BENIGN NEOPLASMS OF THE LUNG*

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Benign tumours of the lung are rare, but their clinical recognition is of considerable importance. Clinically, all tumours of the lung must be diagnosed as malignant on statistical grounds; the diagnosis of benignity can only be established with certainty on histological examination. With benign tumours relief of incapacitating symptoms can be offered surgically with low mortality and excellent long-term results.

In my own Thoracic Surgical Unit we have seen approximately 350 cases of bronchogenic carcinoma in the past 10 years. During this period we have seen only 22 cases of benign tumour, comprised of the following pathological varieties, viz. bronchial adenoma 9 cases, hamartoma 5 cases, leiomyoma 2 cases, fibroma 2 cases, and 1 case each of papilloma of the bronchus, chondroma of the lung, endobronchial lipoma, and haemangioma. Two of these cases, viz. a leiomyoma of the right main bronchus and a fibroma of the pleura, showed histological suspicion of malignancy which, as yet, has not been confirmed by their benign clinical course.

CLASSIFICATION

These benign tumours can be classified in two ways, according either to their anatomical site or to their histological type. Whatever the pathological type of the tumour the symptoms will depend on its anatomical origin.

Anatomical Classification:
(*a*) Endobronchial (*b*) Intrapulmonary (*c*) Pleural
Pathological Classification (in order of frequency):
(*a*) Bronchial adenoma (*d*) Fibroma (*g*) Papilloma
(*b*) Hamartoma (*e*) Haemangioma (*h*) Chondroma
(*c*) Lipoma (*f*) Leiomyoma (*i*) Single cases of bronchial angioma, lymphangioma and neurofibroma have also been described, according to Langston.[1]

Most of these tumours present anatomically either endobronchially or interstitially, e.g. adenoma, hamartoma, lipoma and leiomyoma. The fibromata presents either endobronchially or from the pleura. The papillomata present only within the bronchus. Haemangiomata present only in the lung substance.

BRONCHIAL ADENOMA

In all published series the commonest benign tumour is the bronchial adenoma. There is considerable discussion whether these tumours ought strictly to be included amongst the benign lesions. The British school on the whole consider them to be benign but never recommend bronchoscopic removal; the American school on the whole consider them to be malignant. McBurney *et al.*[2] state that of their 111 bronchial adenomas 9 metastasized, and on a review of the literature they found that of 700 tumours 78 had shown clinical metastases. Goldman[3] states, 'It is unlikely that malignant change often occurs in bronchial adenoma but from its inception it is either benign or malignant'. Kincaid-Smith and Brossy[4] describe a case of bronchial adenoma in a female of 58 in whom a right middle and lower lobectomy was performed and in whom a solitary secondary of similar histological character was removed from the liver 6½ years later.

Pathogenesis and Pathology

In 1938 Womack and Graham[5] stated, 'These tumours are of mixed developmental origin arising from rests of foetal lung'. In this way they explain the tendency in these tumours to a varying histological structure. Willis[6] however, states that they arise from the mucous glands of the bronchial wall. It should be recalled that these glands lie partly outside the cartilages and partly superficial to them in the submucosa. That is why the bronchial adenoma is partly submucosal and partly outside the cartilage ring. The important feature is that, even if a small portion of the bronchial adenoma is visible bronchoscopically, it is often like an iceberg with 9/10ths of the tumour outside the bronchial wall.

Liebow[7] states that there are 2 types of bronchial adenoma, as follows: (1) The carcinoid type comprises 85% of the cases of bronchial adenoma. Columns or groups of cells are seen, separated by highly vascular stroma, and a pseudo-acinar picture may be simulated. The cells are regular both in size and in staining property. This histological group gives the best prognosis. (2) Cylindroma consist of branching, tubular epithelial structures with irregular acini and are more invasive microscopically, with a tendency to mucin formation. They account for about 15% of the bronchial adenomas.

Further subdivision of bronchial adenoma is in my opinion not justified.

Site. Most bronchial adenomas occur in the visible bronchi, the right middle and lower bronchi being common sites.

Age. Most of these tumours occur between the ages of 30 and 40. Sherman[8] in 1956 stated that there had been only 10 cases of bronchial adenoma under the age of 14; Ward[9]

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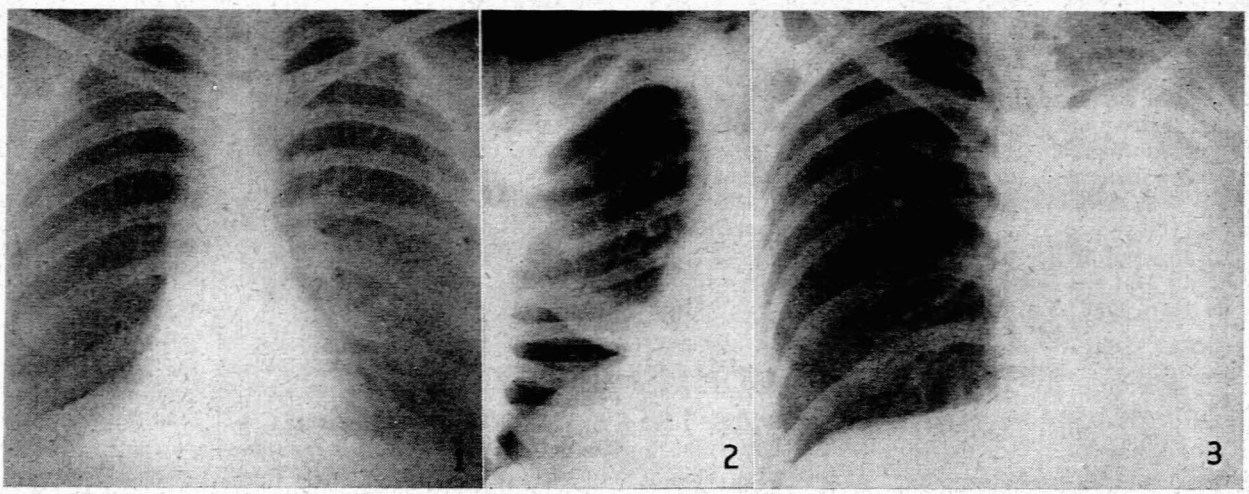

Fig. 1. Note the emphysema of the right lung field as shown by the sparse lung markings compared to those on the left. A bronchial adenoma of the intermediate bronchus has produced atelectasis of the right middle and lower lobes with compensatory emphysema of the right upper lobe.
Fig. 2. Lordotic film showing right middle lobar atelectasis due to bronchial adenoma in a female of 22 years.
Fig. 3. Radiograph demonstrating left-sided pneumonic atelectasis due to bronchial adenoma in a female of 39 years.

reported the youngest in a child of 7 years. We have had an adenoma in a boy of 12, who remains well 8 years after right middle and lower lobectomy.

Sex. Most cases are described in females. Of the 9 cases we have seen, only 2 occurred in males.

Symptoms

In most cases the presence of bronchial adenoma causes definite symptoms, but Good[10] reports that of 100 consecutive cases 17 were quite asymptomatic and were found incidentally on radiological investigation. The symptoms depend on the site of the tumour, whether obstruction to the bronchus is complete or incomplete, and whether infection has occurred distal to the obstruction.

(*a*) *Symptoms due to the tumour:*
(i) Ulceration of the tumour frequently occurs with resulting haemoptysis. This occurred in 6 of our 9 cases.
(ii) A dry troublesome cough due to the presence of the tumour from irritation similar to that caused by a foreign body.
(*b*) *Symptoms due to obstruction:*
(i) *Incomplete obstruction* causes unilateral wheezing from the fact that air is able to enter the bronchus during inspiration but is trapped in the lung during expiration. This obstructive unilobar or pneumonic emphysema can cause marked breathlessness.
(ii) *Complete obstruction.* Sudden atelectasis may cause pleuritic pain and discomfort from the negative pleural pressure. This atelectasis is often accompanied at first by severe breathlessness.
(*c*) *Presence of infection:*
(i) Suppuration occurs distal to the obstruction and a productive cough results.
(ii) Intermittent obstruction results in an abscess distal to the obstruction and produces intermittent expectoration of pus and all its sequelae.
(iii) Infection in an atelectatic lobe may result in bronchiectasis with continual cough, sputum, haemoptysis and pyrexia.
(iv) Infection sometimes spreads to the pleura, causing pleuritis with effusion. Empyema may result, with the development of a broncho-pleural fistula. We have had two such cases which were drained for many months and continued to suffer severe haemoptysis and produce pus. One of these was in a female with a bronchial adenoma and the other in a male with a papilloma of the bronchus. Both of them were treated by extrapleural pneumonectomy and pleurectomy.

The most important symptoms, therefore, are those of haemoptysis, cough, repeated lower respiratory infection and unilateral wheeze.

Radiological investigations:

The diagnostic X-ray appearances of bronchial adenoma are as follows:

(*a*) In 20% of cases the tumour mass itself is seen as the only radiological abnormality on X-ray.

(*b*) By far the larger number of cases are shown on the X-ray by the secondary effects of the tumour, as follows:

(i) Emphysema due to partial obstruction will be seen on screening or on comparing the films of inspiration and expiration.

(ii) Atelectasis will be seen when obstruction is complete. It may be either segmental, lobar (Figs. 1 and 2) or pneumonic. (Fig. 3.)

(iii) The presence of infection will be shown by an abscess, multiple abscess cavities, bronchiectasis, empyema, or broncho-pleural fistula.

A bronchogram, in my opinion, has little place in the diagnosis of the lesion, for it does not supply any pathological confirmation.

Bronchoscopy

This is the most valuable adjunct to diagnosis, for over 98% of these tumours occur in the visible bronchi. The site of the obstruction will be confirmed and pathological examination will be established.

Treatment

Bronchoscopic removal, so ably introduced by Chevalier Jackson, has few supporters today and is roundly condemned by most. McBurney[11] in 1952, stated, 'Since 1948 at the Mayo Clinic no cases have been treated by bronchoscopy as definitive treatment'. Irradiation is also of no value. Treatment is essentially surgical. Ideally, if the tumour is in a main or lobar bronchus and has not yet caused any secondary suppuration distal to it, bronchotomy is the treatment of choice. This was carried out in 1947 by Sir Clement Price Thomas.[12] Thoracotomy is performed, the tumour palpated through the posterior wall of the bronchus, which is opened, and the tumour with a sufficient sleeve of bronchial wall is excised and the bronchus reconstituted. This procedure is of value because it is conservative, without sacrifice of lung tissue. It has not been suitable for any of our cases; in all of them pulmonary resection has been necessary either because of the extent of the lesion or because of gross secondary infection. Seven of our cases came to surgery; in 3 of them pneumo-

nectomy was performed, and in 4 right middle and lower lobectomies.

HAMARTOMA

The term hamartoma was coined in 1904 by Albrecht,[13] who defined it as 'comprising a tumour-like malformation in which occur only an abnormal mixing of the normal components of an organ. The abnormality may take the form of a change in quantity, arrangement or degree of differentiation, or may comprise all three'. Histologically they originate in abnormal mixing of the normal structures.

Pathologically, hamartoma must be differentiated from endobronchial ecchondromas, described by Davidson,[14] which arise from the endobronchial cartilages, are covered by normal bronchial epithelium, and contain no other bronchial-wall elements.

There are 2 types of hamartoma, as follows:

(*a*) *Endobronchial.* These are rare. Donoghue *et al.*,[15] describing unusual bronchial tumours stated that 'of 11,626 patients bronchoscoped in the Mayo Clinic in 10 years there were 5 endobronchial hamartomas'. Paterson,[16] in 1956, reports the 31st case of endobronchial hamartoma. These tumours can cause all the symptoms of bronchial obstruction, though haemoptysis rarely occurs.

(*b*) *Interstitial.* These occur in the lung parenchyma and are usually found accidentally on radiological investigation. In rare cases they impinge on a bronchus, causing obstruction, and even, if they are exceptionally large, cause breathlessness. They are said to occur 3 times more commonly amongst males and have been described from youth to old age. Jones[17] described a rare case in a newborn infant.

The interstitial hamartomata are usually small, varying from a few millimetres to about 4 cm. in diameter, though larger tumours have been described. They are almost invariably subpleural in position and in rare cases may even lie free in the pleural space, being attached to the pleura only by a small pedicle. They are absolutely homogenous, firm to stony hard, and are usually lobulated and encapsulated. They are sharply demarcated from the lung and can usually be shelled out from the lung substance with ease.

Microscopically the bulk of the tumour consists of cartilage, but an abnormal mixture of the elements normally encountered in the bronchial wall are found, viz. ciliated epithelium, glandular epithelium, connective tissue, muscle, fat, and lymphoid tissues. The free surfaces of the lobules are covered by epithelium indistinguishable from the bronchial epithelium which dips down between the lobules in the form of deep clefts.

It is doubtful whether malignant changes have ever been convincingly described in these hamartomata. Simon,[18] however, in discussing a case suggests that certain histological appearances may indicate malignancy.

Incidence. Of 57 benign lung tumours described by Sir Clement Price Thomas[19] in 1954 10 were hamartomata. Rubin[20] found 28 cases in 8,000 routine autopsies—an incidence of 1 in 300. They were all asymptomatic. We have had 5 hamartomas—the youngest patient was a male of 22 and the oldest a female of 65. Of the 5 cases, 3 were females; 3 of the tumours were in the left upper lobe (see Fig. 4) and 2 in the apex of the right lower lobe; 3 showed myxomatous changes. Of the 5 tumours, 3 were shelled out from the lung by very easy enucleation; another was in a patient who was being treated in a tuberculosis sanatorium for a tuberculoma, and as the tumour felt softish and could not be shelled out lobectomy was performed (histologically the tumour showed myxomatous change); the other was in a male of 52, who was treated by lobectomy because there were glands present, and the presumptive diagnosis was that of carcinoma the bronchus.

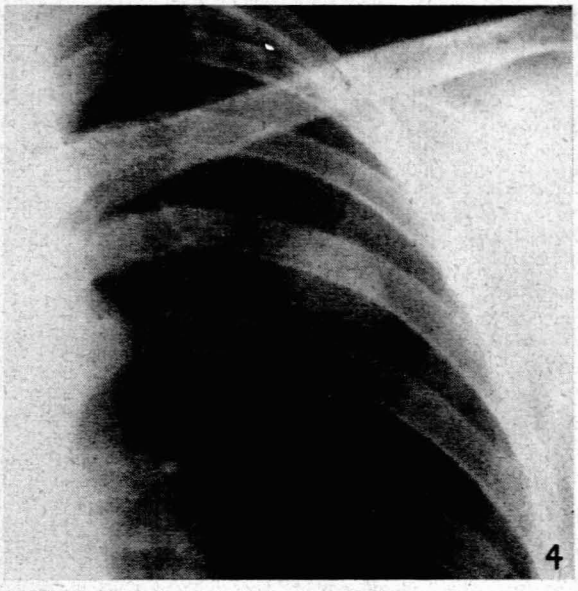

Fig. 4. Radiograph showing a well circumscribed opacity due to an hamartomain a female of 40.

FIBROMA

Fibromata of the lung must be differentiated from those arising from the mediastinum and those of neurogenic origin arising in the paravertebral gutter. Fibromata of the lung appear in two sites:

(*a*) *Endobronchial.* Of these we have no experience. Price Thomas[19] records 2 in his series of benign tumours. He states: 'They show as a lobulated mass growing within the lumen of a dilated bronchus. Histologically they show a rather cellular fibromatous tissue covered by columnar epithelium.' They present with all the symptoms of bronchial obstruction.

(*b*) *Fibromata of the Visceral Pleura.* These present such a characteristic clinical syndrome that they can often be diagnosed pre-operatively. They are sometimes silent from the chest point of view, but occasionally they present with some discomfort or pleuritic pain and a dry cough which later produces a little phlegm, but there is rarely haemoptysis. Early in their history the patients usually suffer from arthralgia, with marked clubbing and pulmonary osteo-arthropathy, which sometimes antedates the chest symptoms. The severe arthralgia disappears dramatically immediately after removal of the tumour.

Macroscopically these fibromata can be seen to arise from the visceral pleura, to which in some cases they are attached by a pedicle; in other cases they compress the underlying lung, from which they arise and from which they cannot subsequently be separated. They are hard, firm, usually smooth, occasionally lobulated, and very well defined. At thoracotomy they can usually be differentiated from malignant tumours with ease. If possible they should be removed without sacrificing lung tissue, or a thin sliver should be

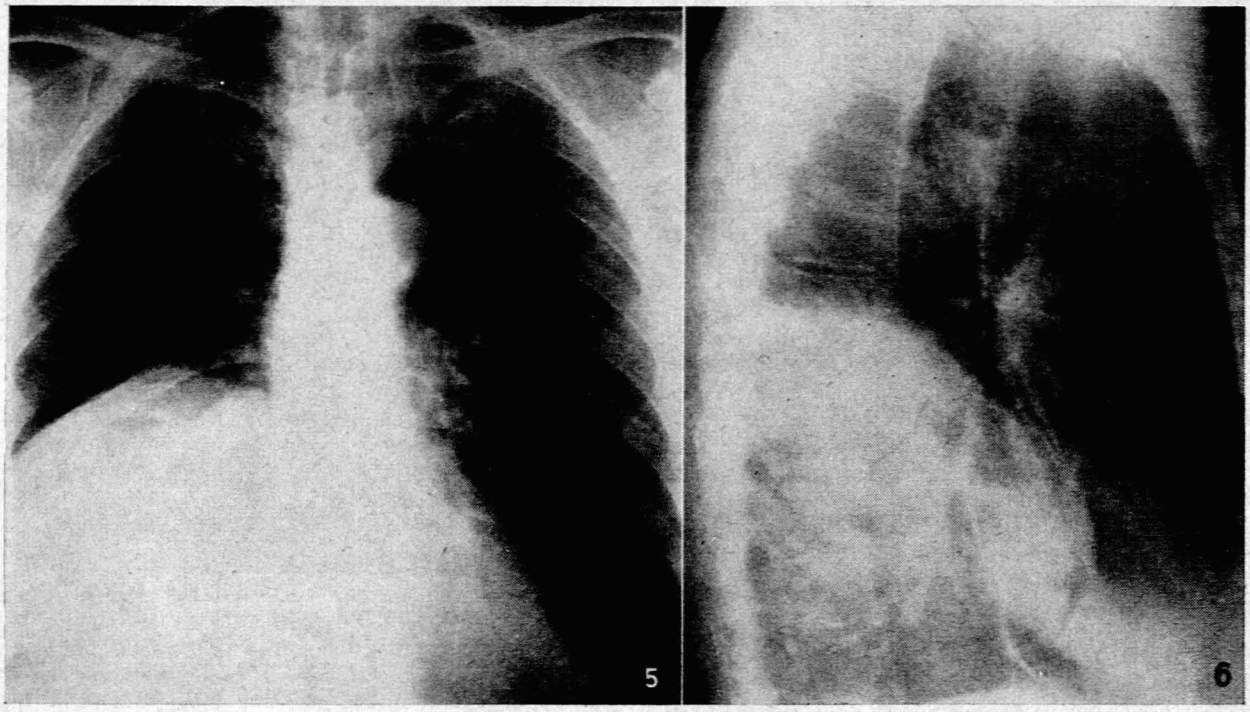

Fig. 5. Radiograph in a male of 54 years showing a dense homogenous opacity at the right base inseparable from and simulating an elevated right diaphragm or an infralobar effusion.

Fig. 6. Lateral film of the same case as in Fig. 5, showing a posteriorly situated fibroma of the visceral pleura.

removed in case there is malignant change. Often, however, the lung has been so compressed by the tumour mass that there is no plane of cleavage and lobectomy has to be performed.

Sir Clement Price Thomas[23] reviewed 6 cases of fibroma of the visceral pleura in 1953.

We have had 2 such cases, both in males. The first occurred in a man of 54 who complained of chest pain but had no clubbing of fingers. Fibroma of the pleura was suspected on radiological grounds because there was a well-defined homogeneous mass lying in the right oblique fissure and bronchoscopy was normal and bronchial washings and smears were negative for malignancy. At thoracotomy a hard, firm, well circumscribed mass arose from the visceral pleura between the two lobes, from which, however, it could not be separated, and as there was an area where the capsule appeared irregular it was felt advisable to perform a right middle and lower lobectomy. The tumour was found to be a fibroma on histological section.

Our second case was in a man of 63 who had a 6 months' history of pain and discomfort in his hands and fingers and gave a 3 months' story of a chest cold with slight productive cough. At the end of this period he consulted a doctor, who thought he had an effusion and unsuccessfully tried to aspirate his chest. He was referred for thoracic surgical opinion when an X-ray showed a huge, well circumscribed mass occupying the whole of the lower right lung field posteriorly (Figs. 5 and 6). The mass could not be distinguished either from the diaphragm or from the paravertebral gutter. Bronchoscopy showed extrinsic pressure on the right-lower-lobe bronchus, and in a Dionosil bronchogram the right lower lobe failed to fill. No evidence of malignant cells were found in the bronchial washings or bronchial slides. The pre-operative diagnosis was that of a fibroma of the pleura, which was confirmed at right thoracotomy, which showed a huge mass arising from the visceral pleura of the right lower lobe, which was grossly compressed; Mr. Denis Fuller performed an uneventful right lower lobectomy with dramatic alleviation of the patient's symptoms and early disappearance of his clubbing.

OTHER BENIGN BRONCHIAL TUMOURS

Lipoma

Intrathoracic lipomata are rare in the lung and those that have been described have mostly occurred endobronchially. Donoghue,[15] writing from an extensive experience at the Mayo Clinic, has described only 4 cases. According to Smart,[21] only 14 cases of intrathoracic lipoma are described in the literature during the period 1927-53. A 15th case treated by transpleural bronchotomy is reported by Brewin[22] in 1952. We include 1 endobronchial submucosal lipoma in our series in a man of 59. This patient had had a dry cough for 2 years, with mucus in the sputum and recurrent attacks of fever. When he was seen in consultation he was producing half a cupful of thick purulent sputum a day but never had any chest pain or haemoptysis. Bronchoscopy had shown a smooth pedunculated tumour arising distally to the left-upper-lobe bronchus. Histological examination showed an intact basal membrane with a large amount of fat underneath. Left pneumonectomy was performed by Mr. Denis Fuller because there was a markedly emphysematous lower lobe, a good deal of pus in the lower-lobe bronchi, and large glands, which fortunately proved to be inflammatory. In addition fibrocaseous tuberculosis was present, with no evidence of activity.

Haemangioma

Some authors differentiate these from arterio-venous aneurysms. Goetz *et al.*[24] state, 'It is a developmental malformation and not a tumour'. Others, however, feel that there is no differentiation between the haemangioma and the arterio-venous aneurysm.

We have had one such case in a female of 43 with exertional dyspnoea, cyanosis, clubbing, and polycythaemia. Clinically and on full investigation at the Cardiac Clinic of the Department of Medicine at the Johannesburg General Hospital the diagnosis was made of aortic stenosis, mitral stenosis, and a pulmonary arterio-venous aneurysm of the left lower lobe. Left thoracotomy was undertaken and an uneventful left lower lobectomy and mitral valvotomy performed. As the pressure gradient across the aortic valve was not sufficient to justify aortic commissurotomy the aortic lesion was not explored. The patient showed marked post-operative improvement in her cyanosis, but she collapsed 48 hours after operation with peripheral circulatory failure which could not be reversed. The immediate post-operative X-ray had suggested no abnormality but on the second day, when she collapsed, there was a suggestion of a mediastinal haematoma. At autopsy the lungs were found to be grossly haemorrhagic and oedematous, and on section showed massive intrapulmonary haemorrhages and ante-mortem thrombosis. The kidneys showed ischaemic renal tubular necrosis. Death was thought to be due to shock from the intrapulmonary haemorrhages and thrombosis, which were probably secondary to the polycythaemia.

Leiomyoma

Although muscle tumours of the lung and bronchi are extremely rare and most that are reported are sarcomatous, we have seen 2 cases of the condition. Amongst the benign tumours reported there have been 3 intrabronchial leiomyomata and 4 intrapulmonary leiomyomata. In addition 2 rhabdomyomata have been reported. Reports of only 4 surgically removed benign muscle tumours of the lung or bronchus are found.[25]

Our first case occurred in a male aged 50 who was quite asymptomatic but was included in a mass X-ray. His film showed a large, well defined, homogenous tumour mass in the right upper lobe (Fig. 7) for which thoracotomy was advised. At operation a hard, well circumscribed mass was found, occupying most of the upper lobe; no enlarged glands were found. It did not feel like a hamartoma and therefore right upper lobectomy was performed. When the tumour mass was cut across it looked like a fibroma of the uterus, and histological examination, for which I am grateful to Dr. Ian Webster of the SAIMR, confirmed that it was indeed a leiomyoma.

Our second case occurred in a 4-year-old male child seen in March 1956 with a 3 months' history of whooping cough. Following this illness the child was breathless and wheezy, and was treated for 'asthma'. During these attacks the child had high fever and scattered bilateral rhonchi were heard. Subsequently there was clinical and radiological evidence of right-lower-lobe collapse and Dr. L. B. Sunn of East London suspected a bronchial tumour. Bronchoscopy showed that the carina was broadened, and occupying the whole of the right main bronchus was a mobile, soft, friable, vascular growth, the appearances of which I thought were those of a bronchial adenoma. Dr. Webster reported that the histological features were those of a leiomyoma, and that no sarcomatous change was observed. Thoracotomy showed that the right middle and lower lobes were atelectatic, and although it might have been possible to do a right middle and lower lobectomy and perhaps remove the tumour by bronchotomy there were fleshy glands surrounding the right main bronchus, and in view of the possibility of sarcomatous change I performed a right pneumonectomy. Thus far the child has remained well apart from mild intercurrent infections.

Papilloma

Although Langston[1] states that benign endobronchial papilloma has been described, several of the papillomata that have been reported have subsequently been proved to be bronchogenic carcinoma, and the others are secondary inplants from papillomata of the larynx; the only substantiated case of primary papilloma of the bronchus is one described by Ashmore.[26] His case was that of a female aged 51 who had an haemoptysis and for whom left lower lobectomy was performed for a tumour which, on histological examination, proved to be a pure primary papilloma.

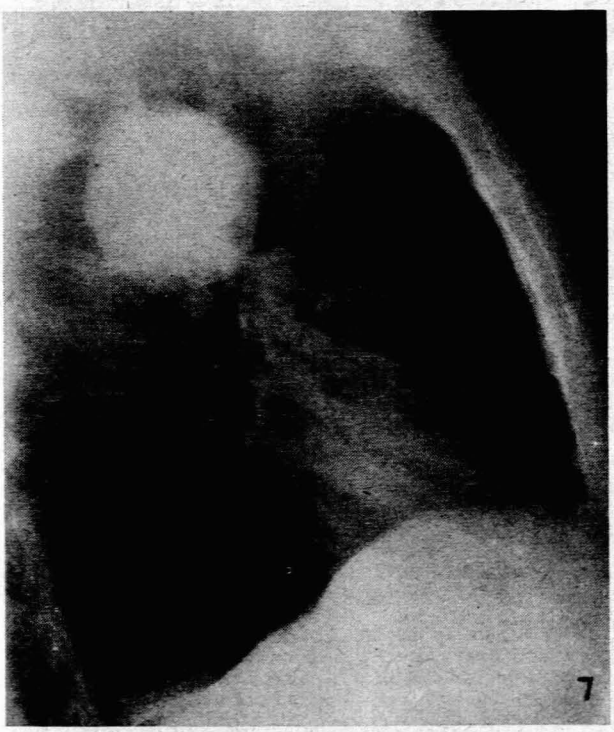

Fig. 7. Lateral film showing, in the right upper lobe of a male of 50 years, a well circumscribed opacity which could be due to any one of many causes. Right upper lobectomy showed growth which proved to be leiomyoma on histological examination.

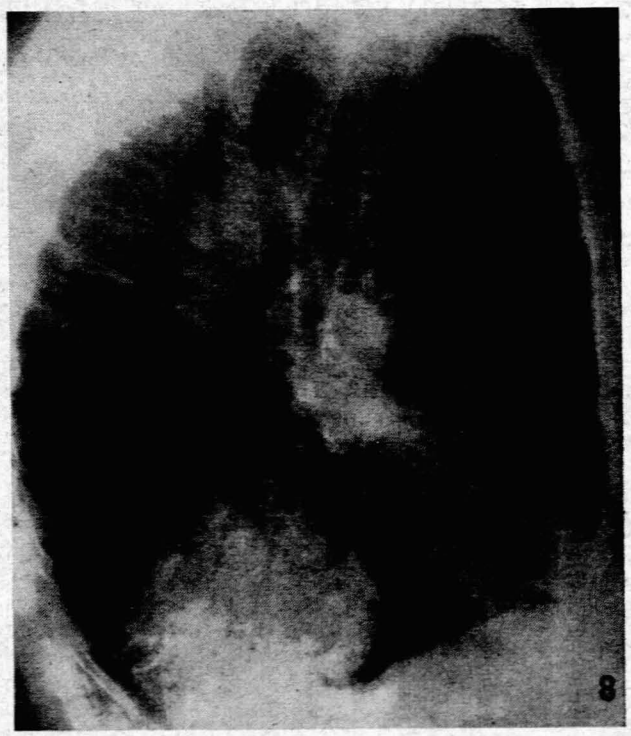

Fig. 8. Lateral film showing a well circumscribed mass with calcification. This was thought to be an hydatid, and on removal by a lobectomy proved to be a chondroma.

We have had one such case, in a male 34 years old, whom we first saw in September 1947 with a 3-year history of cough, left-sided wheeze, and subsequent pain. In 1945 he had developed an empyema for which rib resection was performed and the sinus had discharged for a year. In 1946 Mr. W. L. Phillips, after bronchoscopy, at which he had removed a portion of the tumour for section, had advised left lower lobectomy. In the year following this he continued to cough up to 2 pints of foul sputum a day and intermittent haemoptyses had occurred. An empyema with broncho-pleural fistula was diagnosed and when I bronchoscoped him in 1947 I found a sessile tumour in the left-lower-lobe bronchus. Biopsy of this confirmed the diagnosis of papilloma of the bronchus. A bronchogram unfortunately showed that he now had gross bronchiectasis of the left upper lobe as well as the lower lobe and that he had a broncho-pleural fistula. For this extra-pleural pneumonectomy was performed in September 1949 and he has remained well except for progressive exertional dyspnoea.

Chondroma

These arise from the bronchial cartilages and can be either endobronchial or parenchymal (Fig. 8). They must be distinguished from heterotopic bone formation, which is rarely seen in chronic lung abscess. We have had a pulmonary chondroma in a male of 61, whose chief symptoms were cough, discomfort in the chest, and some pleural pain. Left lower lobectomy was performed by Mr. G. Katz. A year later the patient died, and autopsy showed a squamous carcinoma at the stump of the left-lower-lobe bronchus.

SUMMARY AND CONCLUSIONS

Benign tumours of the lung are uncommon. They are of various types and either show the symptoms of bronchial obstruction or are discovered by chance on incidental radiological investigation. Bronchoscopy for pathological examination is essential if symptoms are present and, in all cases, thoracotomy, and *not* an expectant attitude, is advised, because the benign character of these tumours cannot be assured without full histological examination.

This article classifies the benign tumours of the lung, reviews present knowledge on the subject, and relates the author's experience with 22 cases of benign bronchial tumour seen in the Thoracic Surgical Unit of the Johannesburg General Hospital.

I should like to thank Dr. K. Mills, Medical Superintendent of the Johannesburg General Hospital, for allowing me access to the files of hospital patients.

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