



Systematic Review

# Endovascular and Open Surgical Treatment of Ruptured Splenic Artery Aneurysms: A Case Report and a Systematic Literature Review

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**Abstract:** Background: Ruptured splenic artery aneurysms (r-SAA), although rare, are burdened by high morbidity and mortality, even despite emergent surgical repair. It is suggested that endovascular treatment can achieve reduction in peri-operative death and complication rates, as in other vascular diseases, but evidence of such benefits is still lacking in this particular setting. We report a case of an r-SAA treated by trans-arterial embolization and then converted to open surgery for persistent bleeding, and we provide a systematic review of current results of open and endovascular repair of r-SAAs. Materials and Methods: A 50-year-old male presenting in shock for a giant r-SAA underwent emergent coil embolization and recovered hemodynamic stability. On the following day, he underwent laparotomy for evacuation of the huge intraperitoneal hematoma, but residual bleeding was noted from the splenic artery, which was ligated after coil removal, and a splenectomy was performed. A systematic literature review of the reported mortality and complications of r-SAA undergoing open (OSR) or endovascular (EVT) treatment was performed using the main search databases. All primary examples of research published since 1990 were included regardless of sample size. The main outcome measures were mortality and reinterventions. Secondary outcomes were post-operative complications. Results: We selected 129 studies reporting on 350 patients—185 treated with OSR and 165 with EVT. Hemodynamically unstable patients and ruptures during pregnancy were more frequently treated with open repair. Overall, there were 37 deaths (mortality: 10.6%)—24 in the OSR group and 13 in the EVTr group (mortality: 12.9% and 7.8% respectively, *p*-value: 0.84). There were 37 reinterventions after failed or complicated endovascular repair—6 treated with endovascular re-embolization and 31 with laparotomy and splenectomy (22.4%); there were 3 (1.6%) reinterventions after open repair. Overall complication rates were 7.3% in the EVT group (*n*: 12) and 4.2% in the OSR group (*n*: 7), and did not require reintervention. No significant differences in overall complications or in any specific complication rate were observed between the two groups. Conclusions: Current results of r-SAA treatment show equipose terms of morbidity and mortality between open and endovascular repair; however, in case of hemodynamic instability and rupture during pregnancy, open surgery might still be safer. Moreover, endovascular repair is still burdened by a significantly higher rate of reinterventions, mostly with conversions to open surgery.

**Keywords:** ruptured splenic aneurysms; splenic artery aneurysms and pseudoaneurysms; visceral aneurysms; splanchnic aneurysms; transarterial artery embolization; splenectomy; aneurysm rupture; shock during pregnancy



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## 1. Introduction

Endovascular repair is imposing itself as the first-choice elective treatment of visceral artery aneurysms (VAA) and pseudoaneurysms (VAPA), when anatomically feasible. How-

ever, in urgent and emergent settings, especially in cases of hemodynamic instability, most guidelines and current literature still recommend laparotomy and open surgical repair, depending on the aneurysm location [1,2]. The treatment of ruptured aneurysms of the splenic artery (SAA), in particular, is among the most controversial issues, in the absence of clear evidence favoring open or endovascular repair [3].

SAAAs are the most common visceral artery aneurysm (60% of all VAAs) and the third most common abdominal aneurysm [1–3]. Risk factors for a true SAA or pseudoaneurysm (SAPA) are portal hypertension, liver transplantation, pancreatitis, segmental mediolysis, and other forms of vasculitides. It is more frequent in women, and the risk of rupture is high during pregnancy, with extremely high mortality and morbidity for both mothers and fetus [1,3]. In case of rupture, the prognosis is poor, even in case of prompt open or endovascular treatment [1].

The aim of this study is to report a case of ruptured splenic artery aneurysm (r-SAA) treated by embolization and converted to open repair on post-operative day I, and to revise the current literature reporting on the mortality, complications, and conversion rates of open and endovascular treatment of such diseases.

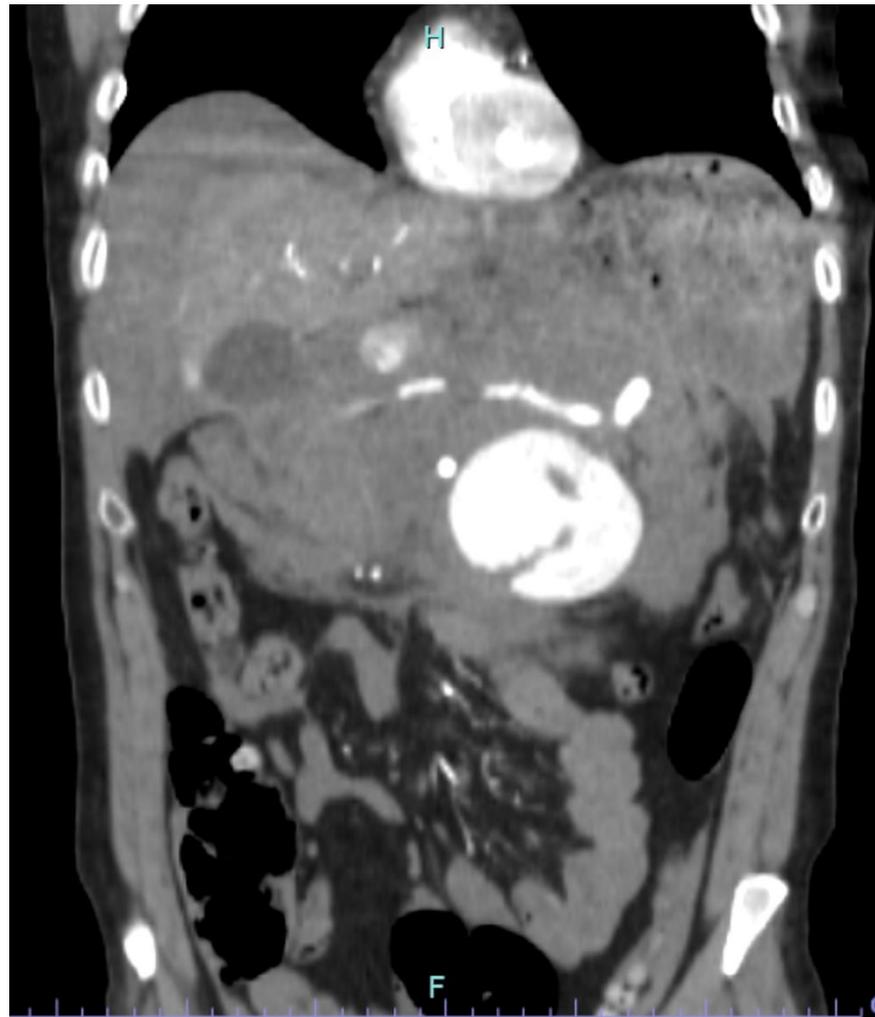
## 2. Materials and Methods

### 2.1. Case Report

A 50-year-old male with no previous cardiovascular history presented to the Emergency Room of our Center with severe abdominal pain and hypotension, which soon evolved into hemorrhagic shock with loss of consciousness. He was an active smoker, but past medical and surgical history were silent. Laboratory tests revealed severe anemia (HB: 7 g/dL) and severe impair of the coagulation (INR: 3, aPTT ratio: 2.2). He underwent emergent Computed Tomography Angiography (CTA), which revealed a giant intraperitoneal hematoma arising from an r-SAA with a maximum diameter of 10 cm (Figures 1 and 2).



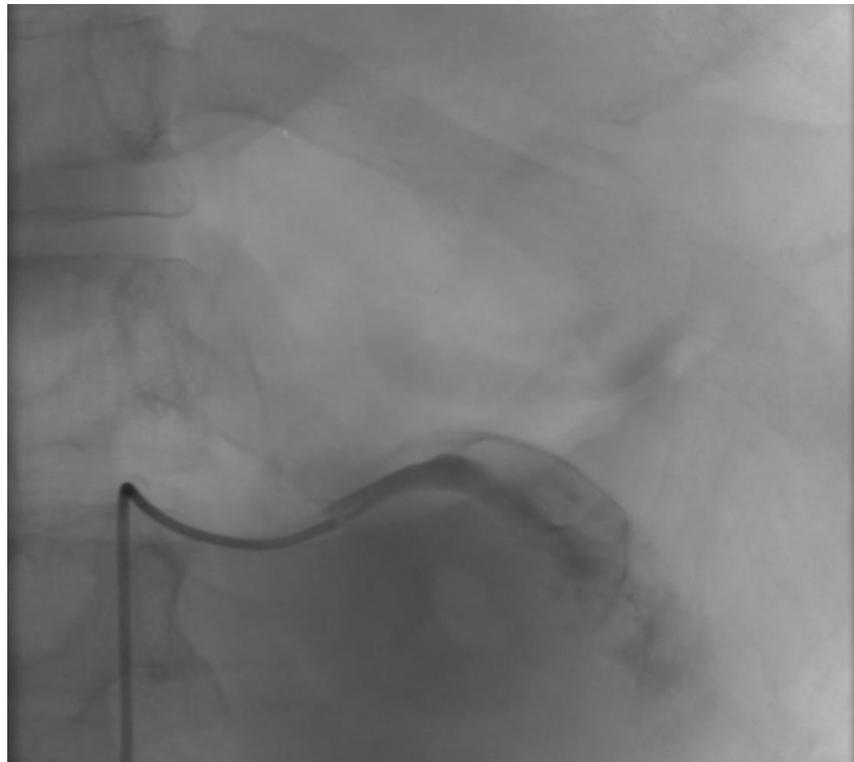
**Figure 1.** Axial view of a huge, ruptured SAA. A: anterior; P: posterior.



**Figure 2.** Ruptured SAA in coronal view.

The patient presented with a severe alteration of all the coagulation parameters, and the CTA showed a huge aneurysm involving the most distal portion of the splenic artery; we thought that outright laparotomy and splenectomy would have made the hemorrhage worse. Moreover, the vascular control of the splenic aneurysm was very difficult to obtain, due to the aneurysm size and the difficulty to isolate the splenic artery. For those reasons, after a multidisciplinary evaluation involving radiologists and general and vascular surgeons, we preferred an endovascular approach, and the patient underwent emergent angiography under general anesthesia. The coeliac artery was engaged through a 6F left femoral access, and after selective angiography confirmed rupture of SA, proximal embolization with 6 detachable metallic coils (Interlock-18<sup>®</sup> and Interlock-35<sup>®</sup>, Boston Scientific, Marlborough, MA, USA), sized 8, 10, and 12 mm, was performed. The angiography showed residual bleeding from a pancreatic branch that joined the distal splenic artery in the most lateral portion of the aneurysm, so we engaged the pancreatic artery through superselective catheterization with a microcatheter and released 3 metallic coils (Interlock-18<sup>®</sup>, Boston Scientific, Marlborough, MA, USA), sized 8 mm, in the distal splenic artery and in the pancreatic artery right above its anastomosis with the splenic artery (Figures 3–5). The hemorrhage was effectively controlled and final completion angiogram from the coeliac trunk showed no signs of contrast extravasation (Figure 6). The patient regained hemodynamic stability, but, due to his severely altered coagulative parameters, the prospected laparotomy to drain the intraperitoneal hematoma and prevent abdominal compartment syndrome

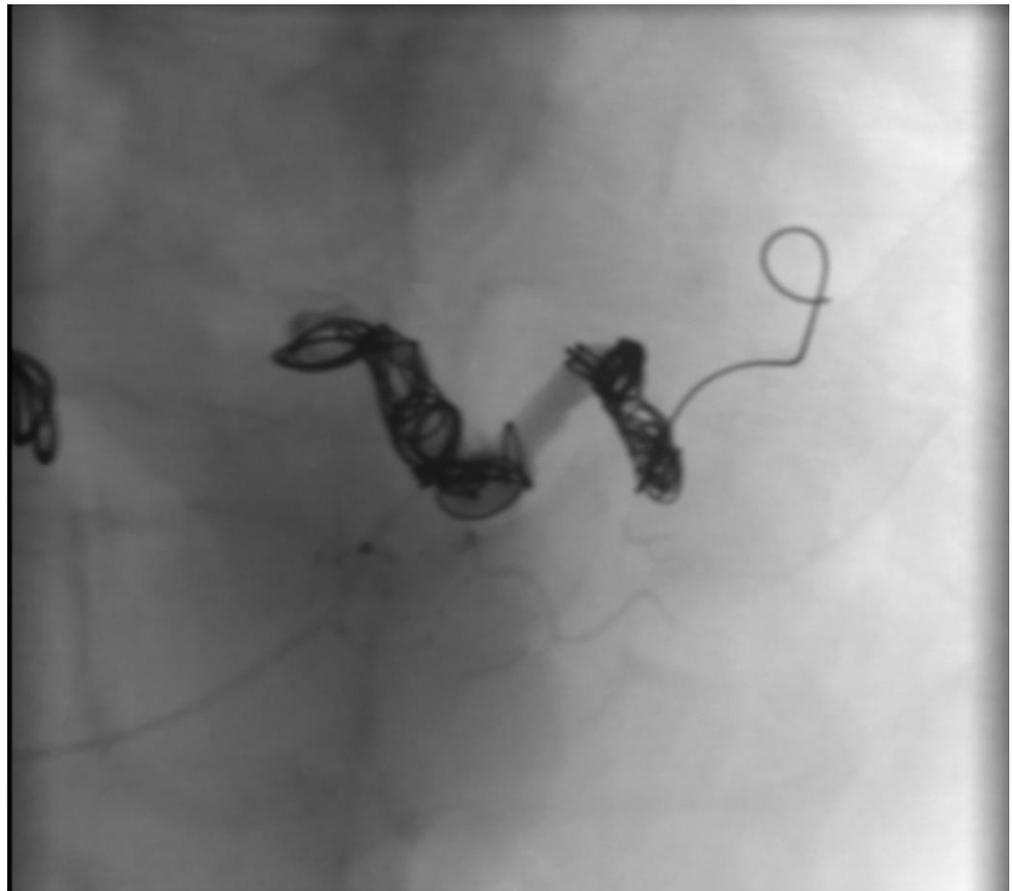
was postponed to the following day. Meanwhile, the patient received red blood cell, fluid, and plasma integration, and remained intubated and stable throughout the night.



**Figure 3.** Selective intraoperative angiography showing a ruptured SAA.



**Figure 4.** Proximal coil embolization of the splenic artery.

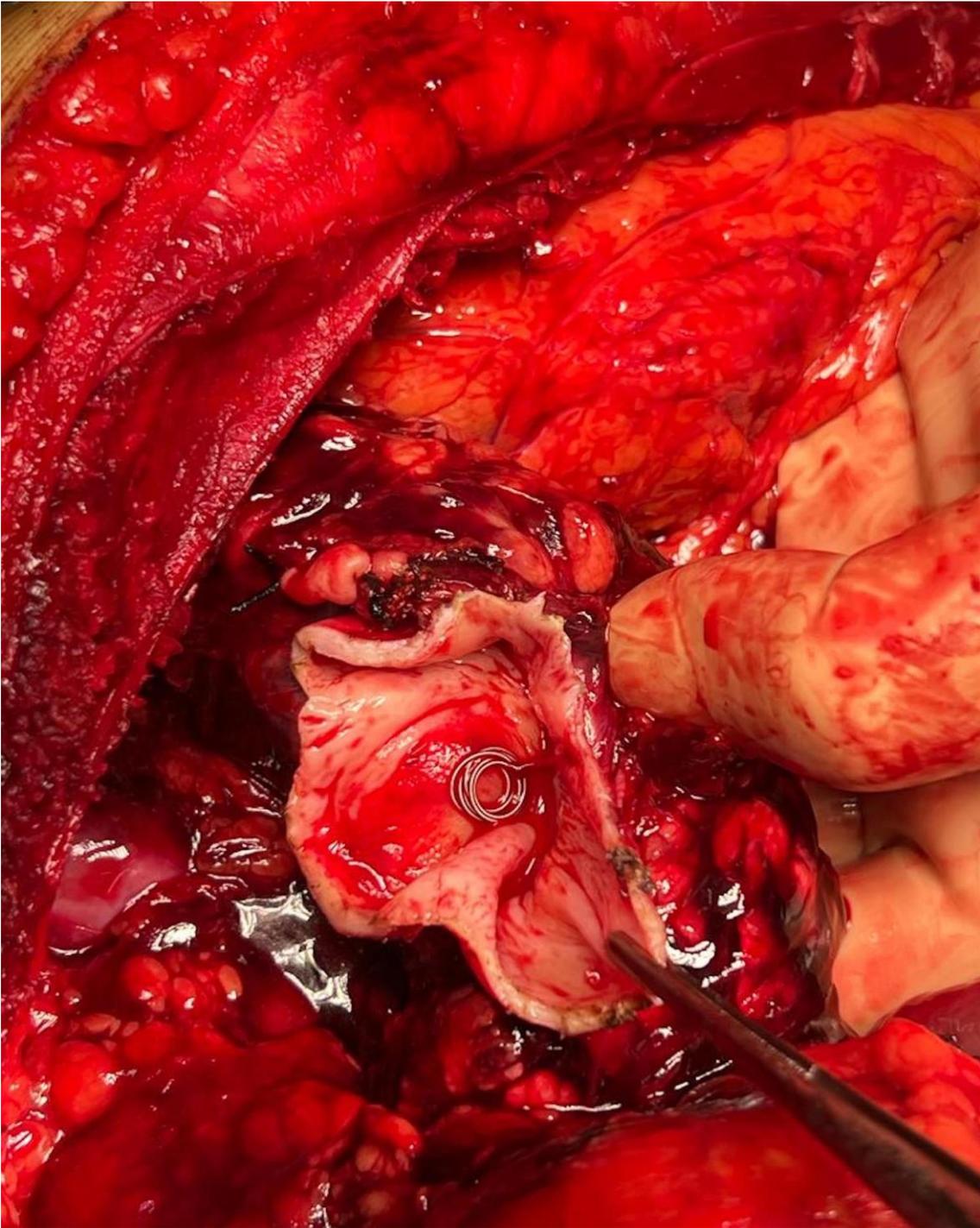


**Figure 5.** Proximal and distal embolization of the SA.

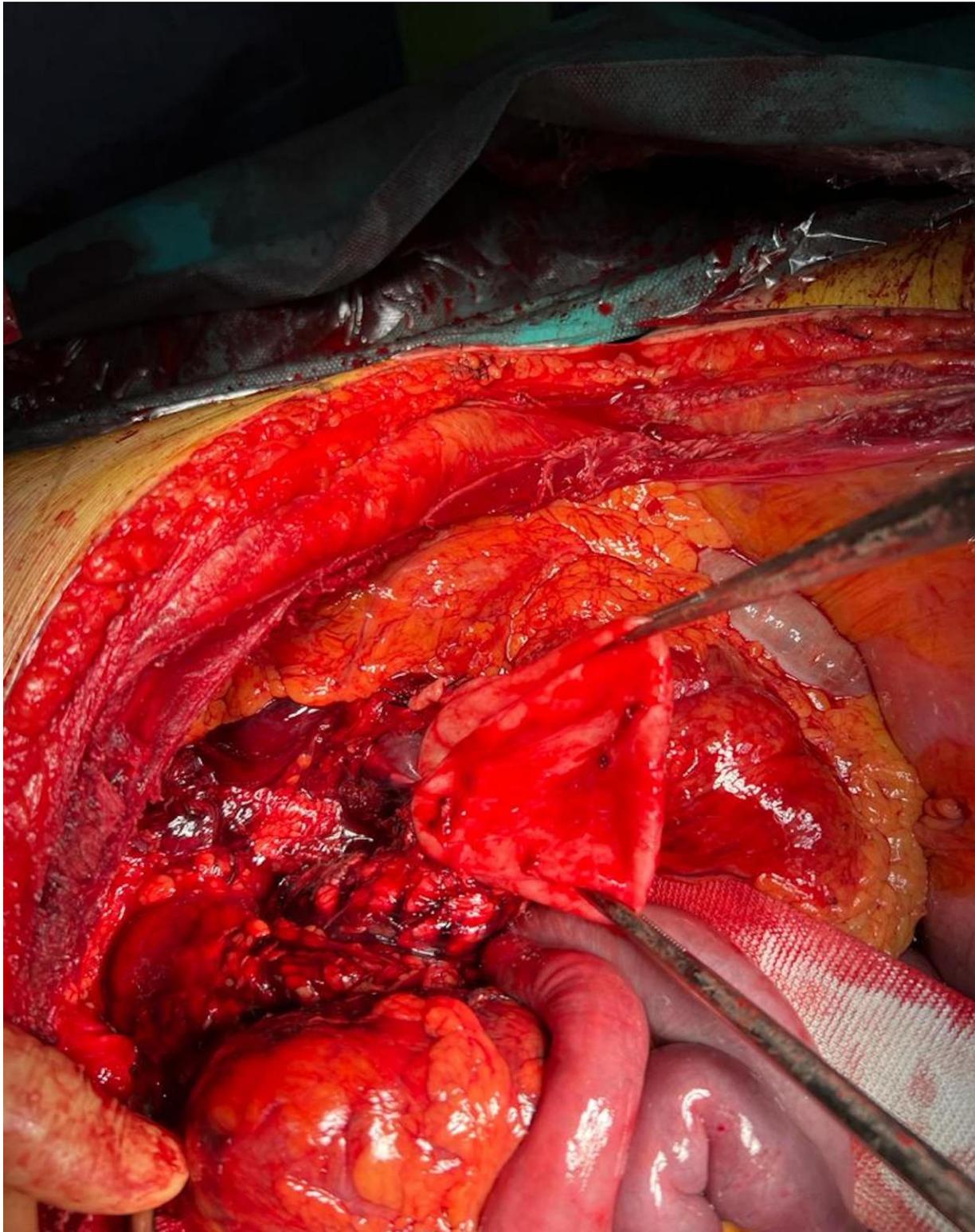


**Figure 6.** Final angiogram showing successful aneurysm exclusion.

On the following day, a chevron laparotomy was performed, but while opening the lesser omentum, massive bleeding was observed that required supraceliac aortic clamping. We isolated the coeliac trunk and the giant splenic aneurysm and ligated its arterial inflow and outflow. We drained the hematoma, removed the coils placed into the afferent and efferent vessels, and resected the aneurysm along with the spleen (Figures 7 and 8). After that, the hemorrhage was fully controlled, and the surgical incision was closed after leaving a drain in the splenic lodge. The patient left the operative room stable, but he deceased a few hours after surgery due to multiorgan failure and severe hyperkalemia.



**Figure 7.** Coil removal after laparotomy.



**Figure 8.** SAA excision.

## 2.2. Literature Review

### 2.2.1. Search Strategy

Cochrane, Embase, and PMC databases were searched using the following search strategy:

“(((visceral OR splanchnic OR splenic) AND ((aneurysm [Title/Abstract]) OR (pseudoaneurysm [Title/Abstract]))) AND ((rupture [Title/Abstract]) OR (ruptured visceral aneurysm [Title/Abstract]))) NOT (((aortic [Title/Abstract]) OR (abdominal aortic [Title/Abstract]) OR (thoracoabdominal [Title/Abstract]))). The last search was made on 1 August 2023.

Study selection and analysis was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) Checklist (Figure 9) [4]. This systematic review was not registered on platforms.

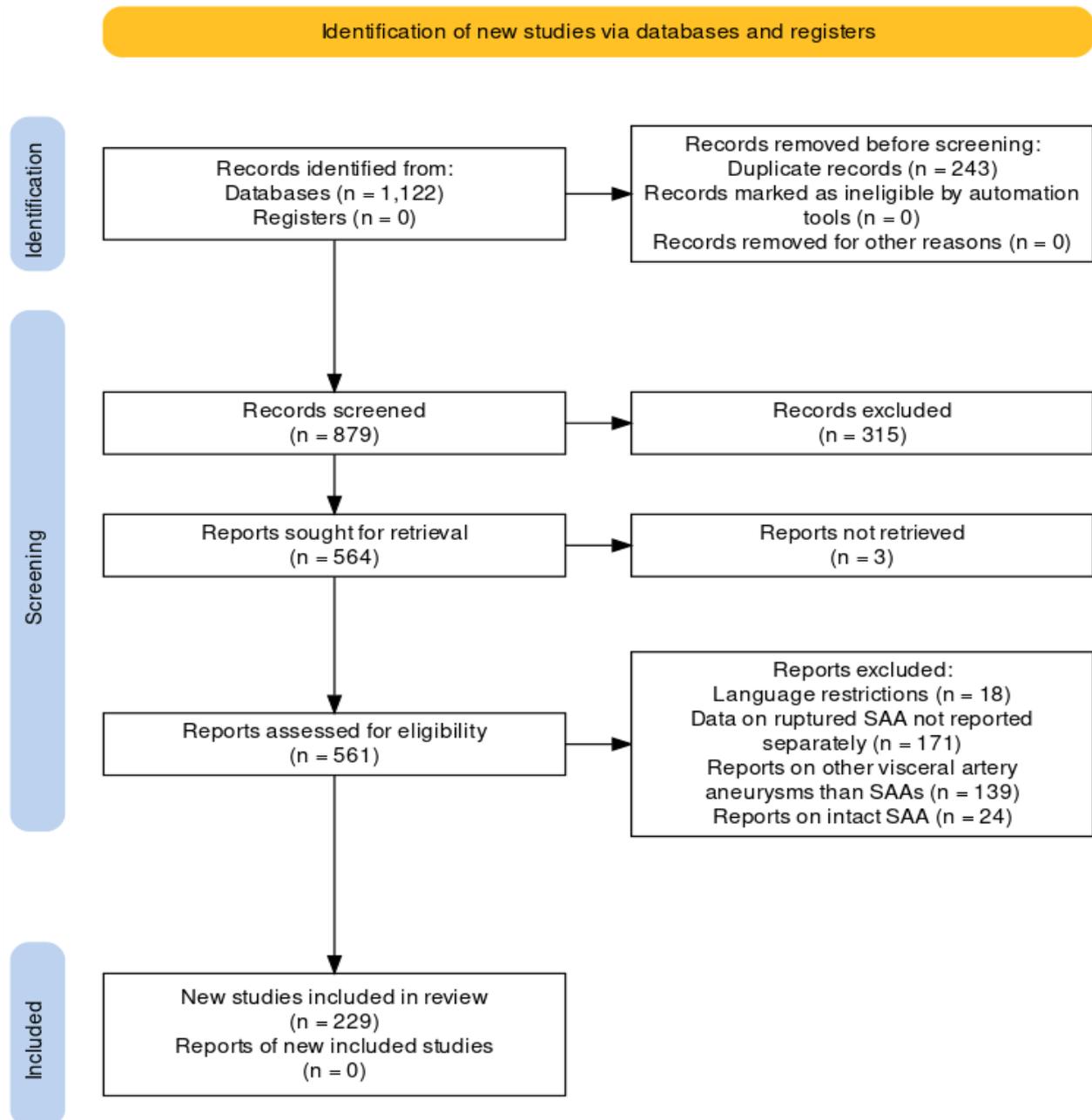


Figure 9. Selection flow-chart of the reports included in the review.

### 2.2.2. Inclusion and Exclusion Criteria

We included all comparative and non-comparative studies published since 1990 in English, French, German, Spanish, or Italian reporting on open or endovascular repair of ruptured splenic artery aneurysms (r-SAAs) and pseudoaneurysms (r-SAPAs). Studies

published in other languages were included only if they had an abstract in English providing all the useful data. All primary sources of research were included regardless of sample size, as long as they reported on mortality, morbidity, and rates of reintervention and open conversion. Studies not reporting on aneurysm location, treatment technique, and peri-operative and post-operative results were not included. Studies reporting on both intact and ruptured visceral aneurysms were included only if separated results for the two groups were provided. The results were screened manually; study selection and inclusion and exclusion criteria were assessed independently by 2 authors (LFR and CB); in case of contrasting results, a third author decided (EMM).

### 2.2.3. Outcome Measures

The endpoints were mortality and reintervention/conversion rates of endovascular and open repair. Mortality was defined as death by any cause reported within 30 days since the intervention, and any surgical-related or aneurysm-related death reported afterwards.

### 2.3. Statistical Analysis

Categorical variables were expressed as percentage, continuous variables as mean and standard deviation. Chi-square and t-test were employed to compare the distribution of nominal and ordinal variables respectively between the two groups. Odds ratios of mortality and reinterventions rates were calculated for patients treated by open and endovascular repair and two-sided *p*-values < 0.05 were considered significant. Logistic regression analysis was performed to ascertain if hemodynamic instability was a predictor of mortality or reintervention.

## 3. Results

### 3.1. Study Selection

We identified 1122 records. After examining inclusion and exclusion criteria and removing duplicates, we selected 227 studies reporting on 350 patients. Most studies (197) were case reports, 29 were series reporting on >2 cases and 3 were comparative retrospective studies.

### 3.2. Findings

#### 3.2.1. Patients

Overall, 120 males, 95 females, and 135 patients whose gender was not reported received open surgical repair (OSR) or endovascular treatment (EVT) for an r-SAA (258) or a r-SAPA (92) [5–231]. Mean age was  $47.2 \pm 17$  years. We found an extreme variability among the reported r-SAA diameters, ranging between 20 mm (mainly PSA) to giant SAAs measuring 14 cm. The mean diameter was 53 mm, with a standard deviation of 21.

Overall, 185 patients received open surgery with ligation and aneurysm excision; splenectomy was performed in 179 cases (96.7%); a distal pancreatectomy and left hemicolectomy were necessary to complete the vascular control in 8 and 4 cases, respectively.

Overall, 165 patients underwent endovascular repair. Most endovascular treatments (160/165) were conducted through transarterial embolization (TAE), mainly with metallic coils or microcoils, liquid agents (12), or a mixture of the two with or without thrombin injection (15) and in one case with an Amplatzer plug. Covered stents were used in three cases, and in two cases the technique was not specified.

Distribution of age, sex, and clinical characteristics between the two groups are reported in Table 1. Patients who had undergone open repair were significantly younger and more likely to be female, to have an ongoing pregnancy, and to present with hemodynamic instability—defined as shock or hypotension at presentation. Pseudoaneurysms were more frequent in patients treated by endovascular means.

**Table 1.** Baseline clinical characteristics at presentation.

	OSR (%)	EVT (%)	<i>p</i> -Value
Male sex	63/185 (34%)	57/165 (34.5%)	1
Female sex	68/185 (36.75%)	27/165 (16.5%)	0.01
Sex non reported	54/185 (29.25%)	81/165 (49%)	-
Mean age	41.5 ± 15.3	53.5 ± 16.9	0.02
SAPAs	23/185 (12.4%)	69/165 (41.8%)	<0.01
Hemodynamic stability	19 (10.6%)	81 (49.1%)	<0.01
Hemodynamic instability	142 (76.5%)	81 (49.1%)	<0.01
Hemodynamic status non reported	24 (12.9%)	3 (1.8%)	-
Rupture during pregnancy	79 (42.7%)	7 (4.2%)	0.01

OSR: open surgical repair, EVT: endovascular treatment, SAPA: splenic artery pseudoaneurysm.

### 3.2.2. Outcomes

Overall, there were 37 deaths (mortality: 10.6%), 35 of them within the first 30 days after treatment (Table 2). The leading causes of mortality were disseminated intravascular coagulation (DIC) in 9 cases, multiorgan failure (MOF) in 7 cases, sepsis in 5 cases, ischemic colitis in 3 cases, cardiac complications in 2 cases, and respiratory distress syndrome in 1 case. In 9 cases, the causes of death were not clearly reported. Overall complication rates were 7.3% in the EVT group (12 patients) and 4.2% in the OSR group (7 patients): in 8 cases, they were systemic (2 sepsis, 2 myocardial infarctions, 1 stroke, 1 respiratory failure, and 2 bowel obstructions, Table 3). Local complications occurred in 7 cases in the EVT group (3 splenic abscesses and 4 splenic infarction) and in 3 cases in the OSR group (1 splenic abscess and 2 pancreatitis) and did not require reintervention. No significant differences in overall complications or in any specific complication rate were observed between the two groups.

**Table 2.** Peri-operative results.

	OSR	EVT	<i>p</i> -Value
Overall mortality	24/185 (12.9%)	13/165 (7.8%)	0.84
Mortality in HD stable patients	0/19 (0%)	7/81 (8.6%)	Logistic regression analysis on HD instability as predictor of mortality: <i>p</i> -value (OSR): 0.02 <i>p</i> -value (EVT): 0.18
Mortality in HD unstable patients	13/142 * (9.1%)	3/81 * (3.7%)	
Early reintervention conversion to OSR	5/185 (2.7%) -	37/165 (22.4%) 31/165 (18.8%)	<0.01 -
Reinterventions/conversions in HD stable patients	2/19 (10.5%)	9/81 (11.1%)	Logistic regression analysis on HD instability as predictor of reintervention: <i>p</i> -value (OSR): 0.30 <i>p</i> -value (EVT): 0.01
Reinterventions/conversions in HD stable patients	1/142 * (7%)	21/81 * (25.9%)	

OSR: open surgical repair, EVT: endovascular repair, HD: hemodynamics, \* HD status non reported in 24 pts in the OSR group and in 3 pts in the EVT group.

**Table 3.** Peri-operative complications.

	OSR	EVT	Total	<i>p</i> -Value
Systemic complications	4/185 (2 sepsis, 1 stroke, 1 ileus)	4/165 (2 MI, 1 pneumonia, 1 ileus)	8/350	1
Splenic abscess	1/185	3/165	4/350	0.8
Splenic infarction	0/185	4/165	4/350	0.3
Pancreatitis	2/185	0/165	2/350	0.7
Total	7/185 (3.8%)	11/165 (6.7%)	18/350 (5.1%)	0.8

OSR: open surgical repair, EVT: endovascular repair, MI: myocardial infarction.

Based on intention-to-treat analysis, the reported mortality rate was 12.9% in OSR and 7.8% in EVT groups, *p*-value: 0.84. The reintervention rate after failed or complicated endovascular repair was 22.4% (37 cases, 6 treated with endovascular re-embolization and 31 with laparotomy and splenectomy) (Table 2). The leading causes for open conversion were recurrent or residual bleeding (26 cases), failure to selectively catheterize the splenic artery (5 cases), and the need to drain an intraperitoneal hematoma to prevent abdominal compartment syndrome (6 cases). Reintervention after open repaired was reported in 5 cases (2.7%), all for hemostasis completion.

Overall, 86 cases of r-SAAs concerned pregnant women, 7 of whom underwent TAE and 79 OSR. The preference for OSR in pregnancy is probably due to the radiation exposure of fetuses for pregnant patients treated with EVT.

Overall maternal and fetal mortality was 16.3% and 32.5%, respectively (14 maternal and 28 fetal deaths). Mortality rates were similar between endovascular and open repair groups: in the first one, 1 maternal and 2 fetal deaths were reported (14.3% and 26.6%, respectively); in the latter, maternal and fetal mortality were 16.5% and 33%, respectively (N: 13 and 26).

### 3.2.3. Regression

Logistic regression analysis confirmed hemodynamic instability as a predictor of mortality in patients undergoing OSR and a predictor of reintervention in patients undergoing EVT (Table 2).

## 4. Discussion

This systematic review found no significant differences in mortality rates between endovascular and open treatment of ruptured splenic artery aneurysms and pseudoaneurysms, but our results confirm that endovascular repair carries higher reintervention and conversion rates than open surgery, as observed in many other vascular diseases, especially in case of hemodynamically unstable patients.

The issue of treatment choice of ruptured visceral vessels is currently object of debate. In fact, although endovascular treatment, especially TAE, has now become the first-line treatment option for ruptured visceral aneurysms and pseudoaneurysms, especially those of the pancreatic arcade and of the distal branches of the mesenteric arteries. The Guidelines are still extremely cautious in recommendations when it comes to ruptured splenic aneurysms.

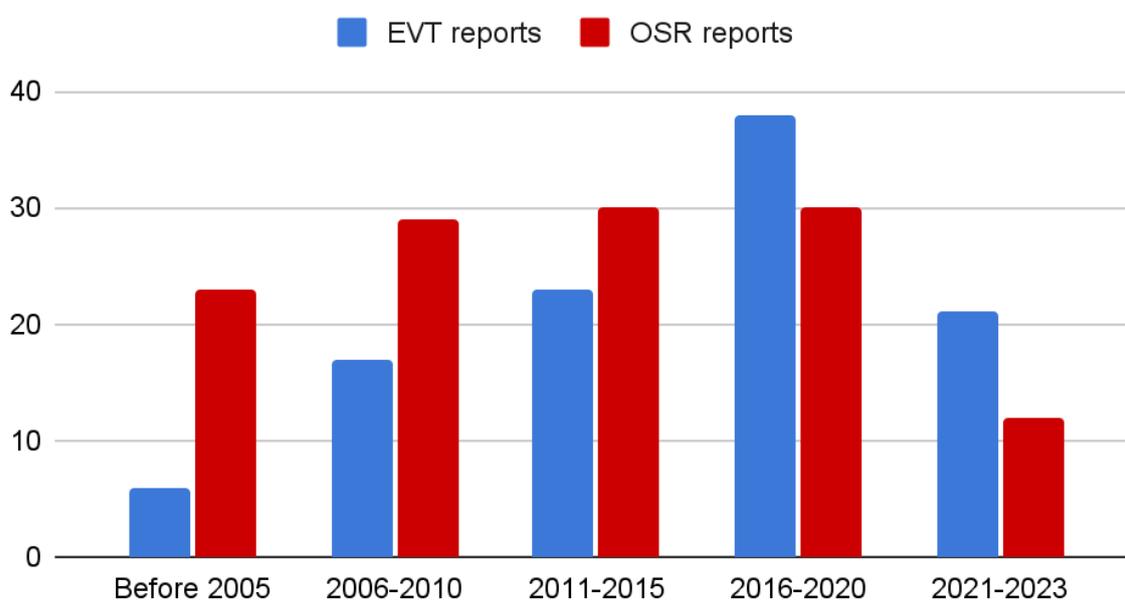
The latest Guidelines issued by the Society for Vascular Surgery, for example, recommend ligation with or without splenectomy of r-SAAs discovered on laparotomy and open or endovascular treatment of r-SAA diagnosed on previous imaging, based on the patient's anatomy and baseline clinical conditions [1].

This is based on the most recent Metanalysis by Barrinuevo et al., which reported high mortality for open repair of r-SAAs (0.29) but provided no data on their endovascular repair [232]. Moreover, that study included an extremely heterogeneous corpus of case

series, most of which did not provide separated outcomes for ruptured and non-ruptured SAAs. This makes it extremely difficult to estimate the actual mortality and morbidity rates related to the two treatment options.

It is known that peri-operative mortality of r-SAAs and r-SAPAs is extremely high, ranging between 20% and 30%, with a considerable risk of fetal and maternal death if rupture occurs during pregnancy [3]. For this reason, urgent endovascular repair is considered a promising possibility to decrease intraoperative mortality, complications, and length of in-hospital stay, but the current data have failed to show better outcomes in the treatment of r-SAA and r-SAPA, and hemodynamic instability is still seen as a setting in which the open approach should be preferable [233,234]. On the other hand, there are many reports in the current literature that describe successful endovascular treatment of ruptured splenic arteries, and their number is growing in frequency in the latest years, as Figure 10 indicates. The uncertainty on this subject is also caused by the rare occurrence of SAAs presenting with rupture and the heterogeneity in outcomes of specific studies reporting on the post-operative outcomes of their urgent repair, which is the goal of this review.

## Records of r-SAAs repair



**Figure 10.** Records of r-SAA repair between 1990 and 2023.

Revising all available reports on r-SAAs and r-SAPAs treated by open or endovascular surgery in the last 30 years, we found a significantly lower overall mortality rate than previously reported (10.9%), which could be attributable to a publication bias concerning the many case reports included in the review [234]. Mortality rates did not differ significantly between the two groups, but endovascular treatment carries a significant higher risk of early conversion or reintervention, consistently with the data reporting on elective repair of SAAs [232].

Notably, we observed that the ratio between r-SAA treated by surgical-first vs. endovascular-first strategy is changing in favor of the latter, in the most recent years, especially when pseudoaneurysms and elderly patients are involved. TAE results by far the preferred technique, with a much lower incidence of ischemic complications than usually reported (4 splenic infarctions; 2.4% vs. 38% according to Cordova et al. [233]), and often without clinical significance. TAE is particularly useful in bleeding and ruptured aneurysms because it achieves faster and usually effective control of the hemorrhage, allowing the patient to recover his stability, and in case of failure it can be easily repeated. The true Achilles heel of

TAE is early and late coil migration, which often require reintervention and open conversion, particularly in hemodynamically unstable patients, like in the case here reported.

Concerning open surgery, splenectomy, although it is not routinely recommended even in emergent r-SAA repair, is still widely employed in emergency surgery in order to optimize bleeding control and prevent infective complications, such as splenic abscesses [1,2]. The main concerns after splenectomy are sepsis from capsulated bacteria, spleno-mesenteric vein thrombosis, and pancreatitis, although the data collected in this regard did not show higher systemic or local complication rates as compared with EVT.

Hemodynamic instability at presentation was confirmed as an important predictor of perioperative death in OSR group, whereas the numbers were too small to prove the same correlation in the EVT group; hemodynamically unstable patients who had undergone endovascular interventions were, however, more prone to have reintervention or open conversions. Conversion could be due to incomplete aneurysm exclusion or abdominal compartment syndrome due to the intraperitoneal hematoma, as shown in our case report. Although the patient was stable after EVT, he still had severe coagulation impairment after the procedure, and we planned decompression laparotomy for the following day. Unfortunately, during this phase, the coils migrated, and the patient started to bleed again. If a conversion is planned to optimize hemostasis, it is better to perform it right after embolization, and that is a lesson learned for us that we would like to share with our readers.

Such observations help to shed a light on mortality and complications data: in fact, although the observed rates between OSR and EVT groups were similar, hemodynamic instability and rupture during pregnancy, which are known predictors of worse prognosis, were significantly more frequent among patients treated by open surgery, suggesting that OSR may be safer than EVT in such circumstances.

#### *Limitations*

This review has several limitations. First, the heterogeneity of the data, which come mainly from case reports, case series, and portions of larger comparative studies conducted on VAAs, from which the data regarding r-SAAs and r-SAPAs were extracted. This also causes a wide heterogeneity in how results were reported and resulted in us excluding many studies in which the specific outcomes of r-SAAs and r-SAPA were not clearly provided [235–237]. In fact, the largest and most interesting studies on this topic report on VAAs considered as a whole, sometimes conducting subgroup analysis by aneurysm location or by symptomatic/asymptomatic presentation; visceral artery aneurysms should be seen instead as separated identities, each with its peculiarities, because of the differences in anatomy and physiology of the abdominal organs they supply. The latest SVS Guidelines, for the first time, endeavor to provide recommendations concerning the treatment choice based on aneurysm locations, but we still need more evidence to strengthen the value of their assessments, especially in the field of ruptured VAAs. To produce high-quality evidence in this respect, reviews and metaanalysis should rely on better quality primary studies reporting separately the outcomes of each aneurysm type and distinguishing between intact and ruptured VAAs [238].

The second limitation concerns the sample size: although 350 r-SAAs and VAPAs are in theory a large number, the number of reported deaths and complications is lower than reported in other studies [233,234]. This may have biased the statistical analysis, especially the logistic regression trying to establish the role of hemodynamic status as a mortality predictor in the EVT group. A publication bias is probably responsible for such discrepancy in mortality and morbidity.

Another important limitation is that the compared patient groups present statistically significant difference in their baseline clinical characteristics, along with the heterogeneity of the reports, but this is a common problem of retrospective studies. On the other hand, the equipoise in mortality, although patients treated with EVT were younger and more

likely to be hemodynamically stable, strengthens the conclusion that EVT is not superior to OSR in emergent setting.

Finally, the review is an attempt to provide new insights on the outcomes of emergent treatment of r-SAAs, focusing on mortality and reintervention, but there are other minor outcome measures that could be considered in a more detailed analysis, such as the length of post-operative recovery and in-hospital stay, the quality of life, and the treatment-related costs. Moreover, mid-term and longer-term outcomes, particularly late conversions, are also worthy of assessment. Unfortunately, such information is seldom reported in primary literature.

## 5. Conclusions

Rupture of a splenic artery aneurysm is burdened by high mortality and morbidity, and its optimal management is still controversial. Although the popularity of endovascular treatment is increasing even in an emergency setting, there is still no evidence of better outcomes in terms of death and complications compared with open surgery, especially in patients presenting with hemodynamic instability and in pregnant woman. Moreover, early and late reintervention and open conversion are still a considerable concern of emergent endovascular repair.

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