

# Long-Term Efficacy of Partial Splenic Embolization in Children

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**Purpose:** To elucidate the role of partial splenic embolization (PSE) procedures, long-term outcome was assessed in terms of the recurrence of thrombocytopenia.

**Methods:** A retrospective study was performed after 41 PSE procedures in 36 patients for hypersplenism owing to portal hypertension. The underlying disease was biliary atresia in 32 patients, extrahepatic portal obstruction in 3, and idiopathic cirrhosis in 1.

**Results:** The average volume embolized was 70.1%. The patients were followed up from 20 days to 182 months (average, 70.8 months). Five patients subsequently died, and 6 underwent liver transplantation. The causes of death or the reasons for liver transplantation were not related to hypersplenism. Eleven patients (30.6%) had recurrence of thrombocytopenia ( $<100,000/\text{mm}^3$ ). There was no significant dif-

ference in the volume embolized or platelet count before PSE between the patients with and without recurrence of thrombocytopenia. The peak value of platelet count after PSE was significantly lower in the patients with recurrence of thrombocytopenia ( $P = .0091$ ). In 17 of 24 survivors without liver transplantation, platelet counts remained normal throughout the follow-up period.

**Conclusions:** PSE is a safe and effective procedure. Hematologic indices improved in all 36 patients after PSE, and its long-term efficacy was shown in 70% of the survivors.  
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**INDEX WORDS:** Partial splenic embolization, portal hypertension, hypersplenism, biliary atresia.

**H**YPERSPLENISM is one of the major complications of portal hypertension. Splenectomy had been a treatment of choice for severe hypersplenism owing to portal hypertension, but because of the risk of overwhelming sepsis after splenectomy, partial splenic embolization (PSE) has been used widely in patients with severe hypersplenism especially in small children. However, long-term efficacy of PSE is still unknown. In this report, we assessed retrospectively the long-term outcome of patients who underwent PSE.

## MATERIALS AND METHODS

From 1984 to 2001, 36 patients underwent a total of 41 PSE procedures in Tohoku University Hospital. PSE was performed 3 times in 1 patient, twice in 3, and once in the remaining 32 patients. The indications for PSE in our institution were as follows: (1) the patient had splenomegaly caused by hypersplenism secondary to portal hypertension; (2) platelet counts were less than  $100,000/\text{mm}^3$  with ongoing thrombocytopenia; (3) the patient had bleeding complications such as epistaxis and gastrointestinal bleeding. We performed PSE in 36 patients who fulfilled all 3 of these criteria. The 36 patients (16 boys

and 20 girls) underwent initial PSE at an average age of 7.9 years (range, 15 months to 22 years).

The mean follow-up period after PSE ranged between 2 months and 15 years (average, 5.9 years). The underlying diseases were biliary atresia in 32, extrahepatic portal obstruction in 3, and idiopathic liver cirrhosis in the remaining 1 patient.

Our previously reported PSE procedures were basically the same as that described by Spigos et al.<sup>1,2</sup> The percentage of embolized volume was precisely evaluated using technetium 99m Sn colloid splenic scintigram.

Follow-up periods, embolized volumes, and the changes in platelet counts were compared between the groups with and without recurrence of thrombocytopenia ( $<100,000/\text{mm}^3$ ). The requirements for a second PSE and liver transplantation and the clinical outcome of each patient were also assessed.

In the statistical analysis,  $\chi^2$  test for categorical variables and Mann-Whitney test for continuous variables were used, and a  $P$  value less than .05 was considered statistically significant.

## RESULTS

Of the 36 patients, 11 (30.6%) had recurrence of thrombocytopenia after the first PSE. The mean follow-up period was essentially the same between groups with and without recurrence of thrombocytopenia (5.9 years).

The percent embolized splenic volume was not statistically different between patients with and without recurrence of thrombocytopenia (68% v. 71%, not significant). Platelet counts before PSE in patients with and without recurrence of thrombocytopenia were  $61,200/\text{mm}^3$  and  $62,600/\text{mm}^3$ , respectively (not significant). The platelet counts significantly increased after PSE in all patients.

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**Table 1. Comparison Between Groups With and Without Recurrence of Thrombocytopenia After PSE**

	Recurrence	Nonrecurrence	P Value
No. of Cases	11	25	
Outcome			
Alive without liver transplantation	6	19	
Dead	1	4	Not significant
Liver transplantation	4	2	
Follow-up period (mean $\pm$ SD, mo)	6.0 $\pm$ 2.7	5.9 $\pm$ 4.2	Not significant
Embolized volume (mean $\pm$ SD, %)	68 $\pm$ 13	71 $\pm$ 14	Not significant
Plt pre-PSE (mean $\pm$ SD, $\times$ 1,000/mm <sup>3</sup> )	61.2 $\pm$ 2.1	62.6 $\pm$ 19.4	Not significant
Peak Plt (mean $\pm$ SD, $\times$ 1,000/mm <sup>3</sup> )	275.6 $\pm$ 92.2	453.1 $\pm$ 194.1	<.01
Final Plt (mean $\pm$ SD, $\times$ 1,000/mm <sup>3</sup> )	75.2 $\pm$ 21.0	180.0 $\pm$ 75.0	<.0001

The peak value of platelet counts of patients without recurrence of thrombocytopenia was significantly greater than that with recurrence (453,100/mm<sup>3</sup> v. 275,600/mm<sup>3</sup>;  $P < .01$ ; Table 1).

In follow-up, 5 patients died, and 6 underwent liver transplantation. The causes of death in 4 patients with biliary atresia were progressive liver failure, whereas the one with idiopathic liver cirrhosis had fatal portopulmonary hypertension at the age of 22 years. Liver transplantations for the 6 patients were performed for severe liver cirrhosis. These 11 patients included 5 with recurrence of thrombocytopenia, but their causes of death or reasons for liver transplantation were not related to hypersplenism.

Twenty-five patients survived without liver transplantation after PSE, including 6 patients (24%) with recurrence of thrombocytopenia.

Of the 11 patients with recurrent thrombocytopenia, 10 had the recurrence within 5 years after the initial PSE. The remaining patient, who had recurrence of thrombocytopenia 10 years after PSE, had an association with hepatopulmonary syndrome and required liver transplantation. Four of the 11 patients who had recurrent thrombocytopenia underwent second PSE, but all 4 patients had recurrence of thrombocytopenia again within 5 years after the second PSE.

There was no correlation between the final value of the platelet counts and the follow-up period. Thus, the platelet counts did not seem to decrease with time after PSE.

A third PSE was performed in 1 patient. He had severe gastric varices as well as recurrent thrombocytopenia at the age of 21 years. The celiac angiogram showed that the variceal blood flow was mainly supplied through short gastric veins. To reduce the variceal blood flow, the third PSE was performed. The gastric varices improved after PSE, and no variceal bleeding has been encountered for 3 years since.

The original diseases of patients who suffered recurrent thrombocytopenia were biliary atresia in 8 patients, extrahepatic portal obstruction in 2, and idiopathic cirrhosis in one. Four of these 8 patients with biliary atresia

survived without liver transplantation, but they had moderate or severe liver dysfunction.

## DISCUSSION

Because the risk of overwhelming sepsis after splenectomy is well known, especially in small children,<sup>3</sup> PSE is a reasonable alternative in patients with severe hypersplenism associated with portal hypertension.<sup>4</sup> We believe PSE should be used rather than splenectomy even in older patients with biliary atresia, because splenectomy may yield excessive adhesions and interfere with liver transplant operation, which might be required later.

Long-term efficacy of PSE and factors influencing the recurrence of hypersplenism after PSE in children still are controversial. According to Kumpe et al<sup>5</sup> splenic regeneration after PSE may occur more frequently in children than in adults. However, according to the long-term follow-up data by Brandt et al<sup>6</sup> hematologic indices returned to and remained normal throughout follow-up in 10 of 13 patients with hypersplenism. Shah et al<sup>7</sup> reported that improvement of the blood picture had been sustained in 7 of 8 patients with initial improvement during the follow-up period from 4 months to 7 years after PSE. The current study showed that 70% of the patients had maintained platelet counts greater than 100,000/mm<sup>3</sup> for more than 5 years after PSE. Although portal hypertension might improve in some patients without recurrence of thrombocytopenia, there was no tendency indicating that patients with ongoing liver fibrosis are likely to have recurrence of thrombocytopenia. Regeneration of the spleen might be prevented by the fibrotic change of the splenic tissue after PSE, but its detail mechanisms are still unclear.

Prediction of the outcome of PSE is an interesting issue from the clinical point of view. Regarding the infarction rate, Sangro et al<sup>8</sup> reported that hypersplenism relapsed in patients who exhibited an embolization of less than 50% of the splenic mass. But we did not see any differences in infarction size between groups with and without recurrence of thrombocytopenia after PSE in the

current long-term study. Four patients who had recurrence of thrombocytopenia after the initial PSE underwent a second PSE, but thrombocytopenia recurred again in all of them within 5 years after a second PSE. The recurrence of thrombocytopenia seemed to be dependent not on the severity of liver pathology but on the regenerative potential of the spleen. Obatake et al<sup>9</sup> reported nonembolized volumetric evaluation was useful in predicting the functional outcome of PSE. Our study showed the peak value of platelet counts was predictive of the long-term outcome of PSE.

Gastrointestinal bleeding is a major sequel of hypersplenism. Although most cases of esophageal variceal bleeding can be treated by endoscopic sclerotherapy

or ligation,<sup>10</sup> treatment for bleeding from the small intestine is much more difficult. We experienced 2 cases of small intestinal bleeding associated with severe hypersplenism, and PSE was quite effective in controlling the bleeding in both cases. PSE may be a good option to control intestinal bleeding in patients with hypersplenism in whom endoscopic treatment is difficult or impossible.

Hematologic indices improved in all 36 patients after PSE, and its long-term efficacy was shown in 70% of the survivors. Long-term efficacy was not expected in patients with a peak value of platelet counts less than 400,000/mm<sup>3</sup> after PSE, and the anticipated benefit of the second PSE might be limited in such patients.

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