

# Management of true splenic artery aneurysms

Two case reports and review of the literature

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Few series of splenic artery aneurysms (SAA) have been reported, but today asymptomatic SAA are detected with increasing frequency. Their importance lies from their potentially fatal consequences as life-threatening hemorrhage. SAA management still remains controversial as reported in this review. Our 2 patients treated with resection of the aneurysms, both located in the middle third of the splenic artery. Some authors demonstrated that when splenic artery has been ligated (or embolized) and the patients remain anatomically splenic, they may not retain any splenic function. Laparoscopic SAA ligation repair appears to be optimal and useful for aneurysms protruding from the pancreas and it is gaining interest because clinical recovery is rapid with a poor morbidity and economic and cosmetic advantages. Transcatheter embolization too offers a temporary control in urgency to stop hemorrhage and go back at later date to make much better elective operation. Endovascular interventions as percutaneous embolization has recently gained popularity: it offers a safe alternative or adjunctive therapy to traditional surgery. We hope in the future instrumentation will likely improve so that this procedure can be done percutaneously by development of prosthetic devices in the 21th century.

**Key words:** Splenic artery - Aneurysm, diagnosis - Aneurysm, surgery - Embolization, therapeutic.

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The first report of splenic artery aneurysms (SAA) was made by Beaussier at necropsy in 1770.<sup>1</sup>

A factor contributing to incomplete knowledge of SAA pathogenesis and natural history is that few series have been reported. To day SAA are detected with increasing frequency, because of the aging population and more liberal use of vessel evaluation with Doppler ultrasonography, computed tomography and arteriography for other pathologies.<sup>2-9</sup> The importance of SAA lies not only on their abundance, but on their potentially fatal consequences.<sup>10, 11</sup> Rupture varies from 5.3 to 46% and is often the first and only symptom which may be associated with life-threatening hemorrhage.<sup>2, 9, 12-16</sup>

Often in clinical suspicion for a hemorrhage in the abdominal cavity a surgical exploration is performed and an asymptomatic aneurysm is incidentally detected.<sup>6, 17</sup>

Large routine necropsy series have reported an incidence of SAA ranging from 0.01 to 10.4%.<sup>2, 5, 12-14, 16-24</sup> The first preoperative diagnosis was made by Hoegler in 1920.<sup>14</sup> Often the right diagnosis is performed only with laparotomy.<sup>10, 25, 26</sup>

In literature about 1800 cases have been

described in about 3000 cases of visceral artery aneurysms and the splenic artery is the third most common site after the abdominal aorta and iliac arteries up to 60%.<sup>5, 6, 8, 12, 16, 27-32</sup>

SAA are from 2 to 5 more frequent in women<sup>3, 7, 8, 10, 12, 24, 27, 32-37</sup> and 92% had been pregnant with an average of 4.5 times<sup>4, 10, 12-14, 16, 23</sup> but in older patients the incidence in each sex tends to be equal.<sup>13</sup> The average age was 50.6 years with the range of 14 to 78 years.<sup>5, 7, 9, 10, 12, 14, 17, 24, 30</sup>

The precise mechanism of aneurysmal dilatation of the splenic artery is unknown. Authors identified two contributing factors: preliminary weakness of the arterial wall and a concomitant rise in blood pressure. Risk factors predispose patients to formation of SAA, such as arteriosclerosis 30-60%, portal hypertension 25-70%, pregnancy 30-70%, arterial fibrodysplasia 13-30%, systemic hypertension 20-30%, infective emboli and arteritis 11-13% and trauma 3-4%.<sup>2-6, 9, 10, 12-14, 17, 19, 22-24, 26, 30, 32, 36, 38-44</sup>

In contrast to aneurysms of large vessels, atherosclerosis is rarely the primary causative factor of SAA,<sup>2, 7, 12, 43, 45</sup> but histological evidence of atherosclerosis is present in almost all cases<sup>12, 14, 16</sup> and they are observed in up 99% of SAA histologically examined.<sup>10</sup>

That circumstantial evidence suggests that atherosclerosis features are secondary in children and in multiparous women.<sup>2, 12</sup> The increased frequency of SAA in women who have had multiple pregnancies<sup>3, 8, 12, 46</sup> is thought to be secondary to increased splenic blood flow, (portal congestion that occur during pregnancy) and the influence of the hormonal changes, particularly those of relaxin on the vessel wall which may promote aneurysmal development.<sup>2, 7, 10, 12, 43, 46</sup>

Increases in splenic artery blood flow are also thought to be an important mechanism which explains in part the increased frequency of SAA as high to 70%, found in patients with portal hypertension and splenomegaly<sup>2, 3, 5, 7, 8, 10, 12, 22, 35, 39, 40, 43, 47, 48</sup> and in they after orthotopic liver transplantation<sup>3, 7, 8, 24, 38, 49, 50</sup> as well as in patients with systemic hypertension.<sup>9, 13, 23</sup> Patients treated with liver transplantation are usually followed with frequent examination with TC and Doppler ultrasound to monitor the size of their SAA.<sup>35, 49</sup>

Size of aneurysms ranges from 2 to 5 cm,<sup>12, 13</sup> but most of SAA are less than 2 cm and are saccular and rupture group has an average size of 5.5 cm; 74-87% were located in the distal third of splenic artery, 22% in the middle third and the others in the proximal third.<sup>3, 7, 8, 12, 13, 35, 51</sup>

Forty percent of patients presented multiple splenic artery aneurysms and 13-20% had 2; 48-71% of all aneurysms are solitary.<sup>3, 8, 9, 12, 13, 24, 32, 35, 51</sup> When multiple, these aneurysms are slightly smaller; 70% are calcified: the presence of calcifications does not prevent rupture as some authors said.<sup>9, 12, 16</sup> Giant aneurysms of splenic artery are saccular with size to 10 cm.<sup>3, 45, 52</sup> The majority of patients (about 60-95%) are asymptomatic when the aneurysm is detected. Frequently SAA often present with classical gastrointestinal symptoms, such epigastric pain, anorexia, nausea and vomiting that can be attributed to a coexistent hiatal hernia, peptic ulcer or gallbladder disease: the diagnosis is generally made incidentally.<sup>12, 13, 53</sup> A palpable mass in the left upper quadrant may be a SAA<sup>3, 13</sup> and a palpable pulsatile mass accompanied by a bruit may occasionally be noted.<sup>10, 14</sup> Symptomatic aneurysms are 8 to 60% and cause epigastric or left upper quadrant abdominal pain.<sup>2, 3, 5, 7, 9, 10, 12, 13, 22, 26, 30, 32, 54</sup> Mode of presentation of rupture is often a left abdominal upper quadrant pain excruciating into the back and it's indicates that a rupture may have occurred, especially when signs of hypovolemia are present.<sup>3, 7, 8, 13, 17, 18, 26, 32</sup> Rupture entails onset of acute abdominal pain with episodes of hypotension: we can see a "double rupture phenomenon" within 48 hours, in which initial aneurysmal bleeding tamponed in the omental sac and it is followed by flooding through the foramen of Winslow into the peritoneal cavity.<sup>3, 7, 8, 10, 12, 13, 16-18, 26, 30, 41, 54-57</sup> This "double rupture phenomenon" was first described by Brockman in 1930.<sup>12</sup> Some patients develop gastrointestinal hemorrhage secondary to erosion of the aneurysm into an adjacent viscus and this event has been described in approximately 13% of patients with a ruptured aneurysm as in stomach, in pancreas, in colon, in the Wirsung duct or the splenic vein creating

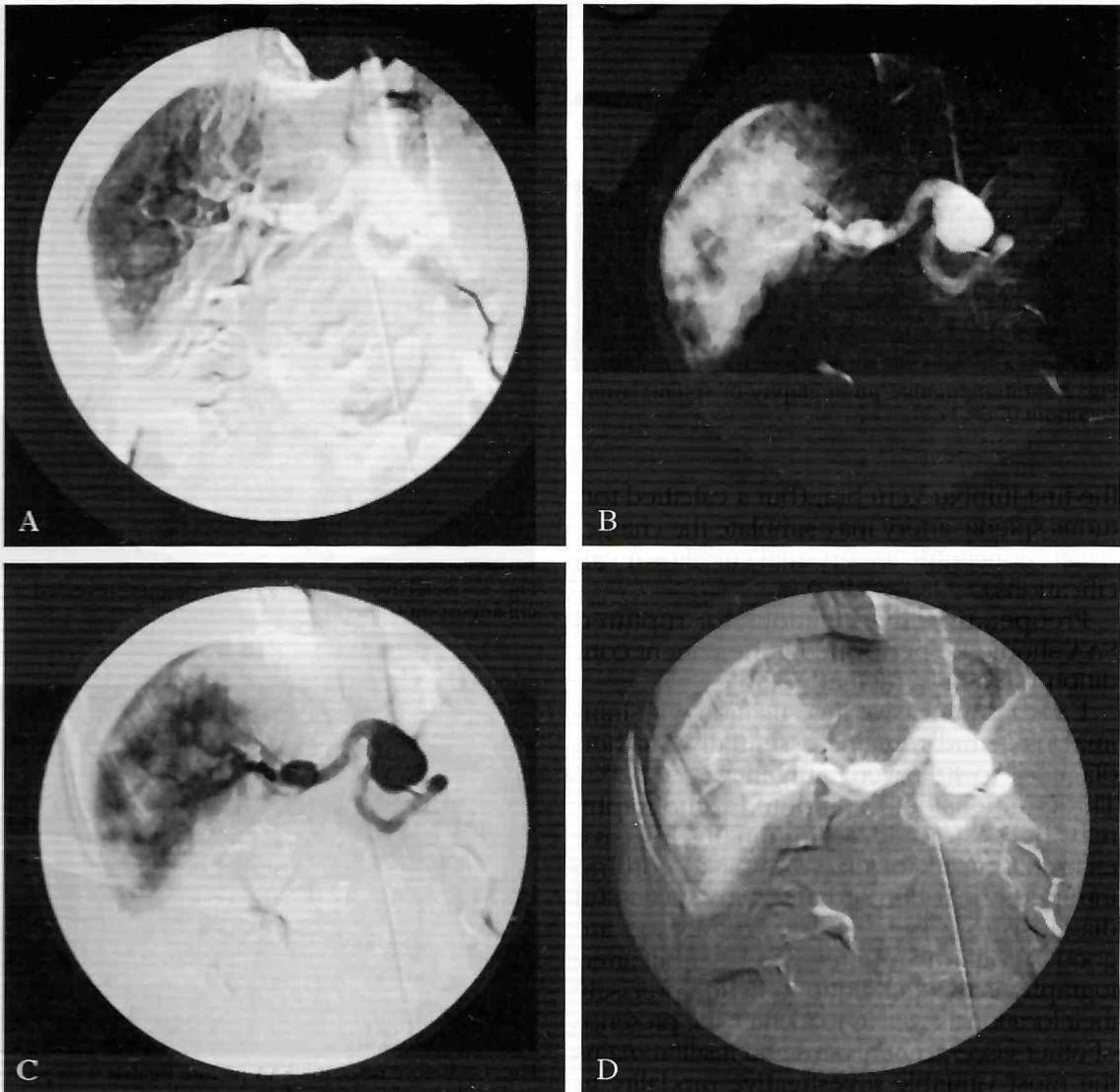


Fig. 1.—Selective splenic artery arteriograms demonstrating a 5 cm saccular aneurysm and one digital subtraction angiogram (Case 1).

an arteriovenous fistula (with associated portal hypertension).<sup>7, 8, 10, 13, 14, 18, 32, 56, 58</sup>

In pregnant patients SAA rupture occurred in the third trimester of pregnancy in 69% and suggested an obstetric emergency such as abruption or ruptured ectopic pregnancy or premature separation of the placenta, or uterine rupture and may cause the immediate death of both mother and fetus.<sup>12, 15, 57, 59</sup>

Rupture not associated with pregnancy has an incidence of 2 to 40%<sup>2, 7-9, 12-16, 35, 36, 60-62</sup> with a high frequency of death in 4 to

36%<sup>3, 7, 9, 10, 47, 51, 54, 55, 61, 63-66</sup> even to 70%<sup>36</sup> and more than 95% in young pregnant women.<sup>67</sup> In asymptomatic patients the risk of rupture is 9.6%.<sup>7</sup>

As already said, these aneurysms are usually identified as incidental findings on diagnostic studies undertaken for other purposes.

SAA are calcified in 72% and are most commonly identified on X-ray film by the presence of concentric calcifications in the left upper abdominal quadrant on the level of

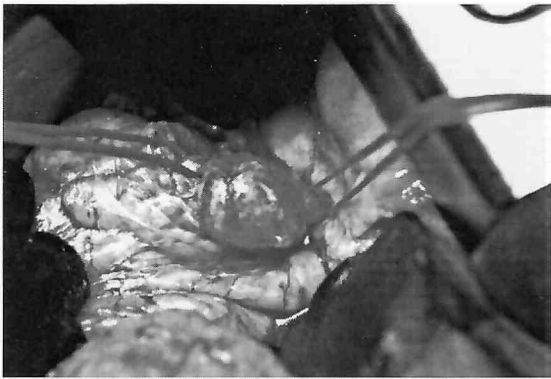


Fig. 2.—Intraoperative photograph of splenic artery aneurysm (Case 1).

the first lumbar vertebra, (but a calcified tortuous splenic artery may simulate the characteristic concentric aneurysmal curvilinear calcifications).<sup>3, 7-10, 12, 13, 18, 25, 68</sup>

Preoperative investigations for ruptured SAA should be performed if the patient condition remains sufficiently stable.

US is a noninvasive investigation with minimal risks and remains essentially on their diagnosis to 40% even in urgency.<sup>3, 5, 25, 41, 55, 69</sup> Three-dimensional computed tomography may be performed as minimally invasive examination to localize SAA or to confirm intraperitoneal hemorrhage with successful diagnosis to 67%.<sup>3, 5, 25, 30, 55, 70</sup> RMN too are good to confirm SAA presence<sup>5, 26, 71</sup> but arteriography is the technique to define precisely their location as well as to identify the presence of other visceral aneurysms and it still remains the most valuable investigative modality to localize the source of bleeding and assess the collateral flow to 100%.<sup>2, 3, 10, 17, 25-27, 40, 43, 55</sup>

Virtual endoscopy enables the creation of endoluminal views by processing spiral TC images, thereby allowing the preoperative and postoperative evaluations of abdominal aortic aneurysms, aneurysms of the celiac and splenic artery, celiac and common iliac arteries.<sup>72</sup>

### Case reports

*Case 1.*—A 68-year-old woman with a history of 2 previous childbirths, complained of upper quadrants abdominal pain. She had no palpable abdominal

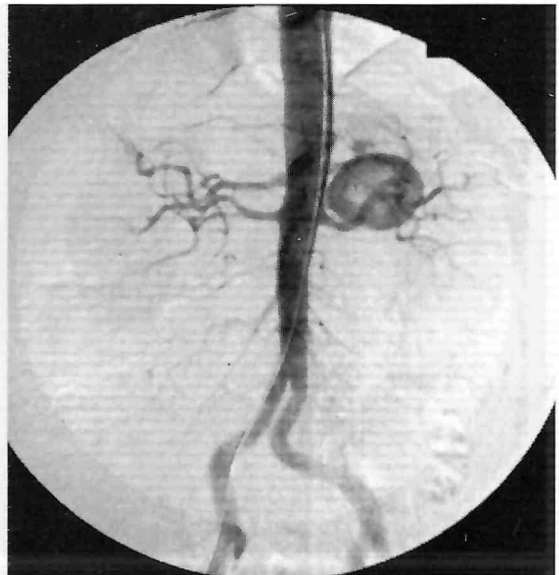


Fig. 3.—Selective celiac artery arteriogram revealed a 3 cm aneurysm (Case 2).



Fig. 4.—Before resection SAA picture treated with proximal and distal ligation of main trunk of splenic artery (Case 2).

mass or vascular bruit. Plain roentgenograms of abdomen showed a calcification: a selective celiac angiography disclosed an aneurysm in the middle third of splenic artery (Figure 1).

Meanwhile lower rectal cancer too was diagnosed in the same patient and the surgical procedure was performed by a traditional laparotomy for a Miles operation. The greater omental sac was opened and the pancreas was exposed: a 5 cm of diameter splenic artery aneurysm protruded from the upper edge of the pancreas (Figure 2). The splenic artery was double ligated and the aneurysm resected out leaving its anterior wall placed on the pancreatic body.

Histological examination showed atherosclerosis of aneurysmal wall and of the splenic artery.

*Case 2.*—A 42-year-old woman who had 2 children was admitted with previous episode of left upper abdominal quadrants pain; by ultrasonography a 3 cm diameter mass was discovered and arteriography showed an aneurysm of the middle third of the splenic artery. At the operation, we saw, in the retrogastric area, a saccular aneurysm located on the body of the pancreas. The peritoneum overlying the aneurysm was incised and the aneurysm was carefully dissected free (Figures 3 and 4) leaving a lozenge of its wall adherent to the pancreas. The splenic artery also was dissected 2 cm proximal and distal to the aneurysm and was double ligated and resected.

As pathological findings, splenic artery fibromuscular dysplasia and atherosclerosis with calcifications of aneurysmal wall were found.

### Discussion and conclusions

There is no unanimity of opinion regarding treatment of SAA and their management still remains controversial, although rupture is the main complication with massive intraperitoneal hemorrhage.<sup>3, 10, 12</sup>

The hemorrhage may be rapidly controlled by dividing the gastrosplenic ligament and reaching the splenic artery through the lesser sac and sometimes cross-clamping of the aorta was required at the beginning of the surgical procedures.<sup>13, 17</sup>

Treatment of SAA is elective in 1-2% of cases and in urgency in 25-65% of cases. If there is a giant aneurysm, operative indication is imperative and if there are dense adhesions to adjacent organs exclusion by proximal and distal ligation is performed.<sup>52</sup> Size, type and location of the aneurysm, patient's age and medical condition at present offer the best guidelines to treatment.<sup>3, 6, 17</sup> Advanced age, even associated with systemic hypertension, should not preclude surgery.<sup>17</sup>

An aggressive approach to the diagnosis and management of this aneurysms is warranted. With an increased index of suspicion and early aggressive treatment of asymptomatic splenic artery aneurysms, mortality has drastically decreased.<sup>11, 12, 17</sup>

Patients presenting with ruptured or symptomatic aneurysm and those more than 2.5 cm in diameter should undergo operation promptly, provided there are no medical contraindications.<sup>2, 8, 10, 12</sup>

Asymptomatic aneurysms discovered in women of childbearing age should be electively resected because the presence of a SAA in a woman of childbearing age represents a serious and potentially life threatening condition.<sup>3, 6-8, 73</sup> Meanwhile treatment of asymptomatic aneurysms <2 cm in diameter is not recommended in the absence of current or contemplated pregnancy.<sup>12</sup> Most splenic artery aneurysms discovered during pregnancy are ruptured (95%),<sup>2, 8, 12, 57</sup> and few women with their neonate survived.<sup>54, 60</sup> Most SAAs that rupture during pregnancy occur during the last trimester of pregnancy with a high maternal and fetal mortality rate because of hormonal changes and increasing blood volume.<sup>18, 29</sup> This increased flow through the SA during pregnancy may lead to rupture, even with a very small aneurysm: treatment may be done before delivery.<sup>3, 7, 8, 10, 12, 13, 33, 74</sup>

It's very difficult to exactly determine the normal arterial size of a young person because their vessels are smaller than those of older patients. Women too have smaller vessels: therefore, if the diameter is twice the size of the proximal artery there is an adequate indication for repair.<sup>30</sup>

A patient of any age with an asymptomatic aneurysm 2 cm or larger in diameter should undergo elective surgery and any patient with an asymptomatic aneurysm documented to be increasing in diameter should have elective surgery. Although the rate of rupture is low the risk of surgical intervention is low and we believe that all SAAs should be treated surgically if the patient is in good health.<sup>12</sup> Some authors indicate treatment even for asymptomatic SAA 1.5 cm in diameter.<sup>7, 12, 17, 27, 40</sup>

In recent years there has been a trend toward conservative management consisting of observation of patients with unruptured SAA.<sup>13</sup> Non operative management in allowing preservation of the immune function of the spleen should be considered, as well as avoiding the morbidity associated with non therapeutic laparotomy identifying risk factors of failure of non operative management.

Other authors documented that SAA less than 2-3 cm may be observed without treatment especially in patients over 60 years old

because in SAA until size 2.5 cm the rupture is very rare: 50% of patients with aneurysms <2.5 cm were followed by some authors without operation and in 10 patients with aneurysms smaller than 2.5 cm followed for 10 years, no aneurysm ruptured.<sup>9</sup> In 18 patients SAAs were followed even to 7 cm of diameter through 11 years without rupture: only 1 occurred in a 52-year-old hypertension woman.<sup>12</sup> In fact a relationship between size and risk of splenic aneurysm has not been established.<sup>2, 3, 7, 8, 10, 12, 17, 18, 24, 27, 60</sup>

Celiac angiography is recommended prior to liver transplantation: if a SAA is found, ligation of the splenic artery should be performed at the time of transplantation to prevent a possible rupture.<sup>75</sup> The rupture of aneurysm of the splenic artery, despite laparotomy in emergency, is often fatal. The mortality after rupture increased to 75-95%<sup>2, 8, 12, 13, 15, 18, 24, 31, 59, 76</sup> while only 1% mortality occurred as a direct result of elective aneurysmectomy<sup>2, 8, 12, 31, 46</sup> The natural growth rate of asymptomatic splanchnic AA is unknown. Although calcification of the aneurysms has traditionally been described as a clinical marker for increased risk of rupture, this has never been shown conclusively:<sup>13</sup> if they are calcified they are well followed with abdominal roentgenography.<sup>12</sup>

It's unclear if it occurs more in hypertension in old age,<sup>8</sup> but for some authors asymptomatic aneurysms in patients less than age 60 with hypertension must be treated.<sup>40</sup>

The choice of operation is determined by location of the aneurysm.<sup>3, 8, 67</sup>

If it is located in the distal third of the splenic artery or at the splenic hilum away from the pancreas, the aneurysm is resected with either the spleen or the spleen and left pancreas if they cannot be saved;<sup>3, 8, 12, 13, 33, 46, 67</sup> if resection of the spleen is necessary, the patient should be warned of the possibility of overwhelming postsplenectomy sepsis and immunizes against pneumococcal infection.

When it is located in the middle third of the splenic artery, given the importance of the spleen in maintaining host resistance, some authors recommend resection with reconstruction of main artery.<sup>26, 36, 77</sup> Endo-

aneurysmorrhaphy is a possible alternative, especially with saccular aneurysms.<sup>25, 62</sup> Simple suture obliteration or excision as performed in our patients need to be considered<sup>67</sup> and if the aneurysm is embedded in the pancreas without resecting the pancreas and/or the spleen:<sup>8, 12, 13, 46, 74</sup> short gastric vessels will prevent an infarction of the retained spleen.<sup>8, 12, 13, 33, 67, 74</sup>

If the aneurysm is proximal it may be treated with aneurysmectomy with proximal and distal ligation without splenectomy.<sup>3, 28, 33, 34, 43, 46, 67</sup>

Splenic preservation however carries a risk of splenic infarction or abscess formation and subsequently the patient may require splenectomy<sup>30, 63, 78-80</sup> as in few cases treated by transcatheter embolization.<sup>32</sup> For some authors the preservation of the splenic artery by repairing the arterial aneurysm does not offer additional benefit to patients and it's not cost-effective to monitor these patients on a long term basis for the possible recurrence of SAA.<sup>81, 82</sup>

Laparoscopic SAA ligation repair appears to be optimal and it's particularly useful for aneurysms distant from the pancreas and protruding from the splenic artery:<sup>43, 67, 70, 76, 82-84</sup> it's successfully performed, according to Roeder's ligation technique.<sup>85</sup> Laparoscopic surgery is gaining increasing interest because clinical recovery is rapid as a result of minimal surgical invasion and for cosmetic and economic advantages because there was correlation in morbidity with operative time, postoperative stay, transfusions and number of trocars of minimally invasive surgery.<sup>26</sup> Authors report a successful laparoscopic approach to treat SAAs with resection of aneurysm in 4 patients with a vascular endoGIA with an average age of 55 and an average size with 3.2 cm; intraoperative ultrasound scanning with Doppler was used to localize the aneurysm and to identify all feeding vessels; the complete cessation of flow within the aneurysm in the case in which the feeding vessels were simply ligated was also documented.<sup>86</sup> Authors think repairing the aneurysm laparoscopically is not the optimal procedure, that is likely to repair just part of the aneurysmal artery, because, although many solitary aneurysms are saccular, there is still a high prob-

ability that a part of aneurysmal artery is being left behind, which may contribute to recurrence of aneurysm.<sup>81</sup> Endovascular intervention as percutaneous embolization has recently gained popularity that transcatheter embolization is the treatment of choice for pseudoaneurysm and has been used more frequently: to day embolization offers a safe and effective alternative or adjunctive therapy to surgery if only one aneurysm is found. Intraluminal occlusion by the introduction of steel coils and other thrombogenic material can be considered for patients presenting high operative risk.<sup>87</sup> Superselective embolization may be performed with Gelfoam sponges (absorbable gelatin sponge), with Ivalon (polyvinyl alcohol particles of 500  $\mu$  size), with stainless steel coils or tungsten coils (0.018 inch diameter) or with combinations thereof.<sup>3, 6, 7, 25, 32, 64, 88-92</sup> Follow-up digital subtraction arteriography was obtained to confirm satisfactory embolization and prospective studies are also needed to assess the efficacy of embolization and the incidence of bleeding or rebleeding and recanalisation.<sup>6, 55, 89, 93</sup> The main splenic artery at the level of the aneurysm is so occluded with sometimes preservation of the spleen,<sup>90, 94</sup> but when it is occluded there is a risk of splenic abscess formation.<sup>32</sup> Transcatheter embolization of aneurysm and feeding vessels down 3 mm may also be an adjunct to surgery as a temporary control to stop hemorrhage, clear up the sepsis and go back at later date to make much better elective operation.<sup>30</sup> For other authors transcatheter embolization is not relevant in rupture of true SAA, since the hemorrhage may be rapidly and safely controlled by surgery: they affirm rupture requires instant surgery and only in the elective phase recommend embolization.<sup>17, 26, 53, 90</sup> Advantages of transcatheter embolization include precise localization of the aneurysm, assessment of collateral flow, lower risk for patients who are not good operative candidates, and easier approach to aneurysms for which surgical exposure would be difficult as small aneurysms difficult to locate.<sup>30, 32</sup> This technique is associated with the risk of total occlusion of the main trunk of the SA especially when the SAA origin is not constricted.

Therefore endovascular SAA repair may be preferable only in selected patients in whom operative therapy entails a high risk.<sup>3, 74, 87, 95</sup> The embolization has some complications as splenic infarction, embolization of other visceral arteries as hepatic artery, sepsis and abscess as pyogenic liver abscess, arterial rupture, toxicity for kidney, incomplete occlusion with recanalization and the dislodgement of the occlusion device.<sup>55, 89, 95, 96</sup> It seems that if embolization is performed in more times the risk of splenic infarction is reduced.<sup>89</sup> Some authors demonstrated that when splenic artery has been embolized proximally, these patients may remain anatomically splenic, but may not retain any splenic function and if they do not, they must be protected against the long term complications of asplenia.<sup>91</sup> If no main splenic artery embolization is performed, but rather branch embolizations, the spleen is preserved in function in at least 50%.<sup>3</sup>

The development of prosthetic devices for use during percutaneous intervention has ushered in a new era. Current available devices for endovascular repair of abdominal aortic and visceral aneurysms require a femoral artery cutdown for insertion. However in the future instrumentation will likely improve to where the procedure can be done percutaneously and this will become a reality in the 21<sup>st</sup> century.<sup>97</sup>

### Riassunto

*La terapia degli aneurismi (veri) dell'arteria splenica. Due casi clinici e revisione della letteratura*

Sono riportati alcuni casi di aneurisma dell'arteria splenica, che oggi sono diagnosticati con maggior frequenza anche per l'uso corrente di ecografie, TC, RMN nel rilevamento anche di altre patologie. La loro importanza sta essenzialmente nel fatto che possono essere causa di emorragie importanti, ma il loro trattamento rimane a tutt'oggi controverso. I nostri 2 casi sono stato trattati, in elezione, con la resezione dell'aneurisma situato in ambedue nel tratto medio del vaso. Alcuni Autori hanno dimostrato che quando l'arteria viene legata o embolizzata, la milza non sarà più funzionalmente attiva. La legatura laparoscopica dell'arteria splenica sembra essere il miglior trattamento per gli aneurismi dell'arteria splenica che sporgono visibilmente dal pancreas ed è certamente di

notevole interesse clinico in quanto il ricovero è di breve durata con bassa morbilità e costi inferiori, oltre che con migliori risultati estetici. L'embolizzazione percutanea in corso di emorragia offre un controllo temporaneo e si può rimandare l'intervento chirurgico a quando le condizioni cliniche saranno migliorate. Anche il trattamento endovascolare oggi è preso in seria considerazione: per tale procedura speriamo di avere una strumentazione sempre migliore, in modo da poter trattare elettivamente questa patologia per via percutanea.

Parole chiave: Arteria lienale - Aneurismi, diagnosi - Aneurismi, chirurgia - Embolizzazione terapeutica.

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