AUTOTRANSPLANTATION OF SPLEEN: SPLENOSIS

CASE REPORT AND PRELIMINARY REPORT OF AN EXPERIMENTAL STUDY IN REVASCULARIZATION OF THE HEART*

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THE ENTITY OF autotransplantation of the spleen, splenosis, is sufficiently rare to warrant publication of a case. This paper will present a case report and a preliminary report of an allied experimental study.

History. The first known reference to the condition of autotransplantation of splenic tissue following rupture of the spleen was made by von Küttner[11] in 1910. He regarded these as accessory spleens and not as implants. Faltin[6] was the first to suggest that these splenic nodules were actually splenic implants.

Albrecht,[1] in 1896, and Schilling,[16] in 1907, reported cases with splenic foci, 400 and 42 respectively, dispersed throughout the peritoneal cavity. Although reported as cases representing accessory spleens, the distribution and number are more consistent with autotransplantation of splenic tissue. A history of trauma was lacking in each report.

Accessory Spleens. Shaw and Shafi[17] distinguished splenic implants from accessory spleens (spleniculi) by their greater number, widespread location and history of trauma. Accessory spleens are usually located in the gastrolienal ligament, but may be found about the pancreas and rarely in the omentum. They are usually two to six in number, but as many as 40 have been found. Accessory spleens are common congenital anomalies.

Splenosis. The term splenosis, autotransplantation of splenic tissue, was first introduced by Buchbinder and Lipkoff[3] in 1939. They described the first case in this country. Microscopic examination of splenic implants show that the blood vessels do not enter through a hilus as is true of accessory spleens, but rather penetrate the capsule around its circumference. The histological pattern is essentially the same as normal spleen.

Parenchymatous cells of no other abdominal organ exhibit similar characteristics except possibly those of uterine endometrium.[8]

For reviews of the literature, the reader is referred to the reports of Shaw and Shafi,[17] Buchbinder and Lipkoff,[3] Hamrick and Bush, [8] Waugh,[21] and recently by Storsteen and ReMine,[19] who collected 22 cases from the literature and added one of their own. The case herein reported will be the twenty-fourth.

Waugh's case is the first report of autotransplantation in an adult following rupture of the spleen and splenectomy. The case reported by Roettig, Nusbaum and Curtis[15] is of interest since intestinal obstruction was attributed to splenic implants. The case report of Stobie[18] deserves comment. Although reported as a case of recurrent congenital hemolytic anemia due to splenosis following splenectomy, several aspects warrant analysis. That is to say, the etiology of the patient's jaundice, anemia

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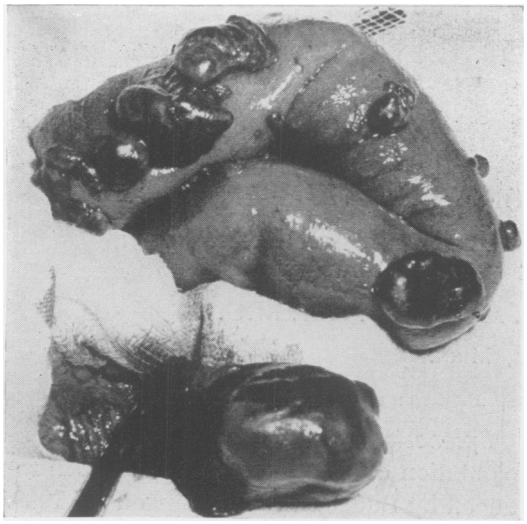

FIG. 1. Photograph of small intestine at time of laparotomy showing multiple splenic nodules of varying sizes on antemesenteric borders of jejunum.

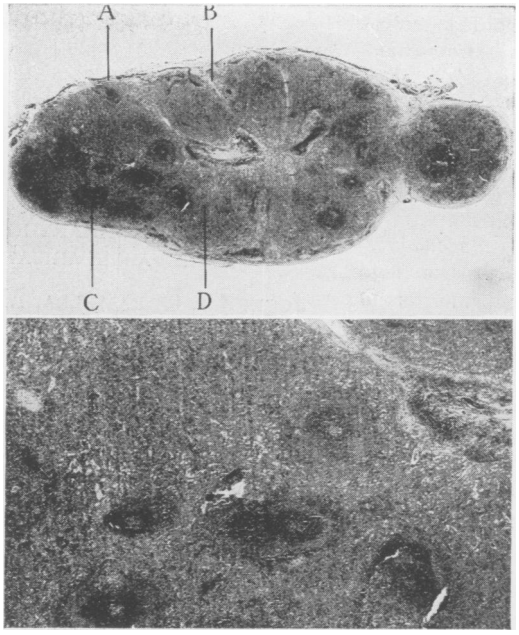

FIG. 2. Photomicrograph (Magnification x 4.6) of one of the splenic nodules removed at surgery. Note the outer capsule (A), Trabecula (B), Follicle (C), Red Pulp (D).

FIG. 3. Photomicrograph (x 23) of same splenic nodule showing follicles and red pulp in more detail.

and splenomegaly is not entirely clear. From the data given, one may conclude that the patient had cholelithiasis, acute cholecystitis, choledocholithiasis, and a stricture of the common duct which required a secondary operation. The diagnosis of congenital hemolytic anemia from the data given is not supported by spherocytosis, fragility tests, nor by a positive family history. The contention, therefore, of those who maintain that splenosis does not follow rupture of a diseased spleen,[5, 11, 17, 21] requires clarification and may remain tenable.

CASE REPORT

A 22-year-old white male (R#94170) was admitted on August 15, 1949, with a 6-day history of pain in the right lower quadrant associated with nausea and vomiting for 36 hours prior to admission. He took castor oil with no relief. The day prior to admission, the pain became severe and he vomited 3 to 4 times. Splenectomy had been previously performed on an undisclosed date for rupture of the spleen following a fall from a horse.

The patient weighed 140 pounds on admission. He was not in acute distress. Physical examination disclosed: T, 37.2° C; P, 82; R, 18; B.P., 110/70. Pertinent findings were the presence of pain, tenderness and spasm in the right lower quadrant. Rectal examination disclosed marked tenderness on the right side.

Laboratory Data: Hemoglobin, 15.3 Gm.; W.B.C., 15,600, 82 per cent neutrophils, 17 per cent lymphocytes, 1 per cent eosinophils. Urinalysis was normal; chest roentgen ray was also normal. The clinical diagnosis was acute appendicitis and laparotomy was performed. The appendix was found to be acutely inflamed. In addition there were multiple, reddish-brown nodules of various sizes on the antemesenteric border of the jejunum, on the right lateral peritoneal wall, and one on the meso-appendix. Appendectomy was performed and two nodules were removed for histologic study.

Pathologic Report. Gross Appearance: Specimen No. 1. Consists of an appendix 7 cm. in length to which is attached a firm, matted meso-appendix. On cut section the appendix is found to be kinked at the distal end and the distal portion is dilated and filled with purulent material. The proximal end shows no dilatation of the lumen, but the serosal surface is involved in the inflammatory process.

Specimen No. 2. Consists of two lobulated pieces of tissue, one measuring 2 x 1.6 x 0.5 cm.; the second measuring 9 mm. in greatest diameter.

On cut section, the tissue is firm and has a lightly mottled, brownish color.

Microscopic Appearance: Specimen No. 1. The mucosa is absent at the tip of the appendix. The lumen is packed with polymorphonuclear cells. The acute inflammatory exudate shows extensive infiltration throughout all layers of the appendix, but is most marked in the region of the mesoappendix.

Specimen No. 2. Sections through the nodule reveals an outer capsule with trabeculae extending from this into the underlying parenchymal tissue. The parenchyma contains scattered lymph follicles separated by tissue having the histological appearance of the red pulp of the spleen with cords of Billroth separating sinusoids that are filled with erythrocytes.

Microscopic Diagnoses. The microscopic diagnosis was (1) acute suppurative appendicitis; (2) splenic implants.

Experimental Splenosis. Experimentally, splenic tissue may be implanted and readily survives. Von Stubenrauch[20] removed the spleen in several dogs and seeded the crushed pulp throughout the peritoneal cavities. Sacrifice of the animals 1 to 3 months later showed multiple implants throughout the peritoneal cavity. Kreuter[10] performed the same experiments in monkeys, obtaining similar results. Jarcho and Anderson[9] reported cases of dogs that had sustained severe abdominal trauma showing multiple splenic implants at autopsy. They stated that Griffini and Tizzoni,[7] as early as 1883, noted spleen-like nodules in the peritoneum following partial splenectomy in dogs. Marine and Manley,[12] Roettig, Nusbaum and Curtis,[15] and Williams,[22] successfully implanted splenic tissue in rabbits, the graft sites being the subcutaneous tissues of the abdomen, peritoneum and ears, respectively. Perla[13] transplanted splenic tissue in the abdominal wall of albino rats. Putschar[14] transplanted splenic tissue in the peritoneum of rats while Calder[4] grafted splenic tissue in the omentum of white mice and albino rats.

Experimental Study. Since splenic tissue is rich in vascularity and serves as a blood reservoir, it was conjectured that this structure, if successfully implanted upon the pericardium, might be effective in protecting the heart with coronary artery insufficiency following the "blood bath" principle of Beck.[2]

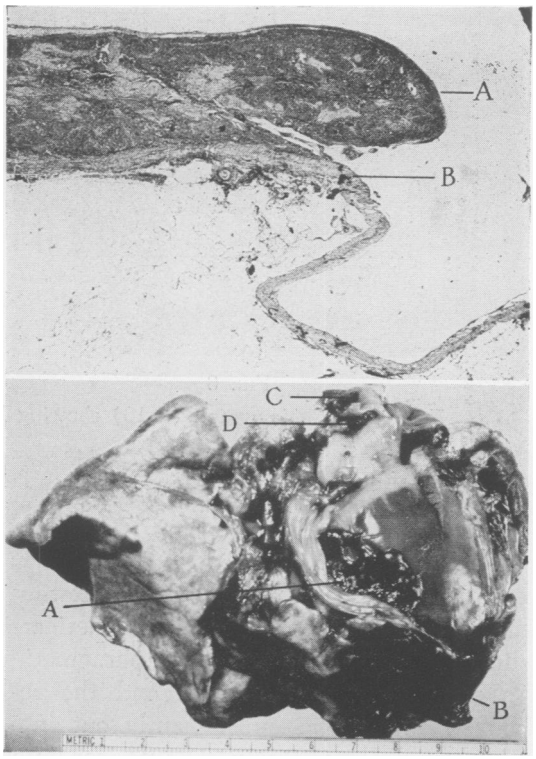

FIG. 4. Photograph of heart-lung specimen of dog. Splenic tissue implanted 23 days prior to exploration. Note layer of splenic tissue growing between epicardium and pericardium (A). The pericardium and epicardium at the apex are fused by a layer of splenic tissue (B). Isolated splenic implants as (C) and (D) are also present.

FIG. 5. Photomicrograph (x 16) showing layer of splenic tissue (A) implanted on pericardium (B).

It was the object of this study, therefore, to determine whether or not splenic tissue could be successfully implanted upon the pericardium of experimental animals.

Method. Dogs not older than one year were used as experimental subjects. Anesthesia consisted of intravenous nembutal, 14 mg. per pound of body weight. An endotracheal tube was employed and connected to a mechanical respirator. Thoracotomy was performed through the seventh inter-

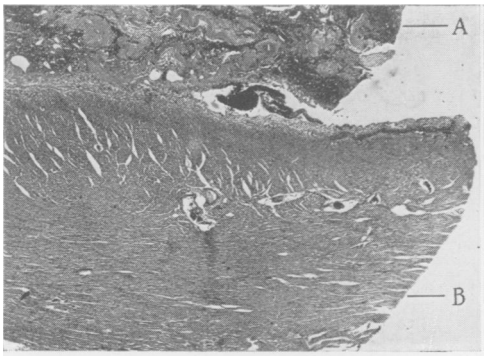

FIG. 6

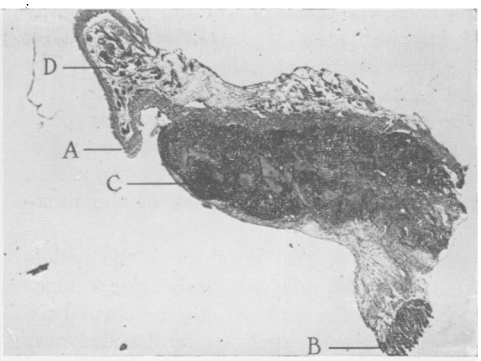

FIG. 7

FIG. 6. Photomicrograph (x 16) showing layer of splenic tissue (A) implanted on myocardium (B).

FIG. 7. Photomicrograph (x 10) of biopsy done 28 days after implantation of splenic tissue in pericardial sac. Note bridging of pericardium (A) and myocardium (B) by splenic tissue (C). Also note the vascularity of the extra pericardial fat (D).

costal interspace on the left, facilitating access to the diaphragm and the pericardium at the same time. Transdiaphragmatic splenectomy was performed and the removed spleen was subsequently cut into fine particles, 1 to 2 mm. The pericardium was incised vertically for a distance of 3 to 4 cm., and approximately half of the finely cut splenic tissue was introduced into the pericardial sac. The pericardium was closed only tightly enough to prevent escape of splenic tissue. Penicillin, 500,000 units, was introduced into the hemithorax and the chest closed. The pericardium was explored three to four weeks later.

Comment. From the data in Table I, it seems clear that splenic tissue can be successfully and consistently implanted upon the epicardium and pericardium of dogs. The coalescence of implants occurs at the dependent portion of the pericardial sac, overlying the left ventricle, principally. Deliberate attempts to produce a more diffuse implantation of splenic tissue and bridging of the epicardium and pericardium by employing abrasion, thrombin solution and gel foam met with only a slight measure of success in these few experiments. The splenic tissue in these latter instances histologically showed a significant increase in fibrous tissue elements. Autopsy of dog number 188 showed bilateral hydrothorax, bilateral atelectasis, compression type, pericardial effusion, and congestion of the liver. Histological study disclosed an acute inflammatory reaction of the splenic tissue in the pericardium. It is felt that this animal developed an infection of the pericardium and succumbed to heart failure secondary to cardiac tamponade.

DISCUSSION

One can only speculate as to the practical application of splenic tissue implanted upon the epicardium and pericardium as a means of revascularizing the heart. The vascular character of splenic tissue and its reservoir function are well recognized, and it would seem that an actively growing structure such as this, bridging the epicardium and pericardium, diverting the extra coronary blood supply, may be more effective than the granulation tissue of surgically induced adhesive pericarditis, the natural history of which is retraction and scar formation. A method to induce a diffuse and uniform implantation of splenic tissue with bridging of the epicardium and pericardium would be desirable. An artificial

TABLE I.

Dog No.	Operation	Exploration	Presence of Implants	Distribution	Bridging of Epicardium and Pericardium by Splenic Tissue
70	3- 4-53	4- 1-53	Yes	Overlying left ventricle, dependent portion area 3.5 x 2.0 cm. Also scattered implants 0.5+ cm.	Yes. At site where pericardium incised initially. Confirmed by biopsy.
62	3-11-53	4- 6-53	Yes	Essentially same as Dog No. 70.	Yes. Essentially same as Dog. No. 70. Confirmed by biopsy.
132	3-18-53	4-22-53	Yes	Scattered implants on epicardium and pericardium.	No.
159	4-29-53 *1	5-22-53	Yes	Pancake layer of splenic tissue most dense over the left ventricle.	Yes. Diffuse bridging overlying left ventricle.
188	5- 6-53 *2	5-14-53 Died	Yes	Extensive.	Yes. Diffuse.
146	5-13-53 *3	6- 5-53	Yes	Pancake of splenic tissue overlying left ventricle.	Yes. Overlying left ventricle. See Photograph.
162	5-20-53 *4	6-11-53	Yes	Layer overlying left ventricle with scattered nodules overlying pericardium of right ventricle.	Yes.

Attempting to induce diffuse implantation of splenic tissue, the operative procedure was modified in the last four experiments.
*1. Pericardium abraded and sterile talc introduced.
*2. Epicardium and pericardium abraded with gauze. Thrombin solution introduced into pericardial sac.
*3. Epicardium and pericardium abraded. Splenic tissue mixed with gel foam.
*4. Epicardium and pericardium abraded. Thrombin solution introduced into pericardial sac.

means of producing an effective hyperplasia of implanted splenic tissue may be practical. The long term behavior of splenic tissue in the pericardial sac bears study. The degree of protection afforded the myocardium by implanted splenic tissue is being pursued.

SUMMARY

1. The subject of splenosis is briefly reviewed and a case report is presented.

2. Instances of experimental splenosis are cited.

3. The preliminary report of the experimental study, transplantation of splenic tissue into the pericardial sac, is presented.

4. In all experimental animals studied (seven), splenic tissue was successfully implanted.

5. The protection afforded the myocardium by splenic tissue is being studied.

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