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Tracheobronchopathia osteoplastica: incidental finding at tracheal intubation

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Subglottic papillomatous growths were observed on routine tracheal intubation for a scheduled colorectal procedure. Fibreoptic bronchoscopy revealed that the lesions extended down to the carina and into the main bronchi. The diagnosis of tracheobronchopathia osteoplastica was made after subsequent bronchoscopy and biopsy in the post-operative period. A brief review of this rare benign condition is given.

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An 83-yr-old man presented for elective surgery for carcinoma of the sigmoid colon. Six months earlier he had had an uneventful inguinal hernia repair, which was performed as a day case under general anaesthesia. The patient had been allowed to breathe spontaneously through a laryngeal mask on that occasion. Apart from a history of trigeminal nerve neuralgia and duodenal ulceration, he was fit.

On the day of surgery, anaesthesia was induced with propofol 200 mg and fentanyl 100 μg, and muscle relaxation was achieved with vecuronium 8 mg. Oxygen saturation when breathing air was 96%. After induction of anaesthesia, the patient was haemodynamically stable and oxygen saturation was 98% when he was ventilated with 50% oxygen in nitrous oxide. On laryngoscopy, the larynx was easily visualized, but when a size 9.0 cuffed tracheal tube was passed through the cords subglottic resistance was encountered. The tube was removed and on further laryngoscopy a small nodular growth was seen on the anterior aspect of the trachea in the immediate subglottic region. An opinion was obtained from a consultant ENT surgeon in an adjacent operating theatre, who was uncertain about the diagnosis, but stated that the growth looked benign. As the patient had no respiratory symptoms, it was decided, after discussion with the surgical team, to proceed with surgery.

A size 8.0 cuffed tracheal tube was passed easily, and before surgery was begun fibre-optic bronchoscopy was performed through the tracheal tube. This revealed superficial nodular lesions, which spread down the trachea to the carina and into the main bronchi. A photograph was taken of the lesions (Fig. 1), but a biopsy was not attempted in order to prevent unwanted bleeding into the bronchial tree.

An uneventful sigmoid colectomy was performed and, apart from partial left lower lobe collapse, which responded to physiotherapy, the post-operative period was unremarkable.

After he had recovered from surgery, the patient was referred to the respiratory physicians. Bronchoscopy was performed and a biopsy of the lesions was taken with some difficulty because of their bony nature. A photograph taken of the subglottic region of the trachea (Fig. 2) shows the extent of the lesions.

Histology reported a diagnosis of tracheobronchopathia osteoplastica. The patient was discharged with a follow-up appointment for the chest clinic.

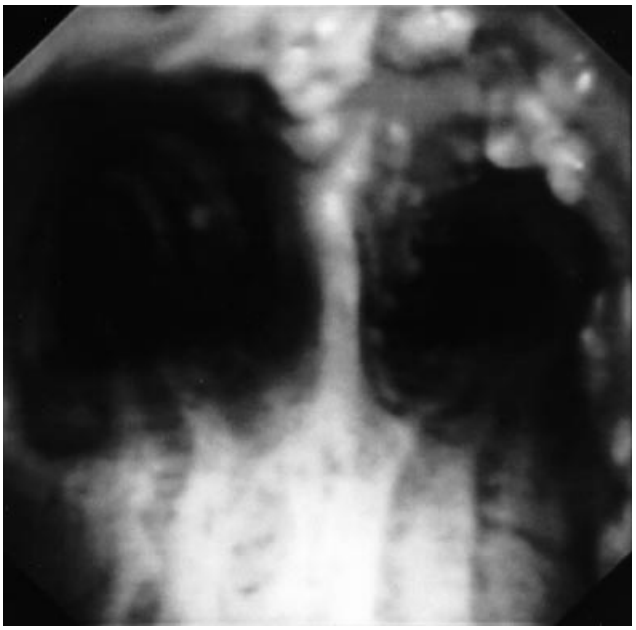

Fig 1 Bronchoscopic view of carina and main bronchi before surgery.

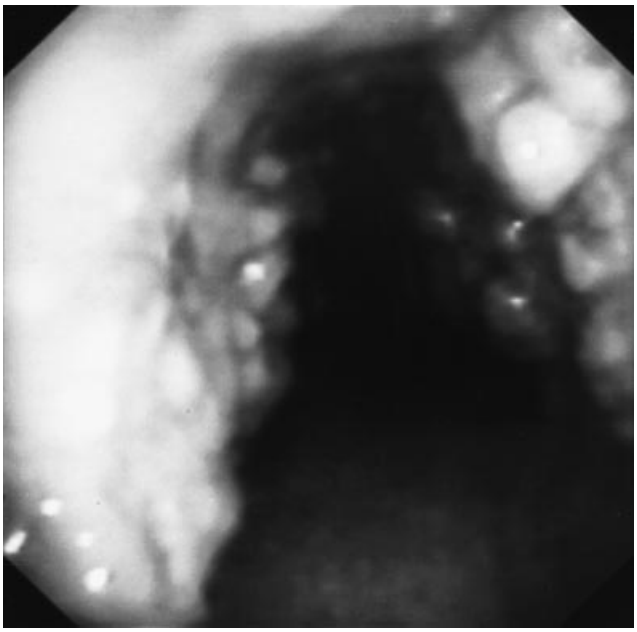

Fig 2 Bronchoscopic view of subglottic region when a biopsy was taken of numerous nodules on the anterior aspect of the trachea. Note the 'cobblestone' or 'rock garden' appearance.[11]

Discussion

Tracheobronchopathia osteochondroplastica is a rare, benign condition and is characterized by the presence of bony and cartilaginous nodules in the tracheal and bronchial mucosa. It was first described by Wilks, physician to Guy's Hospital, in 1857 in a 38-yr-old man who died of tuberculosis. At autopsy, he found the larynx, trachea and bronchi to be 'covered with a number of bony plates'.[1] He also noted that these bony deposits were predominantly anterior to the trachea and lay between the cartilaginous rings.

The cause of the condition is unknown, but several theories have been postulated. Ecchondroses and endochondroses from the tracheal rings were first suggested by Virchow in 1863.[2] In 1910, Aschoff[3] suggested that metaplasia of the elastic tissue may be the cause. More recently, Sakula[4] put forward the theory that tracheobronchopathia might be a form of primary localized amyloidosis of the lower respiratory tract that has undergone ossification. However, histology findings do not support this suggestion. A possible association with atrophic rhinitis and pharyngitis was found by Harma and Suurkari,[5] who suggested that the lesions were due to a build-up of calcium salts within the tracheal mucosa that led to the development of local bony nodules.

The case we describe is consistent with the classical presentation of normal mucosa covering bony or cartilaginous nodules on the tracheal walls, the membranous posterior portion of the trachea usually being spared (Fig. 2). The lesions most commonly occur in the lower two-thirds of the trachea but may extend anywhere from the larynx to the bronchi.[6] Histological findings are typically of bony and cartilaginous nodules (some even demonstrating haemopoiesis) situated in the submucosa, invariably with connections of bone, cartilage or connective tissue to the perichondrium of the tracheal rings.[7]

Most cases are asymptomatic, so it is difficult to estimate the incidence of the disease accurately. Pounder and Pieterse[8] reported the incidental finding at autopsy to be as high as 1 in 400. Primer[9] reported the disease, again as an incidental finding, in four out of 550 bronchoscopies. Males and females are equally affected and age at the time of diagnosis is usually above 50 yr,[10] although some cases have been reported in young adults and children.[5] Those who do present with symptoms typically complain of progressive dyspnoea associated with a chronic cough. This may be associated with intermittent hoarseness, the production of blood-streaked crusts and repeated chest infections.[10]

Diagnosis is confirmed by bronchoscopy and the lesions have been described as having the appearance of cobblestones or a rock garden[11] (Fig. 2). Radiological diagnosis on plain chest X-ray is rare, but computed tomography of the neck has detected even very mild cases.[12] Functional alterations induced by the lesions are variable and depend upon the severity of the disease and the site of the lesion. Spirometry is often normal, but patients with more extensive disease show a mainly obstructive pattern.[13 14]

Only two cases have been described of tracheobronchopathia osteoplastica as a cause of unexpected difficult intubation due to subglottic obstruction.[10 15] In both cases, the disease was extensive and caused significant problems with intubation. One of the cases

was so severe[10] that a size 4.0 microlaryngoscopy tube was eventually passed over a bougie that had been placed through a rigid bronchoscope. In this case, the lesion was removed with a carbon dioxide laser before the patient was extubated the following day.

If this rare condition is discovered at intubation, the extent of the disease and the urgency of surgery affect the decision about whether to continue the operation. It was fortuitous that we had an ENT opinion immediately available, and although the lesion was widespread it was superficial and the obstructive component was minimal. Had the surgery been less urgent, we would not have proceeded but referred the patient to an appropriate specialist.

Most cases of tracheobronchopathia osteoplastica are asymptomatic and the prognosis is generally good. In our literature search, the case described by Birzgalis[10] is the only record of the disease requiring surgical intervention.

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