South African Medical Journal : Suid-Afrikaanse Tydskrif vir Geneeskunde

Cape Town, 1 July 1961 Volume 35 No. 26 Deel 35 Kaapstad, 1 Julie 1961

NON-MALIGNANT OESOPHAGO-BRONCHIAL FISTULA IN THE ADULT

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A perforation of the oesophagus may track in many directions, eventually communicating with other organs. The track may end blindly in the mediastinum; may discharge on the skin of the neck, chest or even the loin; or may enter the pericardium, pleural cavity or tracheo-bronchial tree.

Of the various causes of oesophageal perforation, malignant disease is certainly the commonest and is usually oesophageal in origin, though malignancies of the thyroid, trachea or bronchi have rarely been known to cause fistulae leading into the oesophagus.

Non-malignant causes are varied. Congenital fistulae are usually associated with some form of oesophageal atresia and present clinically soon after birth; these have been well documented. Congenital fistulae without atresia, however, may only present in adult life. The acquired forms may be either traumatic or inflammatory in origin (Table I).

TABLE I. AETIOLOGY OF OESOPHAGO-BRONCHIAL FISTULAE

1. Malignant disease.
2. Congenital: (*a*) associated with oesophageal atresia; (*b*) without atresia, usually presenting in adults; and (*c*) associated with oesophageal diverticulum and possibly other congenital abnormalities.
3. Acquired non-malignant:
   - (*a*) Inflammatory: tuberculosis; syphilis; oesophagitis, e.g. corrosives; actinomycosis; pyogenic empyema; and traction diverticulum.
   - (*b*) Traumatic: oesophageal foreign body; instrumental dilatation; penetrating wounds; operative; and extrapleural 'lucite' plombage.

Relatively few perforations track further than the mediastinal tissues, and even though fistulae into the tracheobronchial system are probably the commonest of those that do go beyond the mediastinum, only 84 reports of acquired non-malignant fistulae of this sort could be found in the literature from 1916 to 1954.[1,2] In 53 the exact site was known. Of these, 25 communicated with the trachea, and those involving a bronchus were sited thus: 14 in the right main bronchus, 6 in the left main bronchus, 5 in the right lower lobe bronchus, and 3 in the left lower lobe bronchus.

The symptomatology is variable and depends partly on the particular aetiology. Thus a diverticulum may give evidence of its presence by the symptom of dysphagia before the fistula is formed. Similarly, the infective causes and some of the traumatic ones may present initially as mediastinitis. However, once a fistula is established, there is almost invariably some pulmonary symptom to indicate pulmonary infection. It is to be noted that infection in the lungs is not uncommonly associated with oesophageal lesions from spill-over at laryngeal level, so that a fistula is by no means always present when oesophageal and pulmonary disease are combined.

The characteristic symptom of a fistula is a bout of coughing associated with the ingestion of food or fluid; this is not, however, always found, particularly in the rare congenital fistulae in adults. It must depend in part on whether the size and direction of the fistula allows material to pass into the lung.

The following are 4 illustrative case reports:

CASE REPORTS

1. G.G., a Coloured female of 19 years, complained of repeated haemoptyses over the previous 3 years; the bleeding had on occasion lasted as long as a month. She had some cough, but little sputum had been produced during this period. Investigation revealed that she had bronchiectasis of the left lower lobe, and on bronchoscopy the degree of inflammation of the bronchial mucosa was marked (Fig. 1). However, no suspicion of any fistula was raised before operation.

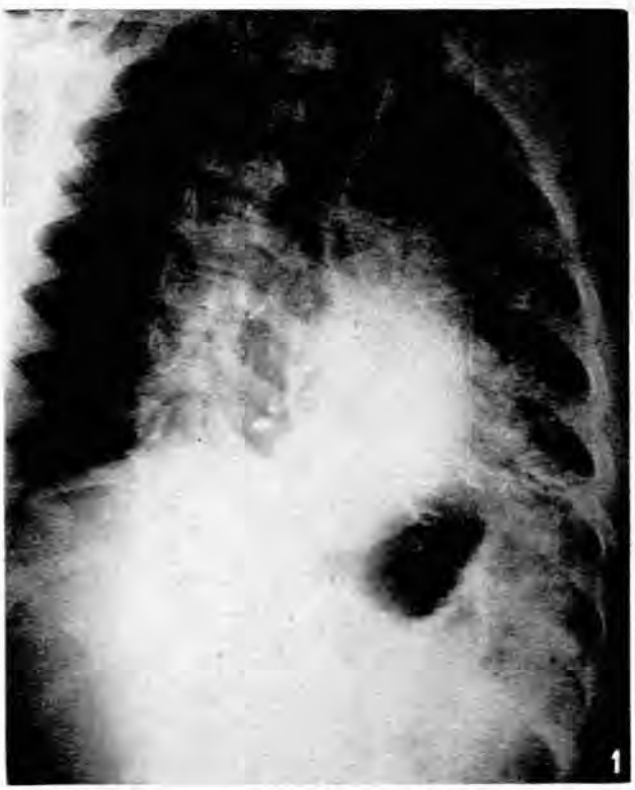

Fig. 1. Case 1. Bronchiectasis of the left lower lobe shown on oblique bronchogram.

At operation the lobe was densely adherent to the chest wall and to the diaphragm. The hilar glands were enlarged and the lobe felt somewhat firm and partly atelectatic. Just above and behind the inferior pulmonary vein a tube-like structure was found, which proved on further dissection to be an oesophago-bronchial fistula, entering the apex of the lower lobe. It was approximately 1 cm. in diameter and was easily displayed, with little surrounding inflammation outside the lobe. In contrast, the lobar hilum was densely matted with inflammatory tissue. A short segment was excised with the lobe and the oesophagus closed with two layers of silk sutures. There was a severe post-operative infective period, almost septicaemic, but recovery was satisfactory.

Histologically, the fistula was lined with squamous epithelium and its wall contained smooth muscle. It can thus be assumed that this was an example of the rare congenital type of fistula without any oesophageal atresia.

2. T.Z., an African female of 26 years, had noticed some dysphagia for about a year with a recent mildly-productive cough. There had been no acute initial episode which could indicate any trauma or infective element, and there was no relationship between the cough and the taking of food. Lipiodol swallow and bronchography showed a small fistula from the middle of the oesophagus entering the right lower lobe. In addition there was an almost complete block of the middle-lobe orifice and the intermediate bronchus was narrowed almost concentrically (Fig. 2).

When endoscopy was performed the fistula could not be visualized, though some bubbling in the oesophagus was noted with respiration at one point. There was no noticeable narrowing of the oesophageal lumen. The bronchographic findings were confirmed, and a biopsy taken from the granular middle-lobe orifice revealed chronic inflammatory tissue.

At thoracotomy the pleural cavity was found to be obliterated by diffuse adhesions and the pulmonary hilum was a solid mass of fibrous tissue, so that the individual structures could not be defined. Eventually, by tracing the oesophagus up from below, it was possible to isolate the fistula. It was less than 1 cm. in length before it disappeared into the fibrous tissue of the hilum, and about ½ cm. in diameter. There was some surrounding inflammatory tissue even at the oesophageal end. The fistula was transected close to the oesophageal wall, carbolized and closed with silk sutures.

The atelectatic middle lobe was removed with some difficulty. Histologically it showed foreign-body giant cells, fat particles, peribronchial inflammatory cuffing and marked fibrosis, consistent with pneumonia on the basis of the inhalation of food particles. Recovery was uneventful.

The exact aetiology in this case is unknown, though the ingestion of some foreign body such as a bone must be quite likely. The hilar fibrosis was presumably due to the chronic non-specific inflammatory process and was not the aetiological factor. There was no evidence of actinomycosis.

3. M.M., an African male of 48 years, had fairly recent dysphagia with cough and sputum. There had been marked loss of weight and his general condition was poor. Barium swallow revealed a large, dependent oesophageal diverticulum in the lower thorax, tracking to the right and eventually communicating with the right lower lobe (Fig. 3). Bronchography showed gross bronchiectasis of the right middle and lower lobes with dye entering the diverticulum; the intermediate bronchus was markedly narrowed (Fig. 4).

When endoscopy was performed, the oesophagoscope passed readily into the mouth of the diverticulum, which was not inflamed; in fact, its appearance was that of a second oesophagus.

Gastrostomy was performed because of the poor general state of the patient and his improvement was slow. It was about 3 months before thoracotomy was deemed advisable. In spite of the delay in operating, there was gross inflammatory change in and around the lung with a completely matted hilum. The fistula was closed distally and the basal segments removed as rapidly as possible, but the patient succumbed soon afterwards. Histology of the removed lung was similar to the previous case.

4. N.B., an African female of 54 years, had had 2 months' dysphagia with loss of weight. On investigation, extremely gross oesophagitis was found with an obstruction at the diaphragmatic

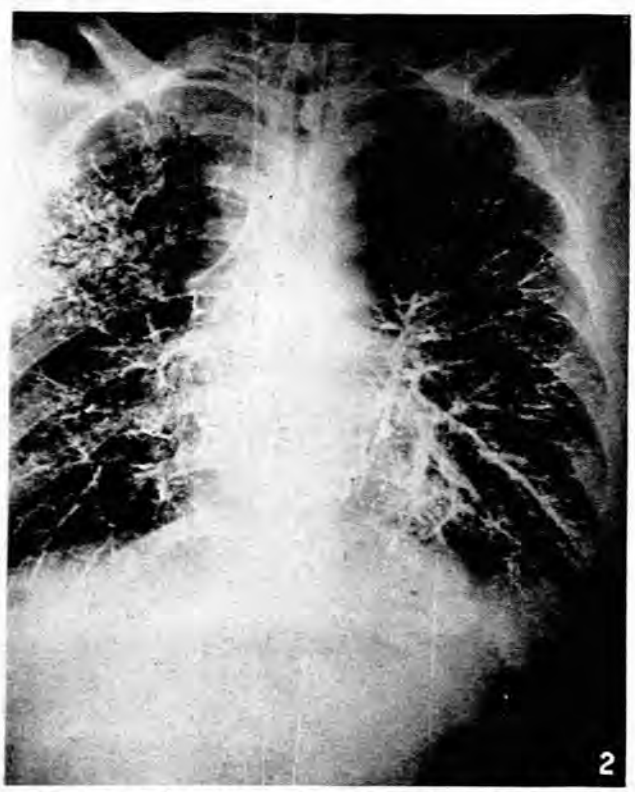

Fig. 2. Case 2. Retouched film of bronchogram showing fistula entering right lower lobe.

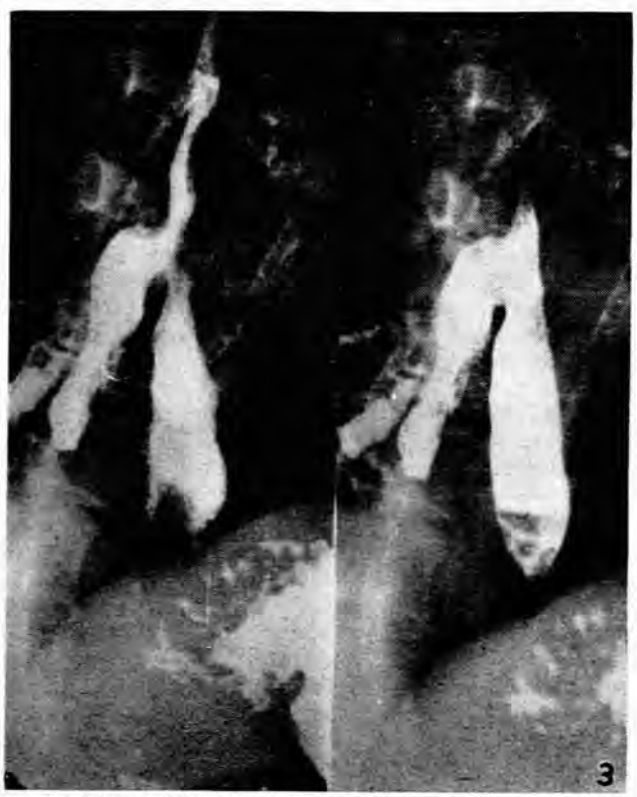

Fig. 3. Case 3. Barium swallow showing large oesophageal pouch with barium in the bronchi.

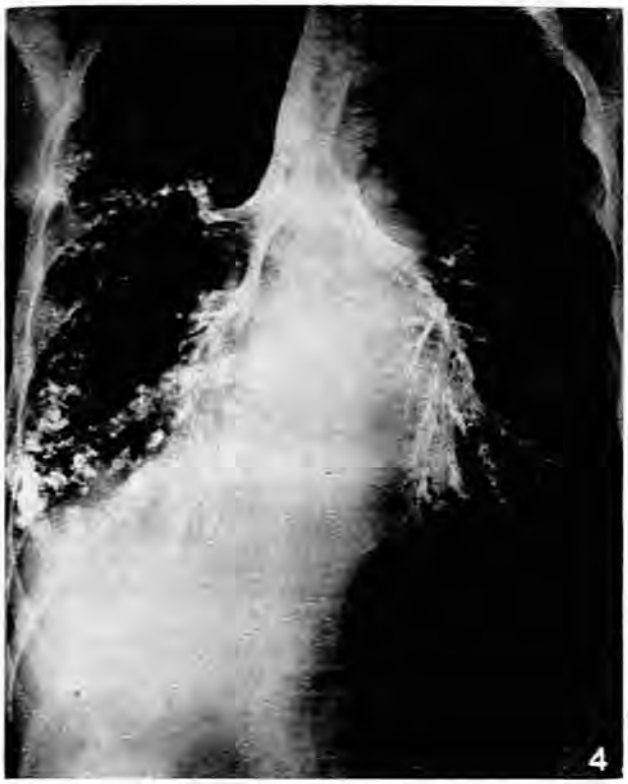

Fig. 4. Case 3. Bronchogram showing right basal bronchiectasis, dye in the oesophageal pouch, and the narrowed intermediate bronchus.

level. About 3 inches above the obstruction a small fistula was demonstrated entering the apex of the left lower lobe. Gastrostomy was performed and the oesophagitis gradually resolved with closure of the fistula. Subsequent operation revealed that the basic obstruction was from a carcinoma of the lower end of the oesophagus. The site of the fistula was not seen; radiologically it was well above the neoplasm and must have been caused by the diffuse inflammatory reaction resulting from stagnation of food.

DISCUSSION

Congenital Causes

Congenital oesophago-bronchial fistula in the adult, though rare, has been well reported by Mullard.[3] Up to 1954, he could find only 11 case reports in the literature which had been adequately documented; there were some 6 others which might have been included, but where the data were incomplete in some way. Of the 11 cases, 2 involved the trachea and the rest one or other bronchus. Only 2 had symptoms dating from infancy, in contrast to the patients who had atresia as well. One patient had no symptoms from the fistula at all, but the majority admitted to cough associated with the ingestion of food at some stage.

Recurrent attacks of pneumonitis usually occur, and perhaps more patients with such symptoms should be investigated with a fistula in mind. Mullard suggested that these fistulae might be found more commonly if such patients were given a lipiodol swallow in the prone position. He also made the point that oesophago-tracheal fistulae could easily be missed at operations for pulmonary resection, and the occasional case of progressive pulmonary suppuration seen after resection could be from such a cause.

Subsequently Lansden and Falor[4] reported 2 cases, both of which had a long history of symptoms, the one dating from birth. One involved the trachea and the other the right main bronchus. This case was found accidentally during a barium-meal investigation on a patient who was found later to have a rectal carcinoma; on further questioning, he admitted to a cough for the previous 40 years.

The majority of the above patients have been treated by operation; this is generally necessary to establish histologically the congenital nature of the fistula. One, however, was treated with apparent success by cauterization. At operation the lung is usually densely adherent and dissection is difficult. This is exemplified by a case reported by Berman *et al.*[5] where a basal fistula was merely ligated as a first-stage procedure and 18 days later a pneumonectomy was performed, since the hilum could not readily be dissected for a lobectomy. However, there have been few fatalities and most patients have been cured.

Various suggestions have been offered why symptoms should so often be apparent only in adult life. The direction of the fistula may be a factor, but the fistulae are usually very short, perhaps ½ - 1 cm. A membranous fold which breaks open later in life is possible, and this could then almost be regarded as a form of congenital oesophageal diverticulum. Movement of the oesophageal mucosa with peristalsis to raise a fold over the fistula would need some explanation of why this mechanism should later fail. It may be that some infective episode sets the stage by producing relative fixation to the chest wall and mediastinum, so that normal descent of the oesophagus and particularly of its mucosa on deglutition is upset.

Some forms of oesophageal diverticula appear to be congenital anomalies of the foregut similar to reduplications. Case 3 in this series presented features which suggested that the pouch was like a second oesophageal tube extending for some inches in a downward direction. These dependent diverticula, seem, not infrequently, to develop infection at their distal tip, presumably from stasis, particularly of solid material, and this may lead to rupture into the lung. Monod[6] reported 3 epiphrenic diverticula associated with lung abscess due to fistula formation. Johansson and Michau[7] found 3 pulmonary fistulae in 7 intrathoracic oesophageal diverticula, all in adult female patients; the fistula in each case was found only at operation. On the other hand, Harrington[8] found no fistula in 8 pulsion diverticula, although one was inflamed at its tip and was adherent to the lung.

A more complicated congenital anomaly with oesophago-bronchial fistula has also been reported. In 2 cases,[9,10] there was so-called sequestration of the left lower lobe associated with an oesophageal diverticulum and the later development of a fistula into the lung. One of these[9] had a track leading up from the oesophagus below the diaphragm for about 10 cm. before entering the left lower lobe; it was first noticed during operation, which was undertaken for bronchiectasis.

Thus there may be varied forms of congenital anomalies associated with an oesophago-bronchial communication, and if it is suspected clinically, it is usually possible to delineate it with opaque media.

Acquired Causes

The acquired varieties of non-malignant fistulae have been reported by Coleman and Bunch for the years 1916 - 1949.[1]

Hughes and Fox[2] extended this to 1954 and, including 2 cases of their own, they found a total of 84 reported cases. In only 53 of these was the exact site known and 28 of these were bronchial.

The aetiology of these 84 cases was as follows: 25 were produced by one or other form of trauma, 14 were tuberculous, 11 syphilitic, 12 associated with a diverticulum, 1 actinomycotic, 2 associated with broncholiths, and 19 were of undetermined origin.

Traumatic causes should be becoming less frequent now owing to the use of antibiotics and to prompt treatment for ingested foreign bodies. Damage due to endoscopy is often anticipated in difficult cases, and radiological assessment afterwards keeps a check on any developments. Infection can usually be quite readily controlled before any abscess or fistula can form.

The various infections are generally on the wane, though mediastinal tuberculous glands are still seen all too commonly. A syphilitic aetiology usually means a mediastinal gumma, though an aneurysm has apparently been known to lead to necrosis of both oesophageal and bronchial walls.

The type of diverticulum in acquired cases is not always clear. The traction type due to infection in a para-oesophageal gland is usually associated with healing of that focus, and is conical with a wide mouth so that stagnation and sepsis are not common. However, Harrington[8] reported a case of a traction diverticulum, due to inflamed mediastinal glands, which later ruptured into the right main bronchus. The tuberculous cases are associated with caseous glands, but need not show the formation of a diverticulum.

Acute oesophagitis may be due to chemical irritation such as the swallowing of dye or the reflux of gastric juice in a hiatus hernia, or to infection from stagnation of food when there is some obstruction, as in case 4 in this series. It is uncommon for such oesophagitis to lead even to mediastinitis nowadays, so that fistula formation into a bronchus from this cause must be very rare.

Case 2 would fall into the undetermined group where one may speculate on the different possibilities. A small foreign body, such as a fish bone, might cause so little initial trauma that the incident is forgotten. Peptic ulceration has been suggested as a possible factor, perhaps caused by ectopic gastric mucosa in the oesophagus.

Rarely, a pyogenic pleural empyema has ruptured into the oesophagus,[11] and passing reference has been made to bronchial fistulae developing in a few such cases. Here the empyema dominates the picture.

In these acquired cases, even more so than in the congenital types, there is a striking amount of inflammatory change in and around the lung, so that any operative procedure is fraught with difficulty. However, in Hughes and Fox's series, of 31 cases treated by various means, 22 were cured; of 14 patients undergoing direct repair, 3 died. Thus the fistula may close on conservative treatment, though this necessitates gastrostomy in many cases (cf. case 4). Particularly, however, if there is any diverticulum of the oesophagus, operative measures will probably be needed. The particular operation depends on the exact pathology present, but in many patients it will entail lung resection in addition to closure of the fistula, since permanent damage in the form of bronchiectasis or chronic lung abscess may have occurred by the time the patient comes to operation.

SUMMARY

The aetiological factors leading to a fistulous communication between the oesophagus and the tracheo-bronchial system are discussed, with particular reference to the non-malignant forms. These may be congenital or acquired.

Illustrative cases of 4 types of fistula are reported.

Some aspects of other cases reported in the literature are mentioned, together with treatment. It is pointed out that most of these non-malignant fistulae can be cured. They are, however, uncommon and only about 100 have been recorded in the literature.

Thanks are due to Prof. J. H. Louw who referred cases 2, 3 and 4; to Mr. W. L. Phillips for permission to publish case 3; to the Superintendents of Groote Schuur and Somerset Hospitals for permission to publish these cases; and to Mr. B. Todt for the radiological reproductions.

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