Surgical Management of Echinococcal Cysts of the Heart and Pericardium

By Alfred Heilbrunn, M.D., C. Frederick Kittle, M.D., and Marvin Dunn, M.D.

With the enlarging scope of cardiac surgery, an increasing variety of lesions is being subjected to operation. Stimulated by difficulties recently encountered in treating an echinococcal cyst of the left ventricle, we have reviewed the surgical management of this unusual cardiac lesion.

Echinococcal infection is rare in North America. Reviewing 519 cases reported in this country, Magath could find only 26 in which the disease had apparently been contracted in the United States. Grulce in 1905, reported the first American case of cardiac echinococcosis. This occurred in an Italian immigrant who suddenly died from multiple pulmonary emboli following rupture of a cyst in the right atrium. The next American patient was described by Mills in 1922, a 36-year-old French woman who also died suddenly. In 1945 Peters reviewed case records of the Massachusetts General Hospital and found five in which echinococcal involvement of the heart was found at autopsy.

Attwood, Verstandig, and Zigmore, each reported a patient in the United States in whom the antemortem diagnosis of echinococcal disease of the heart was made on clinical findings and laboratory studies. Treatment was not mentioned by any of the three authors.

Echinococcal infestation is common in the sheep-raising countries of the world, primarily Uruguay, Greece, Australia, New Zealand, the Middle East, North Africa, the Balkans, and, to a lesser degree, Iceland and the U.S.S.R. The incidence of cardiac involvement is estimated from 0.5 to 3 per cent of all patients with echinococcal disease. Dungan found cardiac involvement in two of 60 persons in an autopsy study in Iceland, whereas Kourias, in Greece, did not find any cardiac lesions in 2,017 patients operated on for echinococcal cysts.

The definitive host of Echinococcus granulosus is the dog. Ova contained in dog feces contaminate grass and water ingested by the intermediate host, usually sheep. The life cycle is completed when the organs of sheep or other intermediate hosts are eaten by dogs. Human infestation usually occurs as a result of contamination from the dog. After ingestion by man, the hexacanth embryo enters the portal circulation. An estimated 70 per cent are trapped in the liver; another 15 per cent pass to the lungs. A few reach the heart after passing both these filters and embolize in the myocardium via the coronary arteries. Cysts are seen in the left side of the heart more often than in the right. Most authors agree that hydatid cysts of the heart are initially primary and single, even when localization in other organs has occurred.

After implantation the hexacanth embryo develops into a hydatid cyst. Reaction to the parasite stimulates a fibrous response in the host myocardium, producing a characteristic adventitial lining.

Once formed in the heart, a cyst may develop daughter cysts, rupture, or degenerate and calcify. Rupture of the cyst produces several clinical entities. Rupture into the pericardium results in additional cyst formation and occasional progression to constrictive pericarditis. The escape of hydatid fluid into the circulation produces an anaphylactoid reaction. The escape of daughter cysts into the circulation produces systemic and pulmonary emboli. Cerebral metastases are common. Death may occur with any of these events although several patients have been described with several episodes of rupture prior to death.
Figure 1
Chest x-ray in June 1961 demonstrating the cyst in the left upper lung and fullness of the left cardiac border.

Case Report
A 27-year-old Iranian student was admitted to the University of Kansas Medical Center on September 4, 1961, with the chief complaint of “hydatid cyst of the heart.”

History revealed that he had been in good health until June 1961, when he noted coryza, cough, fever, occasional night sweats, and bright red hemoptysis in small amounts. Because of these complaints he was admitted to the Menorah Medical Center of Kansas City, Missouri, on June 28, 1961. A complete physical examination prior to his entry into the United States disclosed no abnormalities. A chest film taken in November 1960 was reported as normal, but in retrospect we could see a small opacity in the left upper lobe.

Physical examination was unusual only because of the presence of a grade II systolic ejection murmur in the pulmonic area. The second sound split normally with respiration. X-ray studies showed an ovoid lesion in the left upper lobe (fig. 1). Bronchoscopy, electrocardiography, and angiocardiography gave normal results. Blood analyses showed an eosinophilia of 17 per cent and a white cell count of 9,500.

Left thoracotomy was done on July 19, 1961.*

*Dr. Arthur Adelman, Menorah Hospital, Kansas City, Missouri.

Figure 2
Diagram showing the deep extent of the echinococcal cyst.

A small cyst was removed by wedge excision from the left upper lobe. A 4 by 4 cm. mass was noted in the region of the left atrioventricular groove, and pericardiectomy for further inspection was performed. A cyst was seen within the myocardium, but no attempt at excision was made.

The surgical specimen showed a segment of lung containing a 3 cm. cyst filled with numerous daughter cysts varying from 0.1 to 1.5 cm. in diameter and surrounded by a fibrous capsule 0.2 cm. thick. Examination of microscopic sections confirmed the diagnosis of echinococcal cyst of the lung.

The patient was subsequently referred to the University of Kansas Medical Center for definitive therapy of his cardiac echinococcal cyst.

Physical findings at this time were again within normal limits with the exception of a grade III systolic ejection murmur heard best in the pulmonic area and along the left sternal border. The second sound split normally with respiration. His white cell count was 13,750, with 32 per cent eosinophilia. Chest films showed the previously resected rib and an abnormal convexity in the center of the left heart border.

Cardiac catheterization was done on September 5, 1961. The right ventricular pressure was 45/0

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Figure 3
Daughter cysts evacuated at operation.

Figure 4
Postoperative x-ray showing filling of chest tube and cyst by radiopaque dye.

mm. Hg, and the pulmonary artery pressure 25/10. The pull-through tracing from the pulmonary artery to the right ventricle showed an intermediate pressure area suggesting an infundibular chamber. It was believed that this pressure gradient was produced by the cyst compressing the right ventricular outflow tract. In view of the natural history of echinocoeal cysts of the heart, we advised surgery and the operation was done on September 9, 1961. Extracorporeal circulation apparatus was available.

The pericardium was entered through a midline sternum-splitting incision. A rounded, slightly morulized cystic mass approximately 2.5 by 3 cm. with several 1 to 2 mm. daughter cysts on its surface projected from the lateral aspect of the left ativoventricular groove. The mass was adherent to the pericardium at the previous pericardiotomy. The left atrial appendage was posterior and superior to the cyst. A clear viscid fluid was aspirated from the cyst. Ten per cent formalin was injected into the cyst and allowed to remain for 10 minutes. The cyst capsule was again identified, and dissection was begun to separate it from the myocardium. It was then apparent that the cyst extended transversely across the intermediate sulcus into the anterior and superior portion of the right ventricular outflow tract, posterior to the anterior descending coronary artery (fig. 2). Because of its size and location, we abandoned attempts at total excision, opened the cyst, and evacuated 56 daughter cysts (fig. 3). The interior of the cyst was packed for 10 minutes with a sponge soaked in formalin. The pericardial cavity was rinsed with a small amount of formalin because of possible contamination by the daughter cysts.

The peritoneum was opened at the inferior end of the incision, and the liver was palpated to seek other echinocoeal cysts. None was found.

A 3/16-inch tube was inserted through a separate stab wound on the left and passed through a second incision in the pericardium posteriorly. The cyst edge was sutured to this second opening in the pericardium so that drainage from the interior of the cyst wall could pass to the outside through this tube (fig. 4).

Direct pressure measurements at operation failed to show a right ventricular-pulmonary artery gradient. It was hypothesized that the closed pericardium pressed the cyst into the outflow tract of the right ventricle, thus accounting for the preoperative systolic gradient and the systolic murmur at the pulmonic area.

The patient tolerated the operative procedure well. Subsequent electrocardiograms showed absence of the R waves in lead I, V₅, and V₆ with elevated ST segments in these leads and depression of the ST segments in leads II, III, and aVF. Deep Q waves were noted in leads I and aV₁.
Table 1

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<thead>
<tr>
<th>No.</th>
<th>Author, Year</th>
<th>Country of origin</th>
<th>Sex</th>
<th>Age</th>
<th>Symptoms</th>
<th>Method of diagnosis</th>
<th>Findings and procedure</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>Marten, 1921</td>
<td>Australia</td>
<td>M</td>
<td>49</td>
<td>Congestive heart failure</td>
<td>Chest film</td>
<td>Large cyst at apex aspirated only. At post a 7.5 cm. cyst in right atrium</td>
<td>Died in 24 hr.</td>
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<td>2.</td>
<td>Long, 1932</td>
<td>Australia</td>
<td>F</td>
<td>42</td>
<td>Cough, dyspnea on exertion, weight loss</td>
<td>Chest film</td>
<td>&quot;Goose egg&quot; cyst in left ventricle emptied and drained</td>
<td>Drained slightly 3 mo.</td>
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<tr>
<td>3.</td>
<td>Starr, 1949</td>
<td>New Zealand</td>
<td>M</td>
<td>34</td>
<td>Congestive heart failure</td>
<td>Eosinophilia; complement fixation</td>
<td>Large pericardial cyst with daughters, 1 cm. cyst AV groove evacuamed, formalized</td>
<td>Good, 4 mo.</td>
</tr>
<tr>
<td>4.</td>
<td>D'Abreau, 1950</td>
<td>England</td>
<td>F</td>
<td>50</td>
<td>Hemoptysis</td>
<td>Known splenic cyst; x-ray and ECG</td>
<td>LV 6 x 4 cm., excision</td>
<td>Good†</td>
</tr>
<tr>
<td>5.</td>
<td>Ganickin, 1952</td>
<td>U.S.S.R.</td>
<td>F</td>
<td>37</td>
<td>Cough, chest pain</td>
<td>Eosinophilia; chest film</td>
<td>RV; 14 x 10 x 9.5 cm.; enucleation</td>
<td>Good, 5 mo.</td>
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<td>6.</td>
<td>Larghero, 1954</td>
<td>Uruguay</td>
<td>M</td>
<td>27</td>
<td>Angina</td>
<td>Chest film; angina; ECG</td>
<td>LV, 6 x 4 cm.; evacuation</td>
<td>Good†</td>
</tr>
<tr>
<td>7.</td>
<td>Larghero, 1954</td>
<td>Uruguay</td>
<td>F</td>
<td>40</td>
<td>Asymptomatic</td>
<td>Chest film; angina; ECG</td>
<td>LV, 5 x 3.5 cm.; evacuation</td>
<td>Good†</td>
</tr>
<tr>
<td>8.</td>
<td>Parruel, 1954</td>
<td>Uruguay</td>
<td>F</td>
<td>26</td>
<td>Malaise, dyspnea on exertion, fever, precordial pain</td>
<td>ECG; complement fixation; eosinophilia</td>
<td>LV, 3 cm., evacuated, irrigated</td>
<td>Good†</td>
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<td>9.</td>
<td>Windsor, 1954</td>
<td>Australia</td>
<td>F</td>
<td>48</td>
<td>Epigastric pain</td>
<td>ECG, chest film; eosinophilia; complement fixation</td>
<td>Left AV groove, evacuated, irrigated with saline</td>
<td>Good, 2 wk.</td>
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<td>10.</td>
<td>Canabal, 1955</td>
<td>Uruguay</td>
<td>M</td>
<td>43</td>
<td>Asymptomatic</td>
<td>Chest film, ECG</td>
<td>LV; evacuated and irrigated with ether, 7 daughter cysts</td>
<td>Good†</td>
</tr>
<tr>
<td>11.</td>
<td>Canabal, 1955</td>
<td>Uruguay</td>
<td>M</td>
<td>24</td>
<td>Asymptomatic</td>
<td>Chest film, complement fixation, ECG, positive Casoni</td>
<td>Left apex 6 x 4 cm., evacuated, irrigated with ether, 400,000 penicillin instilled</td>
<td>Good, 1 mo.</td>
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<td>12.</td>
<td>Canabal, 1955</td>
<td>Uruguay</td>
<td>F</td>
<td>40</td>
<td>Hypertension</td>
<td>Chest film, ECG</td>
<td>LV, calcified, evacuated, irrigated with hypertonic saline</td>
<td>Good†</td>
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<td>13.</td>
<td>Cattoit, 1955</td>
<td>Algiers</td>
<td>F</td>
<td>40</td>
<td>Dyspnea on exertion, orthopnea, abdominal pain</td>
<td>Chest film, ECG, hepatic masses</td>
<td>LV, 8 x 10 cm., evacuated, irrigated with hypertonic saline</td>
<td>Good†</td>
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<td>No.</td>
<td>Author, Year</td>
<td>Country</td>
<td>Gender</td>
<td>Age</td>
<td>Symptoms</td>
<td>Treatment</td>
<td>Outcome</td>
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<td>14</td>
<td>Molloy, 1955</td>
<td>New Zealand</td>
<td>F</td>
<td>45</td>
<td>Sudden collapse, shock, rash, incontinence</td>
<td>Eosinophilia; positive Casoni; complement fixation; chest film</td>
<td>Pericardium thickened, 6 cysts, “nut to orange” size, some daughter cysts; wiped with formalin, excess walls excised. Brisk hemorrhage from one cyst</td>
<td></td>
</tr>
<tr>
<td>15</td>
<td>Stojanovic, 1955</td>
<td>Yugoslavia</td>
<td>F</td>
<td>61</td>
<td>Asymptomatic</td>
<td>Chest film, ECG</td>
<td>LV and LA, many cysts; evacuated, irrigated with “rivano”</td>
<td></td>
</tr>
<tr>
<td>16</td>
<td>Houel, 1956</td>
<td>Algiers</td>
<td>M</td>
<td>17</td>
<td>Right upper quadrant mass</td>
<td>Chest film, ECG</td>
<td>Left AV groove, large cyst; 50 ml. aspirated, irrigated with formalin. Liver cyst excised later</td>
<td></td>
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<tr>
<td>17</td>
<td>Karageorgis, 1956</td>
<td>Greece</td>
<td>M</td>
<td>37</td>
<td>Severe congestive heart failure</td>
<td>Chest film, ECG</td>
<td>Huge cyst LV, 100+ daughter cysts, myocardium 1 mm. sudden fatal hemorrhage during operation; Died during operation</td>
<td></td>
</tr>
<tr>
<td>18</td>
<td>Sbarounis, 1956</td>
<td>Greece</td>
<td>F</td>
<td>31</td>
<td>Enlarging mass in chest for 15 yr. 4 mo. pregnant known intra-abdominal cysts</td>
<td>Chest films</td>
<td>LA, 12 X 7 X 5 cm. cyst, evacuated</td>
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<tr>
<td>19</td>
<td>Chalnot, 1957</td>
<td>Algiers</td>
<td>M</td>
<td>30</td>
<td>Dyspnea on exertion, palpitation, retrosternal pain, hemoptysis</td>
<td>Chest film, ECG, eosinophilia</td>
<td>Pericardial cysts, LV, and intramural. Aspirated, injected with formalin. Pericardial cysts excised, intramural evacuated</td>
<td></td>
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<tr>
<td>21</td>
<td>De Vernejoul, 1958</td>
<td>France</td>
<td>F</td>
<td>31</td>
<td>Cyst of appendix at 19; peritoneal and liver cysts at age 22; at 27 episode of fever and CV collapse</td>
<td>Chest film, ECG, eosinophilia, angio</td>
<td>LV cyst (“orange-sized”) evacuated (?)</td>
<td></td>
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<tr>
<td>22</td>
<td>Dobrev, 1959</td>
<td>Bulgaria</td>
<td>F</td>
<td>46</td>
<td>Left subcostal pain, fever, previous laparotomy for liver cyst</td>
<td>Chest film; eosinophilia</td>
<td>“Nut-sized” RV and “apple-sized” LV. Evacuated</td>
<td></td>
</tr>
<tr>
<td>23</td>
<td>Michaud, 1959</td>
<td>North Africa</td>
<td>F</td>
<td>49</td>
<td>Palpitation and precordial pain. Previous laparotomy for liver cyst</td>
<td>Chest film, ECG</td>
<td>Left AV groove “hen’s egg” cyst, injected with hypertonic saline, evacuated and cyst wall removed</td>
<td></td>
</tr>
<tr>
<td>24</td>
<td>D’Allaines, 1960</td>
<td>France</td>
<td>M</td>
<td>30</td>
<td>Left chest pain</td>
<td>Chest film and ECG</td>
<td>Anterior surface LV, evacuated and formalinized</td>
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Table 1 Continued

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<tr>
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<th>Method of diagnosis</th>
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<th>Results</th>
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<tr>
<td>25.</td>
<td>Houel, 1960*</td>
<td>Algiers</td>
<td>F</td>
<td>22</td>
<td>Constrictive pericarditis</td>
<td>Chest film and ECG</td>
<td>Pericarditis; cyst AV groove deep into myocardium. Full of daughter cysts. Autopsy showed additional apical subendocardial cyst</td>
<td>Died hours after operation</td>
</tr>
<tr>
<td>26.</td>
<td>Houel, 1960*</td>
<td>Algiers</td>
<td>M</td>
<td>31</td>
<td>Cough, dyspnea on exertion, hemoptyis, grade IV systolic murmur</td>
<td>Chest film, ECG, eosinophilia</td>
<td>Multiple cysts of LV and lung</td>
<td>Good?</td>
</tr>
<tr>
<td>27.</td>
<td>Romanooff, 1962**</td>
<td>Iraq</td>
<td>M</td>
<td>29</td>
<td>Epigastric and sternal pain</td>
<td>Chest film, ECG, positive Casoni, complement fixation</td>
<td>12 cm. cyst anterior mediastinum; cyst LV; evacuated</td>
<td>Good?</td>
</tr>
</tbody>
</table>

*Dates represent the year that the reports were published. The countries of origin refer to the residence of the patients. Question marks in last column indicate that the length of follow-up is unknown.
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the heart was similarly freed. Ventricular contractions, although initially feeble, became noticeably stronger immediately after the adherent pericardium was excised.

Upon opening the right pleural cavity, we found a 3-cm. echinococcal cyst on the antero-lateral edge of the right lower lobe. This was removed by wedge excision.

Improvement in pulse pressure and increased vigor of cardiac contractions were striking. However, because of continued bleeding from the patient's thoracotomy tubes, we re-explored several hours postoperatively without finding any particular source of bleeding. Two hours after this procedure, while the patient was in the recovery room, he sustained bradycardia with an alarm being given by the electrocardiograph monitor after asystole for 8 seconds. Cardiac arrest responded to external cardiac massage and reinsertion of an endotracheal tube. The patient was maintained on a Bennett respirator, regaining consciousness within 5 minutes. During the next 36 hours several attempts were made to remove the endotracheal tube, but the patient, in an alert conscious state, indicated by writing that he desired the endotracheal tube left in place to assist his breathing. Subsequently the tube was removed and the remaining postoperative course was uneventful.

Signs of congestive failure slowly decreased, and the patient was discharged on December 28, 1961, approximately 1 month after his pericardiectomy. He was followed in the outpatient clinic. When last seen, 6 months after his pericardiectomy, he had resumed normal activities. No signs of congestive failure or other cardiac difficulties were present. At that time blood pressure was 110/75, pulse was 90 and regular. The liver was palpable 2 cm. below the right costal margin, but it was not tender. No murmurs were heard, and there was no evidence of peripheral edema. The eosinophilia was 2 per cent.

**Symptomatology**

Four of the patients were asymptomatic. In at least three other patients, cardiac involvement became apparent during investigation and treatment for cysts in other locations. When symptoms were present, they were either vague (palpitation and substernal pressure), consisted of angina, congestive heart failure, or were secondary to rupture of a primary cardiac cyst. Solitary small cysts were generally asymptomatic. Occasionally rupture of a cyst produced constrictive pericarditis, but in some patients rupture occurred into the adjacent cardiac chamber with development of anaphylactic reactions or peripheral emboli.

**Diagnosis**

The presence of an echinococcal cyst of the heart should be considered whenever a lobular mass is seen adjacent to the left ventricle, particularly if localized calcification is present and if the patient is from an endemic area. In each of the 28 patients operated upon, the diagnosis of cardiac hydatid disease was initially suggested by an abnormal chest film.

The location, the configuration, and the size of a suspected cyst can be further clarified by fluoroscopy and tomography. Eosinophilia over 6 per cent is helpful, but its absence is not significant. The Casoni skin test, although occasionally helpful, may be negative in the presence of proved echinococcal disease (e.g., patient 20).

Electrocardiographic changes, if present, have considerable diagnostic value. The presence of ischemic changes over the left ventricle in a relatively young person, together with an abnormal left cardiac border by radiography, is considered diagnostic in endemic areas. Canabal et al. regard the following as characteristic for electrocardiographic changes: inversion of the T waves in leads I and aVL, upright T waves in aV_R, the presence of normal Q waves, small R waves in the area of the cyst in the precordial leads, with

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an abrupt increase in the R waves beyond the area of involvement, T-wave inversion in the chest leads showing QRS changes, and normal RST segments.

Compression ischemia of the myocardium and thinning of the ventricular wall by the hydatid cyst are manifested by localized ischemic patterns and changes in the QRS complexes. Extent of T-wave changes and reduction in amplitude of the R waves in the precordial leads give an indication of the location and extent of the cyst. Although these changes may be seen with a myocardial infarct, an infarct with these changes is not accompanied by a paracardiac mass.

Specialized studies, including cardiac catheterization and angiography, help confirm the diagnosis and delineate the extent of involvement by the cyst. In at least two patients (patient 26 and ours), pressure gradients have been demonstrated between the right ventricle and the pulmonary artery, both of which disappeared postoperatively. Angiocardiography may be helpful in demonstrating encroachment on the ventricular cavity and thickness of the underlying myocardium. Left ventricular aneurysm may be confused with echinococcal cyst, and in one instance a patient initially reported as having a left ventricular aneurysm was found to have a hydatid cyst.

**Treatment**

Since cardiac hydatid cysts usually rupture with a high incidence of sudden death and widespread embolization, surgical excision is indicated even in asymptomatic patients. Continued growth of the solitary cyst may produce cardiac failure by interference with normal ventricular function. If widespread embolization has already occurred, particularly with cerebral metastases, and a long lifespan is not anticipated, surgical intervention is not justified.

Initially the cyst should be carefully aspirated, avoiding spillage of the fertile hydatid fluid. The cyst should then be injected with a parasiticide. Although formalin (4 to 10 per cent) has been used in a majority of patients, its deleterious effects on the pericardium as evidenced in our patient have led to use of hypertonic (33 per cent) saline solution.

Enucleation is preferred. When this is not possible, the projecting portion of the cyst should be resected, the germinative membrane peeled away, and the residual cavity again treated with the parasiticide. Marsupialization may be desirable when continued drainage is necessary or seems indicated.

**Results**

With the exception of three patients listed in the table, results of surgical management of cardiac echinococcosis have been reported as good. Unfortunately, follow-up in most reported cases is less than 1 year, and it is not possible to obtain statistics as to recurrence. In our patient, at the time of reoperation, 3 months after the initial procedure, no evidence of recurrence of the cardiac lesion was noted, although an additional small cyst was found in the right lower lobe of the lung. This probably had been present initially and did not represent secondary spread.

**Discussion**

Although echinococcal cyst of the heart is rare in North America, it is anticipated that incidence here will be noted more often with increasing world travel and numerous exchange programs involving persons in the affected age groups. The diagnosis is suggested by an abnormal x-ray contour of the left ventricle and characteristic electrocardiographic changes. Eosinophilia and a positive Casoni skin reaction may be helpful signs. A careful search should be made for cysts in other organs, particularly the liver. The recent development of radioactive isotope liver-scan ning technics may be helpful. Extent of the cardiac cyst and its precise location can sometimes be defined by angiography. Hemodynamic changes as a result of the cyst can be evaluated by cardiac catheterization.

Surgical treatment is advised. The experience of our patient illustrates that the use of parasiticides must be carefully controlled to avoid local tissue damage. The danger of
massive hemorrhage through the thin myocardium at the base of the cyst is constantly present and may be fatal during the operative procedure (see patient 17). 32

Summary
Surgical treatment of a 27-year-old man with an echinococcal cyst of the heart is reported, illustrating some difficulties and problems manifested by this lesion.

Twenty-seven other patients operated upon for cardiac hydatid disease are reviewed in tabular form.

Symptomatology, diagnosis, and management of this rare cardiac lesion are reviewed.

References
a propos deux observations. Presse méd. 66: 697, 1958.


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John Cheyne 1771-1831

It is ironic that the founder of the Dublin School had served with the British Army at the defeat of the Irish Rising of 1798 on Vinegar Hill. John Cheyne (1771-1831) entered Ireland first as a surgeon with the British Forces, and only ten years later did he emigrate back to become a practitioner in Dublin. He served as the first professor of medicine at the College of Surgeons and as physician to the Meath Hospital. In addition to his well known description of the respiratory pattern in severe coma, he provided the first report of acute hydrocephalus.—K. M. Cahill, M.D. The Golden Era of Irish Medicine The New England J. Med. 266: 544 (March), 1962.