Endobronchial teratoma

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Intrathoracic teratomata occupying an endobronchial position have been described on only three previous occasions. A further example of this condition is reported and the clinicopathological features discussed.

Case report

The patient, a 5-year-old girl (AM), was admitted to hospital in June 1963, with a one-week history of nausea, fever, and back pain. Physical examination was unremarkable except for evidence of left upper lobe consolidation. A chest radiograph demonstrated diffuse opacification of the left upper lobe with a small area of perihilar calcification. The haemoglobin was 11.2 g/dl, WCC $17 \times 10^9/l$ (70% neutrophils) and the ESR 70 mm/hr. Sputum, urine, and gastric washings were negative for tubercle bacilli on smear, culture, and guinea pig inoculation. The Mantoux test was strongly positive after recent elective BCG inoculation. Bronchoscopy was normal. Pulmonary tuberculosis was diagnosed and treatment started with PAS and isoniazid. This was continued for several months, during which the patient made a slow recovery in hospital. The peripheral radiological changes cleared but a perihilar opacity persisted, suggesting a localised mass. Chemotherapy was eventually discontinued because of a drug rash and the patient was discharged in December 1963. Subsequently, she remained well but the radiographic opacity persisted.

In May 1980 the patient was readmitted (aged 22 years) with a six-week history of pleuritic chest pain, cough, and haemoptysis. Physical examination revealed diminished air entry in the left upper zone but no other abnormality. The previous opacity was still present in the anterior segment of the left upper lobe, with some associated calcification (fig 1). Routine laboratory investigations were normal, the Mantoux test was negative, and no tubercle bacilli were demonstrated on sputum examination. At thoracotomy (£29,580), the left upper lobe was densely adherent to the anterior chest wall and the anterior segment contained a firm mass. Upper lobectomy was performed and the patient recovered uneventfully.

Pathological findings

The anterior segmental bronchus was grossly distended

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Fig 1 Chest radiograph before operation demonstrating left perihilar opacity with foci of calcification

with a $5 \times 4 \times 2.5$ cm smooth, white, polypoid tumour projecting into its lumen. Several coarse hairs protruded from the surface of the lesion (fig 2). The lung parenchyma was congested, with pus evident in some of the smaller bronchi.

Microscopically the appearances were those of a benign teratoma. The tumour had a covering of stratified squamous epithelium with hair follicles, sebaceous and sweat glands. This covering merged with the respiratory epithelium at the site of attachment to the wall of the bronchus. Several crypts within the tumour were lined by well-differentiated intestinal epithelium, and within the substance of the tumour it was possible to identify areas of cartilage, bone, smooth muscle, pancreatic tissue, and neural tissue. There were also several small areas of thymic tissue with recognisable Hassall's corpuscles (fig 3).

Discussion

The clinical features of this case show many similarities to the three previously reported endobronchial teratomata.¹⁻³ All have come to diagnostic surgical intervention in early adult life, the principal symptoms being

Fig 2 Left upper lobe incised to display the teratoma within the dilated segmental bronchus. Hair can be seen arising from the covering epithelium.

Fig 3 Focus of thymic tissue with Hassall's corpuscles (H and E $\times 100$).

chest pain, cough, and haemoptysis, with one patient also noting the expectoration of hair.¹ Generally there has been an interval of some years between presentation and diagnosis, the usual initial diagnosis being of an infective or inflammatory lesion. In none of the previous examples, however, was the age at presentation as young or the period of observation as long as in the present case. This indicates the very slow growth of these lesions, since successive radiographs demonstrated little increase in the dimensions of the mass over 17 years. This tendency to very slow enlargement is also suggested by the pronounced bronchial dilatation which accommodated the growing tumour and by the absence of lobar collapse which is frequently seen with other endobronchial tumours.

Pathologically these lesions resemble other intrapulmonary and mediastinal teratomata in that the constituent tissues represent all three germinal layers. Pancreatic tissue has been a frequent finding,² and thymic tissue, detected in the present case, has been present in previous examples of intrapulmonary teratomata, including the first reported intrabronchial lesion.^{1,4,5} This had led to the suggestion that these teratomata may originate during faulty thymic embryogenesis⁶ with displacement of embryonic thymic tissue within the developing lung bud.^{4,7} The fact that nearly half of all intrapulmonary teratomata, including three of the now four endobronchial lesions, have arisen in the adjacent left upper lobe would accord with this theory.⁸ The reported endobronchial teratomata have all been benign, but malignant change in situ has been noted in intrapulmonary teratomata, although metastatic behaviour has been clearly described on only one occasion.⁷

There is no definitive means of diagnosing such lesions before excision, though the history and presence of a longstanding radiological opacity may be helpful. The dilated bronchus with its intraluminal mass may result in a crescentic translucent area within the radiological opacity,² a similar radiological appearance being seen on occasion in cystic intrapulmonary teratomata. Calcification caused by ossification may be present⁴ but is also seen in other conditions. On only one occasion has such a tumour been visualised bronchoscopically and even then the resultant biopsy was unhelpful.³ It is therefore likely that such lesions will continue to be diagnosed by the pathologist rather than the clinician. In view of the troublesome symptoms, the associated chronic distal infection and the possibility of malignant change, surgical excision would seem to be the treatment of choice even in the unlikely event of a firm preoperative diagnosis.

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