Case report

Ankylosing spondylitis and adenocarcinoma of the lung

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SUMMARY A man with long standing and severe ankylosing spondylitis, treated with radiotherapy, developed a primary adenocarcinoma of the lung in an area of apical fibrosis. The significance of this is discussed and the literature reviewed.

Apical pulmonary fibrosis occurs in patients with ankylosing spondylitis.\textsuperscript{1-4} Adenocarcinoma of the lung and alveolar cell carcinoma are bronchial neoplasms often associated with fibrotic tissue.\textsuperscript{5} The occurrence of adenocarcinoma of the lung arising from an area of apical pulmonary fibrosis in a man with ankylosing spondylitis treated with radiotherapy is therefore unlikely to be fortuitous. We draw attention to this association, anticipating that more cases will be reported.

Case report

A 57-year-old man with ankylosing spondylitis was admitted for physiotherapy. Low back pain had begun in 1944 at the age of 21. In 1948 he had a course of radiotherapy with exposure of 2000 rads each to the neck, upper thorax, lower thorax, lumbar spine, and sacroiliac joints. In 1950 he had a further 500 rads to the thoracic spine, which was repeated in 1952 and 1953. He had been treated with phenylbutazone 300 mg a day and indomethacin 100 mg a day. He smoked 30 cigarettes a day for 39 years but had no respiratory symptoms. Clinical examination showed cachexia with loss of normal lumbar lordosis and restriction of movement of the lumbar and cervical spine. Examination of the respiratory system revealed bilateral fine basal crepitations. There was no clubbing of the fingers. Examination of the cardiovascular and central nervous systems and gastrointestinal tract was normal.

Chest x-ray showed a reticulonodular pattern in both bases with bilateral small pleural effusions and enlargement of the right hilum (Fig. 1). A previous chest x-ray in 1976 was normal. Lung function tests showed a combined obstructive and restrictive pattern. Serum alkaline phosphatase was 1490 IU/l (normal 21–92 IU/l), gamma glutamyl transpeptidase 87 IU/l (normal <50 IU/l). The alkaline phosphatase was mainly hepatic in origin but some bone and bile isoenzyme was also present. The serum amylase was 1127 IU/l (normal 70–300 IU/l). Renal function was normal. Peripheral blood film was initially normal but later he developed a leucoerythroblastic anaemia. Two weeks after admission he developed a massive pulmonary infarct and died.

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Fig. 1 Chest x-ray. Reticulonodular pattern at both lung bases with bilateral pleural effusions and enlargement of the right hilum.
Post-mortem examination showed that both pulmonary arteries were obliterated by ante-mortem thrombus. There was a tumour measuring 15 × 20 mm at the periphery of the upper lobe of the right lung with scarring of the overlying pleura. In addition diffuse tiny white nodules were scattered throughout both lungs, the liver, the kidneys, and the grey matter of both parietal lobes. Enlarged lymph nodes were present in the right hilum and porta hepatitis. Histological examination showed adenocarcinoma, and of the tumour from the right upper lobe an adenocarcinoma arising from an area of apical scarring (Fig. 2). There was no evidence of pulmonary tuberculosis. Lymph nodes from the porta hepatitis and right hilum showed metastatic tumour deposits.

Discussion

Apical pulmonary fibrosis is associated with ankylosing spondylitis. Post-mortem examination showed that both pulmonary arteries were obliterated by ante-mortem thrombus. There was a tumour measuring 15 × 20 mm at the periphery of the upper lobe of the right lung with scarring of the overlying pleura. In addition diffuse tiny white nodules were scattered throughout both lungs, the liver, the kidneys, and the grey matter of both parietal lobes. Enlarged lymph nodes were present in the right hilum and porta hepatitis. Histological examination showed adenocarcinoma, and of the tumour from the right upper lobe an adenocarcinoma arising from an area of apical scarring (Fig. 2). There was no evidence of pulmonary tuberculosis. Lymph nodes from the porta hepatitis and right hilum showed metastatic tumour deposits.

Apical pulmonary fibrosis is associated with ankylosing spondylitis. Initially it was thought to be due to previous pulmonary tuberculosis, but it is now believed to be due to the disease process itself. Patients may be asymptomatic or may have a productive cough, dyspnoea, and occasionally haemoptysis. Cavitation may occur with colonisation by aspergilli. The histological picture is nonspecific apical fibrosis which may obliterate the alveoli, and infiltration with chronic inflammatory cells, predominantly lymphocytes. This patient developed an adenocarcinoma in an area of apical pulmonary fibrosis. Bronchogenic carcinoma arising from apical scars has previously been described. Such scars may be inconspicuous, the first evidence of neoplasia being metastasis or non-metastatic extrapulmonary manifestations. To our knowledge bronchial carcinoma occurring in an area of apical fibrosis associated with ankylosing spondylitis has not been previously reported. This is probably due to the infrequent association of pleuropulmonary manifestations with ankylosing spondylitis.

The development of adenocarcinoma may have been influenced by 2 further risk factors. Firstly, heavy cigarette smoking is correlated with the development of bronchial carcinoma. This was formerly thought to apply particularly to the squamous cell and oat cell types, but there is increasing evidence that adenocarcinoma is also induced. The second factor is the exposure to radiation. Patients with ankylosing spondylitis have a higher mortality than the general population. However, the patterns of mortality change with radiation exposure. In the nonirradiated patients mortality is increased from gastrointestinal disease, accidents, suicide, cerebrovascular, and other circulatory diseases. In the irradiated patients there is not only an increased incidence of acute leukaemia but also an increased incidence of solid tumours at heavily irradiated sites, including bronchial carcinoma.

Adenocarcinoma of the lung tends to occur peripherally and often arises in association with scar formation in the lung. Both adenocarcinoma of the lung and alveolar cell carcinoma can spread by implanted seedlings within the bronchial tree, which would account for the abnormal radiographic appearance in this case. It is most likely that involvement of adjacent structures to the pancreas, namely,
lymph nodes, were responsible for the increased amylase secretion, as there was no macroscopic or microscopic evidence of pancreatic infiltration. However, secretion from the adenocarcinoma of the lung is not excluded.

The occurrence of adenocarcinoma or alveolar cell carcinoma arising in areas of apical pulmonary fibrosis in patients with ankylosing spondylitis is obviously important, particularly in those patients who have been treated with radiotherapy. We suggest that this occurrence is a rare complication of longstanding ankylosing spondylitis and that clinicians need to be aware of its existence.

References
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