THORACIC GASTRIC CYST

ANIBAL ROBERTO VALLE, M.D., AND M. LAWRENCE WHITE, JR., M.D.
CHARLOTTESVILLE, VA.

FROM THE DEPARTMENT OF SURGERY AND GYNECOLOGY, UNIVERSITY OF VIRGINIA SCHOOL OF MEDICINE AND UNIVERSITY HOSPITAL, CHARLOTTESVILLE, VA.

THERE is a great variety of cysts which occur in the mediastinum. One of the more unusual types is of entodermal origin—the gastric cyst. We have found only 16 cases in the literature (Table I) and the rarity of the condition seems to justify this case report.

Case Report.—M. C., a 22-months-old white female child, was admitted for the first time to the University of Virginia Hospital, May 3, 1945, with a history of pulmonary symptoms since shortly after birth.

The patient was born in another hospital, of normal parents, after a normal pregnancy and uncomplicated delivery. She was the first of three children, apparently normal twins being born about one year later. The patient's first two weeks of life while still in the hospital were uneventful, but soon after she was taken home she developed symptoms of a "cold," with a slight cough, nasal congestion and low grade fever. These symptoms persisted until the age of four months when she began to have severe paroxysms of coughing accompanied by cyanosis. Her voice became hoarse, and a diagnosis of "membranous croup" was made by the family physician. A chest film at that time is said to have revealed evidence of pneumonia. These symptoms, wet paroxysmal cough, fever, hoarseness and episodes of cyanosis continued despite several courses of sulfonamide therapy. Serial chest films revealed what was interpreted as a chronic pneumonia on the right side, which, in view of the prolonged illness, was finally decided by her local physician to be a lipoid pneumonia. At the age of seven months, the patient coughed up a large quantity of bright blood, estimated at one pint by the mother. Smaller hemorrhages occurred several times within the next three days. Clubbing of the fingers was noticed at this time by the mother. Since then, until admission to this hospital, seven severe pulmonary hemorrhages occurred, each requiring hospitalization and multiple blood transfusions. Only in recent weeks had the persistently wet cough resulted in the expectoration of grossly purulent sputum.

The child had not developed normally. She had been able to sit alone for several months but still could not stand alone, and had made no intelligible sounds. Her appetite was fairly good, but sleep was interrupted by frequent coughing attacks.

Physical Examination: Examination of the patient on admission revealed a fretful, poorly developed, undernourished, and obviously chronically ill child, weighing 18 pounds. The temperature, pulse and respiratory rate were normal. The skin was cold and moist, and the hair was very scanty. Marked clubbing of the fingers and toes was present. The chest had an enlarged anteroposterior diameter, suggesting emphysema. Auscultation revealed coarse moist râles bilaterally from apex to base, the right side harboring the most marked signs. There was no appreciable dullness to percussion. The abdomen was moderately distended and the liver edge was palpable two centimeters below the costal margin. Nose and throat examination was negative except for much mucus in the pharynx. There were no palpable lymph nodes of significance.

Laboratory Studies: Uranalysis revealed a one plus albumin and 18–20 white cells per high power field of the centrifuged specimen. The hemoglobin was 14 Gm./100 cc., or 91 per cent (Haden). The red cell count was 5.81 million, and the white cell count was 19,000. The differential count of the white cells showed 44 per cent polymorphonuclear cells, 49 per cent lymphocytes, 5 per cent transitional monocytes, 1 per cent basophils, and 1 per cent eosinophils. (Three blood transfusions had been given the week prior to admission.) Stool examination was negative. The Shick and tuberculin tests were both negative. Culture of the sputum revealed a multiplicity of organisms, including *Staphylo-*

TABLE I

THORACIC GASTRIC CYSTS

(*Reported in the Literature*)

Author	Sex and Age	Functional Activity	Treatment	Result
1. Staehelin & Burckhardt, 1909	Female, 9 mos.	Inactive	?	Autopsy finding
2. Mixter & Clifford, 1929	Male, 22 mos.	Active	Drainage and excision	Good
3. Mixter & Clifford, 1929	Male, 7 mos.	Inactive	None	Autopsy finding
4. Smith, 1930	Male, 14 mos.	Inactive	None	Autopsy finding
5. Fischer, 1930	Female, 6 mos.	Inactive	None	Autopsy finding
6. Entz & Orosz, 1930	Male, 11 mos.	Inactive	Drainage	Died, verified at autopsy
7. Poncher & Miles, 1933	Male, 29 mos.	Active	Operation	Died on op. table
8. Boss, 1937	Male, 3 yrs.	Active	Operation	Died on op. table
9. Seydl, 1938	Female, 3 mos.	Active	None	Autopsy finding
10. Nicholls, 1940	Female, 3 yrs.	Active	Drainage and excision	Good
11. Schwartz & Williams, 1942	Male, 23 yrs.	Inactive	Excision	Good
12. Schwartz & Williams, 1942	Male, 4 mos.	Active	Excision	Died
13. Carlson, 1942	Male, 4 mos.	Active	Excision	Good
14. Wyllie & Pilcher, 1943	Female, 12 mos.	Active	Excision	Good
15. Olken, 1944	Male, newborn	?	None	Autopsy finding in stillborn
16. Laipply, 1945	Male, 27 hrs.	?	None	Autopsy finding
17. Valle & White, 1945	Female, 22 mos.	Active	Operation	Died on op. table

coccus aureus nonhemolyticus, *H. influenzae*, unidentified micrococci, *Streptococcus viridans*, and a yeast-like organism never fully identified but apparently nonpathogenic.

Roentgenologic Examination: Roentgenograms of the chest revealed a marked mottled infiltration particularly in the right lower lobe but with some involvement also in the middle and upper lobes. There was a dense elliptical shadow in the right hilar region which was thought to be an enlarged hilar lymph node. No evidence of cavitation was seen. The left lung was essentially clear. A review of the old films taken at intervals since the patient was four months old disclosed similar hilar adenopathy and infiltration in the right lung.

Bronchograms revealed generalized bronchiectasis of the right lung and a large cavity in the posteromesial portion of the right lower lobe. The oil flowed readily into this cavity through a large bronchial communication (Fig. 1). The left side was normal.

Laryngoscopy and bronchoscopy were performed on May 8, 1945. The true vocal cords were reddened and edematous, but the movements were normal. The mucosa of the trachea and right bronchial tree was also subacutely inflamed and copious mucopurulent secretions were aspirated. The left bronchial tree presented a mild inflammatory reaction with a small amount of secretion. There was no evidence of foreign body, tumor or stenosis.

The diagnosis of bronchiectasis with an infected bronchial cyst was made. Since the condition seemed to offer a threat to life, surgical extirpation of the diseased lung was advised in spite of the poor general condition of the patient.

Operation.—May 14, 1945: One of us (A. R. V.) explored the right chest through a long posterolateral incision, entering the pleural space through the periosteal bed of the resected seventh rib. The pleural space was partially obliterated by numerous adhesions but no difficulty was encountered in mobilizing the lung except in the posterior gutter where the lower lobe was very densely adherent. Just above the diaphragm in the lower lobe there was a dense mass, apparently inflammatory, about the size of a hen's egg. This mass was presumed to represent the cystic area seen roentgenographically. With much difficulty this portion of the lung was separated from the parietal pleura, but in so doing the cystic cavity was broken into and the peripheral portion of the cavity wall, densely attached to the diaphragm and posterior gutter, remained behind as the mobilization of the lung was completed. The lung itself was obviously infected, there being areas of dense inflammatory infiltration from apex to base. Pneumonectomy seemed justified and feasible. Hilar dissection was attempted but enlarged hilar lymph nodes

and friable inflamed tissues rendered impossible the individual isolation of the vessels and bronchus. Therefore, the Shenstone tourniquet was placed around the hilar structures, and a transfixion suture of heavy silk was used to ligate the hilum *en masse*. The lung was then amputated. After release of the tourniquet, the bronchus and vessels were individually treated, the former with fine silk sutures and the latter with silk ligatures. The stump was covered with mediastinal pleura. At this stage in the operation the heart suddenly stopped, as if from vagal stimulation, and all attempts to initiate

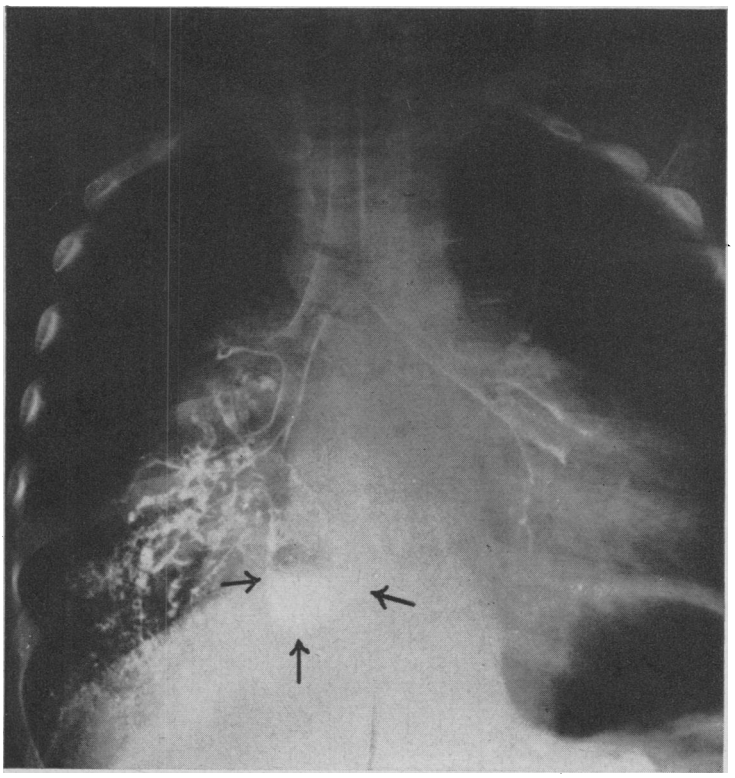

FIG. 1.—Bronchogram showing cyst communicating with the right lower lobe bronchus. Arrows outline the cyst which is partially filled with iodized oil.

pulsations by constant massage and medications for 20 minutes were of no avail. Spontaneous respiration ceased several minutes after cardiac arrest.

It had been our plan to remove the remainder of the cyst wall from the diaphragm and posterior chest wall after the lung had been removed. Postmortem dissection on the operating table of the remaining portion of the cyst proved that complete excision would have been an impossible surgical feat. Closer examination of the lining of the cyst suggested typical gastric mucosa with rugae, and the true diagnosis was first suspected at this time. Further dissection found the remnants of the cyst to extend down through the diaphragm, in intimate contact with the inferior vena cava, the bodies of the vertebrae and the esophagus. Adjacent inflammatory tissue necessitated sharp dissection throughout, and even with the utmost care in order to preserve the specimen the inferior vena cava was entered. The cyst ended blindly about five centimeters below the diaphragm, without anatomic continuity with either esophagus or stomach.

Pathologic Examination: The specimens consisted of an infant's right lung and an irregular piece of tissue, measuring 11 x 5 centimeters. The external appearance of the lung revealed extensive pleural adhesions and a small cavity communicating with

the posterior portion of the right lower lobe. When the lung was sectioned, dilated bronchi were found throughout. A probe passed into the cavity easily entered a branch of the right lower lobe bronchus. The separate piece of tissue grossly resembled stomach wall, having a mucosa with characteristic rugae and apparent muscular layers. The cavity in the lower lobe mentioned above had a similar mucosal pattern.

Microscopic sections revealed bronchiectasis, subacute bronchitis and areas of pneumonitis throughout the lung. Sections through the cyst wall showed the elements of a normal stomach, with typical mucosa and muscular layers (Figs. 2, 3 and 4). One section taken through the bronchial communication with the cyst shows the juxtaposition of the gastric mucosa with the elements of the bronchial wall (Figs. 2 and 4).

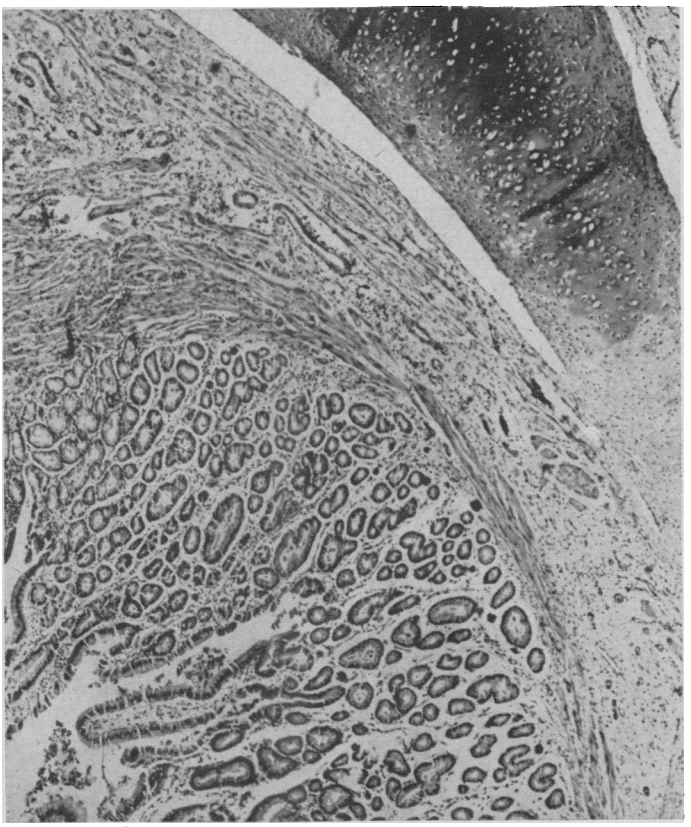

FIG. 2.—Photomicrograph showing typical gastric mucosa and bronchial cartilage.

COMMENT: There are two kinds of gastric cysts, those that are acid-secreting, or functionally active, and those of which the mucosa is without functional activity. Even though the acidity of the cystic secretions was not determined in our case, the early manifestation of symptoms—primarily hemorrhage, is so typical of the proven functionally active cases in the literature, that we feel justified in classifying this case as an acid-secreting or functionally active cyst.[10] From a review of the literature, it is found that there are two constant characteristics of a gastric cyst—it always occurs in the posterior mediastinum and on the right side. The functionally active cysts usually manifest themselves by symptoms at an early age, whereas the inactive

ones may be discovered accidentally in a routine chest roentgenogram later in life.[10]

There are several theories about the origin of endothoracic gastric cysts. They have been ascribed to a pinching-off of an evagination of the embryonic foregut, to an intrathoracic vestige of the omphalomesenteric duct, and to a proliferation of an entodermal germ cell of the esophagus capable of producing gastric epithelium.

The cyst wall has all the elements of the stomach wall. Smith,[12] and

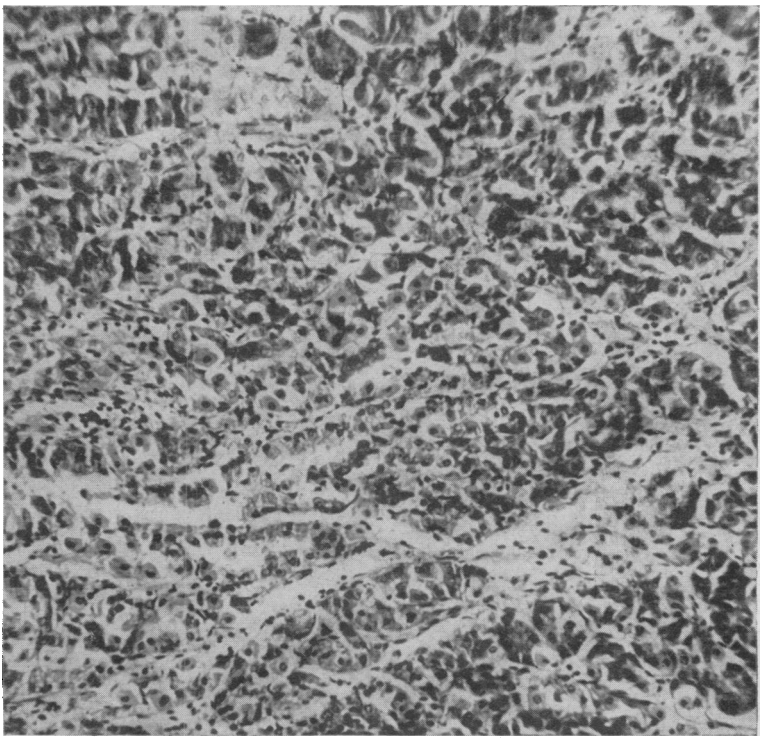

FIG. 3.—High power photomicrograph showing chief cells of the gastric mucosa of the cyst.

Staehelin-Burckhardt[13] have each reported a case which exhibited both gastric mucosa and ciliated stratefied respiratory epithelium in the same cyst. Boss[1] has described a case in which the intrinsic nerves and ganglia of the gastric musculature were identified. In our case, the cyst perforated the diaphragm and terminated in a blind sac about five centimeters below it. To our knowledge this feature has not been reported in the literature. It did not communicate with the stomach or the esophagus. Erosion of bone may be caused by the cyst, as reported in one of Mixter and Clifford's cases.[6]

The most commonly observed symptoms of gastric cyst are dyspnea, cyanosis, cough, dysphagia and sometimes, even in infants, hemoptysis. The most striking symptom of our case was the massive hemoptysis which necessitated admission to her local hospital on nine different occasions, with multiple blood transfusions during each period. Both Boss and Seydl reported

hemoptyses in their cases and attributed them to the presence of chronic peptic ulcers in the gastric cyst. In both cases the ulcers penetrated into the adjacent lung causing fatal pulmonary hemorrhage. Although no peptic ulcer could be demonstrated in the specimen from our case, it is probable that one had been present, since the cyst had eroded into the right lower lobe bronchus and was draining through the bronchial tree. Inasmuch as this

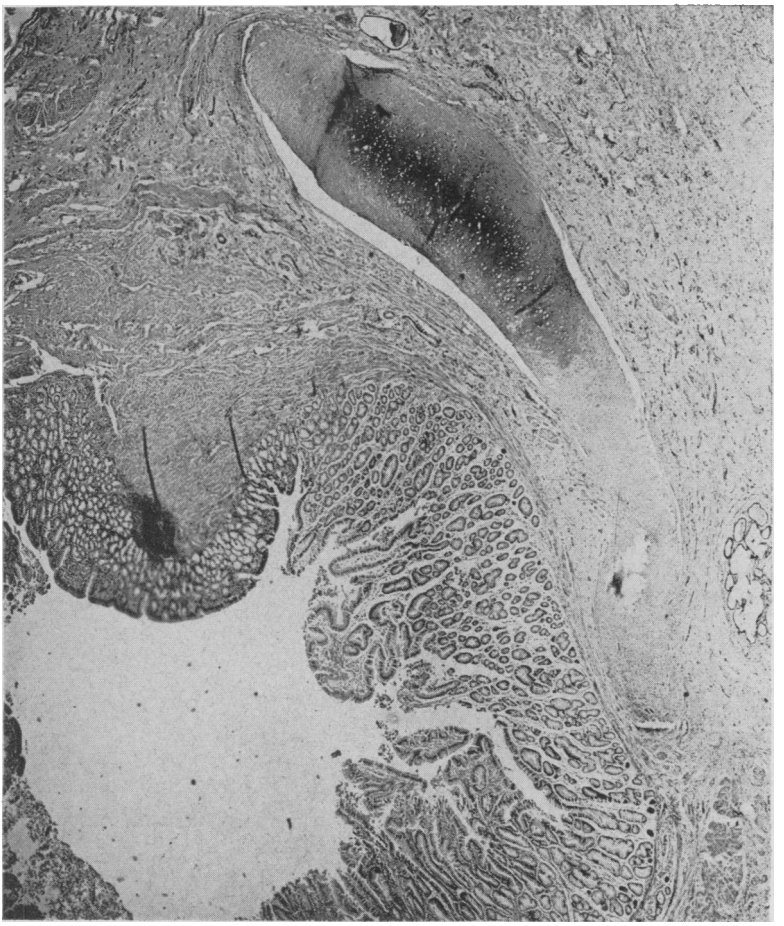

FIG. 4.—Low power photomicrograph of section taken at point where the cyst drained into the bronchial tree.

cyst was functionally active, the hemoptysis was probably due to bleeding from the ulcer itself and to the action of the gastric juice on the lung parenchyma and the bronchial tree.

The physical findings are variable but are usually those of pulmonary atelectasis or pneumonitis. There may be some cardiac displacement, scoliosis, or rarely, bulging of the chest. The findings in our case were primarily those of severe pneumonitis. The generalized bronchiectasis of the right lung, a complication resulting from the rupture of the cyst into the bronchial tree, has not previously been reported.

It is difficult to diagnose a gastric cyst during life with any degree of certainty without exploratory operation. However, the diagnosis is possible if gastric juice is aspirated directly from the cyst.[2] The cyst may contain from a few cubic centimeters of fluid to 400 cc. The fluid may be milky or clear, viscid or thin, or even sanguineous. The reaction is usually acid but may be neutral. Roentgenograms in both the postero-anterior and lateral projections are important, the lateral view particularly so, to show the cyst in the posterior mediastinum. Iodized oil bronchography is of use to show whether or not the cyst communicates with the bronchial tree and if so, to demonstrate its size, shape and location. In our case a diagnosis of posterior mediastinal cyst communicating with the bronchus complicated by generalized bronchiectasis of the right lung was made preoperatively. However, it was not until the typical gastric mucosa was seen when the cyst was opened at operation that the correct diagnosis was suspected.

The only treatment for gastric cysts is complete removal. If only a minute amount of the mucosa remains in the chest it will secrete and cause further complications such as empyema or a draining sinus, with or without excoriation of the skin. To date surgical treatment has not been too successful. Of the 17 cases summarized in Table I, ten had surgical treatment. Five of these survived, representing an operative mortality of 50 per cent. Two of these had a successful two-stage operation (drainage and later excision) and three had a one-stage removal. The high mortality is probably due to several factors. The patients are usually infants; they are usually in poor condition for a major operation; anesthesia is difficult to administer; and the dissection of the cyst from the mediastinum is technically impossible in some cases. The last factor is due probably to adjacent inflammatory reaction from the functional activity of the cyst. In some cases where complete removal of the cyst is impossible in a one-stage operation, preliminary drainage is useful as a palliative measure.

SUMMARY

A case of thoracic gastric cyst with certain unusual features has been reported. The cases reported in the literature have been summarized and a limited discussion has been presented.

REFERENCES

1 Boss, Carl: Virchows Arch. f. Path. Anat., **300**, 166, 1937.
2 Carlson, H. A.: Jour. Thor. Surg., **12**, 376, 1943.
3 Entz, B., and Orosz, D.: Frankfurt. Ztschr. f. Path., **40**, 229, 1930.
4 Fischer, Walther: Virchows Arch. f. Path. Anat., **275**, 711, 1929.
5 Laipply, T. C.: Arch. of Path., **39**, 153, 1945.
6 Mixter, C. G., and Clifford, S. H.: ANNALS OF SURGERY, **90**, 714, 1929.
7 Nicholls, M. F.:Brit. Jour. Surg., **28**, 137, 1940.
8 Olken, H. G.: Am. J. Path., **20**, 997, 1944
9 Poncher, H. G., and Milles, G.: Am. Jour. Dis. Child., **45**, 1064, 1933.
10 Schwartz, H., and Williams, C. S.: Jour. Thor. Surg., **12**, 117, 1941.
11 Seydl, Gunther N.: Frankfurt. Ztschr. f. Path., **52**, 346, 1938.
12 Smith, R. E.: Guy's Hosp. Rep., **80**, 466, 1930.
13 Staehelin-Burckhardt, A.: Arch. f. Verdauungskr., **15**, 584, 1909.
14 Wyllie, W. G., and Pilcher, R. S.: Arch. Disease Child., **18**, 34, 1943.