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Case Report: Tracheobronchopathia Osteochondroplastica

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Tracheobronchopathia osteochondroplastica is a rare benign condition characterized by multiple cartilaginous or osseous submucosal nodules that project into the tracheobronchial lumen. It is infrequently diagnosed on projectional radiography and is usually discovered incidentally at post mortem. We identified a case of tracheobronchopathia osteochondroplastica complicated by right middle lobe collapse and post-obstructive bronchiectasis in which the diagnosis was established by thoracic computed tomography (CT) and confirmed by fibrooptic bronchoscopy.

CASE REPORT

A 74-year-old man with a 15 cigarettes-per-week smoking history presented with a chronic cough of several months duration productive of blood-tinged sputum. The patient had no known allergies. On physical examination, audible expiratory wheezes were present bilaterally with markedly decreased breath sounds on the right. There was no cyanosis clubbing, or palpable adenopathy. The patient was treated with inhaled corticosteroids and β -2 stimulant therapy without significant effect. A chest radiograph revealed irregular narrowing of the intrathoracic trachea, complete right middle lobe collapse, and patchy consolidation of the right lower lobe. Branching tubular soft tissue opacities in the medial right base were suspicious for bronchiectasis and mucous plugging. Incremental thoracic CT (GE Hilight Advantage, GE Corporation, Milwaukee, WI) with intravenous contrast was performed using 10-mm collimation (5-mm collimation through the mediastinum), followed by high-resolution thin sections (1.5-mm collimation) through the regions of abnormality

localized by the conventional images. At CT, scattered mural calcifications and diffuse irregular mural thickening of the trachea and central bronchi were observed (Figs 1 & 2). There was obliteration of the right middle lobe bronchus and complete right middle lobe collapse (Fig. 3). Involvement of the basal segmental bronchi of the right lower lobe resulted in severe proximal narrowing with distal bronchiectasis, mucous plugging and patchy consolidation (Fig. 3). Moderate centrilobular emphysema involved the upper lobe parenchyma. Based on these findings, a diagnosis of tracheobronchopathia osteochondroplastica was rendered. The patient underwent confirmatory fibrooptic bronchoscopy which showed extensive multiple grey-white nodules with scattered calcifications that resulted in areas of stenosis of the distal trachea and in the lobar and segmental bronchi. Nodules ranged in size from 3 mm to 10 mm and were present predominantly along the anterior and lateral tracheobronchial walls, although noncalcified circumferential thickening of the main stem bronchial walls was also noted. A large amount of inspissated secretions was removed from the bronchus intermedius and the right basal segmental bronchi. Bronchial washings yielded normal flora on routine cultures; fungal and tuberculous stains and cultures were negative. No malignant cells were identified. Biopsies were not obtained since the bronchoscopic appearance was considered diagnostic of tracheobronchopathia osteochondroplastica.

DISCUSSION

Tracheobronchopathia osteochondroplastica was first



Fig. 1 – Axial CT scan (5.0-mm collimation) through the trachea shows nodular thickening and punctate calcification along the right lateral tracheal wall (arrow).

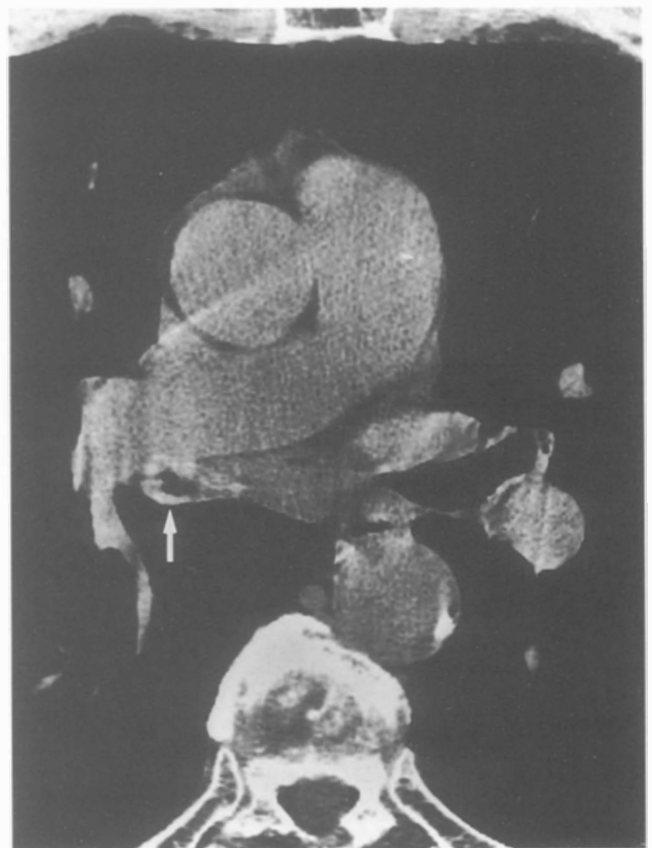


Fig. 2 – CT scan (1.5-mm collimation) at the level of the bronchus intermedius shows circumferential thickening and moderate narrowing of the bronchus intermedius (arrow). Mild uniform thickening of the left main stem bronchus was also present.



Fig. 3 – CT scan (1.5-mm collimation) through the lower thorax shows right middle lobe collapse (arrow). There is significant cylindrical bronchiectasis of the basal segmental bronchi, which are partially filled with secretions.

described by Wilks in 1857, who noted ossific deposits in the laryngotracheobronchial system in post-mortem specimens [1]. The multiple tracheobronchial mural nodules typically involve the lateral and anterior walls, although involvement of the posterior wall may infrequently be seen [2,3]. Punctate calcification occurs within these nodules, but the absence of calcification does not exclude the diagnosis. Involvement of the middle to distal trachea, main bronchi and lobar bronchi is most frequent, although laryngeal and segmental bronchial involvement have also been described [4]. The aetiology is not known but has been postulated to be secondary to either metaplasia of submucosal connective tissue into cartilaginous elements or perichondrial proliferation from pre-existing cartilaginous tissue. Histological examination of the nodules is non-specific, but usually reveals varying combinations of cartilaginous, osseous and fibrous tissue.

Tracheobronchopathia osteochondroplastica is usually identified in patients older than 50 years, and no definitive sex predilection has been established [5]. It is probably underdiagnosed since it has only been diagnosed ante mortem following bronchoscopy, tomography or CT; projectional radiographs and clinical presentation are non-specific. Patients with tracheobronchopathia osteochondroplastica are usually asymptomatic, although insidious development of cough, haemoptysis and dyspnoea have been reported [5], particularly in patients who develop complications from bronchial obstruction. Stridor and hoarseness have been described with laryngeal involvement. Recurrent respiratory infections secondary to bronchial obstruction from the mural nodules may lead to bronchiectasis, as occurred in our patient. Hodges [3] described two cases of tracheobronchopathia osteochondroplastica presenting as right middle lobe collapse, a finding also observed in our patient. There is no evidence of increased risk of malignancy in these patients.

Chest radiographs are usually insensitive to the

laryngotracheobronchial changes seen in tracheobronchopathia osteochondroplastica. In our patient, tracheal irregularity and thickening were suggested on projectional radiographs, but no calcifications were identified. Linear tomography has been useful in identifying tracheal narrowing and nodularity, but is relatively insensitive to the presence of small, punctate calcifications [6]. CT is exquisitely sensitive to calcification within these nodules [7,8] and can accurately delineate both the extent and distribution of tracheobronchial narrowing as well as the complications of post-obstructive collapse, bronchiectasis, and pneumonia. In our case, intravenous contrast enhancement was noncontributory, although the diagnostic benefit of contrast enhancement is not well documented for this condition. Although bronchoscopic biopsy may show the expected osseocartilaginous histology, histological specimens may be nondiagnostic and reveal non-specific inflammatory changes [4].

A number of benign conditions are associated with diffuse nodular thickening of the tracheobronchial tree, including amyloidosis, endobronchial sarcoidosis, relapsing polychondritis, squamous papillomatosis and rhinoscleroma. An association has been reported between end-stage amyloidosis and tracheobronchopathia osteochondroplastica, but the Congo Red staining typical of amyloidosis is absent in histological specimens of tracheobronchopathia osteochondroplastica [5]. In addition, although present in histological specimens of tracheobronchial amyloidosis, calcifications are rarely evident on projectional radiographs. Diffusely infiltrating carcinoma may effect nodular thickening and narrowing of the tracheobronchial tree, but would be an unusual cause of post-obstructive cystic bronchiectasis. Calcification of multiple mural nodules and multifocal sites of narrowing in the tracheobronchial tree are highly suggestive of tracheobronchopathia osteochondroplastica. Some authors have suggested that tracheobronchopathia osteochondroplastica should be the primary diagnosis when CT demonstrates the presence of multiple nodules with punctate calcifications in association with endotracheal or endobronchial stenoses [6,8]. In our case, CT clearly revealed multiple endotracheal nodules with scattered calcifications. Narrowing of the right middle lobe bronchus with right middle lobe collapse was identified by CT and confirmed at bronchoscopy to be secondary to the stenosing endobronchial lesions. Furthermore, in our case, the absence of calcification in the posterior or membranous portion of the trachea and the presence of calcification in the anterolateral portion of the trachea are suggestive of tracheobronchopathia osteochondroplastica. The membranous portion of the trachea contains no cartilaginous tissue, and posterior tracheal involvement with the osseocartilaginous nodules is infrequently observed [6–8]. Thus, in our case, the diagnosis of tracheobronchopathia osteochondroplastica was suggested by the characteristic CT findings.

In summary, CT is an important imaging modality in the non-invasive diagnosis of tracheobronchopathia osteochondroplastica and can precisely demonstrate the extent and distribution of disease as well as the morphological changes that characterize this unusual condition, including nodular thickening of the tracheobronchial wall and associated calcifications. CT may also be important in clarifying the potential complications of tracheobronchopathia osteochondroplastica, including lobar collapse and post-obstructive bronchiectasis.

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Case Report: The CT Features of Orbital Multiple Myeloma

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Orbital involvement by multiple myeloma is rare. We report two cases in whom the diagnosis was considered as a result of the computed tomography (CT) findings. In both instances, CT revealed features characteristic of myeloma, i.e. a large soft tissue mass expanding and destroying bone. In one case, CT also demonstrated compression of the optic nerve by tumour. These CT appearances have not been described previously. Recognition of these characteristic findings may prompt some simple investigations and result in a correct diagnosis without recourse to surgical biopsy.

Orbital involvement is a rare manifestation of multiple myeloma. A review of four series of orbital tumours comprising 1975 patients found only four cases and in the largest review of the world literature on orbital myelomatosis in 1972 the same authors collected only 30 cases, with an age range of 30–89 years [1]. Since then, seven additional cases have been reported [2–7].

We report here two cases of unilateral orbital plasmacytomas associated with multiple myeloma, where the diagnosis had not been considered before investigation by CT. To our knowledge only four similar cases investigated by CT have been previously reported. In these patients the true site of tumour origin was not identified on the CT images [2,6,7].

CASE REPORTS

Case 1. A 46-year-old man presented with a 2-month history of progressive proptosis of the right eye. He was otherwise asymptomatic. CT examination demonstrated a large, enhancing soft tissue mass arising from the frontal bone, associated with marked bony expansion and destruction. The mass extended into the right orbit causing inferior displacement of the globe (Fig. 1). The adjacent muscles were displaced but not involved. Surgical biopsy of the orbital mass revealed histological features characteristic of a plasmacytoma.

Further investigations confirmed a diagnosis of multiple myeloma: an IgG paraproteinaemia of 44 g/l, Bence-Jones proteinuria and a bone marrow biopsy showing a plasma cell infiltration with light chain restriction. No further bony lesions were identified on a radiological skeletal survey.

Following the commencement of systemic chemotherapy, the proptosis gradually subsided.

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Case 2. A 70-year-old woman presented with a 4-week history of left-sided headaches and progressive visual impairment. Examination revealed marked proptosis of the left eye and a palpable swelling over the left temple. Visual acuity was reduced to 6/12 in the left eye and diplopia on left lateral gaze was present. CT examination demonstrated a large enhancing soft tissue tumour mass, arising from the greater wing of the sphenoid bone, associated with marked bony destruction (Fig. 2). Tumour extended into both the posterolateral aspect of the left orbit, compressing the left optic nerve and also into the middle cranial fossa displacing the intact dura.

Further investigations confirmed a diagnosis of multiple myeloma: an IgG paraproteinaemia of 30 g/l, a plasma cell infiltrate within the bone marrow, corresponding to 40% of the nucleated cells and multiple radiolucent areas in the skull and femora.

Owing to the presence of optic nerve compression, detected by CT, the patient was given 5×3 Gy of radiotherapy to the orbital mass prior to commencing oral chemotherapy. Following treatment, the proptosis resolved and the visual acuity returned to normal.

DISCUSSION

Multiple myeloma is characterized by a progressive proliferation of plasma cells within the bone marrow, which secrete paraproteins in the serum and/or the urine [8]. Multiple myeloma, which accounts for 1% of all

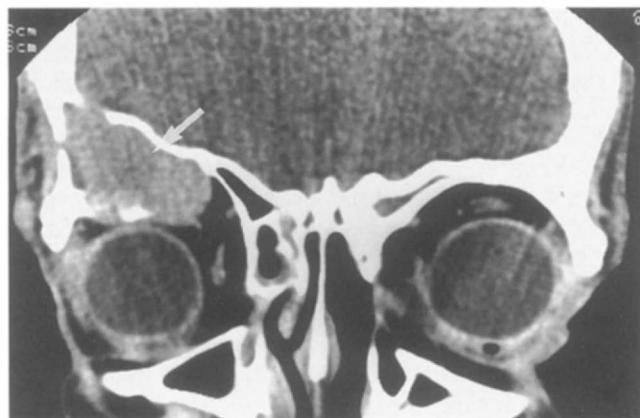


Fig. 1 – Coronal CT following intravenous contrast medium showing an enhancing soft tissue mass (arrow) arising from the frontal bone and extending into the right orbit. Bony expansion and destruction are demonstrated.