INTRATHORACIC DUPLICATIONS OF THE FOREGUT

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PART I

The exact origin, formation and mode of development of duplications of the foregut are still controversial. Some explanations were advanced at a time when the nature of these anomalies was ill understood; other theories were based on the features of a single case or a particular type of abnormality and do not necessarily apply to other types. Explanations that seem to satisfy the morbid anatomy are not always acceptable embryologically. The common underlying principles are best recognized if malformations are studied in perspective, and sufficient literature has now accumulated to make this possible.

Spherical or elongated cyst-like duplications of the foregut may occur at any level from the base of the tongue to the second part of the duodenum. They are frequently classified into intramural cysts of the oesophagus, bronchogenic cysts and enteric cysts. The literature further leads one to believe that the distinction between these types according to their situation, attachments and structural characteristics is easy and accurate. My own study of the clinical, operative and histological features of duplications in 26 patients (excluding all mediastinal cysts of lymphatic and serosal origin, sequestrations, intrapulmonary cysts of bronchial origin, dermoids and hamartomas) does not altogether support these views. Of the 26, 9 were unquestionably oesophageal, 15 arose in the bronchial system, and in 2 it was impossible to ascertain the exact site of origin. A review of the literature showed that certain cases reported as teratoma or hamartoma were histologically identical with duplications described in this paper. The diagnosis in hospital records will therefore depend on the interpretation and experience of the microscopist. Thus 2 cases in the present series were initially called hamartomas.

The distribution and size of the duplications of bronchial, alimentary and undecided origin that are described in this paper, as well as the age and sex incidence of the patients, are shown in Fig. 1.

DUPLICATIONS OF BRONCHIAL ORIGIN

These are commonly called bronchogenic cysts. Laipply[47] collected 34 cases from the literature in 1945. However, these cysts are relatively common; they are mostly diagnosed on routine or mass radiography or at necropsy. They seldom produce symptoms when small, but may give rise to pressure effects when large, especially in young children, in whom they fortunately present less commonly. Usually they increase in size very slowly; several cases in this series were kept under observation for years with only slight enlargement. There was no familial incidence and no difference in the sexes. Contrary to findings recorded in the literature, the cysts occurred mainly on the right side in this series. None became infected.

Only in one case was an associated anomaly found, viz. a high dorsal scoliosis with a congenital abnormality of the spine in a boy of 14 who was X-rayed as a contact for tuberculosis. The cyst was slightly embedded in the muscle layers of the lower third of the oesophagus, but histologically it was a characteristic bronchial duplication. In another case

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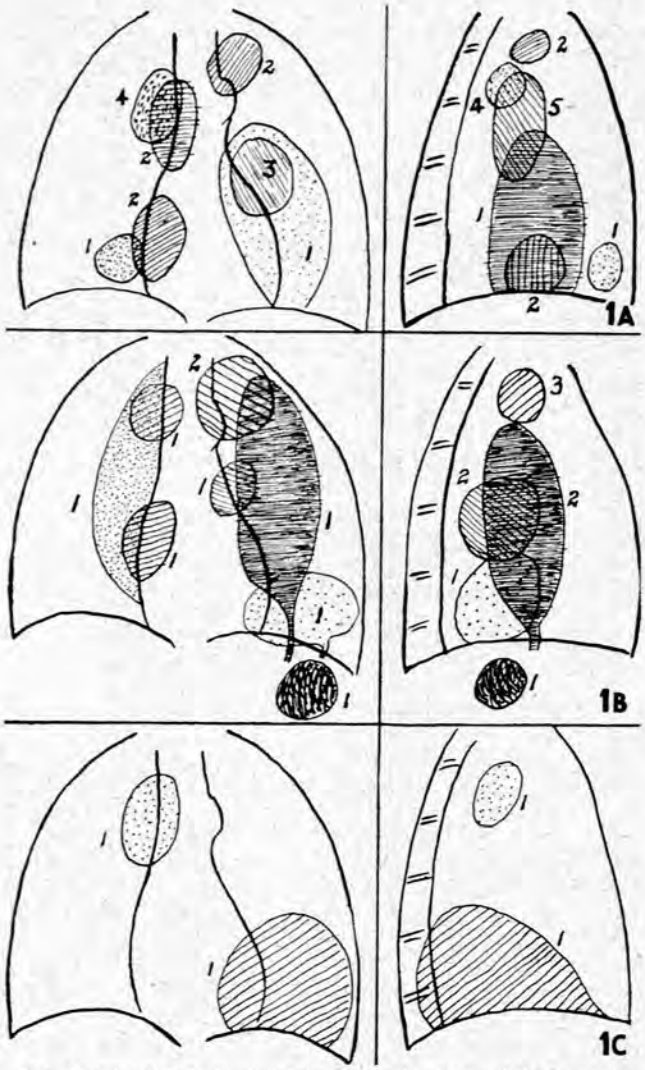

Fig. 1. The distribution and size of duplications of the foregut (diagrammatic AP and lateral drawings) in 26 patients.
(A) 15 duplications of bronchial origin in 7 female and 8 male patients at the ages of 15 months (1), 4 years (1), 5-10 years (2), 10 - 20 (2), 20 - 40 (4), 40 - 60 (4), and 65 (1).
(B) 9 duplications of alimentary origin in 4 female and 5 male patients at the ages of 5 years or under (5), 10 years (1), and between 25 and 55 (3).
(C) 2 mixed duplications in 2 males of 17 and 58 years.

the cyst in the posterior mediastinum was loosely attached to the lung, but was firmly adherent to the heads of the left 4th and 5th ribs and the vertebral column. It is possible that more of these patients had skeletal abnormalities, but in none were any special views taken of the vertebral column or spinal anomalies specially looked for. Minor degrees of rotation, especially in infants and children, make the interpretation of films often difficult, and spinal lesions which are conspicuous on some X-rays may be completely missed on others. Even tomography[79] does not always succeed in showing up the skeletal defects.

The cysts are almost always thin-walled, semi-translucent, single or multilocular, and lie characteristically in the posterior mediastinum in close relation to the trachea and main bronchi. They may be in a superficial extrapleural plane without any apparent connections and shell out with great ease, or be connected by a vestigial fibrous core or rudimentary bronchus to the tracheobronchial tree. In 2 cases the cyst wall was inseparable from the trachea in the mid-line. In one case an oval defect about 1·5 cm. long was present in 4 tracheal rings, where the cyst was adherent to the side wall of the trachea. In 2 other cases the cyst was embedded in the oesophageal wall, lying within the muscle layers of the organ. In one case (Fig. 2) the cyst was wedged in between the trachea and oesophagus so that the muscle fibres of the oesophagus had to be divided and sutured afterwards, and a fragment of the cyst wall had to be left behind on the posterior wall of the trachea, with which it was fused. In none of the cysts was there a fistulous communication with the oesophagus or tracheobronchial tree. The size of the cyst depends primarily on the degree of differentiation of its secretory epithelium and the abundance of deep glands. The contents usually consist of a milky, gelatinous, mucoid material with desquamated epithelial cells and of alkaline reaction.

The histology of these cysts is fairly uniform and the diagnosis seldom presents any difficulty (Fig. 3). The epithelium is predominantly pseudostratified ciliated columnar, but the cells may be flattened in certain areas or the cilia may be absent. It may however be stratified squamous of adult type, or resemble a more primitive stratified pattern. It may be smooth and uniform, or heaped up and irregular, or show such marked papillary formation as to suggest intestinal secretory epithelium. The epithelial lining was sometimes shed in parts or in its entirety.

The epithelium is supported by a loose connective-tissue basement membrane lying on a thin layer of irregular smooth muscle bundles of varying thickness. Fragments of hyaline cartilage were present in varying amounts, but sometimes completely absent. Pale-staining mucous glands were common within the muscle layer, but were not invariably found.

FOREGUT DUPLICATIONS OF ALIMENTARY ORIGIN

Ladd and Gross[46] instituted the collective term 'duplication' to cover the variety of interesting congenital anomalies

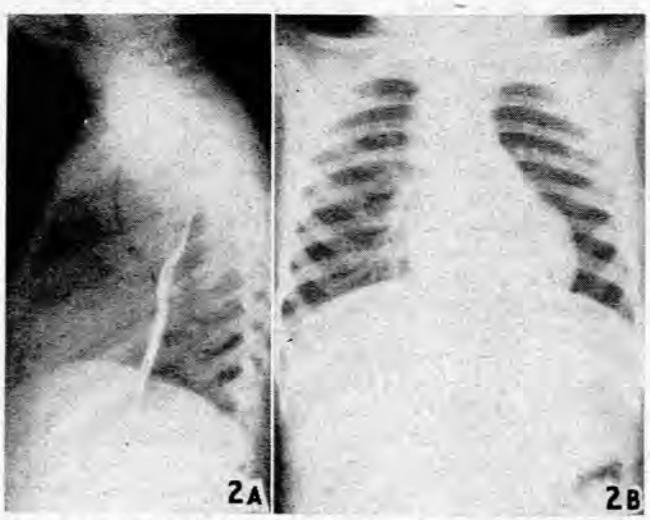

Fig. 2. The AP (B) and lateral (A) radiographs in a girl of 4 years, showing a duplication of bronchial origin in a central position between the trachea and oesophagus.

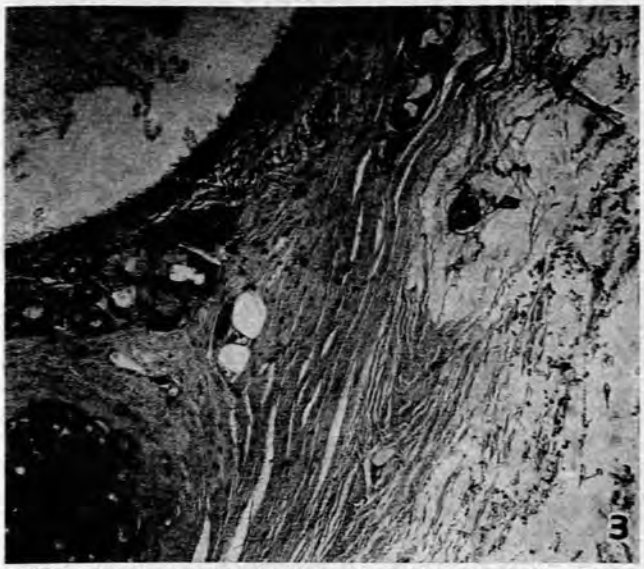

Fig. 3. The typical histological appearances of a duplication of bronchial origin, viz., pseudostratified ciliated columnar epithelium, scanty smooth-muscle fibres, pale-staining mucous glands, and cartilage.

formerly known as enteric cysts, enterocystoma, enterogenous cysts, accessory cysts, accessory thoracic stomach, archenteric cysts, etc. Although formerly regarded as curiosities of morbid anatomy, the surgical importance of these cysts is now fully recognized. Gross's criteria[31] for duplications—structures which (1) possess a coat of smooth muscle, (2) are lined by a mucous membrane similar to some part of the alimentary canal, and (3) are intimately attached to some portion of the alimentary tube—are mainly true for the intestine, but unfortunately they have been applied to the oesophagus without qualification. In contradistinction to the cysts of bronchial origin, which usually follow a benign course and are commonly diagnosed in older subjects on mass radiography, duplications of oesophageal origin frequently give rise to symptoms in infancy or childhood and may rapidly develop disastrous complications if not treated surgically.

In 1947 Lindquist *et al.*[51] collected from the literature 80 cases of intrathoracic cysts lined by alimentary-tract mucosa; 40 were in the mediastinum. All 8 cysts in the present series were mediastinal and posterior (Fig. 1). They varied from a small spherical structure 3 cm. in diameter to large multilocular and elongated sacs which almost filled the hemithorax. Some cysts were in a superficial extrapleural plane unattached to the oesophagus or other mediastinal structures (Fig. 4), but the majority were relatively thick-walled and adherent to surrounding structures. Unattached duplications were normal on barium swallow but others were so intimately connected with the oesophagus that a deformity or filling defect could be demonstrated. One cyst had a fine fibrous pedicle 3 mm. thick, which extended upwards through the thoracic inlet. Two of the cysts passed through the left hemidiaphragm and one was connected to the fundus of the stomach. None, however, communicated with a hollow viscus, although this has been described. The diagnosis is then suggested by a fluid level in the erect position or by outlining the duplication with barium.

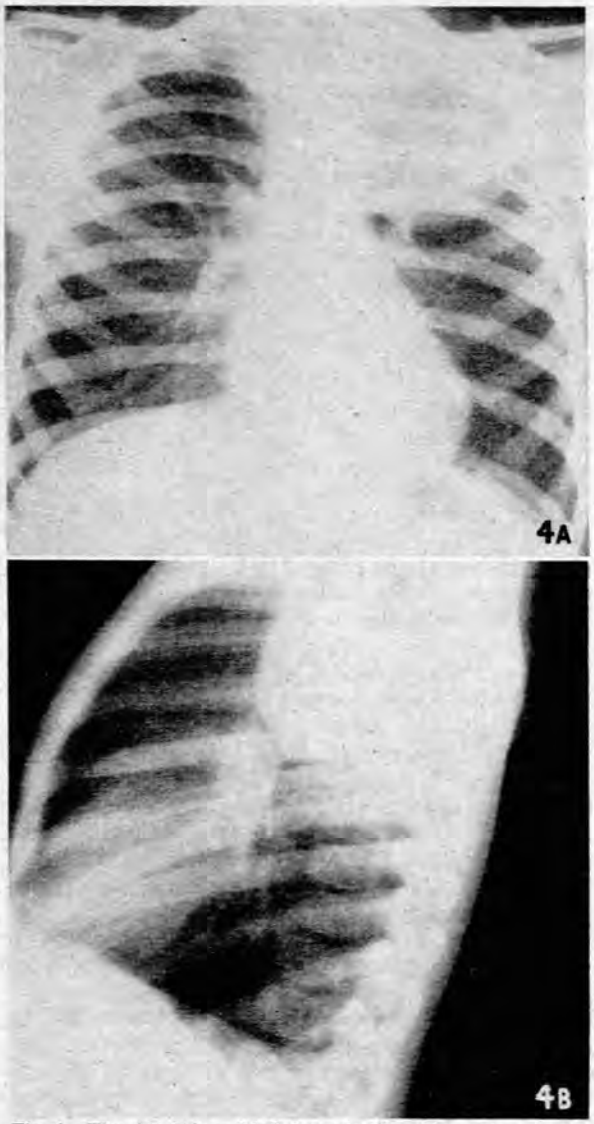

Fig. 4. The AP (A) and left lateral (B) radiographs in a boy of 6 years with a duplication of oesophageal origin.

These cysts are relatively thick-walled and may resemble the wall of the alimentary tube very closely (Fig. 5). The epithelium may be uniform throughout, appearing as a smooth lining indistinguishable from that of the oesophagus or stomach, or it may be irregular and many different types of epithelium may be present in one cyst. The squamous epithelium is often of a primitive type; the cells are large and only a few layers thick, non-ciliated and containing large pale-staining nuclei. This epithelium may become flattened and almost cuboidal, or it may change to typical pseudostratified ciliated respiratory epithelium, or become more differentiated into typical stratified squamous epithelium. In the 2 elongated cysts the squamous epithelium changed above the diaphragm to a zone of typical cardiac glands with short ducts and tortuous tubules, scanty or absent oxyntic cells, and pale-staining mucous glands. This zone quickly changed to gastric mucosa of fundic type, rich in oxyntic cells with long tubular ducts. Differentiation of squamous epithelium into gastric

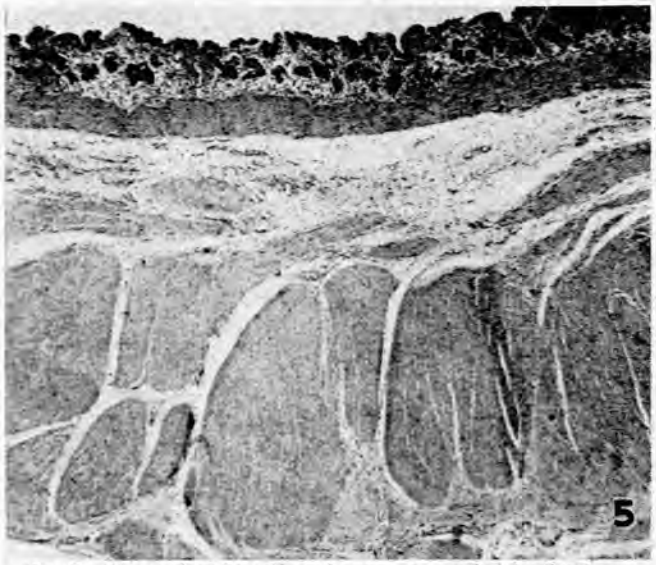

Fig. 5. The typical histological appearance of a duplication of oesophageal origin, viz., a well-defined secretory epithelium, thick muscularis mucosae, and well-developed muscle coat.

epithelium is often incomplete, so that isolated islands of gastric mucosa occur, and in 2 cases there was evidence of ulceration and chronic granulation-tissue formation around the islands of secretory epithelium. In certain areas the entire mucosa was shed. Intestinal or colonic epithelium was not found in the present series but Olenik *et al.*[59] found in a reviewed series (1946) that 12 cysts were lined with primitive oesophageal epithelium, 15 with gastric mucosa, 4 with intestinal mucosa, and 8 with a mixed epithelium representative of both the respiratory and intestinal tracts.

The muscularis mucosae (smooth muscle) is very well developed and commonly consists of 2, 3, or even 4 layers of longitudinal, circular and oblique fibres. The submucosa may be very thick and then contains numerous dilated thin-walled blood vessels and collections of lymphoid tissue, or it may be thin and ill-defined. A myenteric plexus of ganglion cells can often be differentiated, and occasionally large nerve trunks may be identified, suggestive of branches of the vagus nerves. The muscle coat is very well developed and consists of 2-5 layers of circular and longitudinal muscle fibres. The bundles are irregularly arranged, however, and lack the uniform appearance of efficiency which characterizes normal gut.

The contents varied from a mucinous opalescent fluid to a chocolate-coloured paste containing a high proportion of epithelial debris, altered blood and red cells. The pH varied from 7·8 to 3. Hydrochloric acid, enzymes and cholesterol crystals were present in varying amounts.

Associated Abnormalities

Roth,[70] in 1881, described a mediastinal enteric cyst which was firmly adherent to the vertebral column in a patient who also had an ileal duplication and a cyst of the mesentery, again illustrating the principle that congenital anomalies are often multiple. They are usually intestinal or skeletal, or both.

Skeletal abnormalities. In 1954 Fallon *et al.*[23] found 25 vertebral abnormalities (18 of which were spina bifida anterior, hemivertebra or infantile scoliosis) in a series of 61 cases which they collected from the literature, but they also pointed out that the standard chest radiograph is far from ideal for the study or even the recognition of vertebral lesions. Spinal abnormalities in the cervicodorsal area are relatively far cranial to the duplication, so that attention would only be focussed there by observers who are aware of the association. Unfortunately the cervical vertebrae are often not even on the film. The fact that skeletal abnormalities have been missed in some of the cases described in this paper, as well as in other cases of tracheo-oesophageal fistula that I have reviewed, confirms that the incidence of skeletal anomalies is higher than reported by other workers.[80,59,45]

The basic spinal lesion is incomplete anterior fusion. This may vary from minor degrees of cleft vertebra to frank anterior spina bifida, where the halves of the vertebral bodies on each side of the cleft usually diverge to form an oval passageway allowing the spinal cord to come into some form of relation with the gut. Feller *et al.*[24] in 1929 collected 28 cases of malformation which involved the central nervous system and the alimentary canal, and all of them had anterior spina bifida.[24] In my own experience this is also the commonest vertebral anomaly associated with tracheo-oesophageal fistula and the Klippel-Feil type of syndrome. Ladd *et al.*[45] mention a case of mediastinal duplication with asymmetrical thorax and short stunted neck that falls into this category. Incomplete anterior fusion may be manifested by hemivertebrae, often resulting in congenital scoliosis. It may affect a single vertebra or a series, may be on the same or opposite sides, and may co-exist with spina bifida anterior or other defects in other vertebrae. Although most of these vertebral abnormalities are found in the lower cervical and upper thoracic vertebrae, Roberts *et al.*[69] found spina bifida as high as the second cervical in one subject and a hemivertebra on the right at the level of the 7th thoracic together with a more extensive spina bifida defect of the cervical spine in another. Both these cases had elongated mediastinal duplications.

Apart from abnormal anterior fusion of the vertebral bodies there may also be, in addition or quite separately, many other skeletal anomalies. Amongst those described are cases of spina bifida posterior,[60,47] scoliosis with[58] or without[22] spina bifida anterior, and cases of fusion of the cervical vertebrae with and without kyphosis.[37,23] A case described in the present paper showed fusion of the 3rd, 4th and (partly) 5th dorsal vertebrae. Erosion and concavity of the upper dorsal vertebrae were found in one of my cases but as the cyst was large, this effect was probably the result of pressure necrosis and not primarily of congenital origin; Mixter *et al.*[55] reported a similar case in 1929. Abnormalities of the thoracic cage have been reported by Roberts *et al.*[69] and Ladd *et al.*[45] Guillery[32] described a remarkable case showing a distorted vertebra with a large mediastinal duplication on the ventral side of the vertebral column, and a smaller cyst on the dorsal side of the vertebral body; both had funnel-shaped extentions which appeared to run into the vertebral body as if to connect with one another. Other vague references to vertebral abnormalities occur.[77,1] Although there may be no radiological evidence of a spinal anomaly, dense adhesions or even fibrous stalks may connect the cysts to the vertebral bodies, posterior rib ends or paravertebral gutter;[70,5,14] several such cases occur in the present series.

Intestinal abnormalities. Intrathoracic duplications of the foregut may be associated with duplications of the stomach,[27] duodeno-jejunal flexure,[69] and ileum.[61] The present series contains 1 case with duplication of the ileum. Out of 81 cases with intrathoracic duplication reviewed in 1953, Ware *et al.*[80] found 3 with associated small-bowel duplications and 1 with a malrotation; Roberts *et al.*[69] have since reported 2 further cases with malrotation.

Mediastinal duplications may end blindly in the diaphragm[55,37,23] or in the hiatus.[62,69] They may also pierce the diaphragm[48] (there is one such case in the present series), be attached to the stomach (author's case), or open into the duodenum or jejunum.[31,25,69]

A rudimentary fissure completely dividing the apical segment from the basal segments of the left lower lobe was the only other variation from normality found in the present series.

FOREGUT DUPLICATIONS OF UNDECIDED ORIGIN

In the great majority of cases it was fairly easy to decide on the site of origin of the cyst, but in 2 cases this could not be done with certainty (Fig. 1). In one, a cyst the size of a tangerine was lying within the wall of the oesophagus in the superior mediastinum between the oesophagus and the trachea. The muscular wall of the oesophagus had to be incised and repaired after enucleation. The thick smooth muscle coat of the cyst and the mixed muscle fibres of the oesophagus merged into one. There were no bronchial glands, cartilage or muscularis mucosae, and the epithelium was cuboidal and ciliated columnar. In the other case a large multilocular cyst was found in the left lower chest, firmly adherent to the diaphragm and vertebral column and apparently arising from the lower oesophagus. The contents were chocolate-coloured and contained altered blood. The muscle wall was thin. There was no muscularis mucosae or cartilage, but numerous mucous glands had all the characteristics of bronchial glands. The epithelium was columnar, ciliated and cuboidal, and in places papillary processes closely resembled cardiac glands.

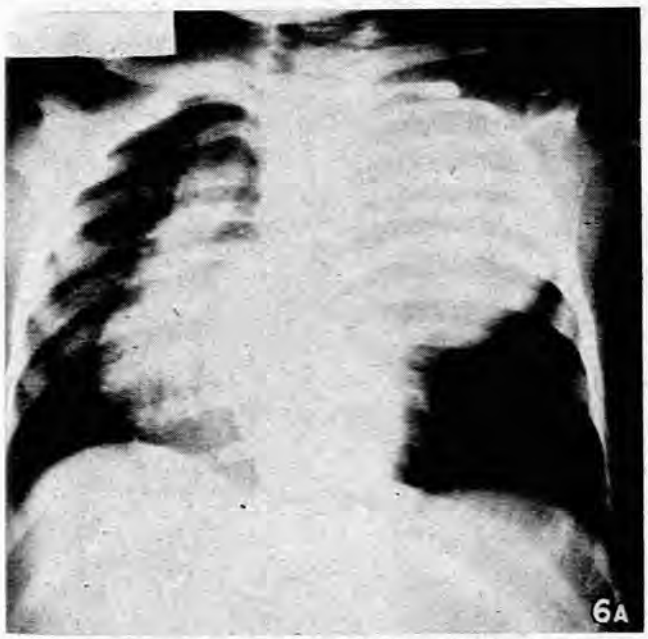

Fig. 6(A). A penetrating AP radiograph of a 2-year-old girl with a foregut duplication in the left upper chest.

ILLUSTRATIVE CASE REPORTS

Case 1

C.G., a girl of 18 months, was referred to hospital with a persistent unproductive cough which became worse after an attack of whooping cough. A chest radiograph showed a right upper mediastinal mass. Barium swallow showed the oesophagus to be pushed to the left and backwards and the trachea displaced forwards. A pre-operative diagnosis of foregut duplication was made. At operation (right thoracotomy) a smooth, thin-walled extrapleural cyst was found between the trachea and oesophagus immediately above the azygos vein. It was slightly embedded in the adventitia of the oesophagus but enucleated easily. Anteriorly the cyst wall fused with the membranous part of the trachea, and this portion had to be left behind after the trachea was inadvertently opened several times. Recovery was uneventful.

The cyst contained a gelatinous opalescent fluid; the wall was smooth and uniform. The histology was characteristic and confirmed that the duplication was of bronchial origin—pseudostratified, ciliated, columnar epithelium, smooth muscle bundles, bronchial glands, and isolated fragments of hyaline cartilage.

Case 2

P.B., a female of 47, was X-rayed for tuberculosis as a contact and a large mediastinal mass was found in the right upper chest. Tomograms showed a sharply defined edge and the diagnosis of a mediastinal cyst was made. At thoracotomy a large, thin-walled cyst was found in the posterior mediastinum, immediately extrapleural, and connected to the trachea by a rudimentary bronchus-like pedicle. The cyst was lined by a uniform smooth mucosa and contained milky mucinous material. In some areas the epithelium was completely shed; in others it was pseudostratified, cuboidal or flattened, with heaped up papillary projections. The further presence of smooth muscle bundles, cartilage, and racemose mucous glands, shows this to be a foregut cyst of bronchial origin.

Case 3

G.P., a full-term female infant of 6½ lb., was first seen at the age of 2 years after an attack of broncho-pneumonia, when a radiograph of the chest showed a left upper mediastinal mass. The Casoni, complement fixation and Mantoux tests were negative, and the blood picture showed no abnormality. Further X-rays revealed a large rounded mass in the left upper chest, encroaching on the vertebral column and eroding the posterior rib ends and the vertebral bodies (Fig. 6A). The mediastinum was pushed over to the right and the upper ribs on the left side were thin, elevated, and asymmetrical. The 3rd, 4th and (part) 5th dorsal vertebrae were fused, giving rise to a slight wedge deformity and scoliosis. A pre-operative diagnosis of foregut cyst or neurofibroma was made.

At operation a large loculated cyst was present, mainly in the superior mediastinum, which because of its size was aspirated to facilitate dissection. A milky sero-mucinous fluid (pH 8) was obtained. Firm adhesions connected the superior surface of the cyst to the extreme apex of the thoracic cavity, but it was mainly adherent to the 3rd and 4th thoracic vertebral bodies. These were concave and partly eroded, but there was no obvious extension of the cyst into the vertebral column. The aorta and oesophagus were displaced to the right. The latter was indentified by passing a bougie, but it was free from the cyst. From the inferior loculus of the cyst a prolongation passed downwards to and through the diaphragm in the neighbourhood of the hiatus, where it was ligated and divided without establishing its connections below the diaphragm.

Pathology. The duplication was 10·5 cm. long and consisted of a large spherical loculus and a smaller oval lower one, connected by a narrow isthmus. One surface was covered by pleura and the other was ragged, with numerous loose fibrous and muscular strands. On cross-section the cyst proved to consist of 3 definite and separate loculi, but intercommunicating by 2 circular smooth

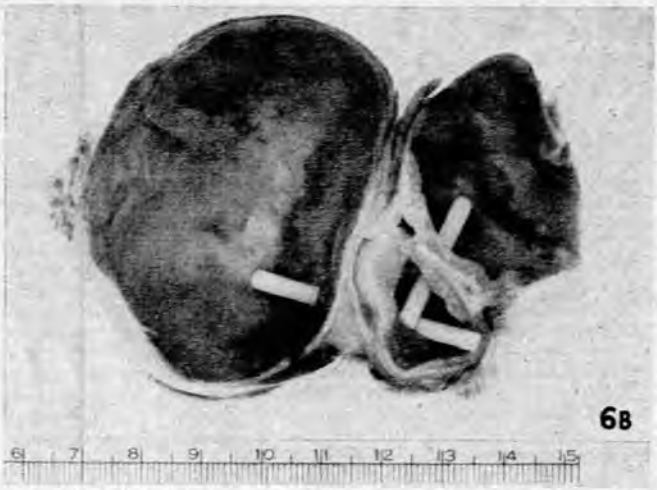

Fig. 6(B). The loculated cyst from the inside. The probes show the holes of communication.

holes 0·9 and 1·2 cm. in diameter (Fig. 6B). The muscular wall of the upper loculus was relatively thin but that of the lower 2 were thick, especially at the isthmus, where the circular muscle layer formed an almost ring-like triangular band. The lining epithelium of the upper loculus was uniform, smooth and pearly white, and vaguely resembled squamous epithelium. Near the upper hole it changed to typical velvety secretory epithelium. In the lower loculus the mucosa further changed and became circumferentially plicated, resembling the folds of Kerkring in the duodenum. Histologically, the upper part contained stratified squamous epithelium and the lower portion secretory epithelium of fundic type.

Follow-up. This child did well after the operation but in subsequent years the mother repeatedly stated that the child complained occasionally of abdominal cramps, and that she had noticed blood in the stools at times. On 2 occasions a barium meal and follow-through gave negative results. When I reviewed the child's X-ray pictures taken when she was 7 years old and

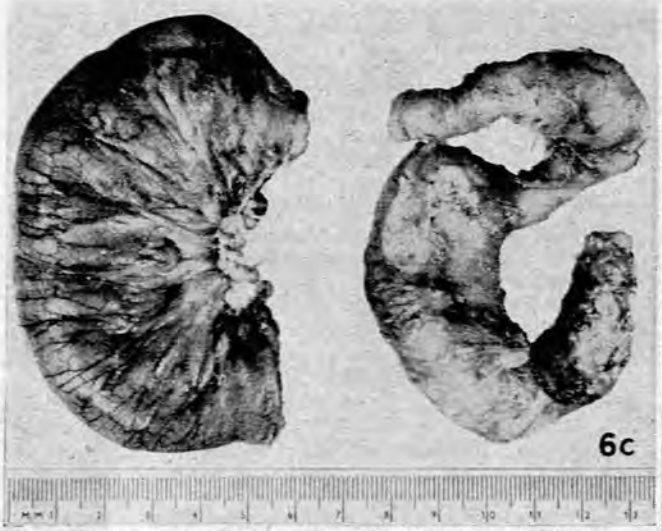

Fig. 6(C). A 10-inch duplication of the small bowel, with and without its mesentery, that was removed from the same patient 5 years after the duplication of the foregut in Fig. 6A was removed from the chest.

saw the spinal deformity together with the suggestive history, a confident diagnosis of an intestinal duplication was made and operation advised. A 10-inch duplication of the upper jejunum (Fig. 6C) was subsequently successfully removed and the child has been perfectly well since.

Case 4

R.M., born normally as a full-term infant of 8 lb., and breast fed, was admitted to hospital at the age of 3½ months. According to the mother he had stridor since birth, associated with a wheeze which she thought was due to bronchitis. Three days before admission the infant suddenly became dyspnoeic, gasped for breath and became blue. On examination he was seen to be well nourished but cyanotic; lower intercostal recession was seen on inspiration, and rhonchi were heard over both bases. A chest radiograph showed a large right mediastinal mass and immediate thoracotomy was considered advisable as an emergency measure. (A review of the films shows anterior spina bifida from C2 to T1.)

At operation the lung was free, and a large loculated cyst, not adherent to the lung or surrounding structures, was present posteriorly in the superior mediastinum. The contents from different loculi varied 'from condensed milk to brown water'. The cyst was opened in order to facilitate dissection. The mucosal lining was red and congested but no ulceration was seen. A thin fibrous pedicle extended upwards through the thoracic inlet.

Pathology. The specimen consisted of a thick-walled cyst, 10 cm. in diameter, which contained thin, brownish fluid with pH 3·9. The cyst wall, 4 mm. thick, showed thick muscular coats lined by apparently normal secretory epithelium. Histologically the epithelium was poor in oxyntic cells.

Case 5

P.L., a full-term normally-born baby girl, was first seen in hospital at the age of 3 months, with feeding trouble. Up to the age of 3 years she was never well. She then developed a left subcostal swelling, which showed up as a large, smooth filling defect of the stomach (Fig. 7A), and was admitted shortly afterwards to another hospital, where a left subphrenic abscess which had ruptured into the left pleural cavity was diagnosed and drained through the chest and abdomen. The post-operative recovery was stormy, but she was much improved for the next 3 years except for periodic vomiting and pain in the left subcostal area. At the age of 6 she again became ill, developing a high fever with signs of left basal pleurisy and pericarditis. The rapidly increasing pericardial effusion was aspirated and brownish offensive fluid obtained, which cultured *B. coli* organisms. Despite antibiotics and aspirations the child died shortly after transfer to the thoracic surgical unit.

Post mortem examination revealed an oval duplication (6 × 4 cm.) on the greater curve of the stomach fundus. A 2-cm.-wide chronic ulcer had perforated into the left posterior subphrenic space, through the diaphragm into the pericardium, resulting in a diffuse

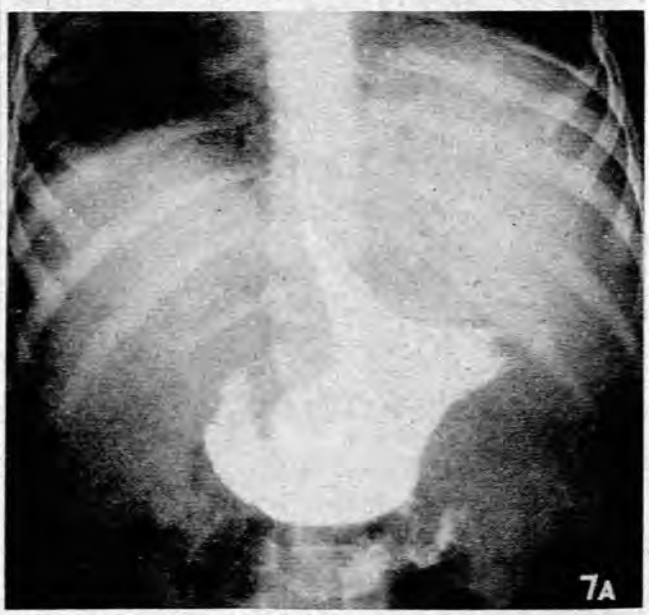

Fig. 7(A). On barium-meal examination a large, smooth, round filling defect of the upper stomach was seen in this 6-year-old girl.

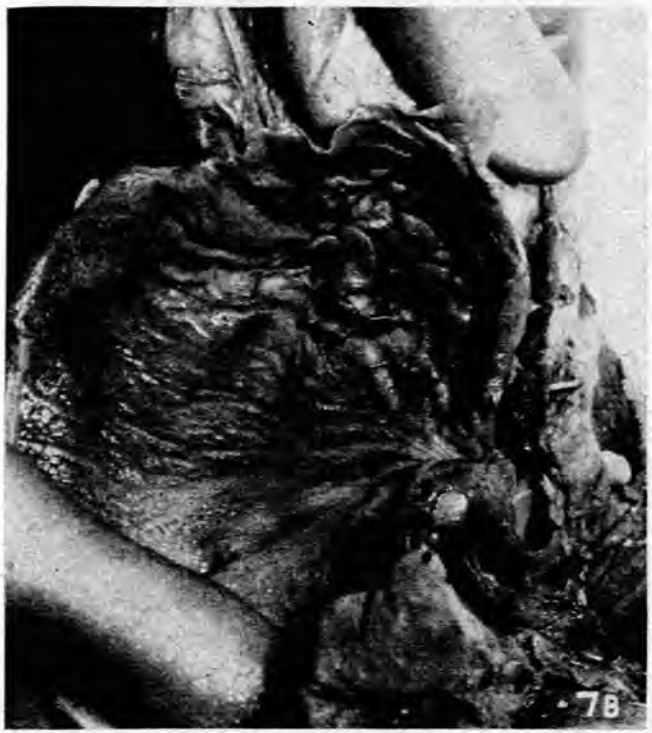

Fig. 7(B). Voluminous mucosal folds are present on the inside of the stomach, where the duplication was attached to the greater curvature.

fibrinous pericarditis. The cyst was covered by peritoneum on the outside and was entirely free from the stomach except at one point, where it was connected by a flimsy fibrous pedicle. On the inside of the stomach at this point voluminous irregular redundant mucosal folds were conspicuous (Fig. 7B). The muscular wall of the cyst was much thicker than that of the stomach. Histologically the mucous membrane was typically of fundic type. The ulcer had all the characteristics of a chronic peptic ulcer. There was no other abnormality and no evidence of a spinal lesion.

Case 6

D.C., a normal healthy boy of 6 years, was X-rayed for tuberculosis as a contact and a mass was found in the left upper chest. Routine investigations threw no light on its nature and a preoperative diagnosis of neurofibroma or mediastinal cyst was made. (Review of the X-ray showed anterior spina bifida in the 4th and 5th cervical vertebra.)

At operation a large spherical cyst was found in the superior mediastinum, close to the oesophagus, but completely unattached to it, the trachea or the spinal column. It enucleated easily, leaving a clean surface.

Pathology. The cyst was thin-walled (about 1 mm. thick in most places) and measured 8 × 5 cm. It contained a brownish fluid, which proved to be partly altered blood. At one point there was a slight thickening in the wall, which presented externally as a boss measuring 1·5 × 1·2 cm. and about 2 mm. above the general surface. Internally it was pinkish, and centrally umbilicated, with the central fossa about 6 mm. in diameter and 2 mm. deep and the plaque raised about 1 cm. above the general internal surface. This island contained velvety mucosa similar to that of the body of the stomach, and rich in oxyntic cells. At the edge of this island were the appearances of a chronic gastric ulcer, with well-marked fusion of the muscularis mucosae and the outer muscle layers. It was this surrounding fibrosis which buckled the patch of the stomach to give the internal concave surface. Auerbach's plexus was well developed. A few centimetres from the boss there was a large nerve, containing ganglion cells; also a small artery, a few ganglia and a lymph node. The epithelial lining of the rest of the cyst was mostly shed; a few islands of stratified squamous epithelium remained. The muscle coat was well developed in 3 layers at the island of gastric mucosa, but the rest of the cyst was rather fibrous, the muscle layers being irregular, thin and undeveloped.

Case 7

E.M., a man of 51, was admitted to hospital because of recent haemoptysis and pain in the left lower chest. Chest radiographs showed a rounded mass posteriorly in the left lower chest.

At operation, a thick-walled cyst was present in the left paravertebral gutter, firmly adherent to the vertebral column, but only loosely attached to the lung. A prolongation extended downwards through the diaphragm, which was incised and found to be attached to the fundus of the stomach. Although the muscular coats fused, the mucosal linings were separate and no communication was demonstrable between the cyst and the stomach. The prolongation was excised from the stomach and the entire cyst removed.

Pathology. The specimen showed a spherical cyst 10 cm. in diameter with a small prolongation on the one side. It was partly covered by pleura. On the inner and posterior aspect there were 2 bosses, 4·5 × 0·4 cm. and 1·4 × 1·5 cm. respectively, separated by a gap of 0·7 cm., and composed of mucus-containing ectopic lung tissue. These 'ectopic lungs' were supplied by a large, thick-walled systemic artery of 2 mm. diameter and two smaller vessels of 1 mm. each. Non-pigmented 'hilar' glands were present where the vessels entered the bosses. The bronchus contained cartilage and racemose mucous glands of bronchial type. There was no actual communication between the cyst and the ectopic masses of lung.

The wall of the cyst was very thick (4 - 5 mm.). The mucosa was smooth but changed in appearance at about the level of the diaphragm. There was no evidence of ulceration. Histologically the cyst was lined by ciliated columnar epithelium. At the diaphragmatic level the cilia were lost, and the mucosa changed to a single layer of columnar cells, which then changed to a secretory epithelium typical of the zone of cardiac glands, and finally to typical fundic mucosa, rich in oxyntic cells. The muscularis mucosae consisted of 3 - 4 layers, while the outer muscle coat contained up to 5 separate layers.

Case 8

S.W., a normally-born full-term baby girl was admitted at the age of 14 months with a history of difficulty in breathing since the age of 4 months. A chest radiograph showed a mass in the right upper mediastinum and barium swallow indicated a smooth uniform filling defect on the right side immediately above the tracheal bifurcation. Below this level opposite the mass the oesophagus was kinked and pulled sharply to the right. A diagnosis of oesophageal duplication was made.

At operation a small cyst was found within the muscular wall of the oesophagus immediately above the azygos vein. It had no connection with the trachea or spinal column, but the muscle layers of the oesophagus had to be incised in order to enucleate the cyst.

Pathology. A spherical, smooth, thin-walled cyst of 3·5 cm. diameter was found containing an opalescent thin mucinous fluid. The mucosa was pale and uniform. Histologically the wall consisted of a small muscle layer of striated fibres and a thin muscularis mucosae. The epithelium was ciliated stratified columnar. Cartilage and bronchial glands were absent.

(To be continued)