

RECURRENCE OF "BENIGN" INTRATHORACIC FIBROMAS

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Intrathoracic fibromas are uncommon but distinctive tumours. They are usually considered to be benign. The origin of these growths, which appear to be primary pleural tumours, has given rise to a diversity of opinion. The literature is confusing, for essentially similar tumours are reported by a variety of names, of which the most common are intrathoracic fibroma, pleural fibroma, and benign localized mesothelioma of the pleura. For purposes of orientation, the subject will be briefly reviewed. The primary concern of this paper, however, is with the liability of these "benign" tumours to recur, an aspect of their behaviour not widely appreciated. Late recurrence especially as a complication is seldom mentioned.

Below we detail the histories of six patients who developed local recurrence after removal of tumours diagnosed as benign fibromas. Every patient with a recurrence has been operated on, examined histologically, and compared with its antecedent tumour. The recurrences have ranged from a pedicled, encapsulated, cellular fibroma to a highly malignant fibrosarcoma. In patients with a succession of recurrences the clinical and pathological pattern has been one of progression towards greater malignancy.

CASE HISTORIES

CASE 1 (Case 2 of Price Thomas and Drew (1953) brought up to date).—In 1944 V. S. M., a woman aged 56 years, began to suffer from pains in the ankles and wrists, accompanied by episodes of fever. A chest radiograph was reported as showing no abnormality. Three years later a large round opacity was apparent in the left lower zone (Fig. 1). She was referred to Mr. N. R. Barrett at the Brompton Hospital.

First Operation.—On November 10, 1947, Mr. N. R. Barrett operated and a huge extrapulmonary tumour was exposed. It was attached by a narrow pedicle to the inferior margin of the left lower lobe, which except for the apical segment was collapsed. With the exception of hard glands in the hilum of the

lung, thought to represent old calcified tuberculous adenitis, the lungs and the rest of the pleural cavity were normal.

The pathologist reported that the specimen was a firm grey-brown flattened mass of approximately round outline, having a mean diameter of 10 cm. and a maximum thickness of 6.5 cm. It had a smooth surface and a definite capsule of 0.02 cm. mean thickness, and its total weight was approximately 500 g. Microscopically, the specimen was a cellular fibroma, showing areas of cellular necrosis with collagen replacement. It had a fibrocytic collagenous capsule covered with wide flattened "epithelial" cells arranged two cells deep.

The patient made a rapid recovery, and her joint pains disappeared in seven to 10 days. She remained well, and a radiograph in 1955 was normal. She returned in June, 1959, when she was 71 years old, and stated that since the previous winter she had had

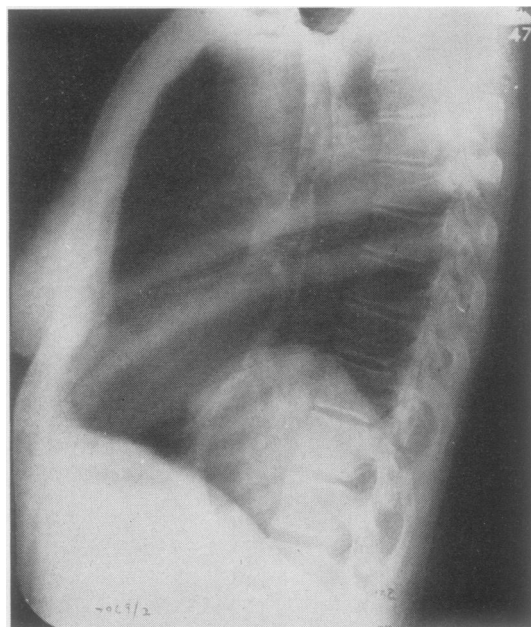


FIG. 1.—Case 1: Left lateral radiograph demonstrating the lesion in 1947.

an ache in the left side, and more recently a return of arthritic symptoms. A radiograph showed another large oval opacity lying medially behind the heart (Fig. 2).

Second Operation.—This was performed on July 16, 1959. This tumour was extrapulmonary, but it arose from the parietal pleura covering the descending aorta. The vessels entering the pedicle could be traced under the pleura as far as the costophrenic sinus. Between the tumour and the lung were filmy vascular adhesions. The tumour and its pedicle were removed. The patient made an uneventful recovery.

Dr. K. F. W. Hinson reported that the tumour was oval, $9 \times 7 \times 6$ cm. in its diameters. The outer capsule was smooth and apparently complete. Microscopically it was a cellular fibroma and benign. The pedicle showed thick-walled arteries and a normal lymph follicle.

The sections of the 1947 tumour are no longer available, but the histological description would fit this tumour.

CASE 2.—H. R., a man of 63 years complaining of cough, breathlessness, and pain in the chest, attended St. Thomas's Hospital in 1950. Radiographs taken at another hospital in 1944 demonstrated a large tumour in the lower part of the left hemithorax. By 1950 it was about one-third as large again (Fig. 3).

First Operation.—On May 16, 1950, Mr. N. R. Barrett operated. On opening the chest, the large lobulated tumour was immediately exposed. It was adherent to the lung and the diaphragm, but it shelled out without much difficulty and with very little bleeding. There was no obvious pedicle, and the only

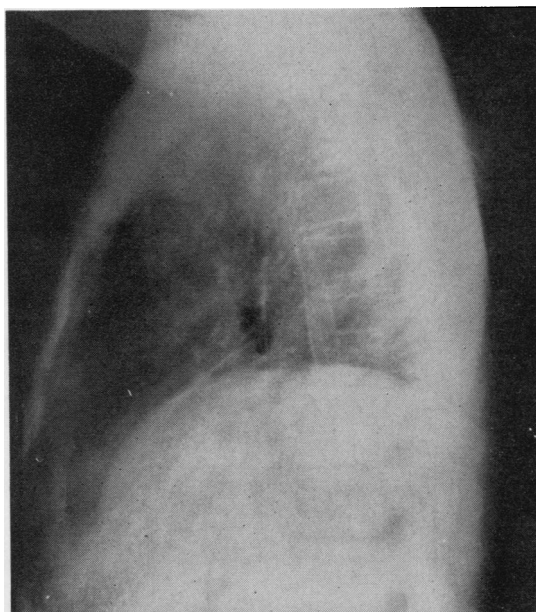


FIG. 3.—Case 2: Left lateral radiograph showing the original growth in 1950. This lesion had previously been seen on a chest film taken in 1944.

place that seemed to be supplying it with blood was the lower surface of the lung.

The tumour measured $17 \times 17 \times 12$ cm., and had a smooth surface except for one or two projecting nodules. It was completely encapsulated. On section it showed oedematous fibrous tissue arranged in irregular trabeculae, with central necrosis, cystic degeneration, calcification, and mucoid degeneration. Microscopically it was a fibroma.

After the operation, the patient's health was greatly improved, and a chest radiograph taken in 1956, when he was readmitted for repair of an inguinal hernia, did not show any abnormality. In January, 1958, when he was 71 years old, he felt generally unwell, and another film revealed a large rounded opacity in the left lower zone (Fig. 4). There was no evidence of glandular or other metastases.

Second Operation.—Mr. N. R. Barrett operated on January 20, 1958. On reopening the chest, it was found that the tumour was mostly inside the lower lobe, although its white, lobulated inferior surface projected and was adherent to the diaphragm and chest wall. A lower lobectomy was performed and the affected part of the chest wall was extensively coagulated with diathermy.

The specimen was the left lower lobe with a roughly spherical tumour partially embedded in it. In most places where the tumour was in relation to the lung, the two tissues were sharply separated by a fibrous capsule, but in or two places the tumour tissue seemed to merge insensibly with fibrous tissue in which there

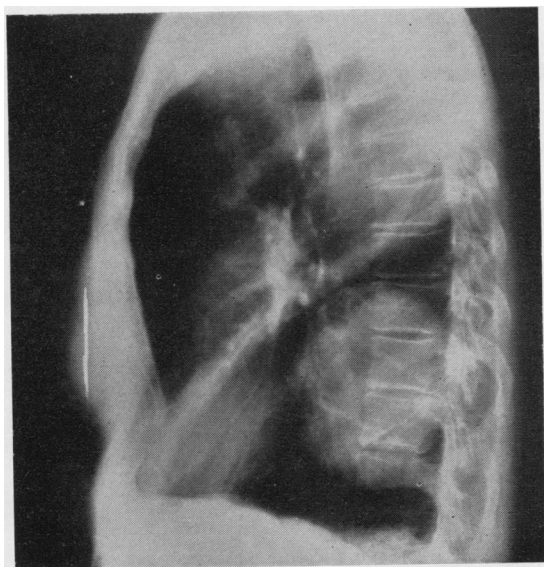


FIG. 2.—Case 1: Left lateral radiograph showing recurrence of the tumour in 1959.

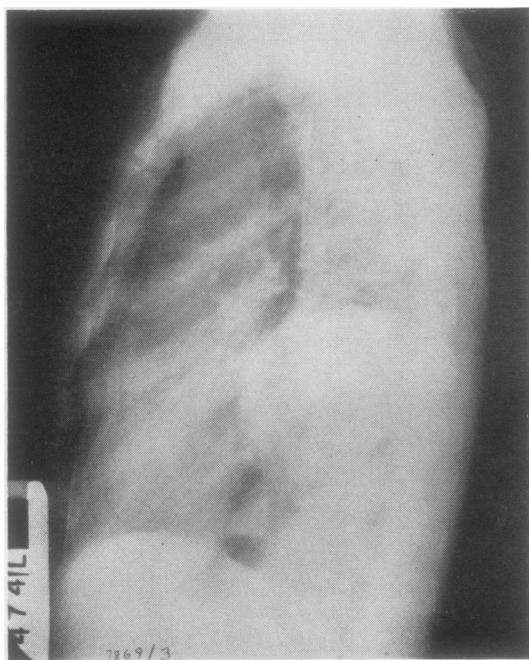


FIG. 4.—Case 2: Recurrence of the lesion in 1958.

were remnants of alveoli. The lung, little more than a cap covering part of the mass, was much compressed, but was otherwise normal. Microscopically, the structure was essentially similar to that of the tumour removed in 1950. In some places, cells were numerous, and of rather embryonic appearance, and mitotic figures were not infrequent. A well-defined fibrous capsule was seen even where on gross examination there seemed to be none. There was no tumour tissue in any of the four extrapulmonary hilar glands. "I have called the tumour a fibroma because, although it was locally invasive, I think it unlikely that it was capable of producing metastases."

In August, 1960, the patient had to be admitted again. There were swellings in the operation scar. Mr. N. R. Barrett operated again, and exploration revealed a growth extending from the chest into the abdomen, where it involved the kidney and the large vessels. It was quite irremovable. A biopsy was taken and the tumour proved to be a fibrosarcoma.

CASE 3.—G. B., a man of 62 years, attended the London Chest Hospital in 1953. Two years previously a febrile illness had led to his being radiographed at another hospital, and an opacity was discovered in the left chest (Fig. 5). He had not felt well since the illness, and had become increasingly breathless on exertion. New radiographs showed that the opacity had increased in size, and bronchoscopy revealed gross distortion of the left main bronchus.

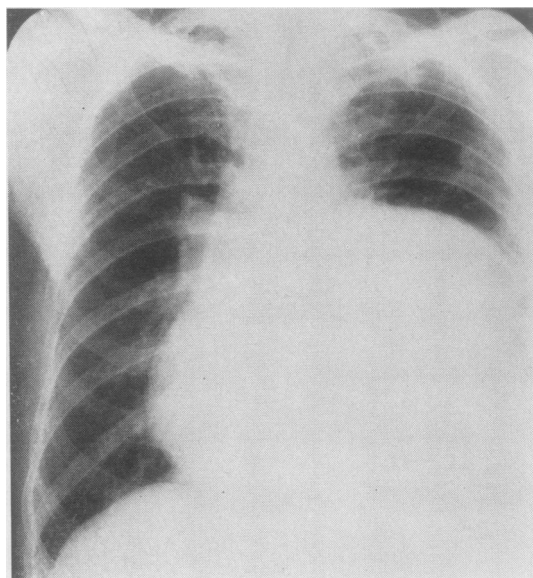


FIG. 5.—Case 3: Large tumour in the left hemithorax in 1953, known to have been present in a radiograph in 1951.

First Operation.—Mr. Vernon Thompson operated on March 25, 1953, when a large lobulated tumour in the anterior mediastinum, adherent to the parietal pleura and pericardium, was found to be causing compression collapse of the left lower lobe. The tumour was well encapsulated, and shelled out satisfactorily.

The panel of the Tumour Registry reported that this was a fibroma weighing 4 lb. 7 oz. (2,047 g.) with extensive necrosis. There was nothing in the histological picture of the preserved areas to suggest malignancy.

The patient, who made a good recovery, was seen yearly. In 1958 he complained of abdominal pain and a non-productive cough. A radiograph (Fig. 6) showed an oval opacity in the right lower zone and a right pleural effusion. A rounded shadow projecting from the left hilum suggested recurrence in the left side of the chest also. Aspiration of the effusion yielded a heavily blood-stained fluid in which no neoplastic cells could be found.

Second Operation.—Mr. Vernon Thompson operated on December 3, 1958. In the posterior mediastinum, and adherent to the lower lobe of the right lung, there was an extrapleural encapsulated tumour, 15 × 5 cm. in size. A smaller, spherical tumour, 2.5 cm. in diameter, was embedded in the anterior segment of the right upper lobe. Both tumours were easily excised.

Dr. K. Hinson reported that the larger mediastinal tumour was irregularly lobulated and spherical. It

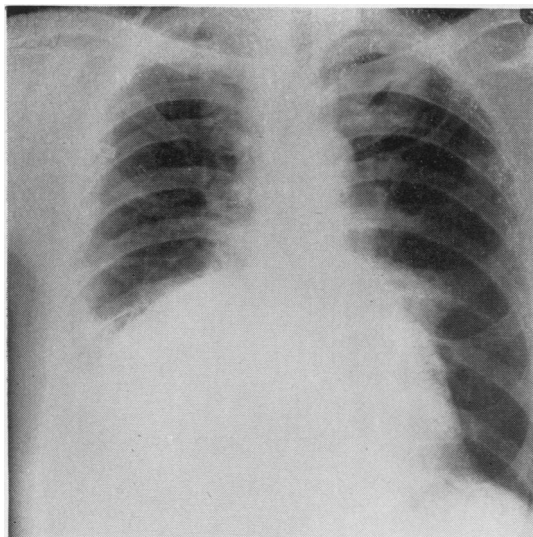


FIG. 6.—Case 3: Recurrence of same tumour in the opposite hemithorax in 1958; note the rounded shadow in the left hilum, suggesting recurrence on this side also.

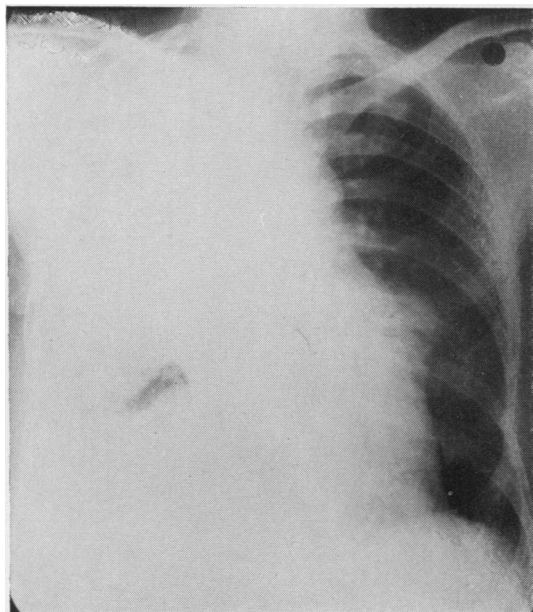


FIG. 7.—Case 3: Second recurrence, this time frankly sarcomatous, in 1959.

had a white, glistening cut surface, except where there was some myxomatous degeneration. Microscopically it was very cellular, but similar to the tumour removed in 1953. The smaller nodular tumour from the lung substance was again similar microscopically. There was probably a false capsule of collapsed fibrosing lung. Both tumours were fibromas.

In September, 1959, the patient was readmitted complaining of constant epigastric pain and severe dyspnoea. Radiographs (Fig. 7) showed a right pleural effusion and multiple opacities in that side of the chest.

Third Operation.—At operation on September 25 the chest wall and the lung each contained many nodules of growth, and the condition was inoperable. The patient died three days later.

Dr. P. Gregory performed the necropsy. The growth involved the chest wall posteriorly and invaded the lung. It also extended downwards into the abdomen behind the liver. Microscopically it was a fibrosarcoma.

CASE 4.—H. A., a woman of 50 years, had enjoyed excellent health until nine months before her admission to the London Chest Hospital. Her first symptoms were a dry cough, chest pain, and marked dyspnoea, followed two months later by dysphagia and loss of weight. A radiograph (Fig. 8) showed a large tumour with some calcification, and a barium swallow demonstrated gross displacement of the oesophagus to the left.

First Operation.—Mr. D. Barlow performed a right thoracotomy on February 2, 1955. Over half

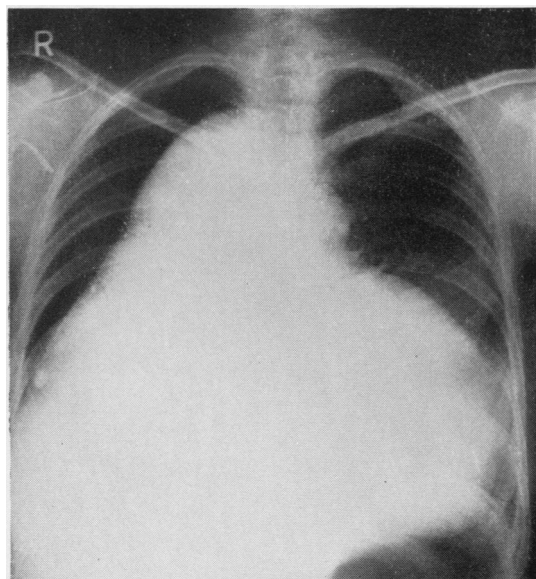


FIG. 8.—Case 4: Large mediastinal tumour extending into both sides of the chest in 1955. The oesophagus was pressed against the left lateral chest wall. Note calcification in the growth.

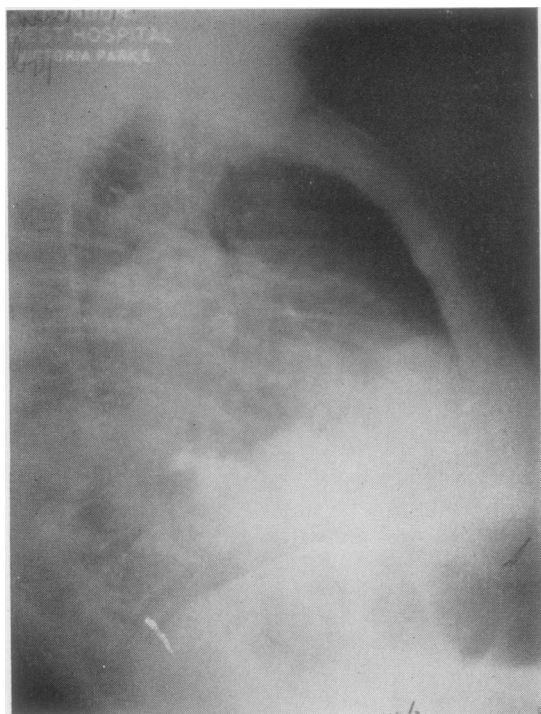


FIG. 9.—Case 4: Right lateral radiograph demonstrating massive recurrence of growth in the mediastinum two years later.

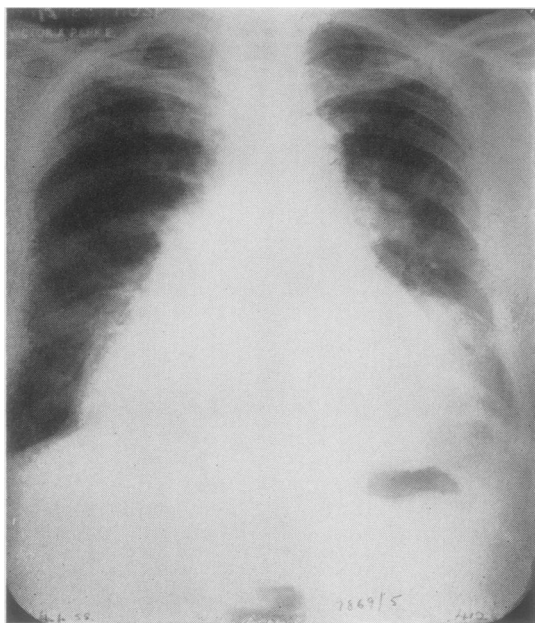


FIG. 10.—Case 5: Radiograph showing mediastinal tumour projecting into both sides of the chest in 1955.

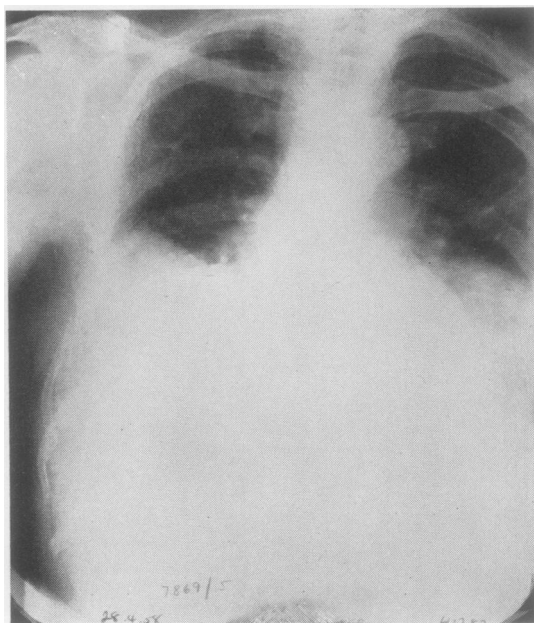


FIG. 11.—Case 5: Three years after removal, recurrence on both sides of the chest.

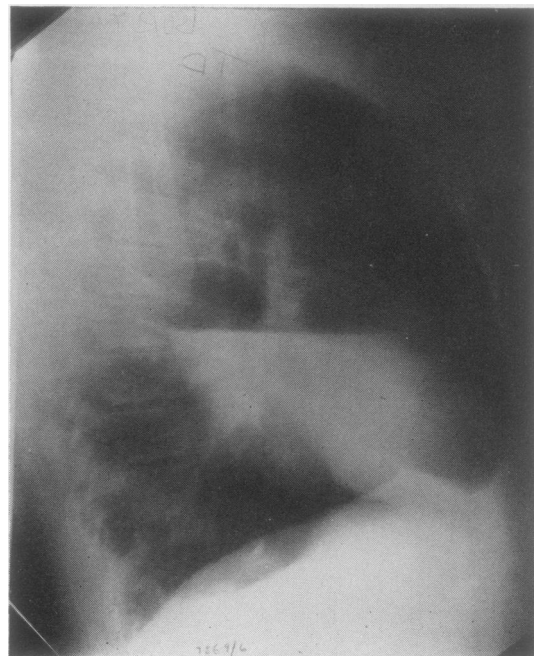


FIG. 12.—Case 6: Right lateral radiograph showing large cyst with fluid level in 1955. This cyst contained a small fibroma.

of the right hemithorax and one-third of the left was occupied by a very large well-encapsulated tumour which displaced the heart forwards and the oesophagus so far to the left that it lay in contact with the lateral wall of the chest. The tumour, which was soft, greyish-yellow, and gelatinous in places, was removed whole with relative ease.

Dr. K. Hinson reported that it was a fibroma weighing 10 lb. (4,550 g.), showing myxomatous degeneration.

The patient made a good recovery and rapidly gained weight. She kept well until July, 1956, when she noticed increasing tightness in the chest and breathlessness. Radiography on January 12, 1957 (Fig. 9), showed a large recurrence in the (posterior) mediastinum compressing the trachea and oesophagus.

Second Operation.—Mr. D. Barlow operated again on January 18, 1957. When the left side of the chest was opened, the mediastinal pleura was seen covering a swelling which extended from in front of the trachea to behind the oesophagus. The tumour consisted entirely of gelatinous material which could only be removed piecemeal.

Dr. K. Hinson reported that the specimen was a collection of gelatinous material. Microscopically there was myxomatous degeneration of a fibroma.

The patient was at first considerably relieved by this last operation, but the growth recurred, involving the chest wall on the right side in the region of the ninth to twelfth ribs posteriorly. She had again severe dysphagia in the terminal stages and died on January 4, 1958.

Necropsy showed that the mass, a fibromyxoma, was constricting the oesophagus in its lower third and extended over the posterior half of the right diaphragm and was infiltrating the right chest wall.

CASE 5.—W. G. B., a man aged 56 years, developed "bronchitis," with cough, sputum, and shortness of breath, 10 months before his admission to the London Chest Hospital early in 1955. Later he had pain in his left shoulder. A radiograph (Fig. 10) showed a large irregular mediastinal opacity, projecting more to the left than to the right, and further radiological investigations demonstrated that the tumour compressed the left main bronchus and displaced the oesophagus.

First Operation.—Mr. T. Holmes Sellors operated on January 18, 1955. On opening the left side of the chest, a firm mass was seen projecting from the mediastinum below the arch of the aorta and the left main bronchus. The tumour was partially mobilized, but it could not be removed from the left side. The right side of the chest was therefore opened as well. The tumour, which projected from behind the pericardium, was intimately adherent to the lung and the oesophagus. Both these structures were injured in the course of the dissection, but eventually the tumour was removed in three pieces.

Dr. K. Hinson reported that the specimen consisted of a series of lobulated encapsulated tumours, with a total weight of 1,250 g. The cut surface was

fibrous with extensive areas of fatty change. In some of the small polypoid excrescences the tissue was completely adipose.

Microscopically the tumour was fibromatous, with marked oedema and fatty change. Ganglion cells were not present, but occasional nerve bundles were distinguishable. One small "polyp" showed fibrous tissue heavily infiltrated with lymphocytes and plasma cells. The tumour was a fibroma (possibly of nerve sheath origin).

The patient had a stormy convalescence but eventually made a good recovery. Three years later, after two months of chest pain, a radiograph showed recurrence of the growth on both sides of the chest (Fig. 11).

Second Operation.—Mr. T. Holmes Sellors operated on April 29, 1958. Through a right thoracotomy, a soft lobulated lipomatous fibroma about 15 × 10 × 10 cm. in size was enucleated from the posterior mediastinum.

Dr. K. Hinson reported that the specimen was a smooth, encapsulated tumour. Microscopically it was a fibroma with fatty changes. The nuclei were irregular and this tumour must therefore be considered malignant.

The patient made a good recovery from the operation, although a portion of the tumour still remained in the left chest. He died suddenly at home three months later. No post-mortem examination was carried out.

CASE 6.—H. B., a man of 35 years, had a radiograph of the chest taken in November, 1955, one week after he had noticed his sputum stained with blood. The film (Fig. 12) showed a large cyst with a fluid level in the right lung, and he was admitted to the London Chest Hospital.

First Operation.—Mr. J. R. Belcher operated on November 21, 1955. A large, thin-walled cyst containing 700 ml. of stale blood lay in the oblique fissure between the middle and the lower lobes. As it appeared to be an emphysematous bulla, only its lateral wall was excised, the medial wall being left attached to the lung surface to avoid air leak.

Dr. K. Hinson reported that the specimen was an emphysematous cyst with an unusual content of blood clot. Within the clot there were the ghosts of alveolar walls. Also with the specimen was a small separate fibroma.

The patient remained well until August, 1956, when he had repeated haemoptyses. A radiograph (Fig. 13) revealed a large rounded opacity at the site of the previous cyst. Aspiration of the chest yielded blood-stained fluid and some mucoid material.

Second Operation.—Mr. D. Watson operated on September 15, 1959. A friable growth in the pleura was diagnosed on frozen sections as a sarcoma. A pleuropneumonectomy with excision of the infiltrated sixth rib was carried out. There seemed to be no involvement of the regional lymph nodes.

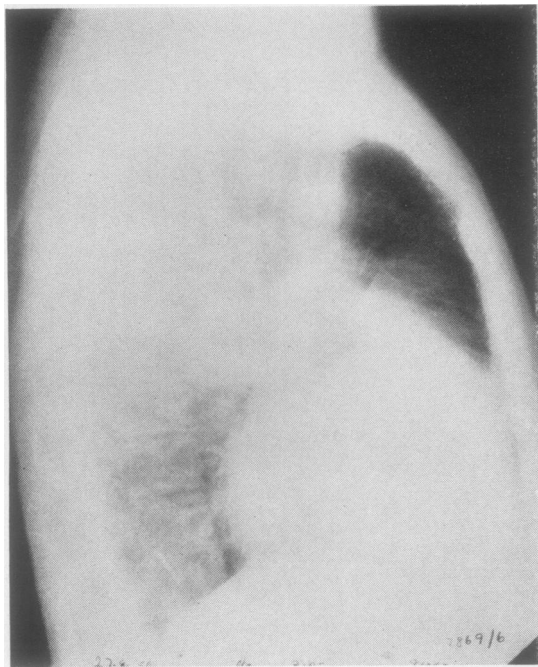


FIG. 13.—Case 6: Right lateral radiograph, one year after excision of cyst together with fibroma, showing large opacity at the site of previous cyst.

Dr. K. Hinson reported on the pleuropneumonectomy specimen in which the anterior basal part of the lung was replaced by necrotic growth which was infiltrating the overlying rib. Microscopically the tumour was an undifferentiated sarcoma of undetermined origin.

The patient remained well until December, 1956, when he noticed swellings along the operation scar. These increased in size, and when he was seen in February, 1957, there were four cystic swellings 0.5–5.0 cm. in diameter.

Third Operation.—Mr. D. Watson operated on March 4, 1957. Cystic metastases extending into the muscle layer were found in the thoracotomy scar. A wide excision, which included the seventh rib and the diaphragm underlying it, was carried out.

Dr. K. Hinson reported a recurrence of sarcoma.

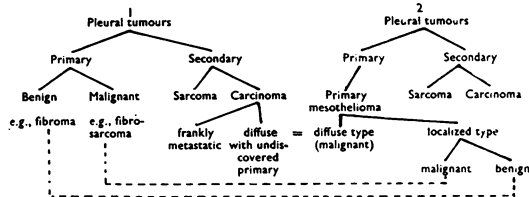
After this operation the patient was given radiotherapy, but he died three months later.

DISCUSSION

ORIGIN OF PLEURAL TUMOURS.—Every part of the body contains mesenchymal elements capable in theory of producing fibrous tissue tumours. In the thorax, with the exception of the intra-bronchial types, these growths are generally believed to arise in the pleura. The other view is that most of these tumours originate in the extra-

pleural connective tissue and that even the pedunculated types have merely acquired a pleural investment. The terms “intrathoracic fibroma” and “pleural fibroma” are used more or less synonymously, but here “intrathoracic” is preferred in that it does not prejudice a controversial issue.

Pleura consists of a single layer of flat serosal cells resting on a sheet of collagenous and elastic fibres with occasional fibrocytes and smooth muscle cells. In the subserosal layer there are also blood vessels, lymphatics, and nerve fibres. The whole structure, with the exception of the nerve fibres, is derived from mesoderm, and its primary tumours are classified accordingly. The view, represented largely by American authors (Klemperer and Rabin, 1931; Stout and Murray, 1942; Clagett, McDonald, and Schmidt, 1952), that all primary pleural tumours are “mesotheliomas” is an attempt to reconcile with mesodermal origin the frequent presence of epithelial tissue in the diffuse malignant tumour affecting the pleura. In their terminology, the fibrous tissue tumours are referred to as benign and malignant “localized mesotheliomas,” which correspond to fibromas and fibrosarcomas respectively. The claim by Stout and Murray (1942) to have shown by tissue culture that the diffuse and the localized tumours are both of mesodermal origin is far from convincing. In common with others (Willis, 1953; Smart and Hinson, 1957), who believe that nearly all the diffuse tumours are secondary to carcinoma elsewhere, we doubt if mesotheliomas exist as a separate entity. The localized mesotheliomas closely resemble mesodermal tumours in other parts of the body in appearance and behaviour, and there is nothing incongruous in calling them fibromas, fibrosarcomas, or lipomas. The opposing views are summarized in the diagram. Further discussion of



this controversy is not called for here, as, apart from tumour identification, it has little bearing on the subject of this paper.

PATHOLOGY OF FIBROMAS.—Fibromas are composed of collagen-forming fibroblasts and are

fairly well differentiated. Varying degrees of cellularity and collagen content are often present in different parts of these growths. In any one tumour there can usually be found all gradations and combinations from dense fibrous tissue to pleomorphic cellular areas with occasional mitotic figures. Metaplastic changes, usually of a mucinous or chondromatous nature, are common. Calcification and lipomatous degeneration also occur.

A well-defined capsule is an invariable feature. It is probably similar in nature to the capsule of any other simple tumour, although it has been suggested that it is derived from the tumour itself, being formed by its own fibroblasts. If this were the case, recurrence should be much more common, for neoplastic cells would be left behind every time a tumour was shelled out.

In the differential diagnosis, tumours of nerve origin provide the greatest difficulty. Stewart and Copeland (1931) presume that many fibrosarcomas, which are not obviously related to nerves, are nevertheless neurofibromatous in nature. The growths in neurofibromatosis, however, are not so well circumscribed and encapsulated, and they tend to infiltrate the surrounding tissues. On thorough microscopic examination, medullated or non-medullated nerve fibres are almost always found. Neurilemmomas and ganglioneuromas are less difficult to identify, recognition being assisted by palisading of the nuclei in the former and the presence of ganglion cells in the latter. The other tumour to be distinguished is the recurrent fibroma or desmoid tumour affecting the abdominal wall. These tumours contain muscle fibres and are not encapsulated. Such tumours occurring in the chest wall have been described (Desaive and Betz, 1954; Dolley and Brewer, 1943).

CLINICAL FEATURES OF INTRATHORACIC FIBROMAS.—The growth is usually silent, and not infrequently it is discovered unexpectedly in the course of routine medical or radiological examination. Sometimes clubbing of the fingers or the joint pains of hypertrophic pulmonary osteoarthropathy are the presenting symptoms (Clagett *et al.*, 1952; Thomas and Drew, 1953). Only occasionally, and then chiefly on account of its bulk, is the tumour responsible for symptoms referable to the chest. The rare intrabronchial fibromas which produce symptoms of bronchial obstruction, while they are still small, are exceptions. The correct diagnosis is seldom made before operation, when a large, sometimes lobulated, well-encapsulated tumour is found.

The growth is either attached by a narrow pedicle to any part of the visceral, parietal, or mediastinal pleura, or wholly or partly embedded in the lung or mediastinum. In the latter case there is no main attachment which might be described as the point of origin. Hilke and Konrad (1958) describe the sites of origin of the 166 cases they analysed as follows: Visceral pleura 68, parietal pleura 24, mediastinum 50, lung 10 (most of them intra-bronchial), and thoracic wall 14.

RECURRENCE OF INTRATHORACIC FIBROMAS.—Very few authors mention recurrence of these tumours after operative removal. Klemperer and Rabin (1931) describe one case where the tumour recurred four years later after inadequate removal. These authors suggest that surgical interference may result in malignant changes. Price Thomas and Drew (1953) felt that too limited a resection was the cause of recurrence in one of their cases. This was a pedunculated fibroma of the visceral pleura which had been under radiological observation for nine years. The growth was removed by division of its pedicle. Within two months there was radiological evidence of recurrence. The lung, containing numerous deposits of fibrosarcoma, had to be resected 12 months after the first operation. The pneumonectomy appeared to have checked the progress of the disease, for there was no further evidence of spread in the next five years. Ehrenhaft, Sensenig, and Lawrence (1960) described another case. Their patient was a young woman who had a "benign mesothelioma" removed two years previously. The second growth proved to be a "fibroblastic tumour within the lung parenchyma, consistent with mesothelioma." In the six years following resection of the lobe which contained it, there was no further recurrence. Four of the 24 cases of "localized fibrous mesothelioma" reported by Clagett *et al.* (1952) developed recurrence. In one of these there was "undeniable evidence of malignancy" in the original tumour, but in the other three the tumours were indistinguishable from those of the series which did not recur. Recurrence occurred in these after six months, four years, and eight years respectively. There is no mention whether these recurrent tumours were submitted to histological examination. Each was the ostensible cause of the patient's death. Stout and Himadi (1951) mention recurrences in three of their eight cases of "solitary (localized) mesothelioma of the pleura." But the original tumours were never classed as truly benign. They believe that pedunculated fibromas are benign, while the

intrapulmonary tumours with broad pleural attachment are histologically more anaplastic, less encapsulated, and therefore tend to behave clinically malignantly.

The diagnosis of malignancy, intimately bound to the question of recurrence, is not easy, because mesodermal growths and fibrous tissue tumours in particular are not sharply divided into the benign and the malignant. The areas of metaplasia, the pleomorphic cells, and the occasional mitotic figures seen in many simple fibromas may transform a benign histological appearance by imperceptible gradations into a malignant one. A seemingly "benign" fibroma can thus exhibit malignant properties, while a malignant-looking growth can behave quite innocently. Dolley and Brewer (1943) maintain that in all fibromas, which are slowly enlarging in the chest, sarcomatous changes have already taken place. Willis (1953) thinks that tumours showing prominently myxomatous changes are best regarded as malignant.

Recurrence of the tumour may therefore be due to one or several of the following factors: (a) The original tumour was not adequately removed. (b) It was potentially malignant, although histologically it was thought to be benign. (c) Malignant change has been provoked by some stimulus. (d) The tumour is one of multifocal origin, e.g., as in neurofibromatosis. (e) The recurrence is an overt metastasis, and this would therefore be indisputable evidence of malignancy in the original tumour.

ANALYSIS OF OWN MATERIAL.—Analysis of the cases yielded a number of interesting points.

Original Tumours.—Five of these were large and produced symptoms for which the patients sought medical advice. Two (Cases 2 and 3) were known to have been present for six and two years respectively before operation, having shown slow enlargement in the intervening periods. The one small tumour (Case 6) was an incidental finding at an operation on a blood-filled emphysematous cyst; it was also the only intrapulmonary tumour. In Case 2 the tumour lay between the lung and the diaphragm, adherent to both. There was only one tumour which had a pedicle (Case 1). In the remaining three (Cases 3, 4, and 5) the growth was embedded in the mediastinum.

Four of the tumours were removed intact in their capsules. In Case 5 the growth was removed in pieces, while in Case 6 the tumour appeared to be complete, but part of the cyst wall, in which it lay, was intentionally left behind.

Each was well encapsulated, with no signs of local extension or distant spread. No extrathoracic primary tumour was discovered in any of the patients during the years they were under observation.

The histology of all the original tumours, with the exception of Case 1, has since been reviewed and nothing has been seen which calls for alteration of the original opinion. Only in Case 5 remains some doubt whether the tumour was a pure fibroma, because of the presence of nerve fibres in its substance.

Recurrent Tumours.—First recurrences appeared nine months to 11 years after the original tumours were removed. All the patients were subjected to operation. Four of them (Cases 1–4) resembled their antecedent tumours and were also diagnosed as fibromas, but after comparatively short intervals three patients (Cases 2, 3, and 4) developed further recurrences which were indubitably malignant in Cases 2 and 3 and very likely so in Case 4. As the first recurrences in Cases 5 and 6 were sarcomatous, the number of patients developing ultimately frank malignant disease is five.

The only patient who is still alive (Case 1) had a pedunculated growth which recurred after 11 years; it again had a pedicle.

In Case 2 the first tumour had been present for at least six years and the recurrence took eight years to appear. This was slightly more active-looking than its predecessor, but it too was well encapsulated and regarded as benign, yet two and a half years later an irremovable fibrosarcoma extended from the site of the previous operation.

The course in Case 3 resembled the above, but was somewhat more rapid. In this instance the cellular fibroma had been demonstrated radiologically two years before removal. After five and a half years localized recurrences were found. These were again considered to be benign, although they were this time situated in the opposite lung and mediastinum. A year later the patient succumbed, the histological picture having changed to that of fibrosarcoma.

Case 4 may be in a different category, although we are regarding it as another example of malignant degeneration. The first tumour was a 4,550 g. fibroma with myxomatous changes. It was removed whole with relative ease. Two years later a large collection of mucinous material was found in the mediastinum. There was no longer a capsule present, the growth being confined by pleura only. The tumour, however, did not

contain malignant cells. Nevertheless the patient died a year later from another recurrence which this time was infiltrating the diaphragm and chest wall. Whether she really had a malignant tumour or whether the condition was more akin to pseudomyxoma peritonei could not be determined. It will be recalled that Willis (1953) tends to regard fibromas with prominent myxomatous degeneration as malignant.

The recurrence in Case 5 took place three years later and was histologically considered to be malignant on this occasion. The diffuse involvement of the mediastinum found at the first operation makes it likely that complete removal was not achieved. The presence of a few nerve fibres, moreover, raises the possibility that this was originally not a pure fibroma but a neurofibroma. He died three months after the second operation from an unknown cause.

The last patient (Case 6), who originally had only a small fibroma in an emphysematous cyst, died of a sarcoma. An almost identical case with similar outcome has been described by Storer and Hooper (1960). There was nothing in the histological appearance of the fibroma to suggest a tumour of high malignancy. The haemorrhagic effusion and the appearance of a sarcoma at the site within a few months make one wonder if an area of malignancy was overlooked in that part of the cyst wall which was left behind.

SUMMARY

Intrathoracic fibromas are usually regarded as benign tumours and recurrences are rarely mentioned. Six cases are reported in which recurrences developed after "benign" fibromas had been removed. Some of these occurred late, but death was not long delayed in five of the patients after removal of the recurrence. In every case it was due to sarcomatous change. A brief review of the relevant facts and opinions about these tumours is given, followed by an analysis of our own cases with particular reference to the problem of determining whether the original tumours were in fact benign.

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