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Giant fibromyxoma of the parietal pleura

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Nwafo, D C, and Adi, F C (1978). Thorax, 33, 520–523. Giant fibromyxoma of the parietal pleura. A 23-year-old Nigerian man with a giant (8·5 kg) benign intrathoracic fibromyxoma presented in respiratory distress with what looked clinically and radiologically like a massive left-sided pleural effusion. The correct preoperative diagnosis was suggested by an almost 'dry tap' together with a solid resistance to the aspirating needle at chest aspiration. Complete removal was successfully accomplished through an extended left thoracotomy. The pathology, clinical features, and treatment of localised pleural fibromas are briefly discussed.

Primary pleural neoplasms are exceedingly rare. The tumours are now widely regarded as mesotheliomas of which two varieties are recognised—a 'diffuse malignant' and a 'localised fibrous' mesothelioma (Clagett et al., 1952). Owing to earlier confusion as to their origin, the localised fibrous variety has been previously described by a multiplicity of names, including intrathoracic fibroma, pleural fibroma, benign localised mesothelioma, and solitary localised mesothelioma. We will use the term intrathoracic fibroma.

A common, though not widely recognised, feature of localised fibrous intrathoracic fibromas is their tendency to recur after resection (Klemperer and Rabin, 1931; Clagett et al., 1952; Ehrenhaft et al., 1960; Kerr and Nohl, 1961). Dolley and Brewer (1943) maintain that all slowly growing intrathoracic fibromas have already undergone malignant change. According to Willis (1953), those tumours in which pronounced myxomatous degeneration has occurred are best regarded as frankly malignant. We report here a typical intrathoracic fibromyxoma that is not only remarkable for its size (8·5 kg) but also in retaining its 'benign' characteristics despite its long history and extensive myxomatous change. Moreover, it illustrates nearly all the features usually associated with an intrathoracic fibroma. So far as we are aware there has been no previous report of a similar case from West Africa.

Case report

A 25-year-old Igbo mechanic gave a four-year history of progressive shortness of breath associated, in the last five months, with a dry unproductive cough and precordial pain. There were occasional febrile episodes and joint pains, but no haemoptysis. He had been treated at another hospital for orthopnoea and swelling of the legs without much improvement. In 1972 he had also been treated for shortness of breath and transient swelling of the legs.

On arrival at the University of Nigeria Teaching Hospital, Enugu (UNTH), he was orthopnoeic and too short of breath to talk for more than a few minutes. His fingers were clubbed. The neck veins were engorged, almost to the mandible. The trachea was markedly deviated to the right and the apex beat located in the sixth right intercostal space at the anterior axillary line. The left hemithorax was stony dull to percussion with absent breath sounds. There were scattered rhonchi over the right hemithorax, but breath sounds were otherwise normal. The spleen and liver were enlarged three finger-breadths below the costal margin. The chest radiograph (Fig. 1) suggested a massive left-sided pleural effusion, but aspiration yielded only 100 ml blood-stained fluid before a solid resistance was encountered with the aspirating needle. The aspirate contained no malignant cells. At bronchoscopy the trachea was compressed anteroposteriorly and markedly deviated to the right. The left main bronchial orifice could not be identified. Pleural biopsy showed inflammation only. At thoracotomy (1 August 1977), an enormous solid, lobulated, and well-encapsulated tumour weighing 8·5 kg (Fig. 2) filled the left chest, compressing the left lung into a small crescent at the apex and pushing the
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Fig. 1 Radiograph showing giant left pleural tumour causing gross mediastinal displacement to right.

Fig. 2 Solid, lobulated, and well-encapsulated fibromyxoma completely removed at thoracotomy (weight 8.5 kg).

Heart into the right hemithorax. The pleura was considerably thickened with large veins coursing over it, and it contained a large quantity of altered blood. Despite a wide thoracotomy only piecemeal removal was possible. This permitted full and sustained re-expansion of the left lung. Recovery was uneventful.

Histologically, the thickened pleura showed only nonspecific inflammation. The tumour itself was a fibromyxoma reported on as follows: 'The pattern is benign, there being no mitotic figures. In parts, there are decussating sheets of connective tissue. Elsewhere a myxomatous appearance is noted (Figs. 3 and 4). The alternative diagnosis of a solid mesothelioma is to be borne in mind, as the latter tumour is occasionally solid, simulating a fibromatous growth'.

Discussion

This case illustrates nearly all the features generally attributed to a benign localised intra-thoracic fibroma. Macroscopically, the tumours are well encapsulated and lobulated. Some are embedded in the lung or mediastinum; others may be pedunculated, and attached to some part of the pleura—visceral, parietal, or mediastinal. As in the present case the cut surface may show a whitish and whorled appearance.

Microscopically, the tumours are composed of collagen-forming fibroblasts. The same tumour may show in different parts a varying degree of cellularity and collagen formation. Metaplastic change—myxomatous, chondromatous, or lipo-
Fig. 3  Note spindle-shaped and stellate cells separated by myxomatous stroma (Haematoxylin and eosin ×120).

Fig. 4  High power view showing cells with some nuclear pleomorphism floating in myxomatous stroma (H & E ×360).
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matous—is common. Calcification, though not noted here, may occur.

 Clinically, intrathoracic fibromas may grow silently for a long time and are not infrequently recognised on routine chest radiography. In the present patient distressing symptoms appeared for the first time five months before admission, although the tumour had been present for at least four years—when the patient first observed shortness of breath and peripheral oedema.

 Sometimes finger clubbing and the joint pains of hypertrophic pulmonary osteoarthropathy are the presenting symptoms (Clagett et al., 1952; Thomas and Drew, 1953). Respiratory symptoms, notably shortness of breath, may as in this patient arise from the bulky tumour interfering with pulmonary expansion or causing mediastinal displacement. The peripheral oedema noted in this patient as well as the hepatosplenomegaly disappeared after removal of the tumour, and they probably resulted from impaired venous return caused by the bulky tumour.

 Episodes of chills and fever may occur (Ehrenhaft et al., 1960)—a notable feature in this patient. Rarely the patient may present with recurrent pleural effusion as in the cases reported by Ehrenhaft et al. (1960) and by Aghina and Boscaino (1956). The latter patient had had a recurrent pleural effusion for six years.

 Haemorrhagic effusions may occur, as in our patient. He had had no previous chest aspiration, and as the pleural fluid contained altered rather than fresh blood, it seems unlikely that the haemorrhage was due to needling. A haemorrhagic effusion signifies neither inoperability nor an unfavourable prognosis (Clagett et al., 1952; Heaney et al., 1957). It is also unusual to recover malignant cells from such effusions. While there are no diagnostic radiographic appearances, a nearly 'dry tap' in the presence of a massive 'pleural effusion' especially if the aspirating needle encounters a solid resistance, suggests a solid intrapleural tumour. We had previously observed this in a patient who was moribund on admission with 'a massive pleural effusion' which was found at necropsy to be a solid intrapleural fibroma. The same finding led to a correct pre-operative diagnosis in the present patient.

 Despite the piecemeal removal of the tumour in our patient, excision was probably complete since the tumour was sharply circumscribed. This view is supported by the histological absence of tumour tissue in the adjacent pleura. A longer follow-up is needed to obtain a reasonable idea of the prognosis. However, many rural Nigerians have to travel long distances to hospital and see little point in attending for follow-up except for distressing symptoms. Our patient has so far attended regularly—and remained clinically and radiologically symptom-free. As Stout and Himadi (1951) pointed out 'the pleomorphism, frequent mitosis, and other histological criteria associated with malignancy do not appear to affect the good prognosis of most patients'.

 References


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