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ACQUIRED AND CONGENITAL OESOPHAGO-BRONCHIAL FISTULAS

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Though abnormal communications between the oesophagus and the respiratory tree seem to have been described as early as the seventeenth century (Ashley, 1941), study of the problem and attempts at correct diagnosis have become of practical importance only with the development of thoracic surgery. The relative rarity of the anomaly evidenced in our first case has prompted us to a summary review of oesophago-respiratory fistula and a brief discussion of the pathological mechanisms involved in this condition, and particularly to oesophago-bronchial fistula. Three patients have been operated upon in the department under the direction of Dr. Dumont, and a fourth one has not yet undergone any surgical treatment.

Oesophago-bronchial fistulas are by far the least frequent of all abnormal communications between the respiratory and digestive tracts, of which they represent approximately one-third (Crafoord and Sournia, 1951), the remaining two-thirds being represented by the tracheo-oesophageal fistulas. The latter are in the majority of cases associated with atresia of the oesophagus; they are unquestionably congenital, and are recognized and treated within the first 48 hours after birth, whereas it is generally accepted that oesophago-bronchial fistulas appear in later life.

From the clinical point of view respiratory-oesophageal fistulas may be classified as those associated with atresia of the oesophagus and those without atresia, distinguishing also between cases requiring urgent treatment in the first hours of life and those recognized in subsequent years and treated mainly for their complications, as well as between the cases of undoubted congenital origin and those of less obvious aetiology.

From the anatomo-pathological point of view, respiratory-oesophageal fistulas can be further classified as follows: (*a*) purely congenital, (*b*) communications through fistulous opening of a congenital diverticulum into the respiratory tree or oesophagus, and (*c*) purely acquired fistulas.

Vogt's (1929) classification of congenital fistulas is certainly the clearest and is briefly recalled here:

I. Atresia of the oesophagus without fistula.

II. Fistula without atresia of the oesophagus.

III. (*a*) Fistula with atresia, the fistula between the upper segment and respiratory tree; (*b*) fistula with atresia, the fistula between the lower segment and respiratory tree; (*c*) fistula with atresia, the fistula between both segments and respiratory tree.

Congenital diverticula of either the trachea or the oesophagus can theoretically form fistulous openings, e.g., through inflammation, into the oesophagus or into the respiratory tree.

Finally, oesophago-respiratory fistulas may be of a purely acquired type, such as traumatic (foreign bodies, chemical burns), cancerous, infectious (specific or non-specific), or arising through acquired diverticula forming fistulous openings.

Obviously, the exact aetiology of any particular case of respiratory-oesophageal fistula will not always be easy to ascertain.

Of all congenital fistulas, type III (*b*) is by far the most frequent. Mullard (1954) gives an excellent summary of Gruenwald's (1940) theory of the genesis of this variant, as well as of the other types of congenital fistulas. Briefly, the oesophagus, trachea, and bronchi are derived from the same embryonal anlage. The tracheo-bronchial tree appears in the 3 mm. embryo as a small bud on the ventral aspect of the foregut. A septum growing into the primitive entodermal sac divides it into a posterior alimentary and an anterior respiratory tube. The splitting of the trachea from the oesophagus takes place when the embryo reaches 5 cm. in length. The next phase of development is a rapid elongation of the trachea. Fistula with atresia would be the result of a failure of co-ordination of the two phases of development, the trachea beginning its elongation before splitting from the oesophagus has been completed.

The oesophagus, its lumen blocked by cells, is reduced to a thin cord or even fragmented.

Since the tracheo-bronchial tree is derived directly from the oesophagus, all types of congenital communications are conceivable without atresia of the oesophagus.

Plass in 1919 and Gruenwald in 1940 described cases of fistula with atresia in human embryos, 18, 8, and 9 mm. long respectively.

As Mullard (*loc. cit.*) very rightly points out, it is surprising to note that the records of major variants of the anomaly, such as atresia alone or atresia with fistula, are far more numerous than those of the minor variants, such as fistula without atresia. In a very thorough review of the literature, he quotes only about 30 cases of fistula without atresia, to which he adds two of his own.

In 1916 Heiderich demonstrated on an anatomical specimen a cord joining the oesophagus to the right stem bronchus; this fistula had a broad base in the oesophagus, of which it contained a few longitudinal fibres. No neighbouring diseased lymph nodes could be found. The fistula was slanting downward from bronchus to oesophagus, and easily allowed a small probe to pass. The congenital origin of the anomaly appears to have been recognized at that time.

In 1933 Clerf reported the case of a woman, 23 years of age, with a history of seven years of cough on swallowing: a fistula between the oesophagus and bronchus could be demonstrated by oesophagoscopy, barium swallow, and radio-opaque bougies. It was treated apparently with success by cauterization with silver nitrate.

In 1937 Moersch and Schmidt reported two further cases. The first was of a woman aged 31, with a history of coughing up particles of food for the previous eight years. A fistula was found between the oesophagus and right lower lobe bronchus. The patient died after operation. Necropsy showed no inflammatory changes which could explain the fistula. The second was of a man aged 52 with a history of strangulation and dyspnoea only on drinking since childhood. A fistula could be demonstrated connecting the oesophagus with the left lower lobe bronchus; the anomaly was obviously congenital.

In 1954 Mullard reported the case of a 54-year-old man with a history of cough and purulent sputum for 11 years, as well as coughing up of food particles in the previous few months. There was a cavity in the left lower lobe, but no tuberculous process could ever be proved. A barium swallow showed the fistula to pass from the oesophagus to the left main bronchus. Pneumonectomy was performed in 1948, the fistula being displayed with very little dissection; it was about 1 cm. long and 2.3 mm. outside diameter at the entry into the bronchus, widening rapidly to 7.5 mm. on entering the oesophagus. The oesophageal defect was closed with linen thread sutures. No post-operative fistula occurred.

In 1950 Morton, Osborne, and Klassen recorded a case of fistula between the oesophagus and the right lower lobe bronchus in a 26-year-old man. The patient did not cough up food particles until adult life. Right lower lobectomy was performed as well as closure at the fistula, which appeared to be congenital.

In 1952 Berman, Test, and McArt reported the case of a 40-year-old woman, who had had symptoms since birth, and in whom pneumonectomy with closure of a fistula passing from the oesophagus to the left lower lobe bronchus was performed. There was a vascular abnormality in this case, viz., two ducti arteriosi.

In 1953 a very large fistula was recorded by Krausey in an African woman aged 38. Death followed impaction of an alimentary bolus in the fistula.

Finally in 1954 Mullard (*loc. cit.*) described his second case of congenital oesophago-bronchial fistula in a woman aged 54, who could give no clear history of coughing or swallowing; she complained of a bad taste in the mouth and had suffered from regurgitation of fluids from the oesophagus, which was attributed to an enlarged thyroid. Thyroidectomy was carried out uneventfully, but without relief of symptoms. Further investigation revealed a fistula from the oesophagus to the right upper lobe bronchus. At operation the fistula was dissected easily, as there was no inflammatory reaction around it; it was funnel shaped, 2 cm. long, 1 cm. broad at the oesophageal attachment and 0.5 cm. broad at its bronchial attachment. Recovery was uneventful. Microscopic section showed the fistula to be lined with squamous epithelium.

Congenital diverticula of either trachea or oesophagus undoubtedly exist; however, proved cases of such diverticula forming fistulous openings seem to be exceedingly rare. Though not absolutely certain, Case 2 of a series of seven oesophago-bronchial fistulas reported by Crafoord and Sournia in 1951, and described by these authors as an oesophageal diverticulum forming a fistulous opening in the right lower lobe bronchus, can reasonably be assumed to correspond to a congenital diverticulum; symptoms appeared at 7

months of age, shortly after weaning, and the anatomical description of the operative specimen tends to demonstrate its oesophageal origin. Jaubert de Beaujeu (1952) considers the congenital origin of oesophageal diverticula very doubtful.

Of purely acquired fistulas, those of traumatic or cancerous origin need no particular comment; the importance of tuberculous adenitis seems to be established in the formation of tracheal diverticula as well as in the subsequent fistulous opening of the latter into the respiratory tree through caseation of the diseased gland. Non-specific inflammations may also lead to peri-diverticulitis, mediastinitis, pleural adhesions, peri-diverticular pneumonitis, abscess, and fistula formation.

As for acquired pulsion diverticula, usually situated near the diaphragm, infections of the diverticulum itself must be considered the main factor in causing the opening, there being no particular reason why external infection should be located at the precise level of an oesophageal pouch of purely mechanical origin.

Finally, the possibility of cystic enlargement of the mucous glands of the respiratory tree must be borne in mind. The observations of Rokitansky (1838) and Gruber (1869 and 1875) are well known. The latter describes a tracheal cyst 4.3 cm. by 2.7 cm. on the posterior aspect of the trachea opening into the latter through three very minute apertures. Such cysts, though smaller in size, may also appear on the bronchi, sometimes reaching the stage of "diverticulosis" of the bronchial wall as described by Duprez and Mampuys (1953). A larger cyst making a fistulous opening into the oesophagus seems possible, though we could find no such case in the literature.

Case Reports

Case 1.—Dev . . . I., an 18-year-old college girl, was admitted to the department of Chest Surgery of the Saint-Pierre Hospital, referred by Dr. Noeninckx from Antwerp.

On admission she gave a history of chronic productive cough, raising a considerable quantity of mucopurulent material. She could not recall any haemoptyses, nor did she ever complain of dysphagia. After the operation, when questioned more accurately, she said that sometimes she had coughed immediately after drinking. Her general condition appeared good. Physical examination only showed ronchi and moist rales in the right lung.

The postero-anterior radiograph revealed a diffuse fibrosis extending from the right hilar region to the base of the right lung with a small annular rarefaction. In the right lateral projection there was an atelectasis of the middle lobe. Bronchography showed extensive bronchiectasis in the right lower lobe and in the medial segment of the middle lobe. The other segment of the middle lobe was not visualized (Fig. 1).

On account of the eight years' duration of the disease, the productive cough, and the atelectasis of the middle lobe with extensive bronchiectasis, a right bilobectomy was decided upon.

On November 1, 1952, the patient was operated upon under general anaesthesia and tracheal intubation in the prone position. The chest was entered

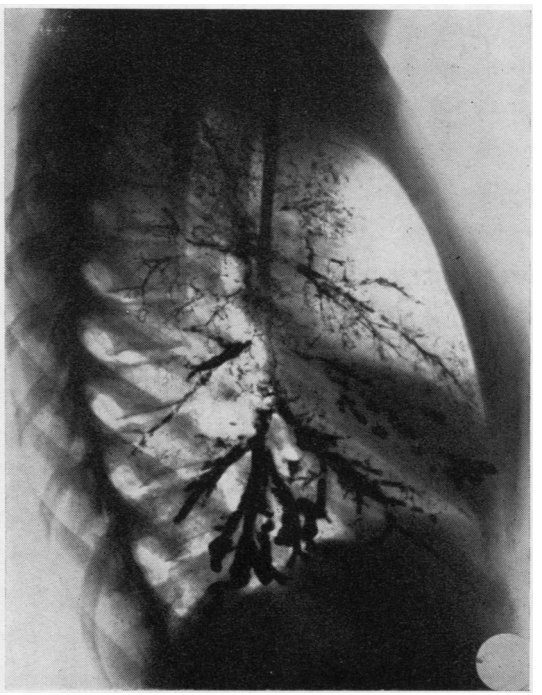

Fig. 1.—Case 1: Bronchography shows bronchiectasis in the basal segments of the right lower lobe and in one segment of the middle lobe. The other segment of the middle lobe is not visualized.

after resection of the sixth rib. The lung was found free from adhesions except in the two fissures. The middle lobe was completely atelectatic, but the lower lobe was normally ventilated. The mediastinal pleura was opened and section of the inferior pulmonary ligament was started. At that point of the operation a channel, 5 mm. in diameter and 10 mm. long, was found extending from the oesophagus to the middle lobe bronchus. This formation was completely free of adhesions, its walls were perfectly supple, and there was no lymph node in the neighbourhood. When it was divided it was possible to explore the oesophagus on one side with a catheter. On the other side no air nor any purulent material was encountered. The oesophageal pouch was closed by two rows of interrupted linen sutures. Bilobectomy was then per-

formed. The bronchus was closed by seven steel wire sutures and covered with a pleural flap. The pleural cavity was drained by means of two catheters, one for fluids, the other for air.

Three cannulas were introduced into the oesophageal communication, the middle lobe bronchus, and the lower lobe bronchus of the specimen. The lower lobe was re-expanded with air until it reached its primitive volume. It was not possible to re-expand the middle lobe which remained atelectatic. "Lipiodol" was then injected through the first cannula into the oesophageal communication. The first radiograph showed a normal segmental bronchus, normal small bronchi, but no bronchioles and no alveoli (Fig. 2). When "lipiodol" was injected in the second cannula, in the middle lobe bronchus, extensive bronchiectasis could be visualized in the other segment (Fig. 3). Through the third cannula lower lobe bronchiectasis was outlined by the "lipiodol." No communication could be demonstrated between the two segmental bronchi of the middle lobe.

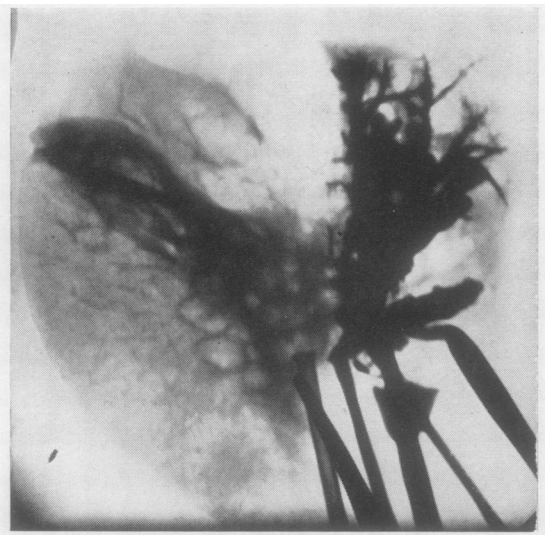

FIG. 2.—Case 1: A radiograph of the resected specimen. Iodized oil has been injected in the oesophago-bronchial fistula and demonstrates the picture of a normal segmental tree.

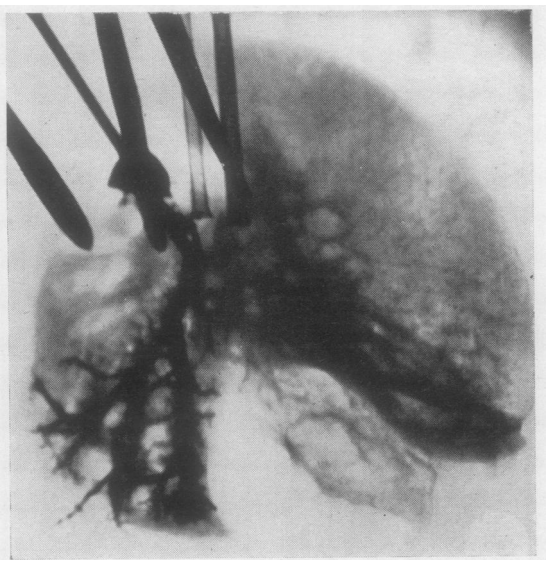

FIG. 3.—Case 1: A radiograph of the resected specimen. A second injection of iodized oil in the middle lobe bronchus demonstrates bronchiectasis in the medial segment in the middle lobe.

The lower lobe was then separated from the middle lobe. It was preserved for large thin sections according to the technique of Gough and Wentworth (1949). The middle lobe was prepared as a bronchial cast (Duprez, 1949).

The thin sections of the entire lower lobe showed typical acquired bronchial dilatations in the basal segment. The middle lobe cast confirmed the information obtained after injections of "lipiodol." Red material was pushed into the oesophageal communication and demonstrated a normal bronchial tree in the lateral segment but no alveoli. Blue material showed large ampullar bronchiectasis in the medial segment. No bronchioles were injected. There was no contact between the blue and red substances at the level of segmental bronchi, but some mixing at the level of the bronchioles, which might have been due to the high pressure of injection.

CASE 2.—A 41-year-old man had had pneumonia in 1925, and a left lung abscess necessitating two months' drainage in 1936. Till 1950 he went on coughing and expectorating moderately; the sputum then became purulent, fetid, and was streaked with blood. In 1952 he was admitted to the medical department (Hôpital Saint-Pierre), where a diagnosis of bronchiectasis was made, and treated with penicillin, streptomycin, and terramycin.

He was admitted to the Thoracic Surgery Department in August, 1953. His general condition appeared to be fair, but he still expectorated from 50 to 100 ml. of mucopus a day, and his breath was offensive. The fingers were slightly clubbed.

Laboratory investigations on admission were practically normal, in particular the B.S.R. (2 mm. in one hour, Westergren) and the total plasma proteins (80.8 g. %). Repeated examinations of sputum for Koch bacilli remained negative.

At operation on August 15 dense adhesions between the left lower lobe and the parietes were encountered and dissected up to the pulmonary ligament. At that point the oesophagus was noted to be strongly attracted towards the left and to present a diverticulum adherent to the neighbouring lung tissue. The diverticulum was entered and showed a communication with the lumen of the oesophagus admitting a No. 14 urethral catheter. The neck of the diverticulum was

tied with two sutures and buried in the wall of the oesophagus. A left lower lobectomy was then performed during which severe haemorrhage from a bronchial artery occurred. Four days after the operation the patient was remarkably well. On September 7 he coughed up blood. However, no other sign of fistula was noted after that incident, and the patient left hospital on September 18.

Injection of iodized oil through the neck of the diverticulum on the operative specimen demonstrated the communication between the oesophagus and bronchiectasis on the posterior basal bronchi. Dissection showed large bronchial dilatations lined with a proliferating mucosa.

CASE 3.—Duymel . . . Magdelena was a young woman aged 33, whose main complaint was persistent cough since the age of 8. She had pleurisy at 11 and 17 years. In March, 1952, she suffered from an affection labelled "grippe," which eventually became a pneumonia with a right lung abscess. From that time on she had a persistent, yellowish, fetid sputum. In July, 1952, a purulent pleurisy was noted on the right side and pus evacuated. The temperature was irregular, reaching from normal to 39° C. (102° F.). In October, 1952, a right lung tomography showed an irregular cavity, probably a sequel of the lung abscess, as well as bronchiectasis, and atelectasis of the right lower lobe. No Koch bacilli could be found in the sputum. Bronchography confirmed the atelectasis of the right lower lobe.

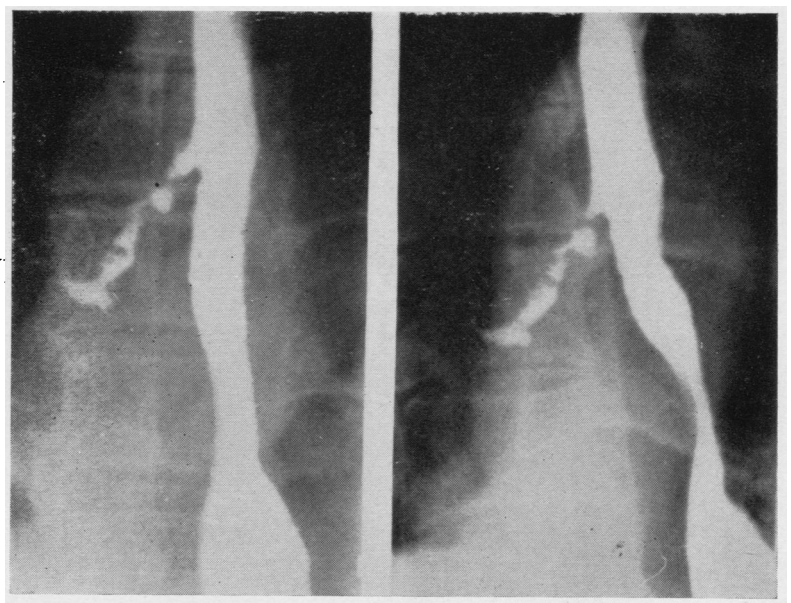

FIG. 4.—Case 3: Iodized oil has been swallowed by the patient and is passing from the oesophagus downwards to the paracardiac segment of the right lower lobe.

The patient was referred to the Hôpital Saint-Pierre for lobectomy in April, 1953, and operated on on May 7. Dissection of the hilus showed a broncho-oesophageal fistula, approximately 5 mm. long and 2 to 3 mm. in diameter, running between dense adhesions above the right inferior pulmonary vein and parallel to it. After section of the fistula the oesophagus was closed by two rows of interrupted sutures, and a right lower lobe lobectomy performed. As the skin was being closed the patient suddenly died from an air embolism.

A bronchial cast of the specimen demonstrated extensive bronchiectasis and a direct communication of the abnormal channel with an enlarged bronchus.

CASE 4.—Ro . . . G., a 57-year-old woman, a housekeeper, had a long history of digestive troubles with a cholecystectomy in 1949.

On admission to the department, in July, 1954, she complained of slow digestion and aerophagia, but without vomiting. Recently she had lost a few pounds in weight.

Bronchopulmonary signs were not very impressive, only some coughing with mucopurulent green sputum, and no dypsnoea or haemoptysis. Sometimes she felt a slight pain in the left part of the chest and in the left arm, but did not observe any exacerbation on effort.

Physical examination was not contributory.

Radiographs of the biliary ducts, after intravenous biligrafin injection, did not demonstrate any abnormality except the absence of the gall-bladder previously resected.

Radiological examination of the digestive tract did not reveal any lesion of the stomach or of the duodenum, but it demonstrated a narrow and tortuous fistula between the lower third of the oesophagus and a bronchus of the right lower lobe, probably the paracardiac. The oesophageal origin of the fistula could have been a diverticulum (Fig. 5).

A radiograph of the thorax was normal.

A bronchogram showed bronchiectasis limited to the paracardiac segment of the right lower lobe, but did not demonstrate the fistula.

The case was diagnosed as an acquired oesophagobronchial fistula after bronchiectasis.

No surgical treatment has been undertaken to date.

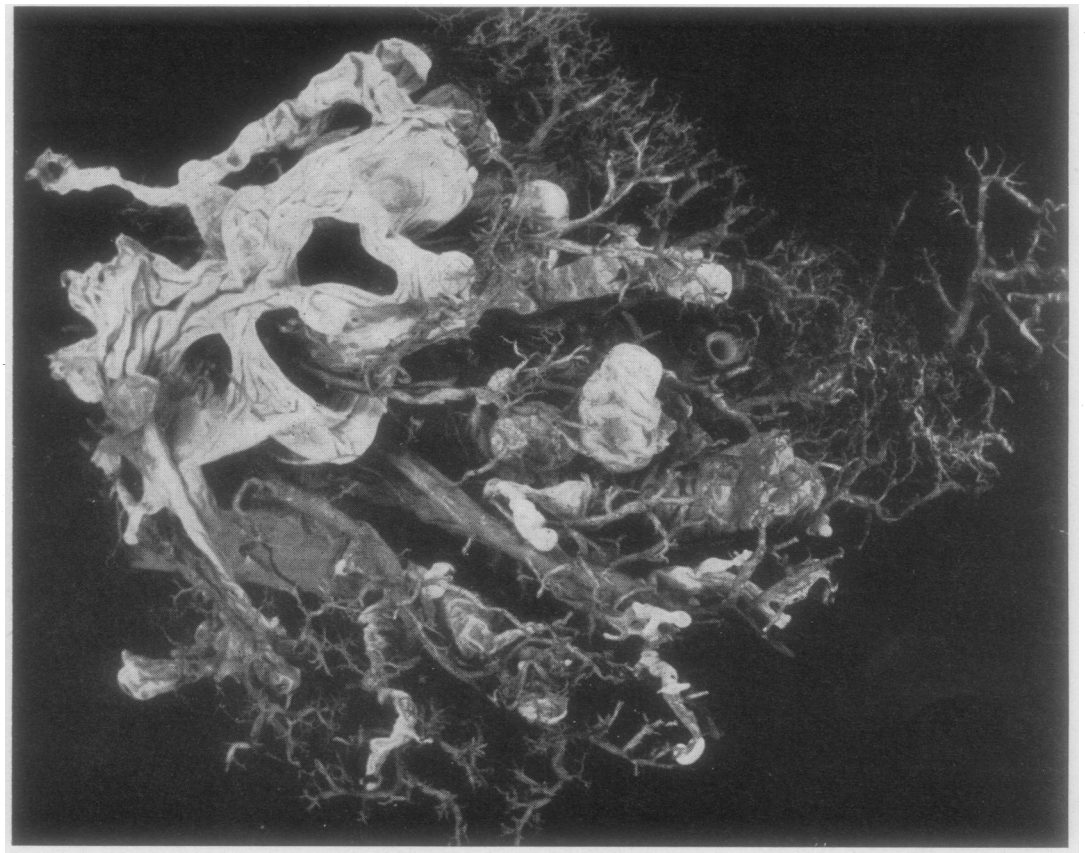

FIG. 5.—A bronchial cast of a resected lower lobe showing extensive bronchiectasis with oesophago-bronchial fistula.

DISCUSSION

Congenital atresia of the oesophagus associated with tracheo-oesophageal fistula is a well-known and relatively common abnormality. More and more cases of fistula between the oesophagus and the respiratory tract without atresia are now recorded. They are diagnosed in adults, but it is often difficult to find out the exact time of onset of symptoms. Most of them seem to appear after an infectious process has developed between the oesophagus and a segment of the bronchial tree. Sometimes it is the result of the growth of malignant tumour, or the destruction caused by a caustic or a foreign body in the oesophagus.

However, the peculiar features evidenced on operation and pathological investigation suggest the congenital nature of the communication between the oesophagus and the trachea or bronchi. Surprisingly enough, it may remain unnoticed for 20 or 30 years, without any dysphagia or pulmonary infection caused by the passage of particles of food. Jackson and Coats gave a simple explanation for the late onset of symptoms, describing a fold or membrane occluding the fistula in the newborn and breaking down in later life (Mullard, 1954). Mullard never observed that membrane. He suggests that the oesophageal orifice could be overridden by a fold of the oesophageal mucosa when the bolus passes in front of it, giving some protection against entry of food particles into the fistula. Later on, the mobility of the oesophagus is interfered with by the adhesions between the affected lung and the mediastinal organs after recurrent attacks of pneumonitis, and the mouth of the fistula opens. The direction of the fistula may be responsible for its patency. However, in our fourth case the fistula was running downwards from the oesophagus to the bronchus, and the patient never complained of dysphagia.

Our first patient has to be regarded as a new case of congenital oesophago-bronchial communication. First, at the time of operation no signs of local inflammation between the oesophagus and middle lobe, such as adhesions, fibrosis or enlarged lymph nodes, were to be seen. The abnormal channel was perfectly isolated and supple. There was no evidence of any oesophageal "traction" diverticulum; secondly, a radiograph taken after injection of iodized oil into the specimen and a bronchial cast prepared after injection of plastic material demonstrated two definite territories in the middle lobe. The first one was the medial segment of the lobe, whose segmental bronchus was branched to the middle lobe bronchus and whose small bronchi were dilated. The second one was the lateral segment, whose bronchi were not connected with the middle lobe bronchus but with the abnormal channel leading into the oesophagus. There was no bronchiectasis to be found in that segment.

The type of pathological examination we chose excluded any histological sections.

Association of middle and lower lobe bronchiectasis is very frequent, but then both segments of the middle lobe are similarly involved. In that case, bronchiectasis, in fact, is a secondary and different disease. It has to be explained on the basis of the general pathology of bronchiectasis. One of us (Duprez, 1954) has put forward elsewhere our hypothesis. Duprez has demonstrated two phases in the evolution of bronchiectasis. The first one is a peculiar type of bronchitis, the "dilating bronchitis," often diffuse in its onset, but leaving deep and permanent pathological changes in localized segments of the bronchial tree. In the segments where adequate drainage of the purulent material is possible, bronchitis resolves without any permanent lesions. In the other ones, the second phase of the disease appears, with its typical picture of narrowing of the segmental orifice, obliteration of the bronchioles, dilatation of the small bronchi between the orificial stenosis, and the obliterated bronchioles. Histological sections show degeneration of the muscles, elastic tissue, and cartilage. Bronchial casts and dehydration of segments of the bronchial wall show diverticulosis of the mucous glands.

The lateral segment of the middle lobe of our case was isolated from the bronchial tree. It represented a sort of sequestrated segment, untouched by the bronchial infection, and so was preserved from bronchiectatic changes.

Our second and third cases were accepted as acquired oesophago-bronchial fistula. Both of them were characterized by the presence of a short channel surrounded by dense inflammatory adhesions in close contact with a bronchiectatic lobe. In Case 2 we observed a typical "traction" diverticulum of the oesophagus.

Inflammatory fistulous openings of the oesophagus are a well-known condition. Abbott (1945) portrayed all its types—oesophago-cutaneous, oesophago-pleural, oesophago-pericardial, oesophago-mediastinal, and, more frequently, oesophago-bronchial.

Santy and his colleagues, and later Jaubert de Beaujeu (1952), reporting eight cases of oesophago-bronchial fistula associated with a "traction" oesophageal diverticulum, incriminated inflammatory lymph nodes as "traction" agents. They even described the passage of the communicating channel through a lymph-node cavity. We have never noticed such an anatomical arrangement, but the inflammatory changes at the level of the fistulous opening were sufficient to explain its development.

Previously, when studying bronchial casts of bronchiectasis we noticed abnormal communications between enlarged bronchi in the middle of severely damaged lobes, in which we also incriminated the extreme degree of the infectious process (Duprez, 1954). The mechanism involved must be the same, either inside an inflammatory lung tissue or in the contiguous mediastinal space.

Our fourth case is only a clinical report, since the patient has not been, so far, operated upon. However, it has many similarities with our two cases of acquired fistula, with a narrow and tortuous communicating channel opening into a bronchiectatic segment of the lower lobe.

Conclusion

Most oesophago-bronchial communications are the result of the extension of an infectious process from the depth of a pulmonary lobe through the contiguous mediastinal space into the wall of the oesophagus. Adhesions, minute abscesses, enlarged lymph nodes, are so many aspects of that progressive inflammation whose cicatricial retraction draws out a "traction" diverticulum of the oesophagus.

Congenital communications are certainly less frequent. We have presented the arguments favouring a congenital origin for the peculiar type of communicating channel, associated with bronchiectasis in the adjacent pulmonary segments, which we found in one case. We have not dis-

cussed the treatment of that condition. Whatever its origin, it can only be cured by surgical excision and suture. This we did in three instances, with one unhappy result.

SUMMARY

Four cases of oesophago-bronchial fistula discovered in adults are presented. Three of them have been operated upon so far.

Operative findings and pathological examination of the three resected specimens lead to the conclusion that one of them certainly represented a congenital abnormality. The others demonstrated a late complication of a severe infectious process in a bronchiectatic lung.

The different theories of the genesis of oesophago-bronchial communications have been reviewed.

Curative treatment of all types of fistula has been restricted to excisional surgery.

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