Recurrent "Pericardial Pain" after Pericardiectomy for Recurrent Acute Benign Pericarditis

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THE CONCEPT of pericardiectomy as a means of definitive therapy in stubborn cases of recurrent acute benign pericarditis was introduced by Zinsser et al. in 1959.1-3 Since then several others have also reported their results with this procedure.4-6 Altogether, a total of 12 cases of pericardiectomy have been reported with the longest recorded follow-up being 5 years. In no case was there a recurrence of "pericardial type" pain after surgery. In all cases, the treatment effected a complete cure. The following two cases were encountered in the space of a 7-month period and were characterized by patterns of recurrent "pericardial type" pain after pericardiectomy—a previously unreported occurrence.

Case Reports

Case 1

P.H., a 17-year-old white male, was first admitted to Mount Sinai Hospital in Minneapolis on July 27, 1964, complaining of chest pain of several days' duration. The pain was located substernally with radiation into both shoulders; it was sharp in nature and was made worse by lying flat and by deep inspiration. About 1½ weeks previously he had had a transient episode of sore throat and rhinorhea. There was no known contact with infectious disease. Physical examination revealed a healthy appearing white male with a temperature of 100.8° F. and a pulse of 100. The throat was mildly inflamed, and there was a small amount of exudate on the left tonsil. A classical pericardial friction rub was heard along the left sternal border. The patient was treated with bed rest and analgesics and became clinically well within 4 days, although the pericardial rub persisted several days longer. The following laboratory data were negative or normal: hemoglobin, leukocyte count and differential, urinalysis, blood urea nitrogen, fasting blood sugar, serum glutamic oxaloacetic transaminase, cholesterol, VDL, 1/1000 old tuberculin, histoplasmin and coccidioidin skin tests, cold agglutinins, stool culture for virus, acute and convalescent complement-fixation titers for mumps, lymphocytic choriomeningitis, and western equine encephalitis, Toxoplasma antibodies, throat culture, and chest x-ray. The only abnormal data were a sedimentation rate of 41 and an electrocardiogram showing mild ST elevation in several leads.

On September 20, 1964, the patient returned with a 3-day history of recurrent pericardial pain and was found to be in acute tamponade with a temperature of 102 degrees. An emergency pericardiocentesis was done, and 230 ml of cloudy yellow fluid were removed with relief of symptoms. By September 28, 1964, the patient was asymptomatic. The following laboratory data were negative or normal at this time: hemoglobin, serum proteins, antistreptolysin-o titer, LE preparation, C-reactive proteins, latex fixation, VDL, blood cultures, and fluorescent antinuclear antibodies. The initial sedimentation rate was 73, subsequently falling to 17; the initial leukocyte count was 15,500 with 86 per cent neutrophils, subsequently falling to normal. The urinalysis showed 4 to 6 white blood cells per high-power field and a trace of albumin. Studies of the pericardial fluid revealed a specific gravity of 1.021, a protein of 5 Gm. per cent, a leukocyte count of 43,750 per mm.3 (with 94 per cent polymorphonuclear neutrophils and 6 per cent mononuclears), and an erythrocyte count of 750 per mm.3 The fluid was negative for tumor cells, and cultures and smears for fungi, Mycobacterium tuberculosis, and other bacteria were negative. The chest x-ray obtained at discharge showed a slight residual enlargement of the cardiac silhouette. The initial electrocardiogram showed changes of acute pericarditis.
with transition to inverted T waves at the time of discharge.

The patient remained asymptomatic until October 11, 1964, when he had recurrence of acute pericardial pain. At this time there was cardiomegaly without a friction rub, and a pericardial knock was heard in early diastole. The neck veins were distended several centimeters above the suprasternal notch at 80 degrees. The patient was febrile with daily temperature spikes to 102 degrees in spite of receiving salicylates to toxicity. There were small bilateral pleural effusions. The electrocardiogram showed diffuse ST elevation with T-wave inversions. Because of the development of a pericardial knock and elevated venous pressure, it was thought that an adhesive and possibly an early constrictive pericarditis was developing. Therefore, on October 21, 1964, a pericardectomy was performed.* At surgery there were adhesions between the left lung and the pericardium. The pericardium was thickened, edematous, and adherent, and there were bilateral pleural effusions. The pericardium was removed from the pulmonary veins on the left to the venae cavae on the right, and from the atrial appendage to the diaphragm. A portion of the diaphragmatic pericardium was also removed. A strip of pericardium adjacent to the left phrenic nerve was not resected. The pathologist’s report of the pericardium was as follows: “The pericardium is focally hemorrhagic and irregularly thickened by both light gray hyaline-like material that also has a somewhat gelatinous quality, and glistening light tan material having a homogeneous appearance. There is no discernible tumor, granuloma or purulent exudate. On sections there is marked thickening secondary to an organizing fibrous and fibrinous pericarditis in which proliferating capillaries, fibroblasts, fibrin deposition, and varying numbers of acute and chronic inflammatory cells are prominent features. There are neither tumor infiltrates nor granulomata, and special stains for fungus, bacteria, and acid fast bacilli are not rewarding.” Cultures of the pericardium for fungi, Myco. tuberculosis, and other bacteria were negative. The postoperative course was benign; no pericardial friction rub was heard at this time. The patient was placed on isoniazid prophylaxis until cultures of pericardium for Myco. tuberculosis were reported negative. Normal or negative laboratory data at this time were as follows: admission hemoglobin (13.9), blood urea nitrogen, bilirubin, antistreptolysin-O titer, urinalysis, alkaline phosphatase, VDRL, throat culture, five LE preparations, Brucella agglutinins, 1/1000 old tuberculin skin test, stool culture for virus, complement-fixation titers for histoplasmosis, blastomycosis, and coccidioidomycosis, and acute and convalescent antibody titers for mumps, western equine encephalitis, lymphocytic choriomeningitis, and St. Louis B encephalitis. Abnormal data were a discharge hemoglobin of 12.3; an admission leukocyte count of 11,750 with 85 per cent neutrophils; preoperative prothrombin times of 18.8 and 18.9 seconds with controls of 14 and 13.8 seconds, respectively; and a preoperative platelet count of 577,000. The patient was discharged asymptomatic on October 31, 1964.

The patient returned on November 4, 1964, stating that he had developed typical pericardial pain on November 1, 1964. He again was unable to lie at an angle lower than 80 degrees and demonstrated rapid shallow respirations. The temperature ranged to 102 degrees, and the pulse was 120. There was a faint apical knock in early diastole, and on the day of admission a definite to-and-fro rub was heard along the left sternal border by two physicians. At this point prednisone, 40 mg. per day, was begun. On this regimen the friction rub disappeared in 12 hours, and the patient became asymptomatic in 24 hours. The prednisone dosage was decreased to 20 mg. per day before discharge on November 11, 1964. Normal or negative laboratory data on this admission were as follows: partial thromboplastin time, clot retraction, fibrinogen index, bleeding time, Lee White clotting time, serum proteins, bilirubin, reticuloocyte count, red cell morphology, Bromsulphalein retention, alkaline phosphatase, cephalin cholesterol flocculation, and cryoglobulins. The following abnormal data were obtained: hemoglobin 11, hematocrit level 36, sedimentation rate 95, leukocyte count 15,750 with 84 per cent neutrophils, urinalysis 6 to 8 white blood cells per high-power field, platelet count 540,000, prothrombin time 17.9 seconds with a control of 13.1 seconds, serum iron 50, and serum iron binding capacity 252. The electrocardiogram showed transitory inversion of T waves in leads II, III, and aV6, in addition to other diffuse, stable T-wave inversions present since the preceding admission.

By December 3, 1964, the sedimentation rate had fallen to 8. By December 17, 1964, the patient’s hemoglobin had risen to 13.2. A chest x-ray on December 31, 1964, was normal. On January 7, 1965, when the prednisone dosage had been tapered to 7.5 mg. per day, a recurrence of typical pain together with a temperature of 100 degrees and a pulse of 120 developed. The symptoms disappeared on increasing the prednisone dosage to 15 mg. per day. The dosage was again slowly tapered and on March 3, 1965, the patient was asymptomatic on 5 mg. daily. On May 1, 1965, the prednisone was discon-

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*Surgery was performed by Drs. Jerome Grismer and David Raab.
continued. At this time the platelet count was 384,000, and the prothrombin time was normal.

**Case 2**

M. R., a 34-year-old white physician, was first seen at the University of Minnesota Hospitals on December 9, 1963. He stated that he had been in good health until June 1947, when he developed pneumonia with pleural effusion, both of which subsided within 1 month. Subsequent to this he had precordial chest pain of mild degree intermittently until 1950 and then had none for the next 2 years. In 1952 the chest pain recurred and was typical of pericardial pain. The pain was still rather mild and came and went until 1953, when he became quite ill with marked dyspnea, orthopnea, pericardial effusion, and electrocardiographic changes of acute pericarditis. He was seen at another medical center later in 1953. The physical examination was negative. Hemoglobin, sedimentation rate, serologic test for syphilis, LE clot test, serum protein electrophoresis, urinalysis, chest x-ray, and upper gastrointestinal series were all normal or negative. However, a second strength PPD was positive. (He had been tuberculin negative prior to this.) The clinical impression was benign recurrent pericarditis. However, because of the conversion of the tuberculin reaction, he was placed on isoniazid for a period of 1 year.

The patient was seen with another acute episode at the same center in March 1959. At that time the hemoglobin was 11.9, and the sedimentation rate was 58. Chest x-rays revealed pericardial and left pleural effusion. Sputum cultures and cultures of the pleural fluid were negative for *Mycobacterium tuberculosis*. A pericardial biopsy was performed in March 1959. This showed changes of nonspecific chronic pericarditis. Cultures of the pericardium were negative for *Mycobacterium tuberculosis* and other bacteria.

Between the years 1960 and 1963, the patient was evaluated at two different medical centers. Both institutions concurred with the diagnosis of recurrent benign pericarditis.

From 1958 to 1963 there generally had been one or two episodes of severe pericardial pain yearly. Recurrences were usually preceded by an upper respiratory infection. Each recurrence was characterized by electrocardiographic changes of acute pericarditis and an elevated sedimentation rate. However, during 1963, remissions of pain lasted only 2 to 4 weeks. The patient had taken intermittent steroids for years. He began treatment with the onset of each acute attack and would stop after about 1 week of therapy. On each occasion the steroids caused subsidence of symptoms within 24 hours.

Physical examination on his initial visit to the University of Minnesota Hospitals was negative with the exception of a blood pressure of 140/90 and the “moon facies” of chronic steroid administration. The following normal or negative laboratory data were obtained: hemoglobin, leukocyte count and differential, sedimentation rate, blood urea nitrogen, fasting blood sugar, urinalysis, VDRL, LE preparations, latex fixation, Brucella agglutinins, cold agglutinins, complement-fixation antibodies for histoplasmosis, blastomycosis, coccidioidomycosis, and psittacosis, chest x-ray, electrocardiogram, and vectorcardiogram. Serum protein electrophoresis was normal except for a serum albumin of 2.9 Gm. per cent.

Because of the intractable pain, pericardectomy was performed on January 24, 1964.* At surgery there was definite evidence of inflammation with greatly increased vascularity of the mediastinum. The pericardium was not particularly thickened, but was adherent to the heart throughout its extent. However, the adhesions were readily separated. The anterior portion of the pericardium back to the left phrenic nerve, and over to the great vessels, including the area of the cava veins was reflected. Also a portion was removed posteriorly off the diaphragm between the two phrenic nerves and further posteriorly almost to the entrance of the pulmonary veins. The only pericardium remaining was the relatively small portion around the entrance of the pulmonary veins posteriorly and that immediately adjacent to each phrenic nerve. The pathologist's report read as follows: "Several portions of pericardium are submitted for pathology. All of these are thick fibrous membranes having a shaggy or dark red lining. Sections show a fibrous membrane lined on one side with a somewhat irregular layer of mesothelium. The membrane is partially hyalinized, and the sections show no remarkable inflammatory infiltration." Cultures of the pericardium were negative for *Mycobacterium tuberculosis* and other bacteria. The postoperative course was benign, and the patient was discharged on February 7, 1964. After discharge the patient did well for about 2 weeks and then began to have recurrent chest pains. Whereas those he had had before surgery were of a crushing and agonizing substernal type which radiated up to the neck and jaws, the pains which have recurred postoperatively are characterized as follows: After a day or so of lethargy, pain of a somewhat dull aching type starts from the left lower chest and builds up to a maximum in about 6 hours with involvement of the entire left chest, shoulder, neck, and cheek. It is pleuritic in nature and is associated with a transiently elevated sedi-

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*Surgery was performed by Dr. C. W. Lillehei.
mentation rate. The pain is less severe 6 hours after resuming steroids and almost completely disappears in 12 hours.

In May 1964, a follow-up physical examination was negative. A repeat hemoglobin, hematocrit, leukocyte count and differential, sedimentation rate, and urinalysis were all normal. An electrocardiogram showed diffusely inverted T waves.

In November 1964, the pattern of chest pain was unchanged. The patient was taking steroids almost continuously and noted that symptoms always recurred within 3 days after stopping them.

**Discussion**

The above cases are typical examples of recurrent acute benign pericarditis. In each, pericardiectomy failed to control the disease. In the first case, the postoperative recurrences were identical to the original attacks. In the second case, the postoperative attacks were different from the preoperative episodes; while it could be argued that the postoperative pains represented a different disease process, their periodicity and response to steroids are more consistent with the preoperative diagnosis. In each case the pericardiectomy was as complete as was technically possible.

The apparent paradox of pericardial type pain in the absence of pericardium is not as difficult to explain as might be suspected. The most highly sensitive region of the pericardium is the lower part of the external surface of the parietal pericardium, especially that in the vicinity of the diaphragm. This portion of the pericardium is innervated by the phrenic nerves, and it is thought that stimulation of these nerve endings is responsible for the shoulder-strap distribution of referred pain which is so common in pericarditis. In performing a pericardiectomy, it is usually impossible to remove completely the pericardium in the vicinity of the diaphragm, and these relatively sensitive remnants might be a source of postoperative pain. Another type of "pericardial" pain has been related to stimulation by an inflamed epicardium of cardiac afferent nerve fibers lying in perid-ventitial layers of superficial coronary arteries. As it is impossible surgically to remove the epicardium, periodic inflammation of this structure might also be a source of recurrent pain. Thus, while pericardiectomy is a highly useful technic in the therapy of recurrent acute benign pericarditis, the above cases point out that even treatment as drastic as this cannot guarantee that the disease will be cured or the symptoms relieved.

**Summary**

Two cases of recurrent acute benign pericarditis are presented. Both were characterized by recurrent "pericardial type" pain after nearly total pericardiectomy, a previously unreported occurrence. A plausible explanation for this paradox is presented.

**References**

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