TRACHEO-BRONCHIAL AND PULMONARY CHONDRO-ADENOMA (HAMARTOMA)

BY

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A discrete pulmonary lesion composed mainly of cartilage was described by Lebert in 1845. In 1934 Goldsworthy, from this hospital, pointed out the constant occurrence of epithelial elements in this lesion and proposed the name "hamartoma chondromatosum pulmonis," which has gained wide acceptance. The condition long remained a pathological curiosity, but the wider application of chest radiography has led to the discovery of an increasing number of cases. Ten cases have been observed in this hospital from 1954 to 1957; in the same period 58 resections for bronchial carcinoma have been performed.

The natural history of these lesions is not well known, since they are either incidental findings at necropsy or, when discovered in life, are promptly excised. In reviewing the cases of this thoracic unit it was noted that in several there was evidence of continued growth during the adult life of the patient. The lesions therefore are truly neoplastic, and the term "hamartoma" seems hardly applicable. Following the suggestions of Adams (1957), the term "chondro-adenoma" is used in this paper.

Most of these tumours occur in the lung parenchyma, but a few occur within the bronchial tree, and present the clinical features and pathological sequelae of bronchial obstruction.

The present series comprises eight examples of pulmonary chondro-adenoma and two occurring within the bronchial tree.

Pulmonary Chondro-adenoma

The significant features of pulmonary chondro-adenoma are summarized in the accompanying Table.

Details of Cases

Case No.	Age	Site	Operative Procedure	Diameter in cm.	Remarks
1	56	Lingula	Enucleation	1·5	
2	50	Middle lobe	Lobectomy	2·0	No change observed in 5 years
3	48	Right upper lobe	Enucleation	1·0	
4	56	Left lower lobe	Lobectomy	2·0	Clear *x*-ray film 8 months before
5	50	Right upper lobe	,,	2·0	Chest clear 7 months before, calcified on section
6	16	,, ,, ,,	Enucleation	2·5	Grew from 1·9 cm. in 14 months (Figs. 1 and 2)
7	59	Middle lobe	Nil	1·0	Present 8 years, slight growth. Angina pectoris
8	64	Left upper lobe	Enucleation	1·8	

All patients were male. There was no operative morbidity or mortality.

Pathology

The tumour may occur in any lobe, but a study of published cases gives the impression they are commoner in the lower half of the chest. Although the tumours vary in size from a few millimetres in diameter (McDonald, Harrington, and Clagett, 1945) to a mass nearly filling the hemithorax (Jackson, McDonald, and Clagett, 1956), the majority are 1 to 3 cm. in diameter. They are slightly lobulated, and are so sharply demarcated from the surrounding lung that it is difficult to cut the lung without dislodging the tumour from its bed.

Inspection of the cut surface gives an indication of the heterogeneous composition—pale-blue translucent cartilage, with areas of yellow fat and pink fibrous tissue. Although the surrounding lung may be heavily anthracotic, the tumour is free of pigment. Microscopically the tumour consists of rounded masses of cartilage, surrounded by a framework of connective tissue through which are scattered clefts of epithelium—cubical, flattened, columnar, or ciliated. In addition, varying amounts of fat, and less commonly lymphoid tissue, may be seen. Calcification is not uncommon, and ossification may also occur.

Although in the majority of these tumours cartilage is the dominant non-epithelial structure, adipose tissue may be prominent as in the case of Brewin (1952) and Case 9, described below, or fibrous tissue as in the cases reported by Scarff and Gowar (1944).

The existence of true chondromata—that is, devoid of epithelial elements—although extremely rare, must be admitted. Examples are those of Edling (1938) and Davidson (1941).

Clinical and Radiological Features

Chondro-adenomata are found at least three times more often in males. They usually occur during middle life; Case 6 is one of the youngest reported. I can find no record of a case in childhood.

Occasionally large tumours may cause slight discomfort. The vast majority are asymptomatic. Radiological examination shows a solid, sharply demarcated, slightly lobulated mass, with no change in the surrounding lung. Punctate calcification may be present, but often requires tomography for its detection.

Evidence of Growth

At least one of these tumours (Case 6) was observed to grow for over a year (Figs. 1 and 2), increasing in diameter from 1.9 to 2.5 cm. Two other cases are said to have had clear *x*-ray films previously, but these were not available for inspection, and the abnormality may have been overlooked. In the remaining case where a previous film is available there appeared to be some increase, but this could be due to technical factors.

Similar observations have been made by others, including Adams (1957) and Good, Hood, and McDonald (1953). The latter reported 25 hamartomas, in five of which a previous chest radiograph had been normal, and one in which the lesion was observed to grow.

Diagnosis and Treatment

The differential diagnosis includes bronchial carcinoma, hydatid cyst, tuberculoma, and metastatic malignant disease. A certain diagnosis may only be made by histological examination, and, since excision is attended by negligible risk, this is the course usually followed. A guide to diagnosis may be given by the presence of calcification, by the rate of growth, and by the age of the patient. Laminated calcification is observed only in granulomata. Punctate calcification occurs in simple tumours and inflammatory conditions, and, more rarely, in large carcinomas.

Although carcinomas may grow slowly, especially in their early stages (Rigler, 1957), a lesion known to have

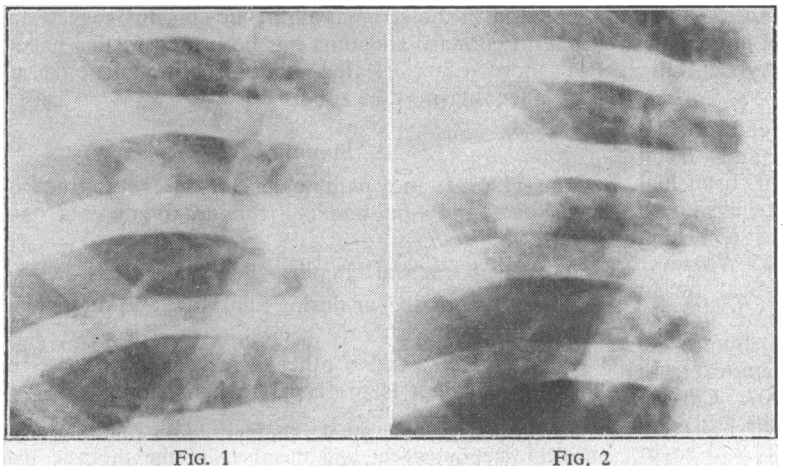

FIG. 1 FIG. 2

FIG. 1.—Case 6. Chondro-adenoma of lung. FIG. 2.—Case 6. The same chondro-adenoma, showing growth in 14 months.

grown should be regarded as malignant. Finally, carcinomas are unusual below the age of 35.

The alternative to excision is careful and repeated observation, which may be irksome to the patient, who may become introspective or who may default. Finally, the presence of an abnormal shadow may be a bar to employment or life insurance. The decision must be made after consideration of all these facts.

A pre-operative diagnosis may be confirmed by the findings at thoracotomy. The tumour is extremely mobile within the lung, and recalls the sensation of a fibro-adenoma of the breast. If confidently diagnosed, enucleation may be performed and the wound in the lung oversewn with catgut. On the other hand, vigorous palpation of a carcinoma is to be avoided.

Thus in Cases 4 and 5, both middle-aged men said to have had a recent normal chest *x*-ray film, the draining vein was ligated and a lobectomy performed because of the strong probability of carcinoma.

Tracheo-bronchial Chondro-adenoma

Case 9

A cane-cutter aged 57 had suffered recurrent attacks of fever, with slight discomfort in the right side of the chest for five years. On admission he had a temperature of 102° F. (38.9° C.), and clinical and radiological signs of atelectasis of the right lower lobe. Bronchoscopy showed a smooth pale tumour in the right lower lobe bronchus, which was removed endoscopically piecemeal. The lobe failed to re-expand, and lobectomy was recommended, but the patient refused and was lost to the follow-up. Histological examination showed fat and epithelium.

The following case of a tracheal hamartoma is reported in detail because of its rarity.

Case 10

A clerk aged 50 was admitted urgently to the thoracic unit of this hospital. While on active service in 1942 the men in his tent had complained that he wheezed loudly while sleeping. His wheeze worsened slightly over the next fourteen years, but he had become so accustomed to it that it worried only the people around him. At their insistence he consulted several practitioners, who treated him for bronchial asthma. As treatment had no effect on the wheeze, and his activity was not limited, he became quite indifferent to it. A few weeks before admission the wheeze became worse and a tracheal tumour was diagnosed on bronchoscopy. A portion of the tumour was removed without change in the clinical state, but a second attempt at endoscopic removal produced severe respiratory obstruction, and the patient nearly died. After 48 hours his condition improved slightly and he was transferred to this unit.

Dyspnoea, orthopnoea, stridor, and rib recession were still present on admission, but as his condition was improving operation was deferred until a full assessment could be made. Plain radiographs of the chest showed evidence of a tumour in the trachea about its middle; the lung fields were normal. Tomography showed the lesion more clearly. A histological report of the tissue removed indicated that cartilage and epithelial elements were present, and the nature of the tumour was suspected. Bronchoscopy was deferred until immediately before the thoracotomy lest obstruction recur.

At operation, bronchoscopy was performed under local analgesia. A smooth, pale, slightly lobulated tumour was seen in the middle of the trachea and almost occluding it. It was sessile and based on the right postero-lateral quadrant of the trachea. It seemed quite impossible to introduce any tube past the obstruction, and it was felt that intubation of the trachea above the obstruction might cause "trapping" of gas beyond it.

A low collar incision was made low in the neck under local analgesia. The isthmus of the thyroid was divided and the trachea opened. With mobilization of the trachea and gentle traction several centimetres of trachea were drawn into the neck. The anterior incision in the trachea was prolonged down under cover of the manubrium to the middle of the tumour. With the extra room thus afforded a Magill tube was then passed through the glottis and guided into the left main bronchus.

General anaesthesia was then induced. The manubrium was split and the incision carried out into the second right interspace. The innominate artery and the left innominate vein were mobilized and drawn down. The tracheal incision was further extended almost to the carina. The mucous membrane surrounding the tumour was incised, and the tumour removed by submucous dissection. Little bleeding occurred. The trachea was closed with steel wire, leaving a Colledge tracheostomy tube in the upper part, and the right chest was closed, with drainage. Accumulation of blood and secretions caused some anxiety for 48 hours, but the tracheostomy tube was removed on the third day and the patient was discharged on the twentieth day. A portion of the resected tumour is shown in Fig. 3.

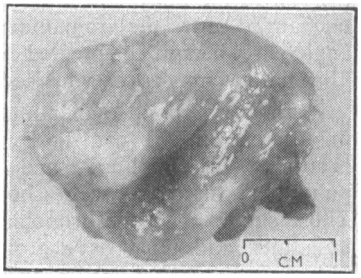

FIG. 3.—Case 10. Chondro-adenoma of trachea after excision.

Endobronchial Chondro-adenoma

Pathology.—The endobronchial type is much less common than the parenchymal. Paterson (1956) found 29 cases recorded and added another. The review is not exhaustive and does not include, for example, four cases described by Price Thomas (1954), or a lipo-adenoma described by Brewin (1952). These tumours have been described in the main bronchi and all lobar bronchi. I have been unable to find a previous report of a case

of chondro-adenoma occurring in the trachea. The histological identity of the endobronchial and pulmonary types has been shown by Sutherland, Aylwin, and Brewin (1953).

Diagnosis and Treatment.—The features of bronchial obstruction in adults—cough, wheeze, and distal atelectasis—must give rise to suspicion of bronchial carcinoma. The bronchoscopic discovery of a smooth pale tumour and a histological report of a benign tumour come as a welcome surprise. Whatever objections may be urged against the removal of an asymptomatic pulmonary lesion, there can be none to operative relief of an obstructed bronchus. Before the wide use of thoracotomy, endoscopic removal was practised; Eicken first performed this in 1907. Carlsen and Kiaer (1950) removed a tumour from the left main bronchus in 1945, and Postlethwait, Hagerty, and Trent (1948) performed a left lower lobectomy for a chondro-adenoma of the left lower lobe bronchus with distal abscess. Excision of the tumour by bronchotomy is preferable to bronchoscopic removal, being easier, safer, and more precise. If the distal lung tissue has sustained irreversible changes it is removed also. The tracheal tumour described above presented unusual and difficult anaesthetic problems, and the means used to overcome them are rarely applicable.

Nomenclature

The name of this tumour has given rise to some difficulty. The word "hamartoma" has been widely used, but has been strongly criticized by Adams (1957).

As originally defined by Albrecht (1904), a hamartoma is a tumour-like malformation (geschwulstartige Fehlbildung). Although Albrecht suggested that benign tumours—for example, fibro-adenoma of the breast—should be included in this class, it would appear preferable, following Willis (1953), to confine the term to a lesion which "is present at birth; grows only with the rest of the body . . . after growth ceases it remains unchanged for the remainder of life." Such are angiomas, benign pigmented moles, and the neuro-fibromatous masses of von Recklinghausen's disease.

Pulmonary chondro-adenomata do not conform to this definition. I can find no record of any case occurring in childhood, and some at least of the pulmonary tumours can be shown to grow (see above). Those occurring in the bronchial lumen occur also only in adults, and must have grown after growth of the bronchus has ceased. Admittedly growth must be slow: both cases here reported have a history of obstruction over many years.

The cases in the newborn reported by Jones (1949) and by Graham and Singleton (1953) are quite distinct from chondro-adenoma, and it would be preferable to restrict the term "pulmonary hamartoma" to these lesions.

The term "mixed tumours of the lung" was used by Möller (1933) and by Ehrenhaft and Womack (1952), who also included bronchial adenomas in this class. These latter, however, are structurally distinct, and malignant change is well known. In chondro-adenoma malignant change is almost unknown, although Simon and Ballon (1947) describe such a case.

Adenochondroma was suggested by Brewer, Brookes, and Valteris (1953). Adams prefers to confine this to bronchial adenomas with marked chondromatous metaplasia in the stroma, and, following his suggestion, the term chondro-adenoma has been used in this paper. For those other related tumours, lipo-adenoma or fibro-adenoma may be used.

Summary

The clinical and pathological features of tracheo-bronchial and pulmonary chondro-adenomata are described.

A case of tracheal chondro-adenoma is recorded.

These tumours occur during adult life, and may grow, although slowly.

For these and other reasons, the term chondro-adenoma is preferred to hamartoma.

I acknowledge with thanks the permission of the general medical superintendent and members of the thoracic staff to publish these cases. Mr. Ian Monk has given great encouragement in the preparation of this paper. Miss M. Simpson, the hospital photographer, is responsible for the illustrations.

ADDENDUM.—Since submitting this paper further observations on the growth of these tumours have appeared in a paper, "Growth Conditions of Hamartoma of the Lung," by K. G. Jensen and T. Schiødt, in *Thorax*, 1958, **13**, 233.

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Paediatrics for the Practitioner, edited by Wilfrid Gaisford and Reginald Lightwood, appeared in 1953–5 (3 vols.). Two supplements (for 1956 and 1957) have already appeared, and a third, for 1958, is now available (Butterworth and Co. (Publishers) Ltd., 140 pp.; 35s.). This comprises original articles on chemotherapy, behaviour disorders, urology, chromatography, and electroencephalography, followed by a short section containing notes on recent advances in paediatrics, and amendments bringing the main volumes up to date.