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 subternally 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 rhinorrhea. 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, VDRL, 1/1000 old tuberculin, histoplasm and coccidiodin 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, VDRL, 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.

From the Departments of Medicine, Mount Sinai Hospital, Minneapolis and University of Minnesota Hospital and School of Medicine, Minneapolis, Minnesota.
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 elev-
ated 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 re-
moved. A strip of pericardium adjacent to the
left phrenic nerve was not resected. The pathol-
ologist's report of the pericardium was as follows:
"The pericardium is focally hemorrhagic and ir-
regularly thickened by both light gray hyaline-
like material that also has a somewhat gelatin-
ous 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 fibrous pericar-
ditis 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, bacte-ia, and acid fast bacilli are not rewarding." CUL-
tures of the pericardium for fungi, Myco.
tuberculosis, and other bacteria were negative.
The postoperative course was benign; no peri-
cardial 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, com-
plement-fixation titers for histoplasmosis, blasto-
mycosis, and coccidioidomycosis, and acute and
convalescent antibody titers for mumps, western
equine encephalitis, lymphocytic choriomeningi-
tis, and St. Louis B encephalitis. Abnormal data
were a discharge hemoglobin of 12.3; an admis-
sion 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 dis-
charged 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 thrombo-
plastin 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 elec-
trocardiogram showed transitory inversion of T
waves in leads II, III, and aVr, 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 pa-
tient'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 recur-
rence of typical pain together with a tempera-
ture 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-

* Surgery was performed by Drs. Jerome Grismer
  and David Raab.
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 Myco. tuberculosis. A pericardial biopsy was performed in March 1959. This showed changes of nonspecific chronic pericarditis. Cultures of the pericardium were negative for Myco. 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, pericardiectomy 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 veins cavae 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 Myco. 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-

* 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 periad-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**


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

BEN GOLDFARB, DAVID GOLD, ELLIOT LATTS, HAROLD WEXLER and YANG WANG

Circulation. 1966;33:283-286
doi: 10.1161/01.CIR.33.2.283

Circulation is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 1966 American Heart Association, Inc. All rights reserved.
Print ISSN: 0009-7322. Online ISSN: 1524-4539

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://circ.ahajournals.org/content/33/2/283

Permissions: Requests for permissions to reproduce figures, tables, or portions of articles originally published in Circulation can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

Reprints: Information about reprints can be found online at:
http://www.lww.com/reprints

Subscriptions: Information about subscribing to Circulation is online at:
http://circ.ahajournals.org/subscriptions/