pain, and the remaining 2 patients presented with upper abdominal pain. These symptoms occurred after vomiting in 8 patients, and the vomiting followed alcohol consumption in 4 of these 8 patients. Chest diagnostic computed tomography (CT) was performed for all patients, and esophagography was performed for 8. The rupture occurred in the left lower thoracic esophagus in 8 patients, the left middle thoracic esophagus in 1 patient, and the right lower thoracic esophagus in 1 patient. Chest CT showed that gastrointestinal contents had perforated into the intrathoracic cavity in 9 patients, whereas the contents were localized to the mediastinum in 1 patient.

Mean time from symptom onset to surgery was 36.2 hours (range, 5–96 hours). Five patients were treated at 24 hours or less and were thus considered the early treatment group, and 5 patients were treated

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**Table 1. The 10 Study Patients with Spontaneous Esophageal Rupture, Case Characteristics, and Surgical Details**

<table>
<thead>
<tr>
<th>Age (years)/Sex</th>
<th>Chief complaint</th>
<th>Precipitating event</th>
<th>Diagnostic method</th>
<th>Time from symptom onset to surgery (hours)</th>
<th>Operative procedure</th>
<th>Location of the rupture</th>
<th>Length of rupture (mm)</th>
<th>Postoperative complications(s)</th>
<th>Postoperative hospital stay (days)</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>52F</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>96</td>
<td>Esophagectomy, late reconstruction</td>
<td>Lower right thoracic esophagus</td>
<td>70</td>
<td>SSI</td>
<td>138</td>
<td>Alive</td>
</tr>
<tr>
<td>51M</td>
<td>Upperabdominal pain</td>
<td>Known</td>
<td>CT</td>
<td>48</td>
<td>Esophagectomy, late reconstruction</td>
<td>Lower left thoracic esophagus</td>
<td>45</td>
<td>Intraperitoneal abscess</td>
<td>228</td>
<td>Alive</td>
</tr>
<tr>
<td>41M</td>
<td>Upperabdominal pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>48</td>
<td>Esophagectomy, immediate reconstruction</td>
<td>Lower left thoracic esophagus</td>
<td>70</td>
<td>Leakage, SSI</td>
<td>128</td>
<td>Alive</td>
</tr>
<tr>
<td>52M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>72</td>
<td>Primary repair with reinforcement</td>
<td>Lower left thoracic esophagus</td>
<td>15</td>
<td>Pneumonia</td>
<td>37</td>
<td>Alive</td>
</tr>
<tr>
<td>60M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>14</td>
<td>Primary repair</td>
<td>Lower left thoracic esophagus</td>
<td>20</td>
<td>Sepsis</td>
<td>61</td>
<td>Alive</td>
</tr>
<tr>
<td>48M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT</td>
<td>16</td>
<td>Primary repair</td>
<td>Lower left thoracic esophagus</td>
<td>15</td>
<td>Leakage</td>
<td>63</td>
<td>Alive</td>
</tr>
<tr>
<td>53M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>8</td>
<td>Primary repair</td>
<td>Lower left thoracic esophagus</td>
<td>20</td>
<td>None</td>
<td>31</td>
<td>Alive</td>
</tr>
<tr>
<td>83M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>7</td>
<td>Primary repair with reinforcement</td>
<td>Lower left thoracic esophagus</td>
<td>30</td>
<td>SSI</td>
<td>37</td>
<td>Alive</td>
</tr>
<tr>
<td>66M</td>
<td>Chest and back pain</td>
<td>Vomiting</td>
<td>CT, esophagography</td>
<td>5</td>
<td>Primary repair with reinforcement</td>
<td>Lower left thoracic esophagus</td>
<td>70</td>
<td>None</td>
<td>22</td>
<td>Alive</td>
</tr>
<tr>
<td>75M</td>
<td>Chest and back pain</td>
<td>Unknown</td>
<td>CT, esophagography</td>
<td>48</td>
<td>Primary repair with reinforcement</td>
<td>Middle left thoracic esophagus</td>
<td>50</td>
<td>Leakage, SSI</td>
<td>40</td>
<td>Alive</td>
</tr>
</tbody>
</table>

CT, computed tomography; SSI, surgical site infection
Table 2. Summary of Clinical Variables in the Total Patient Group (n=10)

<table>
<thead>
<tr>
<th>Variable</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>58.1±12.98 (range, 41–75)</td>
</tr>
<tr>
<td>Sex</td>
<td>Male n=9, Female n=1</td>
</tr>
<tr>
<td>Chief complaint</td>
<td>Chest and back pain n=8, Upper abdominal pain n=2</td>
</tr>
<tr>
<td>Precipitating event</td>
<td>Vomiting n=8, Unknown n=2</td>
</tr>
<tr>
<td>Diagnostic method(s)</td>
<td>CT and esophagography n=8, CT n=2</td>
</tr>
<tr>
<td>Location of the rupture</td>
<td>Lower left thoracic esophagus n=8, Middle left thoracic esophagus n=1, Lower right thoracic esophagus n=1</td>
</tr>
<tr>
<td>Time from symptom onset to surgery (hours)</td>
<td>36.2±31.26 (range, 5–96)</td>
</tr>
</tbody>
</table>

Mean±SD values or number (n) of patients are shown.

Table 3. Operative Procedures and Outcomes in the Total Patient Group (n=10)

<table>
<thead>
<tr>
<th>Procedure</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Primary repair</td>
<td>n=3</td>
</tr>
<tr>
<td>Primary repair with reinforcement</td>
<td>n=4</td>
</tr>
<tr>
<td>Esophagectomy, immediate reconstruction</td>
<td>n=1</td>
</tr>
<tr>
<td>Esophagectomy, late reconstruction</td>
<td>n=2</td>
</tr>
<tr>
<td>Operation time (minutes)</td>
<td>294.7±74.50 (range, 195–390)</td>
</tr>
<tr>
<td>Blood loss volume (mL)</td>
<td>504.2±426.48 (range, 5–1360)</td>
</tr>
<tr>
<td>Postoperative complication</td>
<td>Surgical site infection n=4, Abscess n=1, Sepsis n=1, Pneumonia n=1</td>
</tr>
<tr>
<td>Postoperative hospital stay (days)</td>
<td>78.5±66.05 (range, 22–228)</td>
</tr>
<tr>
<td>Mortality (%)</td>
<td>0</td>
</tr>
</tbody>
</table>

Mean±SD values or number (n) of patients are shown.

beyond 24 hours and were thus considered the late treatment group. Treatment consisted of primary perforation site repair in 7 patients and esophagectomy in 3. A left-sided thoracotomy approach was used in 9 patients, and a right-sided approach was used in 1 patient. Of the 7 primary repairs, 3 were reinforced with stomach fundus, and 1 was reinforced with omentum. Of the 3 patients who underwent esophagectomy, early reconstruction was performed in 1, and late reconstruction was performed in the other 2. Gastrostomy or jejunostomy was performed in 7 patients to facilitate postoperative nutritional support.

Mean operation time was 294.7 minutes (range, 195–390 minutes), and mean blood loss volume was 504.2 mL (range, 5–1360 mL). Mean length of the rupture was 41 mm (range, 15–70 mm). Postoperative complications occurred in 6 patients: surgical site infection (n=4) anastomotic leakage (n=3), intrapleural abscess (n=1), sepsis (n=1), and pneumonia (n=1), with more than 1 complication occurring in some of these patients. All complications resolved with conservative treatment.

Operative details and outcomes are shown for the total patient group in Table 3. Mean postoperative hospital stay was 78.5 days (range, 22–228 days). There was no in-hospital mortality.

Operative details and outcomes are shown for the early and late treatment groups in Table 4. Primary repair was performed in all patients in the early treatment group. In the late treatment group, primary repair was performed in 2 patients, and esophagectomy was performed in 3 patients. Postoperative complications and length of postoperative hospital stay did not differ significantly between the early treatment group and late treatment group. However, postoperative complications were somewhat more frequent in the late treatment group (4 patients, 80%) than in the early treatment group (3 patients, 60%). Postoperative hospital stay also tended to be longer in the late treatment group (114.2 days) than in the early treatment group (42.8 days).

Clinical variables are shown for the patients who underwent primary repair in Table 5. Anastomotic leakage developed in 2 of these 7 patients. No statis-
tically significant differences were observed in the time from symptom onset to surgery, length of the perforations, the use of reinforcement, the creation of nutritional access, or the suturing method used to repair the perforation (1-layer vs. 2-layer suturing). However, the time from symptom onset to surgery tended to be longer for those patients in whom leakage developed (32 hours) than for those in whom leakage did not develop (21.1 hours).

Among the 7 patients who underwent primary repair, leakage developed in 2, and we had reinforced the perforation site in 1 of these 2 patients. Leakage did not develop in the other 5 patients, and we had reinforced the perforation site in 3 of these 5 patients. Thus, the incidence of anastomotic leakage was lower among those patients who underwent perforation site reinforcement.

Nutritional access had been created in 1 of the 2 patients in whom leakage developed and in 4 of the 5 patients in whom leakage did not develop, and thus incidence of anastomotic leakage was lower among patients in whom we created nutritional access.

We performed 1-layer suturing for repair of the perforation site in the 2 patients in whom leakage developed and in 2 of the 5 patients in whom leakage did not develop. Leakage developed in 2 of the 5 patients in whom we performed 2-layer suturing and did not develop in the 3 remaining patients in whom we performed 2-layer suturing. Thus, the incidence of anastomotic leakage was lower among patients in whom double-layer suturing was performed.

There were no postoperative complications in the 3 patients who underwent primary repair, reinforcement with stomach fundus, and jejunostomy.
Case presentation

A 66-year-old man came to our hospital complaining of severe back pain that developed after he had vomited. CT showed left pneumothorax and left pleural effusion, with infiltrative shadows in the left lower lobe. Mediastinal emphysema was noted surrounding the lower thoracic esophagus (Fig. 1). Esophagography showed extravasation of contrast from the lower thoracic esophagus into the thoracic cavity (Fig. 2). Surgery was performed under general anesthesia with the patient in the right lateral decubitus position, and thoracotomy was initiated by opening the left 7th intercostal space. Approximately 250 cc of food residue and turbid fluid had accumulated within the thoracic cavity, resulting in a high degree of contamination. After irrigating the thoracic cavity with physiological saline solution, we confirmed an approximately 7-cm-long perforation on the left side of the lower thoracic esophagus (Fig. 3). The esophagus was taped, and the perforation site was closed by 2-layer suturing (Fig. 4). The perforation site was then covered by gastric fundus (Fig. 5). A drain was inserted into the thoracic cavity, and the chest was closed. We moved the patient to the supine position, created an enterostomy via a mini-laparotomy, and concluded the surgery. The surgery lasted 245 minutes, and the blood loss volume was 5 mL. Postoperatively, the patient was placed on mechanical ventilation in the ICU, but, the ventilator was withdrawn on postoperative day 1. Oral intake was begun on postoperative day 7, and the patient was discharged on postoperative day 22.

Discussion

Spontaneous esophageal rupture was first reported in 1724 by the Dutch physician Hermann Boerhaave and was thus known for quite some time as Boerhaave’s syndrome. We now know spontaneous esophageal rupture as a rare condition, with an incidence of 7.4 per 10 million per year. Rupture through all layers of the esophageal wall occurs due to a sudden increase in esophageal pressure following an episode of vomiting, and the mediastinum and thoracic cavity are contaminated by gastric contents. For this reason, if early diagnosis and treatment are not accomplished, life-threatening complications, such as sepsis resulting from mediastinitis or empyema, may develop, and these must be treated on an emergency basis.

Spontaneous esophageal rupture usually occurs after an episode of vomiting induced by alcohol consumption; therefore, patients tend to be male and aged 40–60 years. In the present study, 9 of the 10 patients (90%) were male, and their mean age was 58.1 years. The mechanism is thought to involve an excessive increase in esophageal pressure that, in turn, causes loss of coordinated movement of the cricopharyngeal part of the inferior constrictor muscle and the lower esophageal sphincter during vomiting. However, a definite cause has not been established.
The site of rupture in approximately 90% of cases is the left lateral wall of the lower esophagus. Rupture occurred in the left lower esophagus in 8 of our 10 patients (80%). The anatomical reasons for rupture at this site include (1) thinness of the muscle layers in the lower esophagus, (2) weaknesses in the circular muscle due to entry of the nerves and arteries from the outside, and (3) lack of surrounding supporting organs.

Initial symptoms after spontaneous esophageal rupture include severe epigastric pain and back pain as well as labored breathing. Thereafter, mediastinitis and pyothorax may arise due to persistent infection. However, early diagnosis is sometimes difficult due to the non-specific symptoms, and the condition is sometimes misdiagnosed as arterial dissection, myocardial infarction, pulmonary infarction, or aspiration pneumonia. The rate of correct diagnosis at the time of initial examination is reported to be low, at 30–50%. The rupture was misdiagnosed initially in 4 of our 10 patients as pneumonia (1 patient), gastric ulcer (1 patient), cholecystitis (1 patient), and epigastric pain (1 patient). These patients were all seen from 2000 onwards, and all 4 patients were admitted to other hospitals and then transferred to our hospital on suspicion of esophageal rupture.

Imaging modalities that are useful for diagnosis of spontaneous esophageal rupture and for determining the subsequent treatment plan include chest X-ray, chest CT, and esophagography. Mediastinal emphysema, enlarged mediastinal shadows, pleural effusion, pneumothorax, and subcutaneous emphysema are observed on chest X-ray. Chest CT will reveal mediastinal emphysema, pleural effusion, and subcutaneous emphysema. In recent years, diagnostic accuracy has increased due to advances in multidetector computed tomography (MDCT), which can detect small amounts of mediastinal emphysema. However, reports indicate that approximately 10% of cases of spontaneous esophageal rupture show no specific CT features, such as mediastinal emphysema or empyema, and that the perforation site and size can be evaluated by performing esophagography with a water-soluble contrast agent, making it a useful study for a definitive diagnosis. We performed CT in all 10 of our patients and esophagography in 8, and a definitive diagnosis was reached in all cases. There are endoscopic procedures that can be used for diagnosis, but with these procedures comes a risk of introducing free air into the mediastinum due to mechanical ventilation.

In 1982, Goldstein and Thompson reviewed the cases of 868 patients with spontaneous esophageal rupture and reported a mortality rate of approximately 30%; all patients who were treated initially...
more than 24 hours after symptom onset died. In 1987, Nebitt et al\textsuperscript{14} reported mortality rates of 0%, 29%, and 40% when treatment was started within 24 hours, between 24 and 48 hours, and beyond 48 hours after symptom onset, respectively. Brinster et al\textsuperscript{2} reported 726 patients with esophageal rupture and that the mortality rate doubled for patients treated beyond 24 hours vs. those treated within 24 hours (27% vs. 14%, respectively). Previous reports give an important indication that introducing treatment within 24 hours of onset decreases the mortality rate\textsuperscript{15}. The mortality rate in our patient group was 0%, but a comparison of our patients treated early and those treated late showed that outcomes, in terms of post-operative complications and hospital stay, were more favorable in the early treatment group.

In terms of treatment, conservative therapy can be considered for patients whose inflammation is limited to the mediastinum and those who have mild clinical symptoms, but surgery is usually selected. The indications for conservative therapy proposed by Cameron et al\textsuperscript{3} include (1) small perforations limited to the mediastinum, (2) good drainage from the rupture wound into the esophagus, (3) mild clinical symptoms and (4) absence of severe infection. Our patient with inflammation localized to the mediastinum was treated conservatively but underwent surgery 2 days after onset because his symptoms worsened. In this case, the imaging study showed inflammation localized to the mediastinum and drainage into the esophagus, but intraoperative examination revealed a large 5-cm perforation, and we presume this is what made conservative therapy difficult. In cases like this, in which inflammation is localized to the mediastinum, we believe it is important to determine whether conservative therapy is truly indicated by accurately evaluating the perforation length by means of CT and esophagography.

Surgical treatment of spontaneous esophageal rupture involves suture closure of the perforation itself, adequate irrigation of the thoracic and mediastinal cavities, and appropriate drainage\textsuperscript{16,17}. Because the rupture frequently occurs in the left wall of the lower esophagus, a left thoracotomy approach is commonly used during surgery. We used a left thoracotomy approach in 9 of our 10 patients. In 1947, Barret et al\textsuperscript{18} reported successful suturing of both layers at the site of perforation, and currently, 2-layer suturing is the recommended method of closure. However, there is a high incidence of postoperative anastomotic leakage in patients for whom 24 hours or more have elapsed since symptom onset, even when 2 layers are sutured. Anderson et al\textsuperscript{2} reports that 2-layer closure is safe within 24 hours of symptom onset.

The anatomical characteristics of the esophagus include poor perfusion and lack of a serosal layer, and we believe the cause of the frequent anastomotic leaks that occur after simple suture closure is the corrosion and inflammation caused by vomit and gastric acid. For this reason, when surgery is performed beyond 24 hours, surrounding tissues, such as the gastric fundus, greater omentum, diaphragm, and pleural membrane, are frequently used to cover the sutures placed during perforation site repair\textsuperscript{19}. The primary repair was reinforced in 4 of 7 patients in our series (gastric fundus in 3 patients and omentum in 1 patient). Two of these patients were treated early, and 2 were treated late. There are also reports indicating that reinforcement of the perforation site is not required in patients treated early. However, we covered the repaired perforation in 2 of our patients who were treated early because the perforations in these patients were long, at 3 cm and 7 cm, and the clinical course was good in both patients; there were no anastomotic leaks. We believe that covering the perforation site should not be performed only in patients treated late, but that it should also be determined on the basis of the perforation length and diameter and the degree of crush injury sustained at the perforation site. When we investigated risk factors for anastomotic leaks in our 7 patients who underwent primary repair, we found that reinforcing the perforation site and suturing 2 layers were effective in avoiding anastomotic leaks.

In cases in which suture closure is difficult due to a high degree of contamination, esophagectomy and late reconstruction are performed\textsuperscript{20}. We performed esophagectomy in 3 of our patients, and for these patients, 48 hours or more had passed before surgery was performed. The hospital stay tended to be longer in these patients, with a mean duration of 133 days, but there were no deaths. We believe, then, that when surgery is performed 48 hours or more after symptom onset in patients with extensive intrathoracic contamination and crush injuries to the perforated tissues, esophagectomy and late reconstruction can ensure survival. Furthermore, gastrostomy or enterostomy creation is useful for patients in this condition, which is associated with a high rate of postoperative anastomotic leakage, so that enteral nutrition can be given postoperatively\textsuperscript{10}.
In recent years, there have been scattered reports of treatment of spontaneous esophageal rupture by video-assisted thoracoscopic surgery (VATS). VATS is minimally invasive and associated with reduced surgical wound infection, but reports suggest that evacuation of intrathoracic contaminants may be insufficient and that a high level of skill is required to suture the esophageal rupture wound. Going forward, we predict that advances in endoscopic surgery will result in VATS becoming more accepted for treatment of spontaneous esophageal rupture. There are also reports of less invasive, non-surgical treatment options, which include placing an esophageal stent (covered metallic stent) within the esophageal lumen at the site of perforation and closure of the perforation with endoscopic clips. Although we have no data on either of these potential options and our patient group was small, we conclude that careful case selection is necessary when we are considering the various treatment options.

Conclusions

Herein, we report on 10 patients with spontaneous esophageal rupture that we treated surgically. The period from symptom onset to surgery was 24 hours or less, and we obtained favorable outcomes in terms of postoperative complications and the duration of postoperative hospital stay, reconfirming the fact that early diagnosis and treatment are extremely important in cases of spontaneous esophageal rupture. Regarding the surgical management of spontaneous esophageal rupture, consistent results were achieved in those patients who underwent primary repair with 2-layer suturing, reinforcement of the perforation site, and creation of a gastrostomy or enterostomy for postoperative nutritional management. These are effective surgical techniques for preventing postoperative anastomotic leakage and may contribute to increased survival.

References

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