ОРИГИНАЛЬНОЕ ИССЛЕДОВАНИЕ

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РЕЗУЛЬТАТЫ ЛЕЧЕНИЯ СИНДРОМА БУРХАВЕ

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Спонтанный разрыв пищевода (синдром Бурхаве, СБ) является редко встречающейся патологией в практике хирурга – не более 2-3% среди всех случаев повреждений пищевода – и сопровождается значительным количеством диагностических ошибок и высокой летальностью. ***Цель.*** Провести анализ качества диагностики и результатов лечения пациентов со спонтанным разрывом пищевода. ***Материалы и методы.*** Проведен ретроспективный анализ историй болезни и результатов лечения 10 пациентов с СБ, находившихся на стационарном лечении в отделении торакальной хирургии ГБУ РО ОКБ (Рязань) в 2007-2018 гг. ***Результаты.*** 4 из 10 пациентов были переведены из других лечебных учреждений. На этапе первичной медицинской помощи шести больным был установлен ошибочный диагноз; двум из них проведена диагностическая лапароскопия с подозрением на наличие острого панкреатита и перфоративной язвы желудка. Среднее время от начала заболевания до хирургического вмешательства составило 71,7±23,4 ч. Ушивание перфорации пищевода выполнено во всех случаях. С учетом сроков оперативного вмешательства все случаи СБ были разделены на 2 группы: раннее вмешательство (до 24 ч, выполнено у 4-х больных), позднее (более 48 ч, 5 больных), еще 1 пациент был оперирован в сроки до 24 ч в лечебном учреждении за пределами Рязанской области. У 9 из 10 больных разрыв пищевода локализовался в типичном месте – в нижней его трети по левой латеральной стенке. В послеоперационном периоде у 8 пациентов выявлена полная либо частичная несостоятельность швов пищевода, что потребовало длительного (54,7±12,1 дней) стационарного лечения. Послеоперационная летальность составила 10% (1 пациент из 10); причина летального исхода – прогрессирующая полиорганная недостаточность и развитие ишемического инсульта головного мозга. ***Заключение.*** Качество диагностики СБ остается неудовлетворительным: в силу редкости данной патологии большинство специалистов первичного звена, в т.ч. хирургов, плохо знакомы с этиопатогенезом и особенностями клинической картины СБ. При оказании первичной помощи количество диагностических и тактических ошибок достигает 60%. Результаты хирургического лечения напрямую зависят от времени, прошедшего с момента перфорации и развития гнойных осложнения. Даже при выполнении оперативного вмешательства в срок до 24 ч с момента перфорации несостоятельность шва пищевода может достигать 75%. Таким образом, успешность лечения СБ определяется ранней диагностикой, своевременной госпитализацией в специализированный стационар и проведением адекватного оперативного вмешательства.

Ключевые слова: *пищевод, повреждение, повреждение пищевода, спонтанный разрыв пищевода, синдром Бурхаве.*

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RESULTS OF TREATMENT FOR BOERHAAVE SYNDROME

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Background. Spontaneous rupture of the esophagus (Boehaave syndrome, BS) is a rare pathology in the surgical practice. Esophageal rupture makes no more than 2-3% of all cases of damage to the esophagus and is associated with a significant number of diagnostic errors and with high mortality. ***Aim.*** The aim of the study was to analyze the quality of diagnostics and the results of treatment of patients with spontaneous rupture of the esophagus. ***Materials and Methods.*** We performed a retrospective analysis of medical histories and of treatment results of 10 patients with Boerhaave syndrome hospitalized in the department of thoracic surgery of the Ryazan Regional Clinical Hospital, Ryazan in 2007-2018. ***Results.*** Four of ten patients were transferred from other medical institutions. At the primary care stage six patients were misdiagnosed; two of them underwent diagnostic laparoscopy for suspicion of acute pancreatitis and perforated gastric ulcer. The average time from the onset of the disease to surgery was 71.7±23.4 hours. Closure of the esophageal perforation was performed in all cases. Regarding the timing of surgery, all patients with Boerhaave syndrome were divided into 2 groups: patients with early intervention (4 patients operated within 24 hours); patients with late intervention (5 patients operated after 48 hours from the onset of the disease). One patient underwent surgical treatment within 24 hours in a medical facility outside the Ryazan region. In nine out of ten patients the rupture was localized in a typical place in the lower third of the esophagus along the left lateral wall. In the postoperative period eight patients had complete or partial esophageal suture failure, which required prolonged inpatient treatment (54.7±12.1 days). Postoperative mortality was 10% (1 patient of 10) and was caused by the progressive multi-organ failure and the development of cerebral ischemic stroke. ***Conclusion.*** The quality of diagnostics of Boerhaave syndrome remains unsatisfactory: due to rare occurrence of this pathology, most specialists of primary care settings, including surgeons, are not well acquainted with the etiopathogenesis and peculiarities of clinical presentation of Boerhaave syndrome. Diagnostic and treatment errors in rendering primary medical assistance reaches 60%. Results of surgical treatment directly correlate with the time from the moment of perforation and development of septic complications. Even with early surgical intervention performed within 24 hours from the moment of perforation, esophageal suture failure may occur in up to 75% of cases. Thus, the success of treatment is determined by early diagnosis, timely hospitalization in a specialized facility, and adequate surgical intervention.

Keywords*: esophagus, damage, esophageal damage, esophageal spontaneous rupture, Boerhaave syndrome.*

Spontaneous rupture of the esophagus has been known since long ago. It was first described in 1724 by a Dutch physician and anatomist Hermann Boerhaave, therefore in literature this disease is known as Boerhaave syndrome (BS) [1]. His patient was a 50-year old Grand-Admiral baron Janvan Wassenaer, who died in 1723 in 1 hour after spontaneous vomiting that led to perforation of the esophagus. The lifetime diagnosis of BS was first made by V. Myers (1858); in 1947 N.W. Frink first documented a case of recovery of a patient with BS after drainage of the pleural cavity; in the same year N.R. Barrett

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performed a successful operation for elimination of the defect of the esophagus in BS [2].

At present BS remains one of the most threatening diseases of the gastrointestinal tract with high mortality. The disease is rather rare – the incidence rate is not more than 7.4 cases per 10 million of population per year [3]. The share of BS among all damages to the esophagus ranges from 2 to 12% [4]. By 1998 only 300 cases of BS were described in the world literature [5]. In the Netherlands only 10 cases of spontaneous rupture of the esophagus were reported over the period from 2003 to 2005 [6]. The share of patients with BS in specialized departments of thoracic surgery does not exceed 0.25% [5].

The leading role in perforation of the esophagus in BS is played by a barogenic-trauma caused by a sudden rise in the intraluminal pressure in the distal part of the esophagus, usually during vomiting, more rarely in coughing or laughter. Besides, also of importance is discoordination and disorder in motor function of the upper esophageal sphincter and spasm of the glottis. Therefore, in literature in the description of the disease the term 'banquet esophagus', 'banquet trauma' is used that reflects a common development of the disease in an attempt to suppress an attack of vomiting [7].

The *aim* of study was to analyze the quality of diagnostics and results of treatment of patients with spontaneous tear of the esophagus.

Materials and Methods

Medical histories and results of treatment of 10 patientswith BS who stayed at the department of thoracic surgery of the Ryazan Regional Clinical Hospital over the period of 2007-2018 were retrospectively analyzed.

The criterion for inclusion into the study was the presence of spontaneous rupture of the esophagus. The criteria for exclusion included iatrogenic damages to the esophagus in endoscopic examination, perforation with the existing tumor, after-burn scarry stricture of the esophagus, traumatic ruptures of the esophagus in wounds and in closed trauma of the chest.

The results were processed by variation statistic methods using Microsoft Excel 2010 program. For each series the mean and standard error of mean were determined. The statistical significance of differences of the compared values was evaluated using paired Student t-test.

Results and Discussion

According to literature, BS most commonly occurs in adults (85-90%) with predomination in males (2:1-5:1) [10]. The average age of patients is 50-70 years, the disease most commonly develops with the underlying intake of alcohol. However, this syndrome is also reported in newborns and in individuals above 90 [10]. According to our data, the average age of patients was 57.10±4.65 years (the minimal age – 27 years, and the maximum – 79 years) which agrees with literature data, however the ratio of males to females was 9:1.

According to the literature, the disease is manifested by pain, dyspnea and shock and is accompanied by intense vomiting. Pain is intense and localizes in the epigastric region, often irradiates to the back, left upper arm, left half of the chest and often enhances after food intake. Another important symptom is dyspnea associated with development of pleurisyor of hydropneumothorax in case of breakage of the integrity of the mediastinal pleura. The classic clinical manifestations are characterized by Mackler's triad: vomiting, intense pain in the chest and emphysema of soft tissues in the jugular fossa. Some authors believea spontaneous ruptureof the esophagus to be a manifestation of stage IV of Mallory-Weiss syndrome [8].

In 6 of 10 analyzed cases the disease started with intense pain in the chest and in the interscapular region. In 3 cases patients presented with pain in the epigastrium and were examined in the surgical hospital with suspicion of perforated gastric and duodenal ulcer. One patient presented with evident dyspnea that was associated with breakage of the integrity of the mediastinal pleura with development of tension hydropneumothorax. In all cases the appearance of symptoms was

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preceded by one or several episodes of vomiting. In 4 patients multiple vomiting developed with the underlying intake of alcohol. The clinical presentation of gastrointestinal bleeding (coffee-ground vomiting and melena) was seen in 2 patients.

The time of admission to hospital varied from 8 hours to 9 days. Four patients were transferred from other medical institutions, 3 of them from central regional hospitals of the Ryazan region. All the patients were delivered to specialized department of thoracic surgery in late periods from the onset of the disease, on average in 109.3±42.3 hours. The rest of patients (n=6) were delivered from home by an ambulance team in earlier periods, on average in 28.2±16.15 hours from the onset of the disease.

The leading methods in diagnosis for damage to the esophagus are X-ray methods. A standard chest X-ray in the anterior and lateral projections permits identification of only non-specific signs, such as left-side pneumothorax or hydropneumothorax and mediastinal emphysema. Besides, in 10% of patients the chest may appear normal [10]. An informative method is intake of a water-soluble contrast substance with simultaneous esophagography that in most cases permits to localize the esophageal defect and visualize entry of the contrast substance into the mediastinum or pleural cavity***. Barium sulfate is not recommended to use as a contrast substance in patients suspected of BS.*** However, it should be taken into account that more than 20% of patients with spontaneous rupture of the esophagus may give false-negative results in radiographic contrast examination [10]. In this case, if there is a substantiated suspicion of BS, the examination should be repeated in 3-4 hours [10]. A highly informative modern method of diagnostics of perforation of the esophagus is X-ray computed tomography (chest CT) [10]. This method permits to identify periesophageal accumulation of gas in the mediastinum indicating perforation of the esophagus before introduction of contrast substance. Postcontrast examination indicates thickening of the esophageal walls at the site of perforation, and direct spread of contrast substance beyond the esophageal contours into thee mediastinum. At present the opinions differ as to reasonability of fibroesophagogastroscopy in diagnostics of BS, because it is associated with a probability for additional injection of air into the mediastinum through the perforation hole and with increase in the size of the defect [10].

In the analyzed cases the examination was conducted in the following volume:esophagography with intake of water-soluble contrast substance in 6 patients, chest CT in 9 patients (in all cases with the additional opacification of the esophagus). Fibroesophagogastroscopywas conducted in 4 patients, in all cases before hospitalization into a specialized department andvisualized the defect of the esophagus in all cases.

Unfortunately, because of rare incidence of the disease and low awareness of the medical personnel of it, diagnostic mistakes are common. Thus, according to J.P. DeSchipper, etal. (2008), the most common incorrect diagnosis is a perforated gastric and duodenal ulcer, then goes myocardial infarction, thromboembolism of branches of the pulmonary artery, dissected aneurism, etc. [6].

This tendency is confirmed by the results of our analysis: on the first referral for medical help, six patients were misdiagnosed. Three patients initially received treatment for acute pancreatitis, in two of them diagnostic laparoscopy was performed.

The majority of authors think it necessary to perform surgical interventions in case of BS [3-6,8-10,12,13]. However, the final choice of conservative or surgical management in the majority of cases depends on how much time passed from the occurrence of perforation. If the diagnosis was ismade within the first 24 hours after the perforation, closure of the perforation is indicated which can be supplemented with fundoplication or with any other method of strengthening of the suture line. However, percent of leakage of sutures applied on the ruptured esophagus still remains high (30% to 90%) [7]. The primary resection or extirpation of the esophagus is

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performed very rarely, not later than in 12 hours after the onset of the disease, in case of extended defects of its wall, usually in case of hydraulic rupture of the esophagus with its tumorous and scarry degeneration or canceration, in dilation of the lumen in achalasia of cardia. Such interventions are traumatic and associated with a high lethality (29-60%)in the early postoperative period. In recent years endoscopic methods of examination have beenused in treatment of BS: installation of self-widening esophageal stents, closure of esophageal defect with endoclips, endoscopic vacuum therapy [11].

In our study, depending on the time of the performed operative intervention, all patients with BS were retrospectively divided to 2 groups: with early intervention (within 24 hours, 4 patients), and late intervention (after 48 hours, 5 patients). One patient wasearlier operated in another medical institution (suturing of the esophageal rupture and drainage of the plural cavity, installation of nasointestinal tube); in our hospital this patient received a conservative treatment, no additional operative surgery was performed.

In view of the rare occurrence of this pathology and its late diagnosis we think it reasonable to give a more detailed description of the surgical tactics and its complications in each analyzed case (Table 1).

Table 1

Peculiarities of Surgical Treatment of Patients with BS (n=10) Performed in Ryazan Regional Clinical Hospital in 2017-2018

Time from Clinical Manifestation to Surgery, h	Kind of Surgical Intervention	Postoperative Complications
44	Laparotomy, diaphragmotomy, suturingofperforation of the esophagus, drainageofthemediastinum, jejunostomy. Drainage of the left pleural cavity.	Purulent mediastinitis, mediastinal abscess, pleural empyema on the left, multiorgan failure syndrome, ischemic stroke.
24	Thoracotomyontheleft, suturingofperforation oftheesophagus, drainageofthepleuralcavity.	Bilateral pneumonia, purulent mediastinitis, pleural empyema on the left.
231	Drainage of the left pleural cavity.	Mediastinal abscesses, pleural empyema on the left, broncho-pleural fistula.
24	Thoracotomy on the left, suturing of perforation of the esophagus.	-
20	Laparotomy, diaphragmotomy, suturingofperforation of the esophagus, drainageofthemediastinum, jejunostomy. Drainage of the left pleural cavity.	Purulent mediastinitis. Pleural empyema.Bilateral pleurisy. Multi-organ failure syndrome. Decompensated pyloric stenosis.
54	Laparotomy, diaphragmotomy, suturingofperforation of the esophagus, drainageofthemediastinum, jejunostomy.	Purulent mediastinitis,bilateral pleurisy.
24	Laparotomy, diaphragmotomy, suturingofperforation of the esophagus, fundoplication, drainageofthemediastinum, jejunostomy. Drainage of the left pleural cavity.	Mediastinal abscess. Mediastinitis, pleural empyema on the left. Bilateral pneumonia.
-	Operated earlier on in another medical institution.	-
96	Laparotomy, diaphragmotomy, suturingofesophagealdefect, fundoplication drainageofthemediastinum, Witzel's gastrostomy. .	Mediastinitis, bilateral pleurisy, bilateral pneumonia. Pleural empyema on the left. Multi-organ failure syndrome.
128	Laparotomy, drainageofthemediastinum, jejunostomy. Drainage of the left pleural cavity.	Fibrillationofatria. Multi-organ failure syndrome. Pleural empyema on the left. Purulent mediastinitis.

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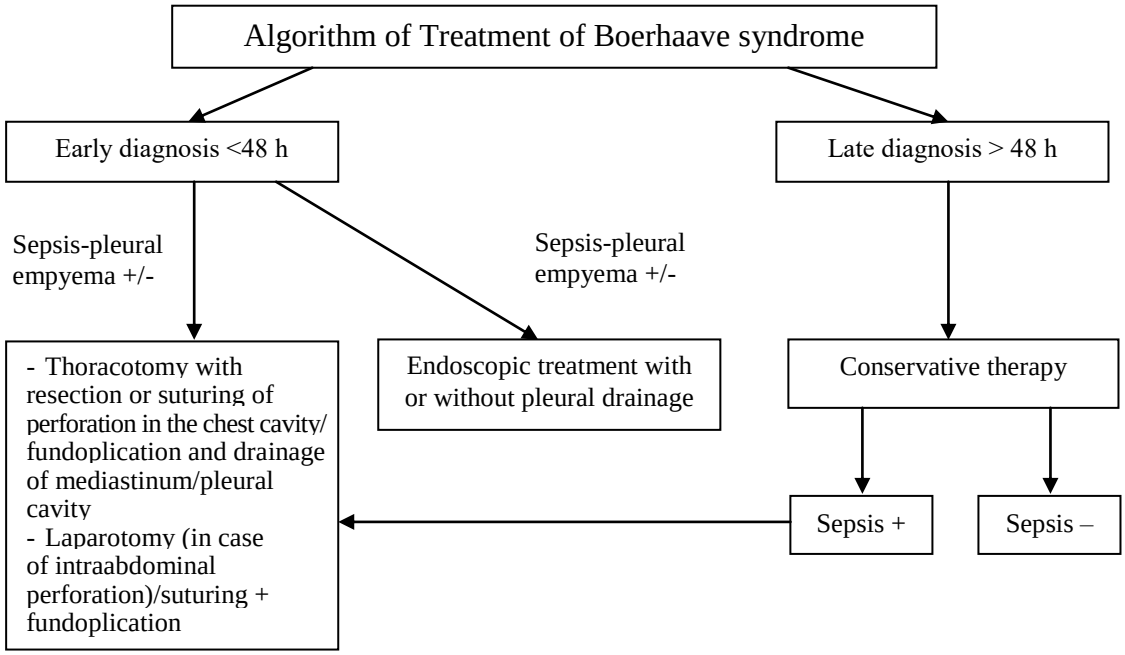

Fig. 1. Algorithm of treatment of Boerhaave syndrome.
Adapted from: De SchipperJ.P., et al. [6]

According to the classic description of BS by V.J. Derbes, et al. (1955), the spontaneous perforation of the esophagus in most cases (85-90%) localizes in its distal aspect along the posterolateral wall which is associated with the peculiarities of the anatomical structure of this part of the esophagus and with weakness of its muscle wall. In most cases the defect is a linear tear with the length varying in a wide range (from 1-2 cm to 4-6 cm and more). With this, in the literature single cases of transverse ruptures of the esophagus in BS are reported [1]. The results obtained by us agree with the literature data: 9 of 10 patients had linear tear localized along the left lateral wall, only in one patient the perforation hole was on the right posterolateral wall; the average length of the defect of the esophageal was 30.2±8.8 mm (maximal – 80 mm, minimal – 6 mm).

The tactics of management of patients with BS in our clinics was determined using personalized approach according to the algorithm given in Figure 1.

It should be noted that in 7 of 9 operated patients closure of the esophageal defect was attempted irrespective of the time passed from the onset of the disease. However, in all patients of the second group operated in late periods, the sutures further developed leaking. In 3 of 4 cases (that is, in 75%) in long-term post-operative periods the sutures failure also developed in patients operated within 24 hours.

In patients admitted in later periods (>48 hours) with purulent complications,closure of the esophageal defect is not always possible and justified. In this case the surgical intervention is directed at the adequate drainage of the mediastinum (in purulent mediastinitis) and of the pleural cavity, and at the 'turn-off' of the esophagus with formation of the esophagostoma, with application of the feeding enterostoma[6,8,10].

According to the results of our analysis, the average period of stay in the inpatient clinic was 54.7±12.1 days (minimal – 12 days, maximal – 117 days). Here, no statistically significant relationship between the duration of postoperative inpatient treatment and periods from the onset of the disease was revealed (R=0.45, p>0.05). In the overwhelm-

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ing majority of patients (n=8) the postoperative period ran a severe course and required intense therapy in the resuscitation department and was accompanied by purulent-necrotic complications and multi-organ failure syndrome.

According to the data of A.F. Chernousov et al. (2000), postoperative mortality among patients with BS ranges from 25% to 85% depending on the time passed after the moment of perforation of the esophageal wall. Unless surgical intervention is conducted within the first 24 hours, the risk of lethal outcome rises above 50% [8]. In our study one patient died in the postoperative period (that is, postoperative mortality was 10%). The lethal outcome happened on the 27th day after the surgery and was caused by progressing multi-organ failure and by ischemic cerebral stroke.

Conclusion

Retrospective analysis of 10 medical histories of patients with Boerhaave syndrome who received inpatient treatment in the department of thoracic surgery of the Ryazan Regional Clinical Hospital, Ryazan in 2007-2018 showed that the quality of diagnosis of this threatening disease remains unsatisfactory because due to the rare occurrence of this pathology most primary medical care physicians, including surgeons, are not well acquainted with the etiopathogenesis and peculiarities of the clinical presentation of Boerhaave syndrome. In rendering the primary medical assistance the number of diagnostic and tactical mistakes reaches 60%.

The results of surgical intervention directly depend on the time passed from the moment of perforation and on the development of purulent complications. Even with the surgical intervention within 24 hours from the moment of perforation the failure of the esophageal suture may reach 75%.

Thus, success in treatment for Boerhaave syndrome is determined by early diagnostics, timely hospitalization into a specialized hospital and adequate surgical intervention.

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Дополнительная информация [Additional Info]

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