Historical Perspectives

The Tale of Spring Water Cysts

A Historical Outline of Surgery for Congenital Pericardial Diverticula and Cysts

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Congenital pericardial diverticula and cysts are extremely uncommon lesions within the anterior mediastinum. Both lesions derive from the pericardial celom and represent different stages of a common embryogenesis. Initial reports date from the 19th century. Surgical pioneers were Otto Pickhardt, who removed a pericardial cyst at Lenox Hill Hospital in New York in 1931, and Richard Sweet, who accomplished the first resection of a pericardial diverticulum at Massachusetts General Hospital in Boston in 1943. These lesions were also called spring water cysts because they usually contain watery, crystal-clear fluid. This history outlines the milestones of evolving surgical management, from the first report in 1837 up to the present time. ***(Tex Heart Inst J 2012;39(3):330-4)***

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Preliminary reports on pericardial diverticula and cysts date back to the middle of the 19th century. During that prosperous period, medical science in general experienced enormous development. In the course of this progress, regular postmortem examinations were established in many hospitals and university institutions. In this manner, pathologists encountered the first documented cases of pericardial diverticula and cysts. However, all 19th-century records of these lesions derived exclusively from autopsy studies. Only considerable advances in radiographic imaging opened up the way for timely detection of mediastinal lesions. With the dawn of thoracic surgery in the early decades of the 20th century, surgical therapy finally became feasible. Otto Pickhardt accomplished the first resection of a pericardial cyst at Lenox Hill Hospital in New York in 1931.[1] Soon other successful operations on pericardial cysts were reported. Initially, causation was controversially debated. The original assumption was that pulsion diverticula were due to increased intrapericardial pressure and that traction diverticula were caused by mediastinal infections. Only in the 1940s were pericardial cysts, as well as diverticula, recognized as congenital malformations. Notwithstanding this successful development, these lesions remained extremely uncommon. Le Roux reported that only 3 cases were diagnosed out of 300,000 people who took part in a mass x-ray campaign in Edinburgh in 1958.[2]

Since then, both surgical techniques and diagnostic imaging have substantially improved. However, the basic principles of the pioneer procedures from the 1930s and 1940s still influence the modern management of congenital pericardial cyst and diverticulum. Therefore, this brief history outlines the major breakthroughs in discovery, diagnosis, and treatment of these rare disorders.

We decided to discuss pericardial cysts and diverticula together, because they represent just slightly different stages of a lesion that has a common embryogenesis. Moreover, the distinction between these lesions in the historic literature is often quite vague, and the difference between a diverticulum with narrow neck and a cyst that communicates with the pericardial cavity through a pedicle sometimes seems arbitrary. When Richard Sweet resected the first pericardial diverticulum, he did not recognize a communication between the lesion and the pericardium until he had the cyst completely shelled out from the anterior mediastinum. Only then did he find a tiny communication. Hence, he defined the cyst as pericardial diverticulum. On the other hand, Le Roux included cystic lesions with free connection into the pericardial cavity in his series of pericardial cysts. Therefore, both indistinct terminology and close embryologic relationship justify a common consideration of cyst and diverticulum.

19th-Century Reports on Pericardial Diverticula and Cysts

The first description of a pericardial diverticulum was presented by T. Hart of the Park Street School of Medicine in Dublin, in 1837.[3] He served as conservator at the museum of the medical school and encountered a "previously unrecorded, pathological condition" upon postmortem examination of an elderly woman. "The anterior mediastinum was occupied by a membranous, pyriform sac of considerable size, lying on and overlapping the pericardium." Moreover, he found a "free communication between it and the pericardium" and a "circular orifice, freely admitting the introduction of a finger."[3] Further reports on pericardial diverticula and cysts soon followed, in the 2nd half of the 19th century. For example, in the *Transactions of the Pathological Society of London* in 1869, Bristowe described a 1.5-inch-long diverticulum with an oval orifice into the pericardium.[4]

A profound pathoanatomic autopsy case series comprising 4 diverticula and 1 cyst was published by Rohn, from the Charles University of Prague, in 1903.[5] He recorded a cystic lesion attached to the pericardium. When the cyst was originally encountered during a postmortem examination in 1891, it had been termed "Cystis mediastini." Only Rohn, upon revision of the specimen, discovered a connection between cyst and pericardium. He concluded that the cyst probably derived from a diverticulum whose communication to the pericardial cavity had been pinched off.[5] In this manner, the transformational relationship between pericardial diverticulum and cyst was recognized for the first time.

From Post- to Antemortem Diagnosis: Advances in Radiology (1920s and 1930s)

All documented 19th-century cases of both cyst and diverticulum were discovered upon postmortem examination. However, the advent of radiography, most especially chest radiography, provided reliable tools for the detection of mediastinal lesions during life. Throughout the 1920s and 1930s, the establishment of standard techniques for the performance of chest radiography enormously improved the available diagnostic measures. Arthur Tudor Edwards, one of the pioneers of thoracic surgery in Great Britain after the First World War, emphasized the importance of chest radiography in 1927: "This method has proved invaluable. Greater precision is being rapidly attained as experience becomes wider."[6]

Moreover, artificial pneumothorax, by introduction of oxygen or other gases, was frequently used to clarify the relationship between an intrathoracic tumor and the lung.[6] The invention of bronchography enabled the radiographic examination of the lower respiratory tract. In 1918, Chevalier Jackson (Philadelphia) started the insufflation of bismuth through a bronchoscope for radiologic visualization of the tracheobronchial tree.[7] Intratracheal instillation of Lipiodol (ethiodized oil) as a radiopaque contrast agent for exploration of the airways was first described by Sicard and Forestier in 1922.[7]

The spectrum of available radiologic tools was further broadened by the introduction of esophagography with barium-contrast swallow, which enabled differentiation between esophageal tumors and other mediastinal masses. An exceptional radiographic procedure was recorded by E.H. Cushing of Cleveland in 1937.[8] He reported on a case in which a large pericardial diverticulum presented itself as soft, fluctuant swelling on the anterior chest wall. After aspiration of "yellowish fluid, . . . 50 cubic centimeters of air was injected into the tumor, and a roentgenogram showed part of the air to be in the pericardial cavity and part beneath the skin."[8] Thus Cushing described the first pneumogram of a pericardial diverticulum.

Further refinement of radiographic methods was soon achieved, and in an increasing number of cases correct diagnosis of a pericardial cyst was obtained from chest radiographs. Wallace Yater (Georgetown University) recorded the radiographic appearance of pericardial cyst and summarized the differential diagnosis in 1931.[9] Walter Addey (Tunbridge Wells, UK) provided a profound report on the same topic in 1940.[10] However, these lesions remained very uncommon. Yater stated that "simple cysts of the pericardium are practically unheard of,"[9] and Addey emphasized that "cysts of the pleura are extremely rare, and are found only accidentally during the routine examination of the chest for some other suspected condition."[10]

Pioneer Operations on Pericardial Cyst and Diverticulum

Improved radiographic means and the dawn of thoracic surgery during the first decades of the 20th century definitely opened the way for successful surgical therapy of pericardial cyst and diverticulum. The first record of operative removal of a mediastinal cyst of at least probable pericardial origin derives from Arthur Tudor Edwards.[6] He reported the case of a young woman who presented with a suspected intrathoracic cyst. Thoracotomy with resection of the cyst was performed at the Brompton Hospital in London on 8 May 1925.[6] Edwards was not able to define the origin of the cyst due to dense adhesions within the pleural cavity. However, he assumed a pericardial lesion: "Or is it a primary diverticulum from the pericardium, the communication of which has become shut off as enlargement occurred? A definite answer is impossible, but its firm attachment to the pericardium is rather in favour of the latter."[6]

Otto Pickhardt from Lenox Hill Hospital in New York is usually credited with the first resection of a pericardial cyst, in 1931.[1] A 53-year-old woman suffered a "sharp pain, knife-like in character, over the precordium for a period of several weeks," and "X-ray examination

revealed the presence of a circular shadow just above the left diaphragm." Pickhardt now performed a thoracoscopy with the patient under local anesthesia and saw "a tangerine-sized, yellowish, smooth, glistening tumor mass in the left pleural cavity."[1] This truly remarkable early use of thoracoscopy underscores the immense progress of thoracic surgery in the United States during the early decades of the 20th century.

Pickhardt then decided to remove the mass via thoracotomy "through the left 8th intercostal space. . . . At the anterior inferior medial aspect, in the costophrenic angle, at the junction of the diaphragm, apex of the pericardium and thoracic cage, there is seen a thin-walled cyst, the size of an orange, with a small, nipple-like projection towards the pericardium." The cyst was "easily shelled out" and the postoperative course was uneventful.[1]

This pioneer deed of Otto Pickhardt was soon repeated by other thoracic surgeons in the course of the 1930s. D'Abreu reported on the successful resection of "a large bluish cyst 4 inches long and about 2 inches wide" in a 33-year-old man at the Royal Infirmary, Cardiff, Wales, in 1937.[11] In the same year, Churchill and Mallory described another case from the Massachusetts General Hospital in Boston.[12] Their 33-year-old male patient had suffered from dysphagia and epigastric pain for approximately 15 years. Radiography suggested a pericardial mass, and hence thoracotomy was advised. A "very thin-walled cyst containing clear fluid" was encountered and easily removed.[12]

The pioneer operation on pericardial diverticulum was also performed at Massachusetts General Hospital in Boston, in 1943. It was reported by Morton Mazer, a professor of radiology, and the operation was carried out by Richard Sweet.[13] The radiographic appearance of the diverticulum was similar to the common appearance of a pericardial cyst. "Examination of the chest revealed a sharply defined, pear-shaped mass, 6 cm in its greatest diameter, lying in the right cardiophrenic angle, against the anterior chest wall."[13] Upon anterior thoracotomy, they found a tumor "obviously cystic in nature. . . . The only attachment of the mass was to the pericardium. . . . The communicating opening was little more than 0.5 cm in diameter," while the whole lesion "measured 8 by 4 by 1.5 cm." Convalescence was undisrupted and the patient left the hospital on the 14th postoperative day.[13]

"Spring Water Cyst": A New Term is Created (1940s)

Due to encouraging reports on the pioneer procedures, more and more thoracic surgeons developed an interest in the diagnosis and therapy of pericardial cysts and diverticula. In 1941, Curreri and Gale (University of Wisconsin, Madison) recorded resection of a cyst, "which was readily peeled from the pericardium."[14] Norman R. Barrett encountered a pericardial cyst at the Horton War Hospital (London) in 1944.[15] "It was a thin-walled translucent cyst which contained clear watery fluid. The cyst was removed without difficulty and the chest closed."[15] Clarence J. Schein (Mount Vernon, NY) reported a similar case in 1948: "The cystic mass was pedicled on the parietal pericardium . . . the lining was tissue paper thin and contained clear fluid."[16]

The appearance of the thin-walled, translucent cysts containing crystal-clear fluid gave rise to calling them "spring water cysts."[17] Greenfield and colleagues (Mount Sinai Hospital, NY) introduced the new term into the medical literature when they encountered a cyst filled with "crystal-clear fluid of watery consistence" in 1943.[17] The observation that pericardial cysts usually contain clear liquid resembling spring water was confirmed by Buyers and Emery (Des Moines, Iowa) in 1948.[18] Thus, "spring water cyst" soon became an established metaphor for pericardial cysts.

Pericardial Cyst and Diverticulum Are Congenital Malformations: Discovery of a Common Embryogenesis (1940s and 1950s)

For many years, the exact origin of pericardial diverticula and cysts remained unknown. The latter were frequently misunderstood as "cystic lymphangiomata" or "cysts probably of lymphatic origin," and diverticula were generally regarded as herniation of the pericardium. Only in the 1940s were both lesions recognized as congenital malformations. Adrian Lambert (Bellevue Hospital, NY) proposed an entirely new causal concept in 1940.[19] He suggested that cyst, as well as diverticulum, derives from disconnected mesenchymal lacunae, which later unite to form the pericardial celom. Failure of one of these lacunae to merge with the others would result in the formation of congenital cyst.[19]

Although the concept of pericardial cyst as a congenital condition was soon widely accepted, it provided sufficient explanation neither for the predominant location of the lesion within the cardiophrenic angle nor for the relationship between cyst and diverticulum. Therefore, Lillie and colleagues (Mayo Clinic, Rochester, Minn) suggested that both diverticulum and cyst derive from the ventral recess of the pericardial celom, rather than from the lacunae.[20] "The discovery that a diverticulum-like structure in the ventral recess of the pericardial celom occurred during development of the pericardium at the site where so many of these lesions were found led to speculation as to their possible relationship." The authors concluded that persistence of the recess results in formation of a diverticulum, while constriction of the proximal part of the persistent recess accounts for either a diverticulum with a narrow neck or a cyst in communication with the pericardial cavity.[20] If the proximal portion of the recess is completely pinched off, an isolated pericardial cyst occurs in the cardiophrenic angle.[20] This concept was soon generally

accepted, and henceforth the term "pericardial celomic cyst" was used.

First Case Series (1950s and 1960s)

Several case series regarding either pericardial cyst or diverticulum were published during the 1950s. In 1959, Le Roux (University of Edinburgh) reported on 20 cases of pericardial cyst that he had encountered at the Royal Infirmary in Edinburgh.[2] All cysts were easily removed via thoracotomy without death. Similar experiences were recorded by Ochsner and Ochsner[21] (Ochsner Clinic, New Orleans) and Weig and Fuge[22] from the University of Buffalo, New York. The encouraging results of these series caused surgical resection to become the generally accepted approach for both diagnosis and cure of pericardial cysts and diverticula.[23-26]

Furthermore, these case series revealed interesting facts about the epidemiology of pericardial cysts. In 1946, Blades reported on a series of 109 patients who had undergone resection of a mediastinal mass at Army thoracic surgery centers throughout the United States during the 1940s.[27] Pericardial cysts accounted for 10 of these lesions and were always asymptomatic.[27] Sabiston and Scott (Johns Hopkins Hospital, Baltimore) collected a case series of 101 patients with primary tumors or cysts of the mediastinum who were admitted to the Johns Hopkins Hospital between 1933 and 1951.[28] This amounted to a prevalence of 1 in approximately 3,400 admissions. Only 2 pericardial cysts were observed among the 101 cases.[28] When the results of a mass radiographic campaign in Edinburgh were published in 1958,[2] Le Roux reported that 3 out of 300,000 subjects were found to have a pericardial cyst. Therefore, he calculated "that about 1 in every 100,000 of the population at large may have such a cyst."[2] Hence, these series confirmed the extreme rarity of pericardial cysts and diverticula.

Should Every Lesion Be Removed?

The indication for surgery in cases of pericardial cyst or diverticulum has been questioned from time to time, because these lesions are always benign and most are asymptomatic.[2,27,29] However, in many cases, doubt regarding the exact nature of the mediastinal mass remains despite all diagnostic measures, a problem discussed by Le Roux in 1959: "The difficulty of making a firm diagnosis without recourse to thoracotomy is emphasized throughout the literature."[2] In 1949, Clarence Schein was convinced that "the cyst is of little consequence except in the differential diagnosis of space-occupying lesions of the mediastinum," and he stated that "in case of doubt excisional therapy is indicated."[16] Because needle aspiration of the cyst has shown discouraging results, it is not widely accepted.

Moreover, case series have frequently revealed such nonspecific symptoms as dyspnea, chest discomfort, and substernal pain.[30-32] Serious sequelae have also been recorded. For example, hemorrhage into a pericardial cyst followed by acute heart failure[33,34] has been reported, as has spontaneous rupture.[35] Infection of pericardial cysts is rather uncommon. Therefore, surgery remains the generally accepted therapeutic approach.

Outlook

The introduction of minimally invasive, video-assisted thoracic surgery revolutionized the surgical management of pericardial cyst and diverticulum. Otto Pickardt had used thoracoscopy to verify a suspected mediastinal mass as early as 1931.[1] However, he lacked the technical means to remove the cyst via thoracoscopy. Now the wheel has turned full circle, and video-assisted thoracoscopy is a promising option for both diagnosis and definitive treatment.

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