Tracheobronchopathia osteochondroplastica presenting at the time of a difficult intubation

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Tracheobronchopathia osteochondroplastica (TO) is a rare and usually benign disorder affecting the trachea and occasionally the bronchi. We describe the case of a 46-year-old woman who was discovered to have TO at the time of a difficult intubation. This case was also unusual since the patient had presented no previous symptoms despite the presence of extensive endotracheal and bronchial lesions.

The incidence of TO appears to be underestimated in the literature in view of the fact that it is usually benign. However, a more accurate estimate of its true prevalence may become available through the use of bronchoscopy and computerized tomographic scanning.

Introduction

Tracheobronchopathia osteochondroplastica (TO) is a rare disorder characterized by the formulation of cartilaginous and/or bone tissue within the submucosa of the trachea and, in rarer cases, the larger bronchi (1). Since the disorder is usually benign, clinical signs are rarely specific; dry cough, dyspnoea, recurrent respiratory infections and occasionally haemoptysis. In exceptional cases, the diagnosis of TO is made during a difficult intubation, as in the present case.

Case Report

A 46-year-old woman, non-smoker, underwent surgery for a benign tumour of the breast on 15 September 1987. She had a past history of primary tuberculosis. During anaesthetic induction, nasotracheal intubation (Portex No. 7) proved difficult. There appeared to be some form of obstruction at the infra-glottic level. This could not have been foreseen as a result of the peri-operative work-up.

Tracheobronchial endoscopy performed after the surgical intervention showed numerous whitish spicules of a hard consistency when grasped with biopsy forceps, extending from the infra-glottic space to the two main bronchi along three-quarters of the circumference of the trachea. The posterior wall of the trachea was not involved and had a normal mobility and movement during either active or passive respiratory manoeuvres (Plate 1).

Histological examination of a biopsy fragment showed a limited area of osteochondroidal tissue situated in the submucosa, surrounded by markedly fibrous connective tissue. It was lined by cartilaginous tissue, non-calcified osteoid lamellae and calcified adult bone tissue. The osteoid lamellae were bordered by osteoblasts. The mucosa had undergone squamous metaplasia; the columnar lining had been replaced by a thin malpighian epithelium, reduced to a few layers of generally dytrophic cells.

Serum phosphorus and calcium levels were normal on laboratory testing. Respiratory function tests revealed a decrease in total lung capacity without signs of obstruction using a flow-volume curve.

A thoracic computerized tomographic (CT) scan showed moderate narrowing of the trachea with irregularity of the walls related to the presence of spicules containing calcium deposits (Plate 2).

Annual clinical and endoscopic follow-up showed no evidence of functional impairment, and confirmed that the endotracheal and bronchial lesions were not progressive (last check-up, February 1994).

Discussion

Tracheobronchopathia osteochondroplastica was first described by Wilks in 1857 and the term
‘tracheo-bronchopathia osteochondroplastica’ was proposed by Muckleston in 1909 and Aschoff in 1910. In 1974, Martin (1) found 245 cases reported in the literature. Tracheobronchopathia osteochondroplastica is usually latent and its most commonly associated respiratory symptoms include: cough; dyspnoea; stridor; recurrent respiratory infections, including one case of an association with an atypical mycobacterial disease (mycobacterium avium-intracellulare complex with no immunodeficiency disease) (2); and haemoptysis. Sometimes the clinical history and the endoscopic appearance can initially remind one of a bronchial carcinoma, as in the two observations by Clee et al. (3). Most cases of TO reported in the literature, however, were diagnosed only at autopsy (4).

The discovery of a case of TO during a difficult tracheal intubation is very rare since only four identical cases have been reported in the literature (6–9). A difficult intubation should be followed by endoscopy in order to confirm the diagnosis. Some authors have recently insisted on using CT-scanning (10–12) to confirm the diagnosis, especially in cases where endoscopy is impossible as a consequence of infraglottic stenosis. Eckhardt et al. (13) have underlined the advantage of using a laryngeal mask during anaesthesia when there is difficulty in intubation for a patient with TO.

The endoscopic findings are diagnostic; whitish, hard spicules projecting into the tracheal lumen from the anterior and lateral walls, with sparing of the posterior wall. In rarer cases, the larynx and main bronchi can be involved (4).

The aetiology of TO remains unknown and several hypotheses have been formulated: metaplasia of the connective tissue; exostosis arising in the cartilaginous ring; the role of chronic irritation by infection; or a link with amyloidosis a degenerative disorder (4). Recently, a familial case has been recorded involving both mother and daughter (14).

The differential diagnosis of multiple nodular lesions of the trachea and bronchi includes papillomatosis, amyloidosis and sarcoidosis. The spirometric parameters of ventilation function are usually normal (15) or show an obstructive pattern (8).

There is no treatment apart from surgery. Neodymium: yttrium-aluminium-garnet laser has been tried but did not show a favourable response (14). The same is true of cryotherapy (16). However, it should be remembered that the condition is usually benign and this partly explains the low incidence of reports.

With the development of bronchoscopy and CT-scanning (17), there are likely to be more frequent reports of this condition which will in turn give a more accurate idea of its true prevalence.

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References